A study examining the views about reproductive screening programmes of young women affected with congenital conditions for which a screening programme is currently offered, compared with those of professionals in the related fields of medicine and disability, and those of young women in the general population.

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SUMMARY

Reproductive screening programmes may be seen as beneficial, within limits, whether in reducing the incidence of certain serious disorders or in offering information and enhancing choice. However, they have been criticised by disability activists as calling into question the value of disabled people and as resting on 'biomedical' values by which living with an impairment is inevitably problematic. While existing studies have examined the views of a number of professional and lay groups towards screening programmes, little is known of the opinions and perceptions of those affected with the conditions which might be seen as 'preventable' or 'avoidable' by the use of such programmes.

The aim of this study was to examine the views of young women with certain congenital conditions for which a screening programme is currently available, compared with those of other groups who have contact with and knowledge of the conditions but who are not themselves affected. The study also aimed to understand affected women's experiences of living with their disorder.

Individual interviews were carried out with fourteen women with spina bifida and fourteen women with cystic fibrosis. Five women with Down's syndrome took part in short interviews about their lives. Further interviews were carried out with twenty professionals in the fields of medicine, disability and support groups, and with twenty-nine women in the general population. An inductive approach was taken to analysis, the focus being on respondents' explanations and values.

Whereas the concern of professionals was with the general issue of the provision of screening programmes, and women in the general population considered the way that women might take decisions on the basis of their perceptions of consequences and resources, women with spina bifida and cystic fibrosis considered both general and personal questions, and issues of both rights and welfare. These women, broadly supporting the principle of choice, did not suggest that the existence of screening programmes was wrong, but a number felt that others might make decisions in a way that they would not. Their concern was mainly that there was a lack of knowledge about the condition which they had themselves, and that public perceptions of the effects were inadequate.

There were differences within groups, especially in relation to personal philosophies of 'activism' or 'acceptance'. There were also differences between the two groups in the way that women described their experiences of living with their disorder and the main causes of difficulties, and also in their views towards screening programmes and the

issues seen as most salient. The majority of women with spina bifida suggested that they would see abortion on the grounds of any abnormality as unacceptable, and also felt that spina bifida was a condition for which prevention might not be justified. Women with cystic fibrosis considered whether or not it was right to offer CF population carrier screening, and divergent views were expressed about the importance of preventing this disorder and its relative severity.

By describing the perspective and the opinions of women whose views are informed by the experience of living with a congenital disorder, and by indicating similarities with and differences from those of others and within the group, this study can offer an important contribution to the existing empirical work and theoretical literature. It is hoped that dissemination of the findings might encourage fuller information to be offered with reproductive screening programmes, but will also help to continue and to broaden the debate about the complex issues which they raise.

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INTRODUCTION

Since the late 1960s, antenatal screening programmes have made it possible for certain conditions, such as Down's syndrome and spina bifida, to be identified prenatally, and a pregnancy terminated. With increasing knowledge of genetics, DNA tests can detect the presence of genes associated with a widening range of inherited traits or disorders, providing the means to identify individuals affected with certain genetic conditions, such as cystic fibrosis, or those who are carriers. Such tests have made it possible for screening programmes for these conditions to be offered either during or before pregnancy.

These possibilities have been viewed both positively and negatively. The debate about screening programmes is complex and has focused on the morality of action; offering such programmes has been seen as beneficial or as or wrong for different reasons. On the one hand, from an ethics of rights, the principle of screening and 'prevention' may be seen as morally wrong, as eugenics or selection, suggesting that people who are affected with a condition, or who are 'different', are unwanted in society. On the other hand, from an ethics of welfare, attempts to reduce the incidence of conditions may be seen as right, if this can prevent suffering or harm. From this perspective, morality is relative and may be seen in terms of outcomes, or costs and benefits; however, the consequences of 'prevention' may be experienced individually or collectively and may be measured in a number of ways. Screening programmes may also be seen as beneficial in offering information which may be used to enhance reproductive choice. It may be felt right, in Western society, that reproductive decision-making should be a matter for the individual (rather than a matter of law or of medical judgement), since people might differ in their circumstances, perceptions and beliefs. However, even when screening programmes are felt to be potentially beneficial, there are concerns about appropriate limits both to autonomy and to the conditions for which prevention is felt to be acceptable, especially where the means of prevention is abortion. These concerns have raised further questions about who should define such limits and on what basis.

Opposition to screening programmes has been most strongly expressed by some disability activists, who have argued that the existence of screening programmes can be seen as calling into question the value of disabled people, allowing them fewer rights than others to be born. Some have also suggested that the logic of seeing 'prevention' as beneficial or right can be seen as resting on or justified by 'biomedical' values or assumptions, by which living with a condition is defined as inevitably problematic. Disability activists have proposed a 'social model' (as an alternative to a 'medical

model') to suggest that the important difficulties faced by individuals living with an impairment should be seen as socially caused by the way that they are seen and treated within society, rather than as inevitable and stemming from the condition. However, the social model of disability has been criticised for an over-theoretical, political approach to the experience of living with an impairment, and as lacking an analysis of subjective perceptions and of physical limitations. Such a model might be limited in its ability to explain the experiences of people living with chronic illness or learning difficulties. Not all those living with an impairment might share the views of disability activists that it is wrong or unjustified to try to avoid the birth of an affected child.

At the time that this study was proposed, there was national and international concern that the pace of change of technological developments, new knowledge and the potential applications of it, were outstripping decision-making about regulation. At the same time, because of the contradictory ethical approaches towards intervention in reproduction and cultural differences, there have been difficulties in deciding whether policy changes were needed or what these should be.

There has been interest in finding out the views and responses of the general public. Epidemiological surveys have examined the uptake of screening programmes. Empirical work carried out by social scientists has investigated the acceptability of screening programmes, and has led to an understanding of particular factors underlying decision-making. However, most research examining attitudes has been carried out from a psychological perspective, using quantitative methods, an approach which has been felt to be inadequate for leading to an understanding of people's views about this complex topic. Sociologists have argued for approaches examining meaning and social context; the few qualitative studies in this area have indicated differences in values between 'professional' and 'lay' respondents, ambivalence around decision-making and the difficulties of defining limits.

Although existing studies have investigated the attitudes of pregnant women, other members of the public, medical professionals, and the relatives of those affected, the views of individuals living with conditions for which a screening programme is offered have rarely been sought. It has been argued that the views and the experiences of this group should be heard, and that they are crucial to the debate. Those living with a condition might have a different perspective from those of others, professional and lay, both about the morality of offering screening programmes, and about whether they would see it as right, beneficial or justified to try to avert the births of others affected with the same condition or impairment as themselves.

The aim of this study was to examine the views about reproductive screening programmes of a sample of young women living with congenital conditions for which a screening programme is currently available. Their views would be compared with those of professionals in the related fields of medicine and disability and with those of young women in the general population. The study population would be young non-pregnant women, as potential mothers and users of antenatal care. The study would also aim to understand affected women's experiences of living with their disorder, in order to consider the impact of their knowledge and perceptions on their views about screening programmes. Similarities and differences in views and experiences of women with conditions which differed in their implications would be investigated. It was decided to look at the views of young women affected with Down's syndrome, with spina bifida or with cystic fibrosis.

It was acknowledged that there might be difficulties with including women with Down's syndrome in the study. There were also a number of more philosophical considerations affecting the design of a comparative qualitative study, such as whether or not it was appropriate to regard those affected with the same condition as a 'group', and the extent to which it was possible to carry out a comparative analysis. An explanation is offered here of the way that findings have been reported.

Early analysis of interviews was carried out using the NUD.IST computer package. This horizontal analysis showed the issues discussed within each group and across groups. A 'vertical' analysis indicated the coherence of individual accounts in terms of philosophies and the way that conflicting values, contradictions or dilemmas were resolved. There were patterns in the data, suggesting some similarities within 'groups' of women, as defined by their being affected or not with each disorder. It was felt that the views of women with spina bifida and those of women with cystic fibrosis towards screening programmes in general and for the condition which they had themselves could not be separated from the way that these women discussed their perceptions of living with the disorder and their quality of life, and that their voices should not be fragmented. At the same time, analysis also indicated the difficulties of constructing chapters around discrete 'themes' or discourses. While rhetoric and explanations could be identified, arguments were often spoken of as being balanced against others, and women's accounts indicated ambivalence and contradictions between different value-positions. Also, while women drew on discourse, they also drew on factual knowledge and personal narrative in explaining their views. For these reasons, and because of the different ways of drawing the sample, a decision was taken to report the findings by 'group', rather than by themes.

This decision has implications for the structure of the thesis. Because interviews with all respondents focused on the same issues, it was not feasible to include background material in the data chapters to any great extent. The literature and the debates are therefore outlined in a long introductory section, Chapters 1-4.

Chapter 1 first describes the development of screening programmes and the implications of new techniques and genetic knowledge. The second part of the chapter focuses on the disorders, outlining some implications for those living with the condition, the means of prevention, and controversies surrounding the screening programme.

The purpose of Chapter 2 is to review the sociological literature in order to show the contribution of sociological perspectives to an understanding of the screening debate. This chapter outlines the arguments of the disability rights movement, the 'social model' of disability and the arguments of disability activists towards screening programmes. The limitations of the social model for examining experience and the contributions of the sociology of the body are also discussed. The chapter concludes by outlining different sociological approaches towards living with a condition.

Chapter 3 summarises the screening debate in terms of conflicting values or rhetoric. It provides a framework for examining, in the data chapters, the extent to which respondents draw on these arguments or explanations. Chapter 4 reviews the existing empirical work. While research has suggested the general acceptability of reproductive screening programmes and some factors felt to be important in influencing decision-making, the lack of studies examining the views of those affected with conditions themselves led to the design of this study.

Chapter 5 outlines the aims of the study and the research questions, and describes the research methods. While it was felt important to find out the views of disabled or affected young women, it was recognised that there were a number of practical and ethical considerations affecting the way that the study was carried out. Particular issues of concern were the possible sensitivity of the topic, who might take part and whether or not women with Down's syndrome should be and could be included in the study. Decisions had to be taken at every stage of the project. The chapter describes the attempts to balance concerns for rights and welfare, especially in making contact with potential participants, and some difficulties in access and sampling.

Chapter 6 outlines the views of professionals in the fields of disability, medicine and support groups towards screening programmes. These respondents were also asked about the way that they saw the needs of young people with the conditions in this study

or disabled young people. This chapter deals with the issue of normality and difference, inherent in the debate. The following chapter, which considers the views and experiences of women with Down's syndrome, also draws on the views of professionals. The first part of this chapter outlines the implications from the theoretical approaches of 'normalisation' and disability rights for the treatment of people with Down's syndrome or learning difficulties. This is followed by an account of the views of professionals consulted for this study and in the literature about the lives of young people with Down's syndrome. The chapter concludes by describing the short interviews which were carried out, and a consideration of whether the attempt to include these women was felt to have been right and worthwhile.

Chapter 8, 'Women with spina bifida', and Chapter 9, 'Women with cystic fibrosis' are similar in structure. Both begin by describing the ways that women with these conditions spoke about their lives, their perceptions of problems and their causes, and the extent to which they felt these might be addressed. Women were asked about the way that they saw their quality of life and the severity of the condition more generally. The focus of these chapters is on the views of women with these conditions towards screening programmes, especially in relation to the condition which they have themselves. The values and philosophies drawn on, women's particular concerns, their knowledge and perceptions, and differences and similarities within each group are discussed. Different issues were suggested as important by women living with spina bifida and women living with cystic fibrosis, and these chapters consider the views expressed against the context of these women's lives.

The sample of women as representative of the general population was drawn from students on child care and nursing courses. The aim was for this group to have comparable levels of education to those of affected young women, and to have some knowledge of antenatal screening programmes and of the disorders being tested for. Chapter 10 describes the way that these women saw screening programmes, and the considerations and values which they suggested as important.

The final discussion chapter draws together the findings from the data chapters to address the aims of the study and the research questions. First, it reports on the varying perspectives of all participants in the study towards screening programmes, relating the views expressed to the arguments identified in the theoretical debate. All respondents drew on the rhetoric of individual choice, and the implications of this are discussed. Next, the chapter describes the views of young women with spina bifida or with cystic fibrosis (although not women with Down's syndrome who could not be asked these questions) compared with those of professionals and other women in the study. While there were similarities in perspective, the differences within 'affected young women' in

views about screening programmes and in the issues suggested as important are discussed, particular differences between women living with spina bifida and women living with cystic fibrosis. The chapter considers the impact that being affected with a condition which might be seen as 'preventable', and perceptions of living with a specific disorder or type of disorder might have on views towards screening programmes. The chapter concludes by noting the limitations of the study, but also suggesting its contribution to the existing literature, and implications for policy and further research.

CHAPTER 1

SCREENING PROGRAMMES FOR DOWN'S SYNDROME, SPINA BIFIDA AND CYSTIC FIBROSIS

Many conditions or disorders are thought to have a biological or molecular basis or cause. It is estimated that 3% of new-born babies have a significant congenital or genetic disorder, and more will be found to have such a condition during childhood and early adulthood (Plouffe and Donahue, 1994). Some congenital conditions can have serious consequences, causing death at a young age or physical or mental impairment (Connor, 1989). With the increasing control of environmental causes of some conditions, there is a growing interest, in the medical profession, in those which have a genetic base, and in the prevention of such disorders (Weatherall, 1985; Connor, 1989).

There are a number of ways in which a condition might be 'prevented'. This can be achieved by primary prevention, i.e. prevention of the occurrence of the disorder; by secondary prevention, i.e. early detection or identification and intervention, which can mean treatment to prevent the condition worsening; or by tertiary prevention, i.e. long-term measures to prevent the debilitating effect (Freeman, 1992). While a number of conditions with a molecular basis can be treated, individuals affected cannot be 'cured'; the condition will always be present.

However, the incidence of such conditions may be reduced by averting the births of those affected. Before the 1960s, the only means of prevention of a condition which appeared to be inherited was by the avoidance of conception, and some genetic counselling was provided for those with a family history of inherited conditions (Royal College of Physicians, 1989). From the late 1960s, the development of screening and diagnostic testing, together with developments in reproductive technology, especially the availability of abortion, has allowed selected conditions to be 'prevented' by intervention in reproduction. New genetic knowledge has widened the possibilities for reproductive decision-making.

This study is concerned with screening programmes for three congenital conditions; Down's syndrome, spina bifida and cystic fibrosis. The first section of this chapter outlines the historical development of screening programmes, and the implications of new techniques and knowledge. The second section focuses on the three disorders, noting some implications for those living with the conditions, the means of prevention, and debates about the appropriateness and benefit of offering a screening programme.

The historical development of screening programmes

Prenatal diagnosis

In the 1960s, prenatal diagnosis of certain conditions became possible with the development of techniques for both sampling and analysing amniotic fluid and foetal blood cells in pregnancy. Early diagnostic tests permitted the detection of haemoglobinopathies, sex-linked diseases and chromosome disorders (Weatherall, 1985).

In the UK, the passing of the UK Abortion Act 1967 offered the means of preventing some genetic disorders and congenital abnormalities by prenatal diagnosis and selective termination of pregnancy. Under the Act, abortion is legally permitted on certain social or medical grounds; one such ground is where "there is a substantial risk that if the child were born it would suffer from such physical or mental abnormalities as to be seriously handicapped" (The Abortion Act, 1967).

Policy relating to abortion on the grounds of foetal abnormality varies between countries and over time, and policy relating to testing is also variable. Diagnostic testing of amniotic fluid is accurate, but the procedure is expensive, and the method of sampling, amniocentesis, carries a small but significant risk of miscarriage, which may be greater than the risk of an affected pregnancy (Nicholson and Alberman, 1992). Diagnostic testing is therefore normally only offered where a pregnancy is perceived to be at high risk.

From 1969, a diagnostic chromosomal test for Down's syndrome could be offered to women where there was a perceived higher risk of an affected pregnancy, whether on the basis of the mother's age or a family history of chromosome disorders. In the 1970s, with the discovery that abnormally high levels of alpha-fetoprotein (AFP) in the amniotic fluid were associated with open neural tube defects (Brock and Sutcliffe, 1972), the availability of prenatal diagnosis was extended to women at a predicted higher risk of a pregnancy affected with these conditions.

Offering diagnostic testing on the basis of family history (and maternal age in the case of Down's syndrome) was found to identify only a small proportion of affected pregnancies, however (UK Collaborative Study, 1977; Chamberlain, 1978; Chard and Macintosh, 1992). During the early 1980s, the discovery that anomalous levels of protein and other markers in the maternal circulation might indicate the presence of a foetus affected with Down's syndrome or open neural tube defects provided the

possibility of developing a screening test based on a blood sample, which could be offered to all pregnant women.

The purpose of a screening test is to identify, among the healthy population, those who are sufficiently at risk of a particular disorder to justify a subsequent test or investigative procedure which might be felt to be in their interests (Wald, 1994b). Policy for offering antenatal screening tests in the UK varies from region to region. In the West of Scotland, a blood screening test for both Down's syndrome and open neural tube defects is offered to all women at antenatal clinics, optimally between 16-18 weeks of their pregnancy (personal communication, West of Scotland Regional Genetics Service).

Because diagnostic tests and screening tests have different purposes, they have different criteria. Diagnostic tests must be able to discriminate between those who have a disorder and those who do not (Reid, 1991). The ability to distinguish between affected and unaffected pregnancies depends on the sensitivity of the test (the proportion of affected individuals it can successfully identify) and on its specificity (the proportion of unaffected individuals it can identify). Diagnostic tests must have a high specificity, so that the number of false positive results is close to zero (Chard and Macintosh, 1992). Because of the need for accuracy, they might need to be more sophisticated and potentially more harmful than would be acceptable for a screening procedure (Mohide and Enkin, 1992).

Screening tests, since they are carried out on the healthy population, should be safe; they should cause no harm. They might be less accurate than diagnostic tests, in that the specificity may be lower; positive results might include some false positives or unaffected individuals or pregnancies. However, there should be few false negatives; screening tests should have a high sensitivity. They should also be simple and cheap to administer.

Implications of prenatal diagnosis

Antenatal screening and diagnostic testing offers information, to both medical professionals and to individuals. This information may be used to prevent the birth of an affected child. Offering prenatal diagnosis might be seen as beneficial, whether to society in general or to individuals, although there are also concerns about costs.

Prenatal diagnosis might offer a number of benefits to individuals both in avoiding having a disabled child and in taking part in the screening programme, such as avoiding the regret from not having done so (Tymstra, 1991). Information in itself may be helpful in reducing uncertainty (Mooney and Lange, 1993); a negative result may offer

reassurance (Farrant, 1985; Tymstra, 1991), while a positive result may be used either to prepare for the birth of an affected child or to consider 'prevention'. However, as with any medical intervention, the benefits of a screening programme to those taking part should outweigh possible harm. The possible advantage of knowledge might be outweighed by concerns about physical safety, unwanted decision-making or psychological or emotional well-being (Marteau, 1989; Tymstra, 1991; Wilkie, 1993). Prenatal diagnosis introduces the possibility of termination of a wanted pregnancy, and the physical harm of an invasive test. Routine screening programmes extends the consideration of having a child affected with a serious condition to those whose risk might be lower, who might have less reason to suspect a high risk, and for whom the implications of a disorder might be unfamiliar (Richards,1989).

It has been stressed that, for a decision to undergo screening to be an informed one, pregnant women should be offered accurate and accessible information about the condition, the reliability of tests, risk, the implications of screening and its voluntary nature (Smith, Shaw and Marteau, 1994). Also, however benefit is defined and to whom, the means of prevention should be feasible and acceptable, and the tests themselves accurate and effective (Scottish Forum for Public Health Medicine, 1994; Brock, 1995). General principles for good screening practice have been developed [see the Scottish Forum for Public Health Medicine (1994), which summarises the criteria drawn up in 1968 by Wilson and Jungner]. Official guidelines covering screening programmes offered in pregnancy emphasise the importance of accurate and accessible information, the voluntary nature of screening, and the need for counselling where results are positive (Royal College of Physicians, 1989).

Some concerns have been expressed, however, about the low levels of sensitivity and specificity of the blood screening test (usually the 'triple test') currently offered. With a high rate of false positives, those with unaffected pregnancies will be offered invasive diagnostic testing, and with a high rate of false negatives, cases are missed. This has consequences for both the overall effectiveness of the screening programme and for individuals. Research and development in antenatal screening programmes has focused on improving the sensitivity and specificity of screening tests, but also on developing programmes which can be carried out earlier in pregnancy and developing less invasive diagnostic tests.

Ultrasound scanning may now be used to improve the efficiency of screening. Ultrasound can check the gestational age and the possibility of complications such as twins. It may be used routinely as a second screening test for congenital abnormalities (Ennever and Lave, 1995), for the direct detection of neural tube defects (Royal College of Physicians, 1989; Nuffield Council on Bioethics, 1993), and, in some areas, as an

alternative to biochemical screening in the first trimester of pregnancy to identify nuchal translucency (Byrne, 1994). Current research in screening focuses on combinations of ultrasound, maternal age and other markers.

There is also interest in developing programmes of screening which can be offered in the first trimester of pregnancy, because of the reduced rate of complications with earlier abortion, and because early diagnosis is felt to be more acceptable (Royal College of Physicians, 1989; Brambati, 1994; Byrne, 1994). Chorionic villus sampling (CVS) of the placental tissue - which shares the genetic make-up of the foetus - enables results to be obtained more rapidly than with amniocentesis, although amino acids such as AFP cannot be detected by this method of sampling (Brambati, 1994). Some concerns have been raised, however, about foetal safety (Mastroiacovo and Botto, 1992; Firth *et al.*, 1994), and about the diagnostic accuracy of this technique, especially early in pregnancy (MRC Working Party on the Evaluation of Chorionic Villus Sampling, 1991). The feasibility, efficacy, and safety of carrying out amniocentesis in the first trimester has also been investigated, although there remains controversy over the reliability of the results (Byrne, 1994; Plouffe and Donahue, 1994). The efficiency of different screening methods in the first trimester is also being investigated (see, for example Wald *et al.*, 1992a).

Another direction of current research is in the development of a simple, non-invasive diagnostic test, by examining foetal cells in maternal blood. Foetal cells were first identified in maternal blood in the mid-80s (Brambati, 1994), and fluorescent *in situ* hybridisation techniques (FISH) have been used to detect foetal trisomy 18 and 21 (Nuffield Council on Bioethics, 1993). A review of experimental developments is given in Simpson and Elias (1994), although the extent to which foetal cells are found to enter the maternal circulation is disputed (Plouffe and Donahue, 1994).

If foetal cells, or material which shares the genetic make-up of the foetus, can be obtained, then diagnostic testing for a growing number of conditions can be based on foetal DNA, or genetics, rather than on the by-products of pregnancy.

The 'new genetics'

Since the discovery of the double helical structure of DNA in 1953 suggested the molecular basis of genes and chromosomes, there has been an increasing interest in the science of genetics and in heredity (Jones, 1993). In 1971, the development of recombinant techniques enabled sections of DNA to be isolated and studied directly. In the mid-80s, the 'Human Genome Project' was established in the US at an estimated cost of £2 billion, in order to sequence and map the entire series of DNA, i.e. every

gene of the human genome (Wilkie, 1993). At the time of carrying out this study, genetic research was being carried out in a number of countries, including the UK, with international coordination.

Progress of research has been rapid. Today, many disorders are thought to have a genetic basis, whether the mutations are to one or to several genes, while the genes associated with the most common single gene disorders have been isolated and characterised (Yates, 1996). A single gene disorder follows a simple or Mendelian pattern of inheritance, whether autosomal dominant, autosomal recessive or X-linked.

It has been suggested that, in the long term, the main benefit of an understanding of certain single gene disorders might be the prospect of treatment by, for example, the replacement of defective genes by gene therapy (Brock, 1996). At the moment, however, the main outcome of research has been the development of molecular genetic tests. Diagnostic tests associated with a wide range of common single gene disorders have been developed (Yates, 1996).

DNA analysis detects the presence of genes, or combinations of genes, and genetic mutations (genotype), rather than the presence of a disorder (phenotype). Analysis is rapid, accurate and relatively inexpensive. Because the genetic makeup of an individual is present in almost all cells and does not change throughout life, direct methods of genetic testing can be carried out on any body tissue, such as a small blood sample, at any stage of life. Diagnostic testing can be carried out on fertilised embryos and on foetuses, but also on new-born infants, children and adults. Molecular genetic tests have a number of potential applications.

Diagnostic genetic testing can indicate the presence of a condition which might not be manifested until later in life. Predictive genetic testing can be offered where there is a family history of certain late onset single gene disorders although, because of the complex implications, this is only offered at specialist centres, and always with counselling available.

The same principle underlies neonatal diagnosis. Although there are ethical dilemmas about the relative benefits of presymptomatic diagnosis for conditions of uncertain severity or late onset where there is no effective treatment (Wald and Morris, 1998), screening is less controversial where (as in the case of PKU) early diagnosis and intervention has been shown to improve prognosis (Scottish Forum for Public Health Medicine, 1994). In the UK, neonatal screening for sickle cell disease is available in some inner-city areas, and has been considered and investigated for Duchenne muscular dystrophy and cystic fibrosis. Neonatal screening can be a means of informing parents

about their reproductive risk, although the usefulness of the information depends on the pattern of inheritance (Modell, 1993; Carswell, 1996).

One important application of DNA testing is in the context of reproduction, where there is the potential to prevent the occurrence of a condition. Geneticists have argued that, in the absence of treatment, reducing the birth incidence of certain conditions offers an alternative approach to management (Brock, 1996).

Since DNA analysis can be carried out on fertilised embryos, a recently developed technique of pre-implantation diagnosis allows the embryos of a multiple pregnancy produced by in-vitro fertilisation (IVF) to be tested, and unaffected embryos selected for implantation (Brambati, 1994). This may enable couples who are known carriers of a genetic disorder to have an unaffected child, although the technique is expensive, time-consuming and carries possible risks to the foetus (McKie, 1988; Brambati, 1994). Research at the present time is experimental, and its use has been restricted to couples who carry a particularly distressing disorder (McKie, 1988). More commonly, prenatal diagnostic testing with the option of abortion may be available, often early in pregnancy, to those who know that they are at high risk of having a child affected with certain genetic conditions (Yates, 1996).

Because DNA analysis detects gene mutations, a further application of DNA testing is in the identification of carriers of recessive or X-linked disorders (heterozygotes) as well as those who manifest the condition (homozygotes). People can learn their carrier status at any stage of life, but knowledge of carrier status is of most relevance in relation to reproduction, since carrier testing can alert individuals to their risk of having an affected child. Where a couple know that they are both carriers for a condition, this knowledge can be used either to prepare for the possible birth of an affected child or to prevent the births of affected children, whether by avoiding conception, by the use of artificial fertilisation with donor germ cells, or by intervention in pregnancy (Wilkie, 1993). Where an individual knows his/her carrier status, another way of avoiding the birth of an affected child might be by choice of partner.

The possibility of a partner being a carrier depends on the incidence of the condition in the population. Also, relatives of those affected with an inherited condition will be more likely than others in the population to be carriers. Carrier screening programmes may be available for families or among communities at high risk, although policies vary. Some have been successful in reducing the incidence of a condition; for example a screening programme in Cyprus has almost eliminated the recessive disorder thalassaemia (McKie, 1988). In the UK, carrier screening is available to communities at high risk of sickle-cell anaemia, thalassaemia and Tay-Sachs disease (Connor, 1989;

Watson, Williamson and Chapple, 1991a), and also for relatives of those affected with an increasing number of conditions (Yates, 1996).

Those who are carriers of genetic conditions may often be unaware of their risk, however, since the birth of an affected child may occur where there is no family history of the disorder. Carrier screening can be offered to those in the population of reproductive age, to inform them of their carrier status and their risk of having an affected child (Modell, 1993; Wilkie, 1993). Such population carrier screening can be offered to individuals, to couples before conception and to couples expecting a child attending antenatal clinics.

In the UK, there is no national, routinely available, carrier screening programme for a recessively-inherited genetic condition. However, at the time of designing this study, pilot programmes offering cystic fibrosis carrier screening were being offered at a number of centres in the UK, both at antenatal clinics and before conception, following the discovery in 1989 of the gene associated with this condition, in order to examine the feasibility and acceptability of a screening programme.

Implications of new genetics

Screening programmes for genetic conditions raise some different issues from prenatal diagnosis. With carrier screening before conception, the means of prevention of inherited conditions need not be abortion. Instead, there are implications for the wider families of those affected, rather than just parents, and for the community. New issues are raised by the range of conditions which can potentially be identified. Also, as with prenatal diagnosis, there are concerns about the efficacy of the tests.

The implications of new genetic knowledge for relatives and those found to be carriers may be perceived either positively or negatively (Richards, 1993). The opportunity to avoid the births of affected individuals may be welcomed or felt to be right (Rhodes, 1998). As noted already, the use of screening programmes to reduce the incidence of certain conditions is acceptable in some populations. The development of some carrier screening programmes has been initiated by families and communities at high risk (Kevles, 1985; McKie, 1988; Wertz, 1992b), and in the UK it has been noted that the availability of genetic carrier screening is generally welcomed by the families of those affected (Royal College of Physicians, 1989; Yates, 1996).

On the other hand, the availability of genetic testing may be felt to emphasise relatives' responsibility (d'Amico *et al.*, 1992) and could be seen as precluding a more fatalistic approach towards having children (see Rhodes, 1998). There may also be more practical disadvantages where others also have access to information about carrier

status. Concerns have been expressed about the confidentiality of information, the way that carriers may be seen by others, and the possibility of stigmatisation and discrimination (Billings *et al.*, 1992; Kitcher, 1996), both in relation to insurance, but also the right to parent (Wilfond and Fost, 1990; Billings *et al.*, 1992; McLean, 1994). Some screening programmes have resulted in discrimination of those found to be carriers (Hubbard and Wald, 1993); a sickle-cell screening programme introduced in Greece led to 'unwanted' marriage partners (McKie, 1988), and in the US, carriers have been denied health and life-insurance (Wilfond and Fost, 1990).

It has also been pointed out that the long-term psychological consequences of knowing one's carrier status, in terms of anxiety or perceptions of health, are unknown (Wilfond and Fost, 1990; Marteau, 1992), and that there are few studies of the way that people deal with carrier status, such as whether they understand and retain the information (Stone and Stewart, 1996), and whether they inform others (Qureshi, 1994), as well as whether they feel more anxious or stigmatised.

Another new issue raised by genetic testing relates to the range of conditions or characteristics which can potentially be identified. Some of these, such as eye or hair colour for example, might be felt to be trivial or non-medical, or might be better defined as traits or behaviour (Nuffield Council on Bioethics, 1993). Concerns have been expressed about whether offering information about such characteristics is necessarily beneficial (Richards, 1989), but also whether this is right. Since screening programmes offer the means of prevention, there are concerns about the limits of conditions for which testing might be felt appropriate, especially in pregnancy (Richards, 1989; Clarke, 1991; Tymstra, 1991). Some have also expressed concerns about a lowered threshold of what might be perceived as a 'serious' condition (Marteau *et al.*, 1992a). It has been pointed out in the medical literature that screening programmes are only useful where disorders are preventable and would cause suffering (Wald, 1994a), and there have been calls for decisions to be taken about those conditions that are felt serious enough to form a screening programme, for which prevention would be seen as medically justified (Byrne, 1994; Wald, 1994a).

It is felt that it is essential, in offering a screening programme, for the public to be well-informed about the condition, and concerns about a lack of knowledge of the implications of conditions for which screening programmes might be offered have been heightened with the increased possibilities of genetic testing (Watson *et al.*, 1991a). Some have questioned the ethics or the benefits of introducing carrier screening to those without a family history of the condition, or for conditions which might be unfamiliar, especially where tests are offered routinely (Richards, 1989; Marteau, 1990). Richards (1993) has pointed out that there might be different perceptions of risk, cultural views

about inheritance or proneness to conditions in different communities, as well as different values about the morality of 'prevention'. He has urged caution about extrapolating the general acceptability of screening programmes from one community, or in relation to one condition, to another.

Concerns have also been expressed about the limitations of genetic tests. While DNA testing in itself is accurate, genetic tests may not be effective in predicting either the risk of inheritance or the prognosis of those affected, for a number of reasons. Since tests might not be able to distinguish all of the mutations of any one gene, diagnostic tests can have low sensitivity and produce false negative results (Beaudet, 1990); it has been suggested, therefore, that genetic 'risks' should be expressed in terms of probabilities (Davison, Macintyre and Davey Smith, 1994). In addition, where mutations can be identified, there may be a number of reasons why the genotype, as tested, might not predict the phenotype or the resulting condition (Lewontin, 1991; Alper, 1996), leading to difficulties in interpreting the results. Genetic conditions can be associated with a wide range of severity, and the severity might not be predictable from diagnostic tests. Many disorders are thought to involve the interaction of a number of genes, and evidence now suggests that even 'single-gene' disorders might be more complex than first thought (Alper, 1996). In addition, genetic mutations may have a number of effects, protective as well as being associated with a disorder (McKie, 1988). Genetic factors might be only one of a number of factors in 'causing' a disorder; there might be a very variable interaction with the environment, even in disorders with a strong genetic component (Lewontin, 1991; Hubbard and Wald, 1993; Davison *et al.*, 1994). Some have noted potential difficulties for those conveying information which is both complex and uncertain (Watson *et al.*, 1991a), and for those offered such information (Marteau, 1990). The need for skilled counselling has been emphasised.

It has been argued, therefore, that because of the range of 'genetic' conditions and because of the lack of effectiveness of some tests, the legal definitions of what might constitute 'substantial risk' and 'serious handicap' (as in the Abortion Act) are unclear (Morgan, 1992). Although some have felt that new developments exacerbate old dilemmas rather than raising new ones (Wertz, 1992a), many have argued that the different implications of genetics raise new issues (Kevles, 1985; McLean, 1994; Harper, 1995), or pose a challenge to existing screening principles (Scottish Forum for Public Health Medicine, 1994). There is international concern about the ethical implications of the rapidly changing possibilities introduced by new genetics and technology (Knoppers and Chadwick, 1994), and there is felt to be a need to establish guidelines covering their use (Kevles, 1985; Wertz, 1992a; McLean, 1994).

In the UK, recommendations covering some of the implications of new genetic screening have been outlined in the report of the Nuffield Council on Bioethics (1993). This has addressed a number of ethical issues, in particular questions of confidentiality and consent, and the importance of counselling, although the authors have noted that questions remain about the limits of conditions for which testing might be offered and about implications for minorities. In response to concerns, the government has published a report (House of Commons Science and Technology Committee, 1995), which has considered issues such as the severity of conditions, the regulation of screening programmes and implications for employment and insurance, and which has recommended the establishment of a Human Genetics Commission. This will be established by the end of 1999, and The Human Genetics Advisory Commission, set up in 1996 to offer the government advice on issues concerned with human genetics, subsumed within the new body.

Summary

This section has outlined the development of screening programmes, considering the new implications of each stage. While developments in prenatal diagnosis and genetics have introduced new possibilities, and while some screening programmes are becoming routine, at the same time, caution has been expressed about the appropriate use of new knowledge and technology.

The complex debate about the morality of screening generally will be discussed later in the thesis. However, even where the principle of screening is accepted, questions have been raised about appropriate limits, especially in relation to the conditions for which it is felt right or beneficial for 'prevention' to be offered. The next section focuses on the conditions being examined in this thesis.

The disorders

This study is considering screening programmes which are currently available, for which the condition is congenital, rather than late-onset, and for which those affected live into adulthood. It was decided to focus on Down's syndrome, spina bifida and cystic fibrosis. All are conditions for which the life-expectancy for those affected has increased over the past twenty years, and for which, because of improvements in treatment or management, or changes in attitudes or policy, the quality of life for those affected might also have improved. There have been changing implications, therefore, not only for those living with the condition themselves, but for others and for service provision.

The following sections outline, for each condition: the prevalence and nature of the condition and possible implications for those affected, the means of prevention, and particular debates about the appropriateness or benefit of offering a screening programme.

Down's syndrome

Implications of the condition

Down's syndrome is a congenital condition which affects about 1 in 800 babies born in the UK and about 100,000 each year world-wide (Wishart, 1993). In 1991, it was estimated that there were over 26,000 people with Down's syndrome in the UK (Scottish Down's Syndrome Association, 1992a).

The number of people with Down's syndrome in the population is rising because of the marked increase in life-expectancy for those with this condition over the past 25 years (Editorial, 1990b; Nicholson and Alberman, 1992; Wishart, 1993). Whereas in 1958, only 47% of babies born with Down's syndrome were still alive after a year (Nicholson and Alberman, 1992), now about 90% survive to five years (Editorial, 1990b), and there is evidence to suggest that over 60% will live to the age of fifty and over 40% to sixty (Nicholson and Alberman, 1992). Although mortality is high in the first year of life, mainly due to congenital heart defects, after infancy mortality rates are similar to the norm (Vyas, 1994).

Individuals with Down's syndrome have certain common physical features, some of which, such as poor muscle tone and facial features, are usually recognisable at birth; they also often have a different growth pattern from the average, tending to be shorter in height. About 50% of children with Down's syndrome have a congenital heart defect (Hallidie-Smith, 1985), half of which require surgery (Down's Syndrome Association, 1992). Some people with Down's syndrome may have sight or hearing problems (Scottish Down's Syndrome Association, 1992a), and there may be other health problems, such as an increased risk of thyroid trouble or of developing Alzheimer's disease in adulthood (Down's Syndrome Association, 1992).

There will also be a degree of learning difficulty, Down's syndrome being the most common cause of learning disability in young children (Wishart, 1993), and accounting for about one in three of all cases of severe mental handicap (Nicholson and Alberman, 1992). However, the degree of learning difficulty is very variable (Booth, 1985; Carr, 1994), and studies have suggested that the degree of retardation may often be defined as moderate or mild (Booth, 1985; Stratford, 1991)

New developments in health care, together with changed values which mean that such care is not withheld (Wishart, 1993), have enabled people with Down's syndrome to live longer and healthier lives. An increasing interest in the psychological and intellectual development of children with Down's syndrome has led to an improvement in the abilities of some (Buckley, 1985). There has also been an increasing focus on the needs and rights of disabled people, and philosophies emphasising normalisation and integration. From these initiatives, in association with the closure of large hospitals and the increasing emphasis on community living for people with learning disabilities, many children and adults with Down's syndrome are increasingly taking part in many of the same activities as others.

It has been pointed out, however, that these possibilities are of little benefit unless they lead to an improved quality of life (Booth, 1985; Editorial, 1990b; Carr, 1994), and that more needs to be known about the experiences of those living with Down's syndrome. A number of studies have examined the experiences of young people and adults with Down's syndrome (Lane and Stratford (eds), 1985; Carr. 1994), and this literature is discussed in more detail in Chapter 7.

The means of prevention

Down's syndrome is known to result from a chromosomal abnormality. There are different types of Down's syndrome, the most common, accounting for 95% of cases, being Trisomy 21 DS, in which an individual has three copies of chromosome 21 instead of the normal two.

Although the chromosomal make-up of Down's syndrome is known, the reasons for the occurrence of the abnormality are not (Nicholson and Alberman, 1992; Stone *et al.*, 1992). There is known to be a correlation with maternal age, and a number of studies have shown that the risk of an affected pregnancy rises gradually until the age of thirty-four, and more steeply after that (Cuckle, Wald and Thompson, 1987; Mutton *et al.*, 1991; Nicholson and Alberman, 1992). Overall, statistics from local and national studies suggest a pregnancy prevalence rate of about 1.4 per 1000 live births (Mutton *et al.*, 1991; Stone *et al.*, 1992; Carothers, 1994).

Since the causes of Down's syndrome are not known, there is no primary means of preventing the condition. Prenatal diagnosis and termination of pregnancy provides a means of secondary prevention. An accurate diagnostic test for Down's syndrome has been available in the UK since 1969, by carrying out a chromosomal analysis on cultured amniotic fluid cells obtained by amniocentesis (Weatherall, 1985). For the reasons outlined earlier, the diagnostic test was initially only offered to women who had

a higher risk of an affected pregnancy, whether because of their age or because of a previously affected child or a family history of a chromosomal disorder (Royal College of Physicians, 1989). A number of studies indicated that offering prenatal diagnosis and abortion on the basis of maternal age reduced the number of Down's syndrome babies by about 10% (Robinson, 1991; Stone *et al.*, 1992), the reason for this small reduction being because the majority of affected babies were born to mothers too young to have been offered the test (Vyas, 1994).

In 1984, work by Cuckle and colleagues suggested that low levels of AFP found in maternal serum between 14 and 20 weeks of pregnancy could indicate the presence of Down's syndrome, and that the difference in levels was great enough to form the basis of a screening test; it was estimated that this test could detect 40% of pregnancies with Down's syndrome, with 6.8% of unaffected pregnancies (Cuckle, Wald and Lindenbaum, 1984). It was felt that an effective screening policy might be to offer amniocentesis to women over a certain age and to those with lowered AFP levels (Cuckle *et al.*, 1987), and it was noted that a blood screening test for alpha-fetoprotein was already likely to be offered (Wald *et al.*, 1988).

Different combinations of markers were investigated to improve the test efficiency (Wald *et al.*, 1988), and the multiple marker maternal serum screening test - now called the triple test - was introduced in the late 1980s. Statistics from a number of studies have suggested that, using this method of screening, the rate of detection of affected pregnancies is about twice that obtained by screening by maternal age alone (Sheldon and Simpson, 1991, Evans *et al.*, 1994). The estimated detection rate of screening with four markers is now given as 59%, and 65% if an estimate based on an ultrasound scan is used (Kennard, Alberman and Gill, 1996). MS-AFP screening (often the 'triple test') is now offered in the UK at between 16 and 18 weeks of pregnancy, although not on a routine basis in every region. In the eight health board areas in the west of Scotland, prenatal screening for chromosomal abnormalities has been routinely offered since 1987 (Crossley *et al.*, 1994).

Controversies surrounding the screening programme

There is debate about whether it is right or beneficial to screen for the presence of Down's syndrome. Concerns relate to the appropriateness of 'preventing' the condition, the effectiveness of the screening test and the implications of introducing a routine diagnostic test.

There are different views about the appropriateness of preventing the births of those affected with Down's syndrome. Learning disability, like mental health problems, has

been shown to cause major burdens on both the NHS and on the social security system (Smith *et al.*, 1995), and some geneticists and medical professionals have argued that a screening programme for Down's syndrome can be justified on the grounds of cost-effectiveness (Sheldon and Simpson, 1991; Wald *et al.*, 1992b). There may also be difficulties for those responsible for caring for someone with Down's syndrome (Booth, 1985), and termination is normally offered on the grounds of benefit to the mother (Bailey, 1996). It has been suggested by those offering a screening programme for Down's syndrome that rates of uptake show its acceptability (Wald *et al.*, 1992b, Piggott, Wilkinson and Bennett, 1994; Alberman *et al.*, 1995). Some have noted that not to offer amniocentesis on the grounds of maternal age, or a screening programme for Down's syndrome, might be construed as negligence (Chard and Macintosh, 1992; Evans *et al.*, 1994).

On the other hand, it has been argued that 'prevention' might be felt inappropriate for a condition which many would not regard as a disease, but rather as a characteristic, an aspect of personhood or a way of being human (Clarke, 1994; Williams, 1995a). More practically, it has been suggested that people with Down's syndrome rarely suffer pain or distress as a result of their impairment (Bailey, 1996), that the quality of their lives may not be poor (Stratford, 1991), and that social factors are important in quality of life (Booth, 1985), although, as discussed, it has also been noted that little is known about the lives of those with Down's syndrome (Editorial, 1990b). The degree of learning disability cannot be predicted from diagnostic tests and may be mild (Birth Control Trust, 1996a); Hubbard and Wald (1993) have noted the improvements in potential in recent years for people with Down's syndrome. Some have suggested that this potential may be little emphasised in the material offered with screening programmes (Stratford, 1991), although more positive aspects of living with the disorder have been emphasised by the Down's Syndrome Association (Down's Syndrome Association, 1992). As with any screening programme, some have argued that pressure towards cost-effectiveness might lead to an over-negative portrayal of the consequences of Down's syndrome (Elkins and Brown, 1993; Williams, 1995a) or that medical leaflets and literature focus on the abstract condition (Bailey, 1996). In 1996, an amendment was proposed to the Abortion Act 1967, stating that the grounds for abortion of 'serious handicap' should not apply to Down's syndrome (Birth Control Trust 1996, a; b), but this was defeated.

Some medical professionals have questioned whether it is necessarily beneficial to offer screening for Down's syndrome to all pregnant women, because of the low sensitivity and specificity of the triple test (Keatinge, 1992). There are particular concerns about false negatives, since about 40% of affected pregnancies are missed by this method of screening (Connor, 1989; Evans *et al.*, 1994). Others, however, have felt that it is right

for screening for Down's syndrome to be offered, since the programme is more effective than offering amniocentesis on the basis of age alone (Sheldon and Simpson, 1991; Nicholson and Alberman, 1992; Wald *et al.*, 1995), and since, on the grounds of equity and informed choice, screening should be available for all who want it (Connor, 1993; Wald *et al.*, 1995). One study of the views of obstetricians has suggested that some confusion exists about the way that the triple screening test for Down's syndrome might best be used in practice (Green, 1994b).

Different issues are raised by the possibility of a routinely offered non-invasive accurate diagnostic test for Down's syndrome. Some have suggested that this might put increased pressure on women to find out information about the unborn child and to terminate a pregnancy (Evans *et al.*, 1994).

Uptake of the screening programme

In the West of Scotland, statistics for the uptake of a programme of screening, diagnosis and termination are available from the Institute of Medical Genetics, Regional Genetics Service. In a large study carried out over a year, the uptake of screening was 80%, and of prenatal diagnosis in those who screened positive for Down's syndrome was 70% (Crossley *et al.*, 1994). Other studies have suggested rates of uptake of prenatal diagnosis of up to 85% (Piggott *et al.*, 1994). The uptake of termination among pregnancies diagnosed as Down's syndrome has been found to be high. In the West of Scotland, data shows that the rates of affected pregnancies diagnosed and those terminated are very similar (Crossley *et al.*, 1994), and, in a national study, Alberman and colleagues found a termination rate of 92% (Alberman *et al.*, 1995). These figures suggest that about half of all affected pregnancies are diagnosed, and a similar number terminated (Crossley *et al.*, 1994). Because of the low sensitivity of the screening test, and because not all take part in screening programmes, it is estimated that the overall rate of uptake of all stages of a screening programme for Down's syndrome might not rise above 60% (Nicholson and Alberman, 1992).

Statistics from national registers which record prenatal and postnatal diagnoses of Down's syndrome have been used to estimate the impact of prenatal diagnosis (Mutton *et al.*, 1991; Crossley *et al.*, 1994; Mutton, Ide and Alberman, 1998). Such statistics suggest that, with the introduction of new screening policies, the rates of prenatal diagnoses and the numbers of terminations of Down syndrome pregnancies are rising (Mutton *et al.*, 1991; Alberman *et al.*, 1995; Mutton *et al.*, 1998), and that the birth incidence of Down's syndrome and the numbers of live births are falling (Carothers, 1994; Alberman *et al.*, 1995; Kennard *et al.*, 1996; Mutton *et al.*, 1998).

Spina bifida

Implications of the condition

Spina bifida is the term for a group of congenital neural tube defects which are caused by the failure of some part of the nerve cord to close before birth. The failure of the upper end of the neural tube to close results in anencephaly, a condition which usually results in stillbirth or neonatal death. A failure of the neural tube or its covering vertebrae to close at the lower end results in a range of congenital malformations known as spina bifida. 'Closed' spina bifida, resulting from the failure of the vertebrae to fuse normally, has little effect, and this condition is not notified as a congenital anomaly. The majority of cases of spina bifida are 'open' spina bifida (Laurence, 1974; Connor, 1989). In this condition, part of the spinal cord herniates into a cyst or sac filled with cerebro-spinal fluid, although the cord itself may or may not be damaged. Where the lesion is more serious, and the spinal cord is damaged and protrudes through the skin (myelomeningocele), the disorder may be associated with hydrocephalus, a build-up of cerebro-spinal fluid in the brain, a condition which may also occur in individuals without spina bifida.

Neural tube defects in general are among the most common congenital anomalies (Howard and Headings, 1991; Stone *et al.*, 1992), although their incidence varies over time and place. The West of Scotland has a high rate of neural tube defects, whether compared with the rest of the world (Anderson and Spain, 1977), with other European countries (Omran *et al.*, 1993, Stone and Dolk, 1994) or with many other regions in the UK (Department of Health, 1992). The pregnancy prevalence of neural tube defects in the West of Scotland (between 1980 and 1990) has been estimated at 3.12 per 1000 (Eurocat Working Group, 1993).

In the UK, there has been a sharp decline in the birth prevalence of neural tube defects since the late 70s, with the increased use of prenatal diagnosis and abortion (Eurocat Working Group, 1993). However, there has also been a more general unexplained reduction in the underlying incidence, in the UK and elsewhere (Leck, 1983; Robinson, 1991; Stone *et al.*, 1992).

Statistics for the prevalence of spina bifida in the population are difficult to obtain. From data from the Congenital Anomaly Register, the birth prevalence rate (live and still births) of spina bifida in Scotland for the 1991 birth cohort has been given as between 0.3 and 0.4 per 1000 total births, and the number of babies born (live and still born) with the condition in Scotland in 1991 as 28 (National Health Service in Scotland Information and Statistics Division, 1992). Data from the Glasgow Register of

Congenital Anomalies gives the number of children under age fifteen with spina bifida (born between 1984 and1988) among Greater Glasgow Health Board residents as 14 with hydrocephalus and 17 without (Scott, *date unknown*).

Until the 1960s, few babies born with spina bifida survived, both because hydrocephalus is a fatal condition if untreated, and because of the infection of exposed nervous tissue. Spina bifida can now be treated at birth by an operation to close the spinal lesion, and by the early detection of hydrocephalus and the insertion of a valve or shunt. Since the 1970s, there has been a marked fall in perinatal mortality (Anderson and Spain, 1977), although the condition can still cause stillbirth or death in early childhood, especially during the first year of life, because of increased susceptibility to infection (Tew, 1987).

Spina bifida is associated with varying degrees of impairment, depending on the level of the lesion and the degree of damage to the spinal cord, but it usually results in the paralysis of muscles receiving their nerve supply from the level of the lesion or below, as well as a lack of sensation. The degree of paralysis in the lower limbs, and hence mobility, will depend on the muscles affected; there may often also be urinary and faecal incontinence. There may also be some limb deformity at birth or later in life, including dislocation of hips and scoliosis (curvature of the spine). The operation to insert the shunt has reduced the danger of brain damage, although hydrocephalus may also be associated with some problems of balance and limb control (Anderson and Spain, 1977), and, some studies have suggested, with unevenness of intellectual functioning (Anderson and Spain, 1977). While operations may be needed or useful in childhood, in adulthood the condition is often stable, although there may be problems such as the sudden malfunction of a shunt (Tomlinson and Sugarman, 1995). The needs of people with spina bifida have been described as complex (Tew, 1987; Corbett, 1989).

The prevalence in the population and the needs of those affected may be affected by treatment policy. It was found that operating on all babies born with spina bifida increased the proportion of survivors who had serious physical and mental impairment and greater vulnerability to infection (Laurence, 1974). Following changes during the 1970s towards a more selective policy of active surgical management, spina bifida has become a less common condition, and is associated with less severe limitations, many of those affected being able to attend mainstream schools, although needing assessment (Tew, 1987).

With the survival of people with spina bifida and hydrocephalus into adolescence and adulthood, there has been an increasing interest in the experiences of those living with

the condition. Early empirical work examining social and practical problems facing families suggested that having a child with spina bifida could have a considerable impact on daily life, and that some problems - with taking part in leisure activities for example - were common (Woodburn, 1972; Anderson and Spain, 1977). While one focus in this early work was on the coping strategies developed by families (Anderson and Spain, 1977), authors reporting empirical studies stressed the effect of social factors in the experience of living with spina bifida, commenting that many problems might have been alleviated with improved resources, but that these were often not available.

Early research considering the implications for young people affected with spina bifida found evidence of social isolation, possible problems of depression, and poor provision for employment or training (see Anderson and Spain, 1977). From the late 1970s, there has been increasing sociological interest in the common experience, and common problems, of disability for those living with certain stable impairments such as spina bifida, and a corresponding focus in empirical work on the experience of disability, rather than on the implications of particular conditions. Theory and empirical studies of disability will be discussed in Chapter 2.

The means of prevention

Despite much research, the causes of neural tube defects are not known (Stone *et al.*, 1992), although it is known that the failure of the neural tube to close occurs within the first few weeks of pregnancy. The causes have long been thought to be multiple and complex, involving both genetic and environmental factors (Anderson and Spain, 1977; Leck, 1983; Tew, 1987). A possible relationship with the level of folate in the mother's diet has been suspected since the 1960s (Leck, 1983; Department of Health, 1992). A multi-centre study of mothers who already had a child with a neural tube defect showed that supplementing diet with folic acid around the time of conception had a significant protective effect (MRC Vitamin Study Research Group, 1991). Since 1992, health authorities in Britain and other countries have advised women who may become pregnant to increase their intake of folic acid (Department of Health, 1992), and there have been calls for food to be fortified (Alberman and Noble, 1999). However, folic acid does not prevent all neural tube defects, indicating that other factors are also involved. Evidence of a higher incidence where either parents or existing siblings are affected suggests an inherited risk factor (Leck, 1983; Department of Health, 1992), and more recent research has indicated evidence of a genetic factor for spina bifida (Van der Put *et al.*, 1995).

Since the causes of neural tube defects are not known, there is no primary means of preventing the condition, although genetic counselling can be offered to parents with an

affected child or where there is a family history of the condition (Tew, 1987). Writing in 1972, Woodburn discussed the effect of having a child with spina bifida on family building, as reported by parents in a large study. Often the child with spina bifida was the first or second born, and fewer than half of respondents had limited their families, although most saw their risk of having another affected child as moderate or serious. While some parents had received genetic advice, this appeared to have been sometimes inaccurate or confused, and Woodburn has noted that some respondents felt that diagnostic testing for the condition would be helpful (Woodburn, 1972).

Since the 1970s, antenatal diagnostic and screening tests have offered a means of secondary prevention for open neural tube defects (Connor, 1989), since the discovery that such lesions were consistently associated with the production of raised levels of alpha-fetoprotein (Brock and Sutcliffe, 1972). In the medical literature, it has been suggested that antenatal screening programmes and abortion might offer a preferable means to 'selection criteria' of reducing the incidence of those severely affected with neural tube defects (Laurence, 1974). A diagnostic test based on raised levels of AFP in the amniotic fluid could be offered where there was a high risk of an affected pregnancy (Chamberlain, 1978), and the finding that there were correspondingly raised levels of MS-AFP in maternal serum offered the possibility of developing a screening test (Leek *et al.*, 1973). It was felt feasible to offer a routine screening test between 16 and 20 weeks of pregnancy (Leighton *et al.*, 1975). The concentration of AFP in the maternal serum is much lower than in amniotic fluid, and varies throughout pregnancy (Connor, 1989), but the level is defined as 'raised' if it is twice the median value for the appropriate gestation (Connor, 1990). However, raised levels can be associated with other causes, including twin pregnancy and, in about 50% of cases, no cause can be found (Royal College of Physicians, 1989). In the 5% of pregnancies with an elevated level, the gestation is confirmed by ultrasound, complications such as twins eliminated, and where the level of MS-AFP is still raised in a repeat screening test, a diagnostic test will be offered (Connor, 1989). Some have proposed that a positive ultrasound scan can avoid the need for amniocentesis (Ennever and Lave, 1995).

The sensitivity of the blood screening test for open spina bifida has been estimated at between 70-80% (Connor, 1989). The sensitivity of the diagnostic test in conjunction with ultrasound has been suggested as 100% for anencephaly and over 85% for open spina bifida (Ferguson-Smith, 1987; Connor, 1989). It has been estimated that 70-80% of cases of spina bifida can be detected by this programme of screening and diagnosis (Wald, 1991; Chard and Macintosh, 1992).

In the UK, MS-AFP screening has increasingly been offered since the early 1980s (Royal College of Physicians, 1989; Nuffield Council on Bioethics, 1993). There is no

national programme of screening for neural tube defects. Local policies vary, but in the West of Scotland, such screening is routinely offered.

Controversies surrounding the screening programme

In the UK, there has been little discussion of the appropriateness of screening for or preventing spina bifida and other open neural tube defects. The cost-benefit ratio of preventing a condition becomes less favourable as the population incidence decreases, and geneticists have noted that, where the pregnancy prevalence of open neural tube defects is high, screening is effective in financial terms (Henderson, 1987). It has also been suggested that the high rates of uptake of screening programmes for neural tube defects, where they are offered in the UK, demonstrate the acceptability of the programme (Ferguson-Smith, 1987; Connor, 1989). However, in some countries, a screening programme for spina bifida is not offered, and some recent cross-cultural work suggests possible differences in perceptions about the condition, and differing views about the importance of preventing its occurrence (see Howard and Headings, 1991; Eurocat Working Group, 1993).

Some concerns have been raised about the uncertain results of a routinely-offered screening test. For neural tube defects, the high rate of false positive levels is a particular concern; about 5% of all women screened will have a raised MS-AFP level, of whom only about 5% will have an affected pregnancy (Howard and Headings, 1991). Some empirical work has demonstrated women's anxiety after raised levels of MS-AFP and with the offer of amniocentesis (Earle, 1981; Fearn *et al.*, 1982, Burton, Dillard and Clark, 1985).

Uptake of the screening programme

Early empirical studies suggested that few women declined screening for neural tube defects (Bennet, Gau and Gau, 1980). Statistics for the uptake of MS-AFP screening have been given as 75-80% (Ferguson-Smith, 1987; Connor, 1989), and the more recent uptake of screening has been noted already in relation to Down's syndrome.

Since the mid 1970s, it has been possible to monitor the effectiveness of the maternal serum screening programme in the West of Scotland. Omran and colleagues have reported that data on births and terminations from the Glasgow Register of Congenital Anomalies and from the Regional Genetics Centre, Duncan Guthrie Department of Medical Genetics, for the period 1976-1986 have shown a rising rate of detection of spina bifida from 33% in 1976 to 73% in 1986 (Omran *et al.*, 1993). The improving sensitivity is thought to be partly due to improved ultrasound. Data from these centres indicates a fall in the annual number of cases of spina bifida over the period. The

statistics also show that, as the rate of detection rises, the rate and number of induced abortions also rises; over the final 3 years of the study, the rate of termination of the total cases of spina bifida was 53%. The average rate of uptake of screening between 1976 and 1986 was 67%, and the rate of termination of affected pregnancies 82.6% (Omran *et al.*, 1993).

Cystic fibrosis

Implications of the condition

Cystic fibrosis (CF) is the most common, inherited, life-threatening condition among the white population in the UK. The birth incidence is approximately 1 in 2500 births (British Paediatric Association Working Party on Cystic Fibrosis, 1988; UK Cystic Fibrosis Survey Report, 1995), about 450 babies being born annually (Goodchild and Dodge, 1985). There are currently about 6300 people with CF in the UK (UK Cystic Fibrosis Survey Report, 1995).

The disorder causes thick viscous mucus to be produced, which blocks the ducts of certain organs, particularly the lungs and the pancreas. The main features of cystic fibrosis are a progressive lung disease together with pancreatic enzyme deficiency. People with cystic fibrosis are prone to chronic chest infections, which may cause a productive cough and shortness of breath; also, because of the inability to break down food, there is malabsorption of fats, which may lead to malnutrition. Some people with the disorder may have more respiratory problems, whereas others may have more digestive problems; there is also considerable variation in the severity of the disorder between individuals. An individual may have periods of stability and periods of ill-health, although the disorder is always present. Early death is normally due to chronic respiratory disorder.

When cystic fibrosis was first recognised, in the 1930s, it was as a condition which was normally fatal in infancy or in childhood (Burton, 1975), whether from respiratory failure or neonatally due to meconium ileus. However, increasing knowledge about the condition has led to improvements in diagnosis and management, and to a dramatic improvement in prognosis.

Cystic fibrosis is thought to result from defects to a single gene, named the cystic fibrosis transmembrane conductance regulator (CFTR) gene. The CFTR gene was identified on the 7th chromosome in 1989. Research suggests that the gene is responsible for the production of protein governing the transfer of salt and water across cell membranes. The improved understanding of the mechanism causing cystic fibrosis has led to improved management of the condition, both by conventional treatment and

by new treatments based on genetic techniques. The identification of the gene has also offered the hope of more radical treatment of cystic fibrosis by somatic gene therapy in the future (Editorial, 1990a; Aldhous, 1995).

The prognosis and survival of those with cystic fibrosis has been steadily improving (British Paediatric Association Working Party on Cystic Fibrosis, 1988); some people are now living into their thirties and forties, and the median life-expectancy of a child born now with the disorder is likely to be 40 years (Elborn, Shale and Britton, 1991). A number of specialist treatment centres for the condition have been established, and there is some evidence that treatment at a recognised centre is associated with increased life-expectancy (British Paediatric Association Working Party on Cystic Fibrosis, 1988). Improved techniques for management enable most people with the condition to manage the disorder at home rather than in hospital.

Studies in social science have investigated the implications for families, and, more recently, for those affected with cystic fibrosis. Early empirical work showed that families faced both practical and emotional difficulties, but that they transcended these by a variety of means or strategies (Burton, 1975; Venters, 1981). A more recent study, in the US, of families and siblings, has similarly focused on the coping strategies employed at different stages of the disorder to preserve a normal way of life (Bluebond-Langner, 1996). One study in the UK examining the implications for affected individuals found that the employment rate of mothers of adults with CF was less than expected in women of their age-group (Walters, Britton and Hodson, 1993).

With improvements in prognosis and life-expectancy, more recent work has focused on consequences for young people and adults living with cystic fibrosis (see The Gallup Organisation, 1996). A number of studies have indicated their normal self-concept or psychological health (Cowen *et al.*, 1984; Shepherd *et al.*, 1990, Blair, Cull and Freeman, 1994). Demographic studies have suggested the increasing autonomy of some adolescents and adults with CF. While the main cause of unemployment is ill-health, authors have also suggested possible variations with educational or class background (Penketh, 1987; Shepherd *et al.*, 1990; Walters *et al.*, 1993; Blair *et al.*, 1994).

The means of prevention

Cystic fibrosis is a recessively inherited condition, thought to result from a single gene. Those inheriting a defective gene from both parents exhibit the disorder, while those with one defective gene are carriers of the condition and have no symptoms. The

probability of any child of a couple who are both carriers being affected with cystic fibrosis is 1 in 4, and of being a carrier is 1 in 2.

The inherited nature of the condition has been understood for a number of years, although the only means of prevention for those at high risk was the avoidance of conception. Early studies of affected families found a low level of understanding about the condition (McCrae *et al.*, 1973), fear of another pregnancy and guilt about conceiving another affected child (Burton, 1975). The authors of one study recommended the provision of improved information about inheritance to families at risk (McCrae *et al.*, 1973), and since that time genetic counselling has been available for those with an affected child or a family history.

Following the identification of the gene for cystic fibrosis, the development of a diagnostic test has offered another means of prevention (Editorial, 1990a; Brock, 1996). In the UK, prenatal diagnostic testing can be offered where there is a known risk of cystic fibrosis (Royal College of Physicians, 1989). DNA testing can also identify those who are carriers for the condition. Carrier testing can be carried out quickly and inexpensively from a mouthwash or small blood sample. In the West of Scotland, such testing (and counselling) is available through the Regional Genetics Service for relatives of affected individuals, but also for others in the population who wish to find out their carrier status (personal communication, 1999).

Population carrier screening can be offered for cystic fibrosis. From the birth prevalence, it is estimated that one person in 25 in the white population of the UK is a carrier for cystic fibrosis, although the disorder is not confined to this group, and that one pregnancy in 600 is at risk of producing an affected child. At present, routine screening is not offered, but there has been consideration of whether this should be introduced.

Controversies surrounding the screening programme

The debate about whether or not it is right and beneficial to offer screening programmes for cystic fibrosis encompasses the different implications of testing for a genetic condition, known to be inherited, as well as concerns about the appropriateness of preventing the condition and the limitations of tests.

As with genetic tests more generally, some concerns have been expressed about the effectiveness of the carrier test, and about how the DNA test should be interpreted. One set of concerns relates to the accuracy of the test in terms of risk (Alper, 1996). Although the test has a high specificity, with a false positive rate of 0.1% (Wald, 1991), it has a low sensitivity, with a high proportion of false negatives. It is based only on the

three or four most common mutations of the defective gene, which, in the UK, are responsible for about 85% of the heterozygotes (Beaudet, 1990; Wald, 1991; Alton, 1996). A test which identifies 85% of individuals will only identify 72% of carrier couples (Beaudet, 1990), and, where reproductive decisions have to be made, it is the screen-positive or screen-negative status of couples which is at issue. A test based on couples will be ineffective in cases of non-paternity (Macintyre and Sooman, 1991). Concern has been expressed about the false reassurance of a screening test which misses a quarter of carrier couples (Beaudet, 1990; Wilfond and Fost, 1990; Editorial, 1992; Modell, 1993). Some have argued that screening should not be introduced until the accuracy of the test has been improved (Beaudet, 1990; Wilfond and Fost, 1990). Others have felt that it would be wrong to withhold information which could enhance reproductive choice for this reason, if the complexity of the information and uncertainty about risk can be carefully conveyed (Editorial, 1990a).

There may be difficulties in interpreting the results of a diagnostic test in terms of prognosis as well as risk (Alper, 1996). While any molecular test identifies genotype rather than phenotype, the prognosis of cystic fibrosis is very variable, the disorder being associated with a diversity of clinical symptoms and a wide range of severity. Some of the many mutations of the gene are thought to be benign (Alper, 1996), or to be associated with less severe disease (Alton, 1996), and research has now suggested that the genetic influence may be complex (Knight, 1992; Alper, 1996; Alton, 1996).

The issue of relative severity relates to a general concern about the appropriateness of 'preventing' cystic fibrosis. As with other conditions, the longer life-expectancy might increase the costs of care, and the medical support needed can be extensive (British Paediatric Association Working Party on Cystic Fibrosis, 1988; Elborn *et al.*, 1991). Some have argued that it is right to try to reduce the incidence of this disorder because it continues to be associated with 'substantial morbidity' (Brock, 1996), and it has been suggested that reducing the incidence of the condition might offer more resources for those who are living (Editorial, 1990a). It has also been pointed out that the provision of screening appears to be wanted, or is approved of, by the relatives of those affected (Koch and Stemerding, 1994).

On the other hand, because of the improved prognosis for those with cystic fibrosis and the hope of further possibilities for treatment, some commentators, including some in the medical profession (Scully, 1985; Editorial, 1992; Alper, 1996), have questioned the appropriateness of averting the birth of an individual affected with CF (Kaback *et al.*, 1984; Evers-Kiebooms *et al.*, 1988), especially by abortion. One study of obstetricians found a range of views about the acceptability of offering an antenatal screening programme for cystic fibrosis, with one in seven not supporting termination

at any stage of pregnancy (Green, 1995). If 'prevention' is felt to be right, therefore, another question is whether screening before conception or during pregnancy offers a more feasible or acceptable approach.

General concerns about introducing population carrier screening for genetic conditions have been discussed already. Some have suggested that the implications of cystic fibrosis may be little understood in the general population (Richards, 1993), and others have raised concerns about possible implications for those identified as carriers, including dealing with uncertain information (Marteau, 1990). Many have made the point that, if carrier screening is introduced, there are a number of reasons for offering adequate information and skilled counselling (Wilfond and Fost, 1990; Watson *et al.*, 1991a; Clarke, 1995), and for learning lessons from communities in which carrier screening has been welcomed and accepted (Editorial, 1990a).

A number of studies investigating the reproductive decisions taken by families with an affected child have indicated that the birth of child affected with cystic fibrosis has a major impact on reproductive plans, and that many parents wish to avoid further pregnancy (Burton, 1975; McCrae *et al.*, 1973; Kabak *et al.*, 1984; Evers-Kiebooms *et al.*, 1988). One study found that the child with CF tended to be the last in the family and that 67% of families underwent sterilisation (Kabak *et al.*, 1984). With the anticipated development of a carrier screening programme, more recent research has investigated parents' predicted use of both a screening programme and abortion. These studies have indicated a range of views, some research suggesting that a proportion of parents might be prepared to take the chance of an affected child (Al-Jader *et al.*, 1990, Wertz *et al.*, 1991, Wertz *et al.*, 1992). Commentators have suggested that the varied attitudes might reflect a number of factors, both personal and cultural (Wertz, 1992b; Green, 1995).

Studies examining views about the acceptability of CF screening have shown that almost all parents of an affected child support the development of diagnostic testing for CF, and feel that this should be made available (Kabak *et al.*, 1984; Evers-Kiebooms *et al.*, 1988; Wertz *et al.*, 1991; Watson *et al.*, 1991a; Conway, Allenby and Pond, 1994). The views and predicted personal use of others in the population have also been studied; the findings are outlined in Chapter 4. In order to determine the acceptability and feasibility of population carrier screening in practice, however, and to assess the cost implications, screening programmes were offered on a trial basis in the UK between 1990 and 1995.

Pilot studies of cystic fibrosis population carrier screening

In the pilot programmes, carrier screening was available for individuals, for couples before conception and for couples during pregnancy, and was offered to those of reproductive age at a number of centres (Wilkie, 1993). Information about cystic fibrosis was provided, couples found to be screen-positive were offered counselling, and, where this was during pregnancy, they were offered prenatal diagnostic testing.

One of the main findings was the wide variation in uptake of screening, depending on the setting and the method of invitation (Watson *et al.*, 1991b; Modell, 1993; Bekker *et al.*, 1993; Brock, 1995). While the uptake of screening before conception in a number of settings was low [see Clayton *et al.* (1996), reporting findings in America], by contrast a consistently high level of uptake of about 69% was found at antenatal clinics (Wald *et al.*, 1993; Livingstone *et al.*, 1994; Brock, 1996). One survey found that 11% of those who declined antenatal screening did so because their partner was unavailable or did not wish to be tested (Wald *et al.*, 1993, see also Mennie *et al.*, 1993b).

Questionnaire surveys suggested that both those identified as carriers and non-carriers found screening acceptable (Watson *et al.*, 1992; Mennie *et al.*, 1992; 1993a) and that information was understandable (Mennie *et al.*, 1993a). However, the number of carrier couples identified through these pilot studies was small, especially outside antenatal clinics (Modell, 1993; Cuckle *et al.*, 1995). In one antenatal centre, 24 couples were identified over a 5-year period (Brock, 1996). Among these couples, the uptake of prenatal diagnosis and of abortion of affected foetuses was high; 22 of the 24 couples identified by the programme requested prenatal diagnosis and all eight affected pregnancies were terminated (Mennie *et al.*, 1992; Brock, 1996).

There is debate about the way that the results of the pilot studies should be interpreted. Many have argued that the feasibility and acceptability of population CF carrier screening in the UK has been indicated by these studies (Modell, 1993; Brock, 1995; Cuckle *et al.*, 1995). Prenatal carrier screening is felt to be cost-effective (Cuckle *et al.*, 1995; Cuckle and Murray, 1998), although it has also been pointed out that the low number of carrier couples identified makes predictions of either the acceptability or the cost-effectiveness of a screening programme difficult (Modell, 1993; Cuckle *et al.*, 1995).

Within this, some have argued that, although testing adults before conception may be theoretically preferable, screening during pregnancy offers the more feasible and effective approach, because of the higher uptake of prevention measures at each stage (Modell, 1993; Brock, 1996). It has also been pointed out that screening in pregnancy

may be more relevant to individuals, that this is acceptable for other conditions, and has been shown to be acceptable for CF (Wald, 1991; Mennie *et al.*, 1992; Brock, 1995). The effectiveness of screening before pregnancy, by contrast, depends on people taking part in the programme and understanding and remembering their carrier status (Editorial, 1992). Others, however, have argued that, since the rates of uptake of screening seem to depend on the setting and the method of invitation, high rates of uptake in antenatal clinics might be evidence of pressure rather than of the acceptability of carrier screening (Bekker *et al.*, 1993; Clarke, 1995), and some have argued for screening before pregnancy (Raeburn, 1994), or for the provision of a variety of screening methods (Modell, 1993).

Others, on the other hand, have argued that the case for offering cystic fibrosis carrier screening outside families has not been made (Marteau, 1994; 1996). They have questioned whether information on carrier status is wanted (Stone and Stewart, 1996), whether the information provided might be pessimistic, or how well-informed decisions to have prenatal diagnosis or terminations have been (Koch and Stemerding, 1994), and have argued that the introduction of CF screening programmes has not been not properly evaluated. Koch and Stemerding (1994), in Denmark, have suggested that both existing antenatal screening programmes and surveys of acceptability can be seen as legitimating the introduction of further tests. From a concern about the unknown long-term effects of population carrier screening (Editorial, 1992; Marteau, 1994), one follow-up study after 3 years indicated some residual anxiety in a few carriers and a slight effect on their perceptions of their own health, but also found that a number of both carriers and non-carriers could not accurately recall their test results (Axworthy *et al.*, 1996). A number of studies have indicated that it is not fully understood that a negative result does not necessarily offer reassurance (Bekker *et al.*, 1994; Mennie *et al.*, 1997).

It has also been suggested that the uptake of testing and termination in the general population might be greater than in families at risk (Marteau, 1996). There is some evidence of differences in response towards screening between relatives and others in the population; those with more knowledge being less anxious about screening than others (Bekker *et al.*, 1994), or showing more interest in learning their carrier status when invited by letter (Super, Schwartz and Malone, 1992). One large study at antenatal clinics concentrated only on high-risk couples identified through the screening programme (Brock, 1996), so that comparisons could not be made. Little is known about relatives' uptake of carrier screening or the use that they make of this information, although one study (Lane *et al.*, 1997) and anecdotal evidence suggests that it may be very variable (personal communication, West of Scotland Regional Genetics Service).

Following the pilot programmes, population carrier screening is being offered at maternity hospitals in Edinburgh (Warner, personal communication, 1999), although not in Glasgow, and not on a national basis (personal communication, West of Scotland Regional Genetics Service). The results and implications of the pilot programmes in the UK are being assessed and a report prepared. A number of geneticists have argued that it is feasible to introduce routine carrier screening at antenatal clinics, and they have advocated its introduction when counselling services are felt to be adequate (Cuckle *et al.*, 1995, Haddow *et al.*, 1999). However, studies of the attitudes of GPs suggest that, while this group approves of the provision of CF carrier screening (Boulton and Williamson, 1995), it has been felt to have a low priority, especially in the absence of family history (Mennie *et al.*, 1998). Another suggestion is that carrier screening in the absence of a family history might be offered privately, with cascade screening and neonatal screening more generally available (Super and Abbott, 1998).

There continues to be debate about whether or not routine neonatal diagnostic testing should be introduced for cystic fibrosis. Arguments for the introduction of neonatal screening (which is supported by the CF Trust) are that diagnosis is distressing and that early intervention might improve prognosis (Al-Jader *et al.*, 1990). Arguments against relate to the uncertainty of the test results and the lack of clear evidence of long-term benefit (Wilfond and Fost, 1990; Cuckle and Murray, 1998; Wald and Morris, 1998). Neonatal diagnosis would, however, also alert some, but not all, couples to their carrier status (Modell, 1993). Although it has been argued that this is a poor method for starting population screening (Modell, 1993), in one study in the UK, a neonatal screening programme has been followed by a reduction in family size. The author has pointed out that, given the apparent desirability of offering population screening and the disadvantages of some other approaches, neonatal diagnosis might offer a useful option (Carswell, 1996).

Summary

This section has focused on the conditions being investigated in this study. Living with Down's syndrome, spina bifida or cystic fibrosis may have particular consequences for those affected and for others, and there may be various implications for services. Different arguments have been drawn on in discussing the appropriateness of trying to reduce the incidence of each of the three conditions, as well as about the costs and benefits of offering a screening programme.

However, while there are differences, the same general debate about the morality and about the costs and benefits of screening programmes apply to all. These arguments, which will be outlined in Chapter 3, include a critique of the principle of screening

programmes and the different legitimating arguments of 'prevention' and 'choice'. The debate is complex, but one fundamental critique is that the rationale for screening programmes - focusing on conditions and solutions - rests on a particular system of values. This argument has been suggested by sociologists and by some disability activists and will be considered next.

CHAPTER 2

SOCIOLOGICAL CONTRIBUTIONS TO THE SCREENING DEBATE

The unquestioned benefit or uncontroversial nature of screening programmes has increasingly been challenged. The previous chapter has outlined some concerns about the appropriateness of 'preventing' certain conditions or of offering screening, but others have gone further to question the principle of screening, or to argue that such programmes contain assumptions about benefit.

Among those most critical of screening programmes have been some individuals in the disability rights movement, who have argued that such programmes call into question both the value and the quality of the lives of disabled people (see Kaplan, 1993). Disability activists have drawn on sociological arguments to suggest that the logic of screening - that impairment should be prevented in both individuals and society - may be seen as premised on the values of biomedicine. They have proposed an alternative 'social model' of disability to suggest that both definitions of 'problems' and the causes of the problems of disabled people can be seen as socially constructed. However, not all those affected with such conditions might concur with the arguments of disability activists. The experience of living with conditions has also been the subject of sociological research.

The purpose of this chapter is to outline sociological perspectives towards both screening programmes and the experience of living with a disabling condition. The first part of the chapter reviews the sociological literature and its contribution to an understanding of screening programmes and the values underlying their use. The second part of the chapter examines theoretical and empirical approaches towards the experience of living with a condition. A first section considers theoretical approaches to the experience of disability, including the 'social model', and the critiques of some disability activists towards screening programmes. This is followed by a discussion of the limitations of the social model in examining experience, and the contribution of a 'sociology of the body'. The chapter concludes by outlining different approaches to empirical work.

Introduction

The interest of sociology is in the interaction of the individual and the social, as Wright Mills has explained, in the way that "the personal troubles of milieu" cannot be separated from "the public issues of social structure" (Mills, 1970). While reproductive decision-making or living with a condition may be seen as private, personal matters,

they may also be understood as matters of public debate and policy. A sociological approach considers the social consequences of actions, and regards actions and experiences as socially and historically situated. Although different theoretical perspectives start from different premises about the way that society and its relationship with the individual should be perceived, and have different implications for method, from a social constructionist approach, both experience and knowledge (or thinking) may be seen as socially constructed (Berger and Luckmann, 1967) by social norms, power relations and material circumstances. Sociology aims to examine, by theory and method, this material and cultural social context.

Sociological approaches to screening programmes

Sociologists have argued that many of the concerns raised by psychologists about the questionable benefit of screening programmes relate to matters of safety or presentation, and can be addressed within the medical profession, by the provision of counselling for example. Van Dyck (1995) has made the point that such critiques use the language of science. From a view that important questions about screening programmes concern the social context in which they are offered and especially questions of values, sociologists have criticised a focus in the literature on concerns about the individual (Lippman, 1992b). They have also pointed out that ethical analyses require an examination of the material and cultural social context within which choices about prevention are offered, or within which moral values are defined (Rothman, 1985; Stacey, 1992; Lippman, 1992b).

Different views have been expressed by sociologists about the influences underlying the development of screening programmes, and their power. Many have argued, however, that a powerful influence on the introduction of screening programmes offered in reproduction has been a technological medical form of thinking.

From the 1970s, the concept of 'medicalisation' has been suggested, from a number of sociological perspectives, to describe the perceived expansion of areas of life claimed as medical, and the increased status of the medical profession and medical knowledge, although different explanations have been offered from different perspectives. Whereas Friedson (1970) has argued that medical dominance rests on its claims to effectiveness, a Marxist analysis has offered explanations in terms of the service of the institution of medicine for capitalism. From this perspective, medical interventions have been suggested as reproducing health inequalities (Turner, 1987), and medical thinking, determined by the economic interests of the ruling class, as providing individualistic explanations for collective problems of ill-health (Navarro, 1976). Medical knowledge

has been suggested from other perspectives as a cultural construct based on only one value-system or rationality, although with different explanations for its power.

The domination of technological medical thinking or 'biomedicine' was suggested by Foucault, who argued that, at the beginning of the 19th century, there was a changed way of thinking about health and illness (Foucault, 1973). He made the case that biomedical thinking as an episteme or discourse was characterised by an emphasis on the rational, the scientific, the analytical and the classificatory, on diseases and disorders as entities in themselves. For Foucault, biomedical discourse also defined doctors as professional 'experts' in relation to individual 'patients', thus legitimating practices of social control, such as scrutiny, surveillance or intervention (Armstrong, 1987). Social control might also be exerted by self-discipline or self-control, since biomedical thinking was defined (normatively) as morality (Turner, 1987).

From this view, the status of medical knowledge has been seen as deriving from the separation of the biological and the social, and from the prioritising of the biological or 'natural' by its representation as scientific or neutral (Stacey, 1992; Oakley, 1993a). Some have proposed that the status of the medical profession, and its perceived immunity from criticism, rests on this assumed objectivity and on unexamined values (Wright and Treacher, 1982). Zola (1975) has described 'medicalisation' as the presentation of an apparently objective and rational biomedical model as not only the norm but also as expertise, a matter of importance, the implications being that not only are problems which might result from social circumstances increasingly defined in terms of 'health' and 'illness', but that an acceptance of these definitions presupposes medical intervention.

It has been suggested that there are a number of questionable assumptions within 'medicalisation', and that the concept can be criticised at different levels (Wright and Treacher, 1982), in some cases from within the medical profession. Medical claims to the safety and efficacy of interventions have been challenged since the 1970s, for example by Illich (1975) and McKeown (1979). As noted already, concerns about the possibly iatrogenic effects of prenatal diagnosis and abortion have been expressed within the medical profession and by psychologists.

Turner (1987) has argued that the separation between technological medicine and the social in 'biomedicine' (discussed within medical sociology) needs analysis at different levels; theoretical analyses of the social organisation of society, the social construction of disease categories, and an analysis of the descriptive experience of illness.

There has been an increasing interest within social science of the experiences of individuals living with disabling conditions. Whereas, in 'biomedicine', the body and its anomalies is the object of analysis, in social science a separation of 'nature' and 'nurture' can be seen as reductionist and determinist. For example, the importance of social and emotional aspects of experience was recognised in the definition of health in the WHO constitution (1948): "a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity". Biological explanations have traditionally been refuted as 'essentialist' in sociological theory, which has emphasised the effect of environmental or socially constructed factors, such as class or gender, on experience. Empirical studies of the experiences of those living with a disorder carried out by social scientists have illustrated the inter-relationship of the physical and the social, and it has been noted that the medical profession, which has drawn on such studies, has long recognised that the consequences of having a disorder cannot be seen narrowly in terms of physical effects (Bury, 1994). As noted in Chapter 1, some scientists and medical professionals have discussed the interaction of genetic and environmental factors in the prognosis of those with a genetic condition (Rose, Lewontin and Kamin, 1984; Lewontin, 1991; Knight, 1992).

While medical professionals have acknowledged the overlap of nature and nurture, and social aspects of experience (see Kerr, Cunningham-Burley and Amos, 1998c), from a social construction perspective, experience - whether of considering taking part in a screening programme or of living with a condition - cannot be separated from the values inherent in conventional thinking. From this perspective, 'problems', 'risks', 'needs' and 'solutions' can be seen as constructed from a value-system which defines what may be seen as right or natural (Lippman, 1991a; Scott and Williams, 1992). It has been argued that sociological challenges to the culturally powerful and apparently value-free nature of biomedical thinking are more difficult to address from within the medical profession (Wright and Treacher, 1982).

Early critiques of the values underlying new reproductive technology came from feminists in the 1970s. With the introduction of amniocentesis, and especially IVF, while concern was expressed about the safety and effectiveness of new techniques (Hanmer, 1984; Oakley, 1993b), more fundamentally, feminist sociologists drew attention to the social context and power relations within which new reproductive technology was introduced, and to the values inherent in medical intervention (Roberts, 1981; Rothman, 1985). They argued that pregnancy was increasingly being defined as a medical rather than a social event, in which women were patients, and pointed out the difficulty of resisting 'medical' assumptions or the expectations of medical professionals (Farrant, 1985; Oakley, 1993a; 1993b). For example, drawing on the work of Foucault,

Petchesky (1987) suggested that antenatal care and ultrasound scans could be seen as an example of the way that physical bodies may be controlled by a powerful medical gaze. Feminists offered different explanations for the 'medicalisation' of pregnancy, suggesting that new reproductive technologies were introduced within a context of patriarchy (Wallsgrove, 1980; Corea, 1984; Hanmer, 1984) or capitalist values (Rothman, 1985). Medical intervention in pregnancy was seen as having the potential to uphold or reflect these values and to recreate women's unequal social position in society or stereotypes of their role as mothers (Denny, 1994), and hence was seen as representing a threat to their reproductive freedom (Hanmer and Allen, 1980).

Whereas early feminists suggested that the provision of reproductive technology could be seen as reflecting certain values (often those of biomedicine), from the 1980s, with a growing emphasis on women's reproductive rights (Hanmer, 1984) and on individual control more generally, the focus of feminists has been on women's control over its use, in line with 'women's right to choose' abortion (Stanworth, 1987). Analyses of the 1970s, suggesting women as victims, have been criticised as over-determinist (Rose, 1987; Denny, 1994; Van Dyck, 1995), although some have continued to suggest the gatekeeping role of medicine as having the potential to limit access to reproductive possibilities, and to reinforce conformist values (Rose, 1987; Henifin, 1993). It has been noted that technology can offer both the means for individuals to control or to be controlled (Comaroff, 1982), and a number of feminist writers have suggested that the implications of new reproductive technology are contradictory for women, potentially both widening and narrowing their choices (Farrant, 1985; Stanworth, 1987; Oakley, 1993b).

Early feminists have been criticised for treating women not only as victims but also as a homogenous group (Denny, 1994; Van Dyck, 1995). In line with a recent emphasis within feminism and within sociology on experience and on individual difference in a pluralist society, it has been argued that the feminists of the 1970s did not speak for all women (Wertz and Fletcher, 1993).

The changing concerns of many feminists have reflected more general changes in views towards the status of medical professionals and the power of medical thinking in modern society. It has been suggested that, at the end of the twentieth century, there is a new form of society, post-modernity. Such a society has been seen as characterised by a scepticism towards grand narratives (Lyotard, 1984), or expert discourses or knowledge; unease rather than certainty about progress (Beck, 1992). New forms of social processes have been proposed, with an emphasis on individual rights, responsibility and self-efficacy, choices and their outcomes, especially in relation to health (Nettleton, 1995). Many have drawn attention to a reduction in the authority of

medical professionals (Williams, 1998), and to the increased control of individuals over their treatment (Lupton, 1997). Some sociologists have argued, following Giddens (1991), that the concept of 'medicalisation', in terms of the balance of control, might now be less relevant, the relationship between professionals and individuals being better seen as one of reflexivity (Williams and Calnan, 1996). While empirical work has indicated some scepticism among the public towards technological medicine and assumptions of progress (Williams and Calnan, 1996), it has also been suggested that there are contradictions in the way that medical values are perceived by the public; an increasing scepticism about medical treatments and interest in alternative medicines, yet at the same time a dramatic expansion of technological medicine, an increasing reliance on medical intervention and an interest in one's health (Stacey,1992).

Many have argued that there are aspects of life in which medical values continue to dominate. The concept of 'medicalisation' is still felt to be relevant in the context of reproduction (Farrant, 1985; Scambler, 1987); Oakley (1993a; b) has claimed that medical values continue to shape both women's experiences and the common understandings of pregnancy, which is seen in a mechanistic way, implying an emphasis on physical and measurable outcomes and an uncritical acceptance of technology.

Medical values or a 'medical model' has also been suggested as defining the way that impairment is perceived (Oliver, 1990). Similarly, many have argued that the continuing power of 'biomedical' or scientific explanations and definitions, and assumptions of value-neutrality or certainty, can be seen in the current interest in genetics and heredity, where - in spite of a general recognition of the inadequacy of biomedical explanations - such definitions or 'codes' appear to offer powerful and natural explanations (Davison *et al.*, 1994; Shakespeare, 1995; Schwartz, 1997). It has been argued that genetic issues continue to be presented in the medical literature in ways which imply benefit or simplistically (Conrad, 1999), and with a lack of acknowledgement of the underlying rationale of science or of alternative values (Cunningham-Burley and Kerr, 1999). Lippman (1992b: 1470) has suggested a process of 'geneticisation' (as a development of 'medicalisation') in which, she argues, "differences within individuals are reduced to their DNA codes, most disorders and behaviours, as well as physiological variations, are defined as at least in part genetic in origin, and the adoption of interventions that employ genetic technologies to manage problems of health is advocated".

From this view, sociologists have argued that biomedical values are inherent in the provision of screening programmes, which implicitly define both genetic problems and suffering, and medical intervention as the 'right' or logical solution (Stacey, 1992;

Lippman, 1992b; Schwartz, 1997). It has been claimed that assumptions of 'prevention' as the outcome of screening programmes are evident in the medical literature (Shakespeare, 1995; Bailey, 1996), but also that the existence of such programmes contains a normative view of what should be done, implying the importance of finding out information about an unborn child (Lippman, 1989; Oakley, 1993; Richards and Green, 1993), as well as the acceptability and appropriateness of abortion on the grounds of foetal abnormality. From this perspective, Lippman (1991a; 1992b) has argued that a lack of analysis of 'medical' or 'scientific' values - or assumptions of objectivity and neutrality - can legitimate prenatal diagnosis and abortion, preventing discussion about ethical issues, such as the widening range of conditions for which prenatal diagnosis may be considered.

Some have gone further to argue that screening programmes and genetic knowledge can offer a means of social control, whether of women, given their social position (Oakley, 1993b; Schwartz, 1997), or of individuals more generally (Lippman, 1992b; Williams, 1998). Genetic screening, offering a 'profile' of individuals, has been seen as surveillance (Hubbard and Wald, 1993), and the classification of 'genetic diseases' and diagnostic counselling as evidence of power relationships, or as indications of rational behaviour (Yoxen, 1982). Biological ideas about inheritance have been seen as linked not only to class and patriarchy, but also to a current cultural emphasis on 'individualism' in modern society (Nelkin and Lindee, 1995; Schwartz, 1997). Williams (1998) has drawn on the work of Zola to suggest that, in a society in which health is seen as a value, or as a moral issue, and a matter of individual responsibility, people may have little choice about reproductive decisions involving genetics.

Sociologists have questioned why, or in whose interests, screening programmes are offered (Farrant, 1985; Lippman, 1991a; Oakley, 1993b), and some have questioned the extent to which new technologies have benefited individuals or women (Oakley, 1993b). Concern has been expressed about the potential of screening programmes to constitute a pressure on people's decisions or on their existing values (Green, 1990; Wertz and Fletcher, 1993). However, empirical studies have indicated a high uptake of screening programmes, as well as women's general approval of their provision (Farrant, 1985; Tymstra *et al.*, 1991; Pryde *et al.*, 1993; Wertz and Fletcher, 1993).

Some commentators have suggested that the findings of empirical work might be seen as illustrating a normative view about the right course of action (Tymstra, 1991). Others, however, have offered explanations for these findings in terms of women's social position or the meaning of their lives (Rothman, 1985; Wertz and Fletcher, 1993; Oakley, 1993b); some have suggested women's material inequality (Rothman, 1985; Warren, 1992; Denny, 1994), and many have pointed out the social reality of women's

role or responsibility for caring (Finch and Groves, 1983; Lippman, 1989). The point has been made that women's choices about abortion more generally cannot be seen as separate from their lives, including their role in the workforce (Luker, 1984). Rothman (1985) has argued that there are material constraints on women's decisions about abortion on the grounds of foetal abnormality, as there are about abortion more generally, since such choices are offered in a context of individual responsibility and the powerlessness of women.

Many have stressed that screening programmes cannot be seen outside a social climate of economic restrictions, an emphasis on cost-effectiveness generally, and increasing individual and family responsibility for health care (Rothman, 1985; Brodsky, 1990; Morris, 1991). For example, Rothman (1990) has pointed out the economic costs, in the US, for families or individuals living with ill-health or disability, and the possibility of lessening medical and social support. From this view, it has been claimed that prenatal diagnosis can be explained, like individual health care, as both constructed as a problem of individual responsibility and as experienced as such, in a climate of cuts in social provision (Lippman, 1989; Morris, 1991). It has been pointed out that scientific knowledge must be seen within the values of the existing social context (McLean, 1994), or that medical definitions can be seen as reflecting more general societal norms or needs.

This section of the chapter has outlined sociological critiques of screening programmes, in particular the view that prenatal diagnosis may be seen as resting on 'biomedical' values, which define both 'problems' and 'solutions'. This perspective has been drawn on by some disability activists who have been in the forefront of critiques of screening programmes.

The main focus of the disability rights movement has been to suggest the way that the experience of living with an impairment might best be understood. Activists have drawn on sociological analysis, and on a political approach, to propose a 'social', rather than a 'medical' model of disability, to suggest that both definitions of the 'problems' of impairment and the causes of problems of disabled people can be seen as socially constructed rather than as inevitable. However, the social model has been criticised for a lack of discussion of differences in the experiences of those living with an impairment, the subjective and the physical. The experience of living with a disabling condition or illness has been considered by sociologists in a number of ways, both theoretically and in a body of empirical work. Since this study is examining the way that people affected with a disorder perceive both screening programmes and their experience of living with the condition, these different approaches in the literature are outlined next.

Sociological approaches towards the experience of living with a condition

With changing patterns of illness, increasing numbers of people are living with long-term or disabling conditions which are a constant presence in their lives. Epidemiological surveys have measured the prevalence of conditions in order for services and benefits to be provided, but, with the recognition of the distinction between a condition and its consequences, there has been an increasing interest in the experiences of those affected. Early research in social science focused upon the consequences, mainly material and psychological, for families where children were surviving into childhood and adolescence, authors of studies generally noting the importance of parents' reactions and family coping strategies, since these have been suggested as crucial factors in adjustment (Burton, 1975; Anderson and Spain, 1977). More recently, the emphasis in research has been on the experiences and perceptions of those affected with certain conditions.

Early empirical studies, including government surveys (Harris, Cox and Smith, 1971), suggested the negative experiences of those with certain impairments, and the effect of social factors on their lives. A sociological approach to experience reflects the fact that illness may be seen as a social phenomenon, culturally situated (Williams and Popay, 1994), although theoretical perspectives vary in the extent to which experience may be seen as socially constructed, and in the relative importance of cultural and material factors. From a number of perspectives, the concept of disability has been proposed.

Disability

From a social interactionist perspective and the sociology of deviance, Goffman (1968), against medical and psychological thinking, theorised the effect of powerful cultural norms on the experience of those with certain conditions, who might be seen, by themselves and by others, as disabled. Goffman's concern was with the breakdown of social interaction and with the effect of societal reactions, or labelling, on experience (Williams, 1987b). For Goffman, from a societal perspective of 'normality', those with certain attributes, which were seen as unwanted or stigmatised, were perceived or characterised as abnormal and unacceptable types of person. Goffman argued that this ascribed negative or stereotyped social identity might affect interaction and could legitimate discrimination, whether of prejudice or inappropriate treatment.

Goffman proposed that individuals might share these dominant norms, and suggested the effect of the stigma-bearer's acceptance of such norms on individual response, including the effect on identity. Whether the attribute was obvious or hidden, Goffman (1968) argued that real or anticipated societal reactions, and shared cultural norms,

might affect the individual's ego or felt identity, which he suggested would be 'spoiled', as would their social identity. Goffman suggested individual responses in order to manage (or counter) 'spoiled' identity, the construction of an alternative, positive identity, whether for the purposes of interaction or for psychological adjustment. He suggested that, where the stigmatising condition was visible - the individual was perceived as 'discredited' - an individual might develop 'covering' strategies to manage tension in interaction. Where the condition was hidden, and the individual might be defined as discreditable, 'passing' strategies might be used for the management of information. Following Goffman, Scambler (1984) has developed the concepts of 'felt' and 'enacted' stigma, the former affecting perceived identity and the latter referring to discrimination.

Although Goffman analysed the 'management of a spoiled identity', it can be argued that he was not advocating the use of strategies to attempt to deny difference or approach the normal, but was rather emphasising the pressure exerted in society generally or by health professionals towards 'good adjustment' or normality (Williams, 1987b). His interest was in the negative experiences of those in minority groups, and he pointed out the need to examine the wider social structure, as well as problems of interaction (Goffman, 1968).

Goffman's view that social prejudice can legitimate discrimination, and his suggestion that acceptance of social norms can affect identity, has been drawn on by minority groups in the human rights movements of the 1970s. In particular, his ideas have been developed by Wolfensberger in relation to the treatment of people with learning disabilities. As Emerson (1992) has explained, Wolfensberger (1972; 1983) proposed the principle of 'normalisation', a prescriptive approach which emphasises that people with learning disabilities should be treated equally in terms of rights and services, and equally valued in society. The implications of this approach are discussed in more detail in Chapter 7. However, 'normalisation' has been seen as a moralistic approach to disability (Dalley, 1992), and functionalist assumptions of shared values have been criticised by sociologists. From structural perspectives, normalisation has been seen, as have interactionist approaches more generally, as lacking an analysis of material factors in experience, culturally determined social position, or an explanation of the origins of social norms (Chappell, 1992; Dalley, 1992; Whitehead, 1992).

Early empirical studies carried out by sociologists drew on various theoretical perspectives in examining the experiences of those living with certain disabling conditions, and in considering the extent to which such experience might be common. From an interactionist perspective, Strauss (1975) focused on the work done by individuals living with a chronic illness in maintaining as normal a life as possible,

strategies of coping with a condition and social consequences. Blaxter (1976) and Locker (1983), drawing on both micro and macro theory, suggested the consequence of socially constructed disadvantage or disability, while examining different meanings of 'disability' for the individual. Zola (1982) a disabled sociologist, aimed to investigate and present the experience of disability, including both personal experience of physical limitations and the powerful effect of cultural norms in a society in which disability is defined as 'invalid'. At the same time, he emphasised the limited possibilities for people with impairments, and argued for a collective approach towards disability.

The disability rights movement and the 'social model' of disability

A collective approach to disability has been emphasised by the growing disability rights movement. Activists have drawn on the civil rights movements originating in the US, and on sociological theory, in particular Goffman's work on the position of minority groups in society, and Marxist explanations of social structure, power, and the hegemony of dominant groups. Disability has increasingly been seen as socially constructed exclusion. Both the consequences of living with a disorder and the characteristics which are termed disabling may be seen as socially determined (Asch, 1989). However, as Shakespeare (1996) has explained, disability can be seen as socially constructed in a number of ways, implying different interventions in the sense of collective or social solutions.

A cultural approach to disability suggests that those with impairments are seen as abnormal in society. Shakespeare (1994), for example, has argued that societal norms are implicit in negative or stereotypical images of disabled people. Others have drawn attention to the use of negative adjectives, such as defective or dis-abled as nouns (Zola, 1982; Finger, 1984; Peters, 1990), suggesting a contrast with an 'able' society. Social classifications or categorisations, constructing certain individuals as 'other', have been seen as arising from a need for order in society and as suggesting social norms (Douglas, 1970b).

Anthropologists have proposed the concept of liminality, of being on the margins, for those who are seen as being both the same and as different from others, or on the boundaries of classification (Murphy *et al.*, 1988). Such people are felt to be threatening to others, making interaction problematic, and often socially invisible. Murphy and colleagues have argued that the concept of liminality might offer a more useful model for viewing disability than deviance, because most disabled people do not grow up among other disabled people, nor are they brought up by disabled parents. Drawing on these arguments, Shakespeare (1994) has stressed that the experiences of disabled people can be influenced by societal attitudes towards normality or by the way

that they are perceived, both because individuals can be objectified by attitudes that they evoke fear, and because cultural attitudes influence expectations.

From a cultural approach, it has been argued that impairment ought not to be seen as unnatural, but rather as part of the diversity or continuum of the human condition (Shearer, 1981; Shakespeare, 1995). Collective pride, or a celebration of difference, has been proposed as one solution to being perceived as negatively different. Other social solutions have been advocated in terms of education and social inclusion, in order to counter fear and prejudice, to change attitudes within society towards impairment and to widen definitions of 'normal' (Flaker, 1994).

Cross-cultural analysis has shown that what counts as a disability is culturally-specific (see Ingstad and Whyte, 1995), and some have questioned why certain conditions - such as mobility and IQ for example - should be deemed important (Stacey, 1992). From a Marxist perspective, Oliver (1990) has argued that cultural definitions can illustrate the social organisation of society; from this view, what is defined as a problem in the UK may be seen as related to ideas about social productivity within which physical impairments are defined as important (Oliver, 1990; Abberley, 1992).

For Oliver, disability should be understood, not as a personal tragedy, but as a relationship of social oppression (Oliver, 1984); disability can be explained in terms of a failure by the state to meet the needs of people with impairments (Oliver, 1990). From this perspective, the 'problem' of disability is located in the social structure of society, the way that society is organised, and in its processes of inclusion and exclusion (Rioux, 1993), those with impairments sharing a common social position of socially created inequality or disadvantage. A Marxist approach suggests that both negative individual consequences, and explanations in terms of individual problems can be regarded as socially caused; hence 'problems' such as dependence may be seen not as inevitable but as amenable to social change.

A materialist approach acknowledges problems stemming from impairments but argues for social, rather than individual, solutions or responsibility. Oliver (1990) has argued for a political response to disability, which would identify the structures and institutions within society which disable people with impairments, and would work towards the eradication of such barriers (attitudinal, physical or economic), ensuring that the needs of disabled people are met. From this perspective, the emphasis is on the special needs of disabled people as a group; societal solutions are seen in terms of a change in the way that society is organised, changes in policy or legislation, increased economic support, and improved opportunities. From a view that independence and employment

might be achievable with improved intervention and support, there has been a focus on goals of independent living.

While there are differences within the disability rights movement, all activists have emphasised social solutions to disability in terms of inclusion in society and equal rights, but also in terms of examining and challenging existing societal definitions or explanations. It has been argued that such definitions might not only have consequences for the individual, but might also preclude other ways of defining or perceiving the problem (Oliver, 1990). Many have argued that the prevailing or conventional view of disability is a 'medical model', based on a system of unacknowledged 'biomedical' values. It has been argued that impairments, traits or conditions are 'medicalised' or defined as medical. The implications within a 'medical model' are that the consequences of living with a condition are inevitably problematic and that difficulties stem directly from the condition (Oliver, 1990; Asch, 1994; Bailey, 1996).

However, within this critique, 'medical' has been interpreted in different ways and challenged on different grounds. Some have argued that medical definitions, such as Down's syndrome, imply a focus on the condition rather than the person. Such definitions may also be seen as stereotyping labels, implying a lack of 'perfection' (Booth, 1985; Brown and Smith, 1989). It has also been argued that a physical difference or anomaly is perceived as medically unnatural or as 'tragedy', implying the importance of medical intervention (Shakespeare, 1995). From this perspective, some disabled people have questioned the idea of inevitable adaptation, or rehabilitation (Peters, 1990) as adjusting to wrong ideas about normality (Shearer, 1981; Shakespeare, 1995). On the other hand, from a Marxist perspective, it has been argued that medical or 'official' definitions imply individual rather than collective problems, and individual solutions in terms of responsibility for health, rather than social support (Stacey, 1985; Oliver, 1990, Morris, 1991).

Disability activists have proposed a 'social model' of disability as an alternative to a 'medical model'. A social model of disability challenges assumptions of inevitable individual psychological or medical problems resulting from impairment (Finkelstein and French, 1993), indicating that many of the problems faced by disabled people might be seen as collective and socially caused, rather than individual, and also that 'problems' may be seen as defined as such. The social model suggests the potential for social change (Kaplan, 1993), whether in terms of the provision of increased material support, improved rights or changed attitudes and a redefinition of disability (Asch and Fine,988a; Morris, 1991).

The disability rights movement, in the UK and elsewhere, has campaigned, on the basis of the social model of disability, to improve the rights of disabled people, and to change attitudes within society. An emphasis on needs and equal rights has been influential on policy and thinking. Activists have argued that, in fundamental ways, people with impairments should be treated in the same way as others; for example, they should have the same right to medical treatment (Asch and Fine, 1988b), and their sexuality and reproductive needs should be acknowledged in the same way as that of others (Finger, 1984; Saxton, 1984).

At the same time, some have considered the implications of difference. Within this, from a focus within feminism on difference, some have examined the particular experience of being a disabled woman (Asch and Fine, 1988a; Morris, 1992). Asch and Fine (1988a) have argued that policy regarding disability has focused on disabled men, and that both disabled people and women may be seen as disadvantaged by social position rather than by biology. Others have suggested social or cultural norms in the way that each of these groups may be defined and valued (Finger, 1984), and there are felt to be similar issues for disabled people and for women concerned with cultural images of a perfect body (Blackwell-Stratton *et al.*, 1998). Asch and Fine (1998a) have cited evidence that disabled women are less likely than non-disabled women to be in a permanent relationship. They have argued that disabled women are considered to be unfit as sexual partners, but also as mothers, because of fears (often irrational) about inherited conditions and about their ability to care for a child.

Some disabled feminists have also argued that the women's movement has ignored the concerns of disabled women (Asch and Fine, 1998a; Morris, 1991), noting for example that issues about the rights of disabled people to parent have been little discussed by advocates of reproductive rights (Finger, 1984). Finger has also made the point that there has been an unconsidered acceptance of the values inherent in prenatal diagnosis and abortion on the grounds of foetal abnormality, because of an emphasis on women's choice. Some in the disability rights movement have argued that debates about women's responsibility for caring, and women's independence and rights, have wrongly constructed disabled people as the 'problem', suggesting that the needs of disabled people and those of their families, carers or women are in conflict (Finger, 1984; Keith, 1992). Many have drawn attention to the lack of community support or services for those bringing up a disabled child (Blackwell-Stratton *et al.*, 1988; Morris, 1991), and it has been recommended that a focus on the equal rights of disabled people might offer a broader approach to discussions about caring and responsibility (Keith, 1992).

Critique of screening programmes by disability activists

Among those most critical of screening programmes have been some in the disability rights movement, for example Finger (1984), Saxton (1984), Davies (1987), Asch and Fine (1988b), Asch (1989), Morris (1991), Shakespeare (1995), Bailey (1996). They have argued that the logic of offering such programmes implies the avoidance of disability in both individuals and society (Saxton, 1984; Asch, 1986; 1994), and that this logic can be seen as resting on certain societal value-systems in which impairment is seen as problematic and as something to be eradicated. Some have argued that the existence of screening programmes or policies may be seen as evidence that disorders, or those with impairments like themselves, are felt to be problematic (Asch, 1986; Shakespeare, 1995), and hence that 'prevention' may be assumed to be both beneficial and right.

Within this rhetoric there are different arguments, which will be examined in more detail in Chapter 3. One argument relates to the wrong use of knowledge and information, the other relates to wrong assumptions about individual benefit. Both rest on a critique of the way that impairment - or those with impairments - might be generally perceived, and (to different extents) on a critique of a 'medical model'.

To imply that society would benefit if certain people - who might be seen as 'different' - did not exist, suggests that such people are unwanted or that their lives have less value. Screening programmes may be seen as a means of selection, and criticised as discriminatory or wrong. Some in the disability rights movement have argued that disabled people are already defined as problematic or unwanted within society (Finger, 1984), whether because they are seen as 'abnormal' or imperfect, as socially unproductive or burdensome (Morris, 1991), or whether from medical standards of fitness and health (Shakespeare, 1995; Bailey, 1996).

To imply that prevention may be more individually beneficial suggests inevitable problems or suffering for those affected or for others. A number of disabled people have suggested that such arguments may be based on unjustified assumptions, which may be seen as medical (Asch, 1989; Barile, 1990; Goundry, 1990; Morris, 1991). As Asch (1994: 13) has expressed it, "the whole genetic enterprise is permeated by the medical model of disability - linking every difficulty to the physiological characteristics of the condition and not to any characteristics of the society in which people with the condition live their lives". Not only might experience be a subjective matter, but - from the social model of disability - both definitions of 'problems' and the causes of the problems of disabled people can be seen as socially constructed (Shakespeare, 1995).

From this perspective, 'prevention' might not be felt to be beneficial or necessary, whether for the sake of affected individuals or for others (Asch and Fine, 1998b). The social model also suggests the possibility of different values, focusing on the rights of disabled people, rather than welfare.

However, not all those with 'preventable' conditions might share the views that it is wrong and possibly unjustified to avoid the births of others like themselves. For example, the Genetic Interest Group, a body which supports individuals and families affected by genetic disorders, campaigns against discrimination but does not believe that sanctioning termination for abnormality leads to discrimination against disabled people. The goal of the Genetic Interest Group is to promote awareness of genetic issues so that services are available for those who need them; these services include screening programmes. The group takes the view that individuals should be free to make their own decisions, and supports a woman's right to terminate a pregnancy

Although the views towards screening programmese of activists and of representatives of organisations have been expressed in the media, little is known about the attitudes of affected individuals. While the views of those affected might be shaped by the experience of living with a disorder or a type of disorder, it has been suggested that the social model of disability is inadequate for offering an understanding of experience. The chapter next considers theoretical critiques of the social model, in particular the contribution from 'the sociology of the body', before reviewing the different ways in which the experience of living with a disabling condition has been examined in emprical work.

Critiques of the 'social model'

The social model of disability is a theoretical or political model, based on an analysis of society, and it has been criticised in a number of ways. For example, Davis (1995), as a disabled writer concerned with the power of a 'normalising society', has argued that binary divisions of 'disability' or classifications of disabled people as a group are not necessarily helpful, since they can be used by others to define people with impairments, as well as by disabled people themselves. Others have argued that this type of model can be seen as inappropriate in a pluralist society (Chappell, 1992).

The social model of disability focuses on the way that disabled people are seen and treated, and suggests the potential for different treatment, with attention to special needs and equal rights. It has been suggested that there is ambiguity in the social model in the means prescribed for addressing the problems of disabled people; that goals are suggested of both 'equal value' and of 'overcoming' (Zola, 1982), or that there are

contradictory approaches towards independence (Williams, 1993). To an extent, this alleged ambiguity about whether the theoretical focus should be on equal humanity and rights, or difference and special needs, is a more general difficulty in society; this issue has also been considered within feminism (Brown and Smith, 1989), and is a concern of the medical profession. This tension between normality and difference, the person and the condition, is fundamental to the debate about screening programmes.

While the social model of disability, from a focus on the way that people are treated, suggests the negative experiences of individuals living with impairments, such a model is inadequate for considering subjective aspects of experience and quality of life. Kaplan (1993) has pointed out that little is known about the quality of life of those who might identify as disabled. The balance between theory and empirical work, the objective and the subjective, has been considered within feminism and sociology. The social model has been criticised by some for an over-determinist, over-theoretical or over-socialised approach to experience (Pinder, 1995; Bury, 1996). It has been pointed out that individuals may have different perceptions of the experience of disability (Asch, 1989; Morris, 1991). Commentators reporting empirical work have pointed to differences among disabled people, both in their views and in their experiences; Blaxter (1983) has questioned assumptions that disabled people perceive themselves as a group, and Williams (1996a) that there is a 'disabled experience' or voice.

As Morris (1992) has emphasised, a focus on the subjective and on experience can imply attention to difference or to the condition or impairment. Many commentators, including disability activists, have pointed out that there might be limits to the extent to which important problems experienced by those with impairments might be felt to be socially caused or defined as such (Asch, 1989; Morris, 1991). It has been argued that the social model may be inadequate for aiding understanding of all the problems experienced by people with learning disabilities, for example (Corbett, 1989). Also, pain and limitations may also be part of experience, and for many might be perceived negatively (Morris, 1991; French, 1993a; Davis, 1995). It has been suggested, therefore, that disabled people, those with impairments or illness, are in a different position from those in other minority groups. Thus there is debate within the disability rights movement between those who emphasise the common social position of disabled people and who advocate a political approach to disability (Finkelstein, 1993; Oliver, 1996), and others who suggest that the social model would be enhanced by an analysis of the physical in subjective experience (Morris, 1991; French, 1993a; Crow, 1996). This debate reflects a more general consideration, within sociology, of the way that physical aspects of life might be theorised, or explored in empirical work.

Sociology of the body

From the philosophy of science, there are biological realities, external to our perceptions or interpretations (Sheeran, 1995), and there has been an increasing interest in the way that such material realities, which cannot be seen as constructed, might be included in social science and in sociology (Freund, 1988; Frank, 1990). It has been argued, most notably by Turner (1984; 1992; 1996), that a comprehensive sociology should include an examination and analysis of embodiment, both of populations and individual social actors (Turner, 1984). The relationship between the physical and the social has always been problematic for sociological theory (Armstrong, 1987), and Turner has argued that an analysis of the body has been left out of sociological theory because of anxiety about essentialist thinking. Because of this, sociology can be criticised for 'sociological determinism', or for perpetuating the same Cartesian dualism as has been criticised in the biomedical model (Turner, 1996). Turner has argued that such a separation of the physical and the social has created difficulties within philosophy, phenomenology and post-modernity.

A number of sociologists have suggested the importance of an examination and analysis of the inter-relationship between the physical and the social at the present time. They have pointed to the increasing emphasis on difference and diversity, subjective experience, emotions and psychology, the centrality of the body in the post-modern interest in consumerism, representation and images (Featherstone, Hepworth and Turner, 1991; Shilling, 1993). Commentators have particularly noted the growing interest in the 'medicalised body' (Frank, 1990), the 'interior of the body', 'nature' and genetics (Bury, 1995). Birke (1986) has emphasised the need for sociologists to engage with the issue of the role of the body in experience, in order to challenge both scientific practices and biological explanations.

There is currently debate about the way that the body might be theorised, or embodiment explored in empirical work; these issues, and the relationship between the personal and the collective have been debated within feminist sociology (see Fuss, 1990; Bordo, 1993). Some have questioned whether there can be simply 'a sociology of the body' (Frank, 1990). Turner (1996), noting an increased interest in the work of Goffman, has suggested that the body might be seen in a number of ways; as subject (I am a body), as object (I have a body, perceived body, societal response to body), and also as project (I do a body). For Turner, some aspects of life are more open to social construction than others; he has argued that it is not necessary for sociologists to deny embodiment, although it is necessary to make a distinction between the ontological and epistemological critiques of the biomedical model. As discussed earlier, he has argued

that, in order to examine the interaction of the social and the body - both as 'subjective lived experience' and as 'socially constructed objective presence' and the tension between them - different perspectives are needed, both theory and empirical research, a social construction approach and phenomenology (Turner, 1996).

Theory and empirical studies about the body already exist in medical sociology (Freund, 1988; Bury, 1995). With the increasing number of people living with a chronic illness or a disabling condition, and with an emphasis on 'lay knowledge' rooted in experience, rather than the views of 'outsiders' or experts (Conrad, 1990; Williams and Popay, 1994), a body of sociological work has examined the experiences of those living with a disorder. Research examining experience has been carried out from different perspectives (Frank, 1990; Turner, 1996), and varies in the extent to which it draws on theory and in approaches to analysis (Scambler, 1987). A review of this literature is beyond the scope of this thesis. However, since the experiences of those living with a disorder might be relevant to their views about screening programmes, this section concludes by considering the different ways in which the experience of living with a condition has been examined in empirical work.

Sociological approaches to empirical work

A number of studies, often of chronic illness, have increasingly taken a phenomenological approach, drawing on the interactionist perspective of Strauss (1975), the theoretical work of Goffman (1968) about social value and management of identity, and the interest in anthropology in the narrative, and the inter-relationship of the body, the self and society (Kleinman, 1988; Frank, 1995; Pinder, 1995). While, from this approach, the focus is on psycho-social aspects of experience, the social meaning for the individual (Strauss, 1975; Fitzpatrick, 1984) and individual coping strategies, the body is not taken for granted (Frank, 1990), and some have proposed the central importance of the physical in the experience of living with a condition (Kelly and Field, 1997).

Qualitative studies have suggested the complexity of 'lay views' or perspectives, but also the coherence of strategies and beliefs (Williams and Popay, 1994). A naturalist approach to health and illness, however, has been seen as lacking an examination of the wider social context (Bury, 1991), the social and cultural implications of living with a condition, and the extent to which concepts might be common or generalisations made from experience (Anderson and Bury, 1988; Conrad, 1990). Some in disability studies have criticised an analytical approach focused on 'coping' as both prioritising individual experience and pathologising disability (Shakespeare and Watson, 1997), and sociologists generally have argued that examinations of experience, meaning and

identity should be carried out within a collective framework (Williams, 1996a), strategies or social action understood against a social system in which physical competence is important, and cultural definitions of disability are negative (Bury, 1988). A number of qualitative studies have drawn on the early work of Blaxter (1976), Zola (1982) and Locker (1983), discussed already, in examining the experience of disability (see for example Williams, 1987a; Anderson and Bury, 1988; Pinder, 1996; Thomas, 1997). Some research has been initiated by disabled people, and several studies have examined the experiences of disabled women (see for example Fine and Asch, 1988; Lonsdale, 1990; Morris, 1991; Morris, 1996).

From a Marxist perspective, Oliver (1990) has argued that, because of the focus on the subjective in both phenomenology and medical sociology, neither approach is appropriate for examining the collective experience of disability. From the 1970s, surveys examining the prevalence of certain conditions have been widened to include examination of social consequences of disability, in terms of disadvantage, inequality or a lack of opportunities (Harris *et al.*, 1971; Martin, White and Meltzer, 1989), and some have been carried out on behalf of organisations of disabled people (Barnes, 1991). A focus on needs and rights has implied an attention to the transition to adulthood, and on the opportunities available; a number of studies have looked particularly at the needs of young people and young adults, and at the extent to which they might achieve the objectives, or important aspects, of adult life (Centre for Educational Research and Innovation, 1986; Doyle, Moffat and Corlett, 1994; Hirst and Baldwin, 1994).

Empirical studies and personal accounts have suggested that the social model of disability reflects the experiences of many people with impairments (Lonsdale, 1990). Research has confirmed that disability is often associated with disadvantage and inequality, social isolation and lack of employment (Centre for Educational Research and Innovation, 1986; Barnes, 1991; Doyle *et al.*, 1994; Hirst and Baldwin, 1994), and relative poverty (Blaxter, 1976; Asch and Fine, 1988a; Abberley, 1992). Important problems experienced by disabled people are often also felt to derive from others' perceptions and attitudes rather than from the condition and its limitations (Finger, 1984; Morris, 1991; Keith, 1996). However, studies have also suggested that 'disability' is not a homogenous category (Blaxter, 1983; Hirst and Baldwin, 1994).

While Shakespeare (1997) has argued that empirical work should allow for a variety of theoretical sociological perspectives, there has been debate about an appropriate methodological approach for examining the experience of living with a disabling condition (see Barnes and Mercer, 1996). Bury (1991; 1995) has criticised approaches based on the social model as over-determinist, taking assumptions of negative

consequences as unproblematic, avoiding consideration of the physical experience of chronic illness, and lacking an analysis of action; he has argued that the body of work within medical sociology has usefully indicated the main problems that people living with certain conditions face, and the main responses that they adopt for overcoming them (Bury, 1996). Others within medical sociology, however, have emphasised the importance of questioning assumptions and including social constructionist definitions of the body in analysis (Williams, 1996b). Some have considered whether the social model might be developed in order to examine illness (Pinder 1995), or how to develop an approach to empirical work which is neither over- nor under-socialised (Williams, 1996a).

A number of recent studies have taken a more realist approach to qualitative work, examining 'lay views', beliefs or perceptions in the context of cultural beliefs or discourses about health and illness (see Davison, Frankel and Davey Smith, 1992; Radley, 1993). Narrative studies have been seen as indicating not only different cultural perceptions of groups, but also more dominant cultural views or rhetoric in society (Williams, 1993). Analysis has included an examination of 'medical' values or ways of thinking. However, the point has been made that there is not a 'gulf' between 'medical' and 'lay' beliefs or values but interaction (Fitzpatrick, 1984; Davison *et al.*, 1992), since people may 'borrow' discourses (Radley, 1993). It has been felt that 'lay' views must be seen and analysed in a more complex way (Pierret, 1993; Pinder, 1996).

Questions raised about an appropriate approach to empirical work, such as the way that a 'group' should be defined, or the way that analysis should be carried out, have implications for the design of this study, which will be returned to later in the thesis.

Conclusion

The thrust of this chapter has been that screening programmes offering 'prevention' can be regarded as expressions of values. This chapter has outlined a critique of a 'biomedical' value-system. It has been argued that such values - which can define both the way that conditions or people with conditions may be seen in society and appropriate solutions - may be unexamined, and that there may be alternative definitions of problems. Such a critique of screening programmes has been expressed by sociologists and by disability activists. On the other hand, limits to or challenges to this view have also been suggested.

The next chapter will place these arguments in the more general debate about screening programmes. The debate is complex, and concerns the question of choice as well as 'prevention'.

CHAPTER 3

THE DEBATE: CONFLICTING VALUES

The first chapter of this thesis has outlined the development of prenatal diagnosis and genetic testing and some considerations about the appropriateness of offering reproductive screening programmes for certain conditions. The previous chapter has presented critical perspectives offered by sociologists and disability activists, who have suggested that the rationale for seeing 'prevention' as beneficial can be seen as premised on certain systems of values, in particular a 'biomedical model'. The chapter also suggested limits to this view, and the possibility of conflicting perspectives. However, the debate about screening programmes is complex and also concerns the issue of autonomy. This chapter outlines this debate.

Questions about screening programmes mainly concern the morality of action. To offer a screening programme may be seen as right or wrong for different reasons. This issue has been debated in the media, in medical and academic journals, by professionals and activists in various disciplines. One aspect of the debate is the conflict between the moral perspectives of rights and welfare. From an ethic of justice and rights, intervention in reproduction to prevent the births of individuals affected with any condition can be seen as morally wrong, as eugenics or selection. From an opposite ethic of welfare, care and responsibility, it may be felt to be right, within limits, to attempt to reduce the incidence of conditions if this can prevent harm. However, the provision of screening programmes may be justified not only on the grounds of reducing the incidence of certain conditions but also on the grounds of offering information and enhancing autonomy (Lippman, 1991a; Macintyre, 1997).

This chapter outlines these moral perspectives. Each of the three sections summarises the main arguments or rhetoric and the way that costs and benefits are perceived, but also counter-arguments and concerns, especially about appropriate limits. The chapter illustrates the tension between the different value positions. It concludes with a discussion of the relevance of these arguments to policy (and questions about whether, or for which conditions, it is right for screening programmes to be offered), to individual reproductive decision-making and to this study.

The arguments of the debate outlined in this chapter offer a framework. The data chapters will consider the extent to which those interviewed for this study, particularly women affected with genetic disorders, draw on or suggest the different values as important. The thesis will examine where there are similarities and differences in the

way that rights and welfare arguments, general and personal concerns, are balanced by different individuals or groups.

Prevention is wrong: eugenics, social values and rights

There is a general acceptance in Western society that all human beings have equal value; this might be felt to be the basis of religion and law. From a philosophical position of rights, and from concerns about social values, screening programmes aimed at reducing the incidence of certain disorders in society may be seen as offering a means of selection, or eugenics. To prevent the existence of those affected with impairments or certain characteristics might suggest that such people - who may be seen as 'different' - should not exist, that they are unwanted, or that their lives have less worth (Finger, 1984; Morris,1991; Wertz, 1992a; Kaplan, 1993). From this view, the principle of screening and 'prevention' can be seen as a wrong use of knowledge. Wolfensberger (1994: 395) has claimed that there has been a collapse of moral values in society, describing abortion on the grounds of foetal abnormality as, "societal death-making ... aided by a corrupt and intellectually dishonest bioethics culture".

From this perspective, some have argued that the existence of screening programmes can be explained in terms of societal prejudice, or premised on value-judgements about normality, cultural values of perfection, or on views about the improvement of the race (Kevles, 1985; Brodsky, 1990). Other explanations have also been suggested, such as an unwillingness to support those who might be less productive (Morris, 1991) or to implement their rights (Barile, 1990), medical assumptions about quality of life or a misguided concept of an individual 'right to fitness' (Shakespeare, 1995; Bailey, 1996).

Concerns about the eugenic implications of screening programmes have been raised particularly by some disability activists, who have argued that the logic of offering such programmes implies the avoidance of disability both in individuals and in society (Saxton, 1984; Asch, 1986; 1994). As noted already, many have suggested that disabled people are already defined as problematic or unwanted within society for a number of reasons (Finger, 1984; Morris, 1991).

From this perspective, the condition itself is irrelevant, and no new issues are raised by the introduction of genetic tests. Shakespeare (1995) has pointed out that, while genetic tests might have highlighted concerns about the conditions which can be tested for, and might have generated discussion about 'social problems' or discrimination, selection on the basis of any physical impairment is a matter of value-judgements about normality. As he has argued, "the biological determinism and eugenic implications of the Human

Genome Project are simply yesterday's bad practice, with better technology" (Shakespeare, 1995: 30).

Kevles (1985), with others, has made the point that all intervention in reproduction can be seen as eugenics, from the sterilisation of those deemed 'unfit', the directive genetic counselling of the '50s and '60s, to diagnostic testing and selective termination. Population carrier screening has been seen in a similar way (Hubbard and Wald, 1993). For example, Finger (1984) has described sickle-cell carrier screening, like sterilisation, as an attempt to achieve human perfection by technological and social manipulation. From this view, preventing the births of certain individuals can be seen as based on value-judgements, whether or not intervention is defined positively, and whether or not decisions are a matter of state policy or coercion, or of individual choice (Kevles, 1985). Such a change in policy may be seen as simply representing a 'new eugenics' (Kevles, 1985) or eugenics cast in individual terms (Hubbard and Wald, 1993).

It has been suggested that those individuals who would avoid the birth of a disabled child may have a personal 'desire for perfection' (Kaplan, 1993), and concerns about 'designer babies' have been expressed in the media (see Wilkie, 1993). However, from a sociological approach, the choices of individuals may be seen as shaped by the values and constraints of society (Rothman, 1985; 1986; Morris, 1991; Stacey, 1992). Certain choices may be normatively acceptable (Stacey, 1985; Hubbard and Wald, 1993), or can be seen as explainable in terms of the social context. Rothman (1985) has argued that children are becoming seen as products, in terms of their characteristics, describing this as a process of 'commodification' of children, and has pointed out that prenatal diagnosis offers a means for controlling the quality of a child, acceptance being conditional on quality. For Rothman, however, the concern of individuals to have a non-disabled child cannot be seen separately from the social context of individual responsibility for children and for disabled people, in which decisions about prenatal diagnosis must be taken, thus explaining the social acceptability of screening.

Concerns have been expressed not just that people might choose in ways seen as wrong, but that, although reproductive choice may appear to be a private matter, individual actions - such as a willingness to discard 'imperfections' (Richards,1989; Wertz, 1992a) - may have negative social consequences (Kevles, 1985; McLean, 1994).

The existence of screening programmes has been seen as having the potential to change social values. Rothman (1986) has suggested that the introduction of prenatal diagnosis has had an effect on the way that pregnancy is perceived, and has proposed the concept of the 'tentative pregnancy', one not discussed with others until the absence of abnormality has been confirmed. Others have suggested an increasing acceptability of

abortion on the grounds of foetal abnormality and a corresponding reluctance to accept uncertainty (Marteau, 1992), raised expectations of being able to control impairment (Bailey, 1996) or changing attitudes to normality or acceptability. Some have argued, therefore, that to offer prenatal diagnosis and abortion is to perpetuate or legitimate societal values perceived as wrong (Wolfensberger, 1994).

It has been suggested that the existence of screening programmes may also have negative social consequences for those with 'preventable' conditions. Not only might such screening be offensive (Scully, 1985; Clarke, 1991), but, if certain conditions are seen to be preventable, less money might be spent on services (Richards, 1989; Morris, 1991), and attitudes towards those affected might become more negative. Since attitudes or values can be enshrined in policy, a number of concerns relate to the potential for discrimination (Morris, 1991; Shakespeare, 1995), particularly in relation to the rights of those with impairments to parent or reproduce, and their right to life (Finger, 1984; McLean, 1994).

As discussed already, some have expressed concerns that disabled people can be treated differently from others in that parenting may be felt to be inappropriate (Finger, 1984; Saxton, 1984; Asch, 1989; Lonsdale, 1990). Although some commentators have noted a lessening of disapproval towards the rights of disabled women to parent (Lonsdale, 1990), it has also been pointed out that they may continue to be offered abortions inappropriately (Lonsdale, 1990), or be discouraged from using technology which could benefit them (Peters, 1990). It is still not unusual for women with learning disabilities to be subject to sterilisation (Walmsley, 1991), or to be given Depo-Provera with little analysis of its side-effects (Finger, 1984). Where an impairment may be inherited, doctors' reactions towards women having children have been found to be variable (Saxton, 1984; Goundry, 1990; Lonsdale, 1990), and with the introduction of genetic tests, there are concerns about the rights of those with a wider range of conditions, or carriers, to have children (McLean, 1994; Billings *et al.*, 1992).

Most arguments about the eugenic implications of screening programmes, however, relate to the equal right to life of those with conditions which can be prevented; as Morris (1991), a disabled woman, has pointed out, what is at issue is the assumption that the prevention of disability through screening is a legitimate goal. The concern of many has focused on abortion as a means of prevention.

Abortion on both social and medical grounds has been legally permitted in the UK since the introduction of the Abortion Act 1967, although there are controls over its use. Abortion on the grounds of foetal abnormality may be seen as wrong because of a general view that any abortion is murder and is morally wrong. Some anti-abortion

campaigns, particularly in the US, have linked concerns about abortion to disability rights, suggesting that the rights of affected foetuses should not be seen differently, legally or morally, from the rights of affected living people; some disability activists have taken this view (Davies, 1987). However, foetal rights may be seen in different ways; others have argued that there are differences between the rights of foetuses and those of living people, and that it is possible to support termination for foetal abnormality and equal rights for disabled people (Birth Control Trust, 1996a).

Abortion on the grounds of foetal abnormality raises different moral issues from abortion on social grounds (Asch and Fine, 1988b); as Hubbard and Wald (1993: 30) have pointed out, there is "a marked difference between a woman having an abortion because she does not want a child and having an abortion, although she wants a child, because she does not want this one". A number of disability activists have made a distinction between concerns about abortion on the grounds of foetal abnormality and a 'right to life' position. Although the clause in the Act allowing abortion on medical grounds can be seen as introducing choice or as calling into question the right to life of those with certain conditions (Nuffield Council on Bioethics, 1993; McLean, 1994), it has been pointed out that this 'eugenic clause' was relatively uncontroversial when it was introduced (Finger, 1984; Kevles, 1985; McLean, 1994). Some women with disabilities have argued that, in fact, the history of the abortion debate suggests that handicap has seemed to offer a strong argument for abortion (Finger, 1984; Morris, 1991).

The introduction of the Human Fertilisation and Embryology Act (1990) has heightened concerns about foetal rights, however. Under this Act, the time-limit for legal abortion has been reduced from 28 to 24 weeks, but can be over-ridden in cases of severe handicap; the Act allows the termination of a foetus for which this would not have been considered if it were not for disability (McLean, 1994). As Morris (1991: 75) has argued, "it is outrageous that, under legislation just passed in Britain, a foetus of more than 24 weeks gestation is treated as having rights as a human being but loses those rights once it is diagnosed as being disabled". It has been suggested that the more explicitly eugenic policy in the UK since 1990 represents a response to the increasing ability of the medical profession to detect genetic disorders (Morgan, 1992).

The philosophical basis of the eugenics argument - that 'prevention' can be equated with discrimination - has been disputed by Harris (1995), who has asserted that there is no necessary connection between discrimination against disabled people and preventing the birth of a disabled person. Making a distinction between the impairment and the person, Harris has argued that it is not morally wrong to prefer to produce, or even to

be, a non-disabled rather than a disabled person, although it is wrong to prefer a non-disabled to a disabled person.

Others have questioned the weight placed on the eugenics argument. Although some have taken an 'absolute' position that to prevent the birth of a child affected with any condition can be seen as discriminatory and as always wrong (Wolfensberger, 1994), many who have expressed concerns about the eugenic assumptions in screening programmes and abortion laws have felt that these concerns should be balanced against others. For example, Wertz and Fletcher (1993) have argued that there is a danger that valid concerns about moral judgements might be used to restrict women's choice at a time of women's changing roles. A number of disability activists have defended a woman's right to decide about termination (Finger, 1984; Saxton, 1984; Morris, 1991; Shakespeare, 1995); Asch (1986), for example, has argued that women need to live in the world as it is.

Others have discussed the difficult moral balance between concerns about the right to life and concerns about welfare, noting that preventive action can be ethically indicated if it will prevent suffering, whether for a mother or a child, and that there are difficult questions about what might constitute a 'serious' disorder (Kaplan, 1993; Bailey, 1996). Morris (1991) has also made the point there might be different views, individual or cultural, about the appropriate balance between rights and welfare.

Prevention can be morally right to avoid harm

From an ethic of welfare and care, to prevent the birth of an individual affected with certain conditions or to try to reduce the incidence of such conditions may be seen as right or morally valid, if this can prevent harm (Post, 1991; Kitcher, 1996). Some have argued that, in relation to prenatal diagnosis, rights arguments are unhelpful (Dunstan, 1988). This opposite value-system is expressed in the view of the Birth Control Trust that programmes of screening are not eugenic in intent but morally defensible; as they argue, they are "motivated by the same spirit that leads us to try to cure disease" (Birth Control Trust, 1996a).

However, from a perspective of welfare or minimising suffering or harm, arguments about right and wrong are relative. Morality may be considered in terms of costs and benefits. Benefit or harm may be experienced by individuals, by groups, by institutions in society or collectively, and the consequences of offering a screening programme or reducing the incidence of a condition might be perceived and measured in a number of ways (Wertz, 1992a; Chadwick, 1993).

Societal benefit

Reducing the incidence of certain genetic disorders might be felt to have general or collective benefit. Kitcher (1996) has pointed out the need to consider screening programmes and genetic knowledge in a global context of an increasing world population. Societal benefit can be seen in terms of improving public health but also in terms of costs (Sheldon and Simpson, 1991). Many have made the point that the increasing life-expectancy of people with long-term chronic conditions or impairments, new possibilities for treatment and care, as well as changing ethical perceptions and increasing attention to their rights, might cause financial burdens on service provision (Davison *et al.*, 1994, Bluebond-Langner, 1996). Some attempts have been made to calculate the financial implications for services of different conditions (Smith *et al.*, 1995).

Estimates have been made of the effectiveness of screening programmes in terms of the proportion of affected births prevented, but also in terms of their cost-effectiveness (Evans and Chapple, 1988; Henderson, 1987; Sheldon and Simpson, 1991; Cuckle *et al.*, 1995). Some have been justified in cost-effectiveness terms; for example, the screening programme for Down's syndrome has been described as "substantially less than the cost of lifetime care" (Wald *et al.*, 1992b: 391).

However, the inevitable dependence of those with certain conditions has been questioned (see Kaplan, 1993), and even those calculating the cost-effectiveness of screening programmes have discussed the impossibility of assessing 'value for money' (Cairns and Shackley, 1994). Cost-benefit analyses have largely been criticised as crude measures of effectiveness, ignoring ethical and personal issues, other values and measures of benefit (Clarke, 1990; Sheldon and Simpson, 1991; Elkins and Brown, 1993). Some have further argued that cost-effectiveness arguments contain assumptions about societal benefit (Clarke, 1990) and about prevention as the logical outcome (Shakespeare, 1995), and might constitute pressure on doctors to practice defensive medicine or to portray conditions negatively during pregnancy (Clarke, 1991). It has generally been argued that the criteria for evaluating collective benefit should be in terms of medical benefit, reducing suffering and ill-health (Clarke, 1990; Post, 1991).

At the same time, with conflicting health and welfare needs, and with competition for scarce resources in health care, the priority that should be given to programmes of screening and prevention has also been debated. Some have questioned whether money for screening programmes might be more appropriately spent on social interventions or

services for those affected (Wilson, 1993), or whether 'public health' goals might be achievable by other means (Kaplan, 1993). It has been argued that priorities for a population might be more basic needs, such as addressing poverty (Lippman, 1992b) or general ill-health (Henifin, 1993), and some have gone further to suggest that the availability of new reproductive technology might increase inequalities both in access to health care and in allocation of resources (Lippman, 1991a; Wertz, 1992a; Henifin, 1993).

Individual benefit

However, genetic conditions might have serious consequences for those affected and for others, especially family members. Advances in genetic knowledge have been seen as beneficial in offering hope for families of those affected with genetic disorders (McKie, 1988), and screening programmes providing the means to avoid the birth of an affected child as preventing individual harm or reducing suffering (Connor, 1989; Wald, 1994a). The UK Abortion Act (1967) is framed in terms of individual welfare, abortion on the grounds of foetal abnormality being offered in the interests of those affected with certain conditions or of other family members, especially the mother.

At one time the collective and individual benefit of avoiding the birth of an affected child were seen to coincide, and the question of 'whose benefit' was not discussed, screening programmes being described simply as 'potentially beneficial' (Chamberlain, 1978). More recently, the emphasis in the literature has been on individual benefit, genetics being described as 'personalised medicine' for example (McKie, 1988). A number of geneticists have pointed out that, although there are always concerns about the potential for eugenics in applications of genetic knowledge, the beneficiaries of new techniques and information are individuals (Weatherall, 1985; Jones, 1993; Wald, 1994a).

The interests of parents and families

From this perspective, Connor (1989) has argued that, while antenatal screening programmes may be assessed in terms of their effectiveness, the main beneficiaries are pregnant women. Screening programmes may offer information and choice, but also the means of prevention, and 'prevention' may be felt to be in the interests of parents and family members. Where a child is born with a seriously disabling or life-threatening condition, there may be emotional, psychological and economic consequences (Clarke, 1990). These may be long-term when the affected individual needs continued care, especially where child-care is a private or individual matter

(Rothman, 1985; Blackwell-Stratton, 1988; Morris 1991). Empirical research has indicated the possibility of strain.

However, studies have also indicated the importance of individual factors and strategies on perceptions (Burton 1975; Venters, 1983; Ekwo, Kim and Gosselink, 1987). Disabled people have also pointed out that assumptions of benefit for women in avoiding the birth of a disabled child, who may be seen as a 'burden', may be felt to be not only insulting to those with conditions, but also unjustified (Saxton, 1984; Bailey, 1996). They have commented that there is a focus on the condition and on the negative in the literature offered with screening programmes, and a failure to acknowledge either the positive aspects of having a child or the potential of many affected with impairments. As discussed already, it has also been pointed out that, while there may be difficulties for individuals caring for a disabled child, some problems, such as dependence or in others' attitudes, might seen as socially caused and not inevitable (Saxton, 1984; Barile, 1990; Oliver 1990; Keith 1992).

In addition, even where there might be psychological and economic benefits for parents in preventing the birth of an affected child, there may also be negative emotional and psychological consequences where a wanted pregnancy is terminated (Richards, 1989; White-Van Mourik, Connor, and Ferguson-Smith, 1992). Those taking part in screening programmes may experience "iatrogenic anxiety" (Lippman (1991a: 31) caused by the risk of amniocentesis, unwanted decision-making or the uncertainty of the test results (Richards, 1989; Green, 1990; Marteau, 1992). It has been pointed out, therefore, that the costs and benefits to individuals of preventing the birth of a child affected with a condition by prenatal diagnosis and abortion are a complex matter (Mooney and Lange, 1993).

The interests of the affected individual

It may also be felt right to prevent the births of those affected with certain conditions in their own interests, where an argument can be made that their suffering would be such that it would have been better for them not to have been born (Annas, 1994). Kitcher (1996) has suggested that such a decision may be felt to show respect for human life. The question of appropriate clinical interventions, where people are affected with conditions which cannot be cured, is fundamental to medical ethics. In the medical literature, 'prevention' by prenatal diagnosis and abortion has been seen as right for a serious condition for which treatment is ineffective (Brock, 1995), although some reservations about the means of prevention have been acknowledged; McKie (1988) has described the link between genetics and abortion as only a stage, rather than 'good

medicine'. Changes in thinking about appropriate means of prevention over time have been discussed earlier in relation to spina bifida (Laurence, 1974; Tew, 1987).

As noted earlier, it has been suggested that there have also been changes in thinking in the value attached to health, and that the current interest in genetics and 'prevention' accords with an increased importance attached to securing individual well-being at the present time (Stacey, 1992; Bailey, 1996). Some have gone further to suggest that there is currently felt to be a 'right to health' (Hubbard and Wald, 1993).

While the goals of medical practitioners may be to improve or preserve health, concerns have been expressed in the medical literature about whether or when 'prevention', rather than treatment, might be felt to be the most ethical course of action (Cairns, 1995). Concerns about the appropriateness of preventing the existence of those affected, rather than other interventions, have been heightened with new possibilities for treatment, which might include advances in foetal medicine (Bradley, 1995), treatments based on genetics, and new techniques for management, which can extend the life expectancy for those with a number of conditions (including those in this study), and which might improve quality of life. In the medical literature, it has also been noted that the outcome for those with a congenital condition might be improved with the provision of improved services as well as with treatment (Nicholson and Alberman, 1992; Clarke, 1993).

If quality of life might not be poor, then to prevent the births of those affected in their own interests would not be justified or necessary. This raises the question of how quality of life should be evaluated and who should be the judge. Some in the medical field have noted that scientists' definitions of 'quality of life' can be questioned (McKie, 1988), or that the question of whether it might have been preferable not to have been born is complex and not easily answered (Wilfond and Fost, 1990). As discussed earlier, empirical work carried out by social scientists has indicated that the consequences for an individual affected with a disorder can be seen in social as well as medical terms, that well-being or perceived 'quality of life' is a subjective matter and can be complex, and that experience can be affected by social factors as well as medical interventions.

This latter point has been emphasised by some disability activists, who have stressed that, while disabled people do face problems, important consequences of disability - such as physical barriers, a lack of welcome - should be seen as socially caused, and as potentially changeable by changes in society, whether in terms of societal support or changed attitudes (Saxton, 1984; Asch and Fine, 1988b; Morris, 1991). From this perspective, some have questioned the view that it is right and beneficial to avert the

births of those affected with an impairment in their own interests (Saxton, 1984; Bailey, 1996). They have argued that assumptions about reducing suffering can be seen as resting on a 'medical model' of living with a condition, in which, as Bailey (1996: 148) has argued, "level of function is intrinsically linked to quality of life".

Limits to the appropriateness of 'prevention'

Within a medical or 'welfare' perspective, a range of views has been expressed about the potential benefits or the appropriate use of screening programmes and genetics. A number of professionals in medical and scientific fields have expressed enthusiasm about the possibilities of genetic knowledge for reducing the incidence of disorders (McKie, 1988; Bodmer, 1990); new developments have been described as "exciting and extremely promising" (Plouffe and Donahue, 1994: 738). Medical publications have emphasised progress in genetics (British Medical Association, 1992), and attention has been drawn to the success of certain screening programmes in reducing the incidence of conditions (McKie, 1988). At the same time, the importance of fully evaluating the costs and benefits of applications of genetic knowledge, including screening programmes has been emphasised (Plouffe and Donahue, 1994).

For a number of reasons, the unquestioned benefit of screening programmes offered in reproduction 'prevention' has increasingly been debated. At one time, it was felt that the overall benefits of prenatal diagnosis for genetic abnormalities were uncontroversial, both for economic reasons and for those to whom it was offered who regarded abortion as acceptable (Chard and Macintosh, 1992). It was argued that empirical studies of uptake showed that a view of screening programmes as beneficial was shared by pregnant women (Kings Fund Forum, 1987; Connor, 1989). However, medical literature now suggests that some concerns expressed by social scientists, ethicists and disability activists about over-optimism, assumptions of benefit and coercion have been acknowledged. A generally more cautious approach has been noted over recent years towards screening programmes for genetic conditions, especially where the means of prevention is abortion (Green, 1995; Harper, 1995).

Concerns about good medical practice in relation to the morality of offering screening programmes have been heightened with advances in genetic knowledge (Cairns, 1995; Harper and Clarke, 1997). Because of the widening range of conditions for which 'prevention' may be possible, the implications for relatives and for carriers, and the limitations of genetic tests, many have expressed reservations about their appropriate clinical use (Editorial, 1996; Harrap, 1997). There have been calls for decisions to be made about the limits of conditions for which 'prevention' may be felt to be right or beneficial (Byrne, 1994).

Testing for genetic conditions is felt to have raised not only the question of what appropriate limits of testing and prevention should be, but also of who should take decisions about the criteria for offering a screening programme (Annas, 1994). As Annas has noted, historically, medical professionals have defined what should be seen as a 'serious condition' but, with critiques of 'medicalisation', this concept is increasingly being debated. One argument, which underlies this study, is that the experiences of those affected are relevant to the debate, and some have claimed that there might be 'medical' assumptions about problems, or that difficulties should not be seen as inevitable.

Another view is that ethical limits to conditions for which 'prevention' might be felt to be right should perhaps be according to what is wanted (Richards, 1989; Annas, 1994). From this perspective, Finger (1984) has argued that 'medical' discussions about the conditions for which abortion might be felt appropriate ignore the argument that women have the right to control their own bodies. However, the point has also been made that difficulties of defining appropriate limits to screening programmes have been magnified and made more complex, not only with the introduction of genetic tests, but also because they have been introduced in a climate of both diverse values and increasing autonomy (McLean, 1994).

Debates about the morality of 'prevention' have highlighted the question of why screening programmes are offered. While one reason might be that they offer a means of reducing the incidence of certain conditions, screening programmes have also been seen as potentially beneficial because they offer information and enhance individual reproductive choice. The argument for autonomy is considered next.

The value of choice

In Western society or liberal democracies there is a strong emphasis on values of individual autonomy, rights and benefit, and individual difference (McLean, 1994). In modern society, there is an increasing emphasis on this perspective, and a focus on responsibility and self-efficacy, information, choices and their outcomes. Modern society has been seen as characterised by a scepticism towards 'expert' views and assumptions of progress, but also by an increasing emphasis on emotions rather than 'rationality' (Turner, 1996), and consumption rather than production (Featherstone *et al.*, 1991; Turner, 1996). The emphasis on the individual and choice has been indicated in definitions of modern society as an 'information society' (Lyon, 1988), a 'knowledge society' (Giddens, 1991), or a 'risk society' (Beck, 1992).

Policy changes in the UK in the 1990s have reflected an increasing emphasis on the individual and on his or her needs, on individual difference, and on choice, rights and responsibility. These are the values or the social context within which new reproductive technology and new genetic tests have been introduced (Rothman, 1985; 1990; Morris, 1991; Lippman, 1992b; McLean, 1994).

Changes in the status, authority and autonomy of medical professionals, discussed earlier (Williams and Calnan, 1996; Lupton, 1997; Williams, 1998), have implications for the provision of screening programmes. Commentators have discussed a potential pressure on medical professionals to practise 'defensive medicine', or to err on the side of prevention or offering screening. Such pressure has been seen as stemming not only from an emphasis on cost-effectiveness, but also from a focus on individual health (Hubbard and Wald, 1993; Shakespeare, 1995), individual benefit, and concerns about litigation (Tymstra, 1991; Chard and Macintosh, 1992). Individual rights and choices may be backed up by legislation; in the United States, a child can sue doctors for 'wrongful life', and parents can sue for the 'wrongful birth' of a disabled child (McLean, 1994). However, screening programmes may be seen as potentially beneficial for individuals not only in offering the opportunity to avert the birth of an affected child, but in more generally offering information and reproductive choice.

It has been argued that it is right for reproductive decisions to be a matter for the individual for two reasons. First, in line with an increasing emphasis on control of one's life, and on 'women's right to choose', individuals are seen as having a right to take reproductive decisions (Warren, 1992; Bradley, 1995). They are also felt to have the right to information, both for its own sake and in order to take such decisions. As the Birth Control Trust has expressed it, "women should be able to take advantage of antenatal screening, should have access to accurate and objective information about abnormalities and should be able to end an affected pregnancy if they so wish" (Birth Control Trust 1996a). Some have argued that the rights and interests of women should take precedence over those of a foetus or child (Richards, 1989).

Secondly, it has been argued that it is right for reproductive decisions to be taken by individuals, rather than being a matter of law, because people and their circumstances differ (Wertz and Fletcher, 1993). For example, literature from the Birth Control Trust stresses that only individual women know whether they can cope practically or emotionally with the consequences of having a child affected with an abnormality (Birth Control Trust, 1996a). Individual reproductive decisions may be taken in the light of varied material circumstances, perceptions of the consequences, relationships and emotional resources, desires or priorities or personal values.

Many have argued that, in spite of concerns about social values or equal rights, and while some arguments of disabled people that problems may be socially caused may be true, nevertheless, since parents raise children and take the consequences, it is right that they should take reproductive decisions (Rothman, 1984; Wertz, 1992b; Bradley, 1995). Kaplan (1993) has noted that disability activists emphasise rights, needs and difference, and that many support the principle of autonomy.

It has also been argued that women can be trusted to make "reasonably sound choices" (Warren, 1992: 118), and that individual decision-making and consideration of the consequences might offer an ethical approach to concerns about screening programmes. Kitcher (1996) has pointed out that there is a moral spectrum of views, and there are conflicting values, regardless of new developments in genetics. For Kitcher, there are concerns about limits and about unforeseen consequences with 'idealist' positions and - since it might not be realistically possible to offer total 'solutions' - interventions may be partial and inefficient. He has argued therefore that, in spite of concerns about autonomy, a pragmatic approach might be a means of addressing the difficult ethical issues of reproductive technology.

The goals of screening programmes

However, many have noted the inherent potential for a clash of goals where screening programmes are voluntary, as in the UK (Wertz, 1992a; McLean,1994; Macintyre, 1997). In the medical literature, there has been increasing discussion not only of the fact that individual and societal benefit might not coincide, and a critique of cost-benefit and logistical approaches (Editorial, 1992), but also about whether 'autonomy' or 'prevention' should be the goal of screening programmes. As Wertz (1992a: 502) has observed, "if public health programmes in genetics are aimed at prevention, as many appear to be, they come into direct conflict with non-directiveness". Some have argued that it is necessary to clarify the goals of screening (Royal College of Physicians, 1989; Sheldon and Simpson, 1991).

The different goals of 'prevention' and 'autonomy' imply different approaches to offering a screening programme, as discussed earlier in relation to cystic fibrosis population carrier screening. If the goals of screening are seen in terms of reducing the incidence of a condition, then 'success' may be defined in terms of effectiveness and measured as the proportion of affected births prevented (Wilfond and Fost, 1990; Brock, 1995). Geneticists have stressed that a screening programme should be feasible and effective before it is right for it to be offered, pointing out that the overall effectiveness is affected by the acceptability of the means of prevention (Brock, 1995). For a screening programme to be effective in reducing the incidence of a disorder, it has

been felt important that it should reach the whole population, that the tests themselves should be effective (especially in regard to sensitivity) and that there should be a high uptake of prevention measures at each stage (Chamberlain, 1978).

At one time, since the goals of individuals taking part in screening programme were also felt to be 'prevention', it was felt appropriate only to offer amniocentesis to women who agreed beforehand to terminate an affected pregnancy (Farrant, 1985). However, the emphasis more recently has been on autonomy. Clarke (1991; 1993), among others, has argued that maximising autonomy should be the goal of screening programmes, since goals of 'prevention' may not only have the potential for eugenics, but may also be inappropriate in containing assumptions about suffering. If the enhancement of reproductive choice is the goal, then the aims can be seen as achieved when all those who want to know have been informed of their reproductive risk (Wilfond and Fost, 1990). Information might or might not be used to prevent the birth of an affected child; it may be seen as beneficial in itself and it might be felt wrong to withhold it (Raeburn, 1994; Clarke, 1995).

One concern is that individuals' decisions about family building or interventions in pregnancy should be informed ones. It has been emphasised that the information offered with screening programmes about risk, about the implications of taking part and about the condition being tested for, should be understandable and accurate. In this context, some disabled people have suggested that the consequences of living with a condition might be portrayed negatively in the material offered with screening programmes (Saxton, 1984; Morris, 1991; Williams, 1995a); they have argued for the best possible information to be offered, so that women may be offered a 'real choice' (Saxton, 1984; Asch, 1994).

On the other hand, some reservations about the benefits of information and choice have been expressed. As noted already, psychologists have pointed out that the provision of information and choice in pregnancy may or may not benefit individuals. While perceived benefits might include cognitive or psychological benefit (Tymstra, 1991; Mooney and Lange, 1993), there might also be negative emotional or psychological consequences of unwanted decision-making and anxiety (Tymstra, 1991; Marteau 1992). From these concerns, some medical professionals have expressed caution about offering information which is uncertain (Beaudet, 1990), although others have emphasised the rights of individuals to knowledge (Connor, 1993; Cuckle, 1993).

Official guidelines have emphasised the importance of informed choice and the accessibility and accuracy of the information, and have aimed to address concerns about anxiety and about coercion, stressing the voluntary nature of screening programmes and

non-directive counselling. The goals of guidelines, official reports and recommendations about prenatal diagnostic screening and testing are clearly stated in terms of autonomy and enhancing reproductive choice (Royal College of Physicians, 1989; British Medical Association, 1998).

Limits to choice

Concerns have been raised, however, about autonomy as the goal of screening or about a pragmatic approach; for example about how people might choose, the extent to which choice implies responsibility, whether what is wanted or acceptable should be an ethical basis for offering a screening programme, and whether some choices may be seen as intrinsically wrong. As McLean (1994) has argued, although liberal Western tradition emphasises individual rights and values, these interests and rights might be in conflict with the perceived 'greater good' of the community, and might have social consequences in terms of both values and service provision. Stone and Stewart (1996), from a public health perspective, have questioned the ethics of a 'paradigm shift' in which 'prevention' is not felt to be a goal of screening and information is regarded as beneficial in itself regardless of outcome. Similarly, Chadwick (1993) has suggested that autonomy should be the means rather than the goal of screening programmes, and has considered whether goals of genetic health should be more carefully explained.

Some have questioned, therefore, whether there should be limits on the provision of choice, whether screening should be withheld for trivial conditions, or to establish foetal sex for example, or whether this might be criticised as paternalistic (Tymstra, 1991). Many have expressed the need for an appropriate ethical balance between protecting and limiting autonomy; those supporting the principle of autonomy are agreed that there might need to be restrictions on choice (Chadwick, 1993; Clarke, 1993).

Some limits on choice have been suggested in a BMA publication, written to give guidance to professionals and the public. The objectives of prenatal diagnosis are stated as being, "to allow the widest possible range of informed choice to women and couples at risk of having children with genetic disorders, within the boundaries established by society" (British Medical Association, 1998: 49). From this view cultural values can provide a consensus about decisions which may be seen as inherently wrong or unacceptable. Others, however, have argued that guidelines assuming shared values both fail to analyse whether societal values might be discriminatory and lack attention to minority views (McLean, 1994).

The BMA publication has also stressed the link between choice and responsibility, increasingly emphasised politically; as the cover notes, "the book confronts the potential for conflict between individuals' choices and their moral responsibilities for other people". The guidelines suggest that choices might be limited by a sense of duty: "When contemplating a decision that has profound moral consequences, the decision-maker should take the foreseeable consequences into account. Bringing new life into existence is one of the most important acts individuals undertake and, ideally, all intending parents should think about how to ensure the health and welfare of their future child" (British Medical Association, 1998: 21). From this moral perspective, Rhodes (1998) has emphasised interpersonal responsibility and has argued that to choose not to know about genetics is not a tenable moral position. On the other hand, the concept of genetic 'responsibility' has been criticised as an 'ideal' of perfect health (Nelkin and Lindee, 1995). It has also been pointed out that such a view of 'reproductive responsibility' can be seen to suggest a lack of understanding of environmental causes of difficulties (Kitcher, 1996).

How to choose

The question of the way that people should or do take decisions about moral issues, and the factors which might influence their views, has been considered from a number of theoretical perspectives, and in theory and empirical work. As Giddens (1991) has pointed out generally, the provision of information - 'informed choice' - is of little use when decisions concern values. In relation to screening programmes and genetics, the difficult issues are felt to concern values (McLean, 1994).

From a philosophical perspective, Gilligan (1977) has suggested that women take decisions about social abortion on the basis of responsibility, both towards a child and towards themselves. Bewley (1994), however, has outlined the view that there are three moral frameworks for taking ethical decisions; duty-based, rights-based or goal-based. She has explained that context-specific value-systems can underlie the ways that people make decisions, although decision-making might be based on intuition or reason as well as on beliefs.

Sociologists have also argued that discussions about 'choice' require an examination of the cultural and material context in which such choices are offered, or in which risks or problems may be defined as such (Rothman, 1985; Lippman, 1992b; Macintyre, 1995). Within sociology, however, there are different theoretical views not only about the factors influencing choice and the extent to which there is felt to be a consensus, but also about the relationship between action and structure in modern society. Sociologists

have expressed a range of views about the extent to which choice about prenatal diagnosis may be seen as shaped by the social context.

Some have argued generally that structural sociological analyses can be seen as over-deterministic and inappropriate in post-modernity or a pluralist society, and in which there is less emphasis on ideology or experts. Suggesting the need for a more complex analysis of social action in modern society, Giddens (1991) has proposed a relationship of 'reflexivity' between individuals and institutions. For Giddens (1991: 201), people choose according to a "project of the self", according to their identity and their own values, and this individual control of the relationship with the world can be seen as substituting for morality. In relation to screening programmes, Richards (1989; 1993), drawing on anthropology, has noted the current diversity of cultural views and values. He has argued that there might be variations between populations, communities and families in perceptions about the effects of different conditions, in values about the acceptability or morality of 'prevention', and in perceptions of risk.

From other sociological perspectives, it has been argued that the work of Giddens has over-emphasised the role of agency in choice, or that control is a middle-class concept (Williams, 1995b). 'Reflexivity' has similarly been seen as a concept of the intelligentsia, or an over-optimistic view, especially in relation to new scientific developments (Lash and Wynne, 1992). A number of sociologists have argued that reproductive choice should be seen as shaped or constrained by the material or cultural context in which they are offered, and that an examination of this context can explain why people behave as they do (Davison *et al.*, 1994). Lippman (1991a) has suggested that questions should be asked about why reassurance is sought or about how risk groups are generated.

As discussed in the previous chapter, many have proposed the powerful influence of the values of 'biomedicine' on individual choice. It has been suggested that the values or goals of medical professionals might differ from those of individuals offered screening (Farrant, 1985), and that, in spite of guidelines, the information offered about conditions in pregnancy can be partial or biased, and presented as suggesting a normative view about appropriate action (Lippman, 1989; Marteau *et al.*, 1992; Harper and Clarke, 1997). Some empirical work has suggested that prenatal diagnosis has been presented in ways which encourage uptake (Marteau, Plenicar and Kidd, 1993).

Others have argued that individual decisions can be influenced by the opinions or by the status of such 'experts' (Clarke, 1991; Richards, 1993). Direct coercion may not be the issue, but the difficulty of resisting expectations or assumptions. From this view, the concept of non-directive counselling emphasised in guidelines has been seen as

unrealistic, not only because of the status of professionals, but also because of assumptions about suffering or harm contained within the existence of screening programmes and reflected in the guidelines themselves (Clarke, 1993). The implications are that screening programmes are worthwhile, that 'prevention' or selective abortion is the right or appropriate outcome or action (Finger, 1984; Farrant, 1985; Clarke, 1993), that impairment is a tragedy, and that it would be better for those affected with disorders not to be born (Saxton, 1984; Asch, 1986; Morris, 1991). It has been argued, therefore, that there might be little discussion of whether choices are appropriate or acceptable (Bailey, 1996); those offered screening programmes in the context of antenatal care might assume that they are aimed at relieving suffering (Stacey, 1985), and that taking part is in their interests or in those of their child (Farrant, 1985; Lippman, 1989; Richards and Green, 1993). As Macintyre (1997: 1099) has pointed out, "the public may assume that these tests would not be offered unless there were clear cut benefits or actions that could be taken on the basis of them".

Many have also suggested the powerful influence of social pressure or social norms of what is 'natural' or right on choice, arguing that individuals tend to choose in a conforming or socially acceptable way (Tymstra, 1991; Hubbard and Wald, 1993). It has been suggested that prenatal diagnosis and selective termination are seen within society as culturally acceptable (Rothman, 1985; Bailey, 1996). Some explanations for this acceptability have been offered in terms of societal perceptions of disabled people, who may be seen as having little value (Finger, 1984), or powerful cultural beliefs about 'normality' (Morris, 1991) or about 'problems' (Saxton, 1984). For example, Saxton has suggested that, in a culture in which health, beauty and independence are defined as important, people's fears of impairment may be important factors influencing their decisions.

The point has also been made that, since women are held responsible, within society, for the health of their children and for bringing a child into the world, there may be social pressure to decide in a medically responsible way (Rothman, 1985). As discussed earlier, decisions about prenatal diagnosis can also be seen as shaped by economic concerns, where there are material consequences of caring for a disabled child, or in a social climate of individual responsibility (Rothman, 1986; 1990; Morris, 1991). Overall (1992) has argued that women may perceive that they have few choices about abortion on the grounds of foetal abnormality; women have abortions not because they want them but because, given their social circumstances, the alternatives are worse. From a view of choice as structured, Rothman (1984) has questioned whether the provision of information and choice can be seen as giving women control.

A number of sociologists have examined the concept of 'individual choice'. Many have suggested that the currently powerful rhetoric of choice is itself a social construct, and may be seen as an unexamined value or benefit, or neutral idea, in the same way as 'risks' may be constructed as such (Scott and Williams, 1992). 'Choice' has been seen as constituted as a private matter (Davison *et al.*, 1994), and it has been pointed out that if the concept of 'choice' is perceived as unproblematic, the context or values in which choice is offered is more difficult to examine (Morris, 1991).

However, others have taken a more determinist view. Beck (1992) has suggested that, in a 'risk society', individuals have to choose, since to choose not to choose also incurs consequences or risks. Individuals must also choose with a lack of moral or expert guidance, since experts can be seen as responsible for some risks. From this perspective, the right to choose becomes a compulsion to choose (Beck, 1992) or a moral or normative obligation to know and to act (Scott and Williams, 1992). An obligation to choose removes the possibility of fate, uncertainty or chance (Post, 1991; Davison *et al.*, 1994). From a view that needs and demands can be seen as constructed, as can social consequences of choice such as disadvantage or blame, a rhetoric of choice has been seen as legitimating expert or existing discourses about how to choose (Lippman, 1991a; Hubbard and Wald, 1993; Koch and Stemerding, 1994). For Beck (1992), a rhetoric of choice implies individualism; individual risk and responsibility. From this determinist perspective, there are consequences for individuals in terms of social control, but also for society in that other social values might be lost.

Some have argued that concerns about the language of 'choice' becoming a compulsion to choose, and about the dominance of a risk-assessment approach or scientific values of rationality at the expense of other values, are particularly relevant in relation to screening programmes and new developments in genetics (Wilson, 1993; Davison *et al.*, 1994). It has been felt that the choice not to know, or not to choose or to control the quality - to accept the child as it is - may be lost (Finger, 1984; Rothman 1984). As discussed already, where health and inheritance is seen as a matter of moral obligation or individual responsibility, the apparent freedom to take decisions about genetics has been seen as illusory, because of the need to endure social, political and economic costs (Rothman, 1985; Schwartz, 1997; Williams, 1998). From this view, individuals may be seen as blameworthy for not taking part in screening programmes defined as beneficial (Clarke, 1990; Tymstra, 1991; Lippman, 1992b; Macintyre, 1997), or for not 'preventing' conditions which are 'preventable'. Lippman (1991a; 1992b) has argued that the concept of control can be seen as a marketing strategy to make prenatal diagnosis attractive; rather than offering control, genetics might be the instrument by which individuals become controlled.

Discussion

This chapter has outlined the conflicting arguments in the debate about the morality of screening programmes. From one view, the provision of screening programmes may be seen as offering selection, as suggesting that some have fewer rights to be born, and hence as discriminatory or wrong. On the other hand, 'prevention' by this means may felt to be beneficial, whether in the interests of the affected person or others. The provision of choice about reproductive decision-making may also be felt to be right, although there are concerns about limits both to autonomy and to the tests which are offered.

These different arguments or value-positions may need to be taken into account in policy decisions about whether or not screening programmes should be offered, to whom or for which conditions, and in considering why they are offered. Policy may depend on the values within a society (Topliss, 1982; Oliver, 1990; McLean, 1994), on the way that society is organised, what costs society can bear, what counts as progress, and what can be left to individuals (Tymstra, 1991). However, systems of moral values, beliefs about right and wrong, the way that 'rights' and 'welfare' might be defined, ethical beliefs about the balance between individual and collective good or harm, views about the conditions which it is felt right to prevent may vary between cultures and over time (Morris, 1991; Bewley, 1994). For example, sex selection of children by testing and selective abortion is a value position which may be acceptable in some cultures but not others.

In a pluralist society, different cultural views about autonomy and values of rights and welfare co-exist within the same society (Wertz, 1992b). Communities, cultural groups, families or individuals might have divergent views about rights and welfare, how these may be defined and balanced, or whose rights or welfare should predominate (Morris, 1991). This chapter has illustrated the conflicting and often incompatible approaches towards the morality of offering screening programmes in the UK. There is a complexity of views. As Morris (1991) has pointed out, while screening programmes have been seen as discriminatory and perhaps unnecessary by some in the disability rights movement, they might be seen by other individuals or groups as beneficial; it might be seen as discriminatory not to try to prevent sickle-cell disease for example.

Different 'cultural views' - whether those of medical professionals, disability activists, communities or families - about whether 'prevention' might be felt to be right or wrong can be seen as deriving not only from values and philosophies, but also from knowledge, experience and perceptions of the consequences (Richards, 1989). As

Richards has pointed out, those closest to conditions may have different views from others about whether they might see 'prevention' as right, beneficial or justified, or as unnecessary and perhaps wrong.

The conflicting values outlined in this chapter may cause ethical dilemmas not only for policy-makers, but also for individuals in taking reproductive decisions. Individuals might need to balance conflicting values, general views and more personal feelings, concerns and perceptions of costs and benefits, and they might make decisions within different social contexts. Tensions between value-positions might be resolved in different ways by different individuals.

Because of the conflicting theoretical views about screening programmes and because of the widening range of genetic conditions for which 'prevention' might be possible, there has been increasing interest in finding out the views and perceptions of the public and of groups within the public (Macintyre, 1995). There have been calls for empirical research and analysis in order to find out whether academic views and concerns are shared or not, and whether there are alternative views or other concerns (Richards, 1993; Davison *et al.*, 1994; Macintyre, 1995). This study is part of this initiative. The findings from existing studies are outlined in the following chapter.

CHAPTER 4

EXISTING EMPIRICAL WORK

New developments in reproductive technology and genetics have been the subject of empirical research. Much of this has examined the 'acceptability' of antenatal screening programmes, whether by looking at general views or personal use. Statistical data has indicated the uptake of different stages of screening programmes, as outlined in Chapter 1. However, although geneticists have claimed that the uptake of screening programmes indicates their acceptability, others have argued that this cannot be assumed because of concerns about pressure to take part, and because of a lack of knowledge of the experiences of those undergoing screening (Green, 1990). Social scientists have increasingly been involved in research in this area, and both quantitative and qualitative studies have examined people's views about screening programmes and their reasons for taking part.

There have been changes in the focus of research over time. While early work examined the experiences, decision-making, and sometimes the views of pregnant women, the scope of recent research has broadened to include the attitudes of others, both 'professional' and 'lay'. There has also been increasing interest in the way that new developments in technology and genetics have been presented, both in the media and to individuals.

This chapter outlines the main findings from research which has examined people's views towards screening programmes; first, studies examining general views about the provision of screening programmes and about appropriate limits, and secondly those which have looked at individuals' actual or predicted use of such screening (usually prenatal diagnosis), and the factors felt to be important in decision-making. Familiarity with a condition has been suggested as an important influence on the uptake of screening programmes, and a third section reports on research which has investigated the attitudes of relatives. Most research in this area has been carried out from a psychological perspective, using quantitative methods, but the contributions of a qualitative approach are noted. The chapter concludes by summarising the reasons for the current study; while existing empirical work has examined the attitudes towards screening programmes of various groups within the population, those affected with 'preventable' conditions - those for which screening programmes are offered - have rarely been asked for their views.

The provision of screening programmes

While some have criticised the principle of offering screening programmes, there has also been concern about the widening range of genetic conditions for which 'prevention' might be technically feasible. A number of studies, therefore, have examined individuals' attitudes towards the development and provision of screening programmes, and their views about appropriate limits.

Empirical work has overwhelmingly indicated the approval of a number of groups within the general public towards the provision of screening programmes. Studies of the views of pregnant women have found that respondents are positive about prenatal diagnosis (Bennett *et al.*, 1980; Farrant, 1985; Faden *et al.*, 1987; Wertz and Fletcher, 1993), although they might not necessarily anticipate undergoing screening themselves. Similar views have been expressed by other members of the public (Watson *et al.*, 1991a; Marteau *et al.*, 1992; Evers-Kiebooms *et al.*, 1993). For example, in a telephone survey, Singer (1991) found that, although information about technology was not well known, respondents' attitudes towards prenatal diagnosis and genetic testing were overwhelmingly favourable, perceiving such tests as doing more good than harm. As noted in Chapter 1, research has indicated the support of parents (Wertz *et al.*, 1991; Conway *et al.*, 1994) and relatives (Watson *et al.*, 1991a) of a child affected with cystic fibrosis for the development and availability of carrier screening for this condition.

Approval of the provision of a screening programme might indicate that it is seen as beneficial to try to avert the birth of an individual affected with the given condition. However, such a view might also suggest that it is felt right for information or 'the right to choose' to be offered. Where screening programmes are offered in pregnancy, the only means of 'prevention' is abortion, and research has examined views towards abortion, or abortion on the grounds of foetal abnormality.

Attitudes towards abortion may vary between individuals but also between countries, and there are changes over time in the extent to which abortion is seen as culturally acceptable. In the UK, research carried out by the Birth Control Trust and by Social Trends has suggested an increase in support for legal abortion over recent years; a longitudinal study, carried out by MORI, has found that 64% of people now agree with the statement that "abortion should be made legally available for all who want it" (Birth Control Trust, 1997).

General and personal views about the acceptability of abortion may depend on the reason for abortion. Studies of the views of the public and of pregnant women have indicated that foetal abnormality is felt to be an acceptable reason for abortion, whether

or not respondents might consider this themselves (Faden *et al.*, 1987; Green, Snowdon and Statham, 1993a), although less acceptable than maternal health or rape (Green *et al.*, 1993a; Birth Control Trust, 1997). Commentators on the MORI poll have drawn attention to changes over time in attitudes towards abortion on the grounds of foetal abnormality, and to variations between groups. As they note, "there has been a significant increase in the proportion of those who disapprove of abortion on grounds of physical and mental disability. ... Those under the age of 25 were far more likely to disapprove of abortion when the child would be mentally or physically handicapped than other age groups" (Birth Control Trust, 1997).

A number of studies have examined the views of medical professionals towards screening in reproduction, since their attitudes might affect its availability and use (Lippman-Hand and Cohen, 1980; Drake, Reid and Marteau, 1996). Research has suggested that the provision of screening programmes is generally supported by medical professionals (Farrant, 1985; Boulton and Williamson, 1995; Kerr *et al.*, 1998c), although there have been changes over time. In one early study based on quantitative and qualitative methods, Farrant (1985) reported being struck by the enthusiasm of consultant obstetricians for prenatal diagnosis, and by their optimism about its potential for reducing the incidence of the conditions for which it was offered. The large majority saw the benefits of screening as outweighing the costs; benefits were seen as accruing to mothers in avoiding the birth of a handicapped child, but also to society. A comparable survey in 1993 showed the same emphasis on identification of abnormality as the reason for screening, although an increasing focus on individual choice (Green, 1995).

In a recent qualitative study, Kerr and colleagues found that scientists and clinicians were concerned to make a distinction between new genetics and eugenics, and that they refuted concerns about eugenics in a number of ways. New genetics was perceived as scientific, neutral and associated with diagnosis and treatment, in contrast to 'unscientific' or politically motivated discrimination. Actions affecting individuals were seen as different from attempts to affect society or the gene pool. Many also drew on arguments of the 'right to choose'; this was seen not only as an important value but as a safeguard against abuse or the wrong use of knowledge (Kerr *et al.*, 1998c).

One study comparing medical and 'lay' views towards the acceptability of screening programmes found that professionals were more positive than the general public (Michie *et al.*, 1995). However, new developments in genetics evoked both positive and negative feelings in all groups. All respondents felt that some blame attached to mothers who declined screening and gave birth to an affected child, and all endorsed the use of prenatal diagnosis and termination for serious conditions (Marteau and

Drake, 1995). On the question of who should decide which screening programmes should be made available, there were differences in views between medical and lay respondents; while 78% of professionals felt that decisions should be taken by an advisory group, 54% of the public felt that professionals should not be involved (Michie *et al.*, 1995).

A number of quantitative studies have investigated views about the limits to conditions for which it might be felt right for a screening programme (usually in pregnancy) to be offered. Singer (1991) found a lack of approval among the public for screening for foetal sex. Research examining the opinions of medical professionals, both within the UK (Marteau and Drake, 1995) and cross-culturally (Fletcher and Wertz, 1987; Shiloh, 1996), has indicated a lack of consensus about conditions felt serious enough to warrant termination, and one study has found very varied views about the appropriateness of abortion for cystic fibrosis (Green, 1995).

In a qualitative study examining the attitudes of the public towards genetics, a concern raised in all focus groups was 'where to draw the line', both in relation to limits to the conditions for which tests should be available and limits to autonomy (Kerr, Cunningham-Burley and Amos, 1998b). The authors found that further discussion did not resolve these questions, but rather highlighted the difficulties of drawing clear boundaries, and the reasons why this was the case. A clear distinction could not be drawn between 'medical' and 'non-medical' conditions, and 'quality of life' was felt to be problematic as a criterion for taking decisions about prevention, since it was seen to be affected by social processes.

With concerns about the introduction of screening programmes for conditions which are unfamiliar (Richards, 1993), some surveys of those offered screening have included questions about respondents' knowledge of the disorder, and other research has examined the knowledge and perceptions of the public. One study looking at people's understanding (including the risk of inheritance) of five disorders, including Down's syndrome, spina bifida and cystic fibrosis, found that most respondents were more familiar with Down's syndrome than with the others. The authors noted a correlation between knowledge and educational level, experience of working with ill or handicapped people in a professional context, and age, younger people being better informed than older ones (Decruyenaere, Evers-Kiebooms and Van den Burghe, 1992b). Several studies examining people's attitudes towards the provision of cystic fibrosis carrier screening have assessed respondents' knowledge. This research has suggested that knowledge about the disorder among the public is low (Williamson *et al.*, 1989; Cobb *et al.*, 1991; Watson *et al.*, 1991a), although most felt that it was right for screening to be offered. Another study of general practitioners found that, while the

majority supported carrier screening, many had a poor understanding of the genetics involved (Boulton and Williamson, 1995).

To summarise, empirical work examining the views of a number of groups has indicated the general acceptability of screening programmes, although suggesting little about appropriate limits. However, a view that it is right to make such programmes available might differ from a view that it is appropriate to undergo screening oneself to avoid giving birth to an affected child. Another approach to the limits of acceptability, therefore, has been to examine which screening programmes people use in practice, but it has been pointed out that statistics showing the uptake of screening programmes often do not distinguish between those not offered screening and those refusing (Nicholson and Alberman, 1992), that an epidemiological approach cannot examine the reasons for people's actions (Stone *et al.*, 1992), and that such studies cannot examine the possibility of pressure (Wertz and Fletcher, 1993). Social scientists have increasingly been involved in research. While early work examined the experiences and the views of those offered screening, more recently the focus has been on the views and the predicted personal use of screening programmes of other groups within the public.

Personal use of screening programmes: real and hypothetical decisions

With the introduction of prenatal diagnosis, a number of small-scale studies, mostly American, examined the uptake of amniocentesis, women's experiences of prenatal diagnosis and their reported reasons for taking part. Some early work indicated that the risk of miscarriage and the possible need to take a decision about abortion caused anxiety (Beeson and Golbus, 1979; Verjaal, Leschot and Treffers, 1982). With additional concerns about the uncertainty of blood screening test results, and about extending testing to those who had no reason to suspect risk, several studies have found raised anxiety after abnormal AFP results (Earle, 1981; Fearn *et al.*, 1982; Robinson, Hibbard and Laurence, 1984; Burton *et al.*, 1985).

Although there has been a limited contribution from a qualitative approach, a few studies have examined the experiences and the views of women taking part in prenatal diagnosis. The work of Farrant (1985) and Rothman (1986) suggested women's ambivalence about screening; Rothman found that respondents reported both positive and negative feelings about taking part in amniocentesis, and Farrant noted that, while holding generally positive views towards the concept of screening, some women reported negative experiences. She also found that those women who were identified by a screening programme as at low risk of having a child affected by Down's syndrome and neural tube defects found the experience much more distressing than

those who had realised they were at high risk before they were pregnant (Farrant, 1980).

As Green (1990) has noted, findings from this empirical work illustrated the stress caused by having doubt cast on the pregnancy, but also indicated some differences between women in anxiety (Robinson, Tennes and Robinson, 1975), in their attitudes towards prenatal diagnosis (Beeson and Golbus, 1979; Marteau *et al.*, 1989), and in the perceptions, beliefs and attitudes which might influence decision-making.

Following this early work, although some research has continued to examine the views of those offered screening, it has been argued that questions investigating acceptable limits to the use of screening might more appropriately be asked where respondents are under less pressure and might judge more freely (Evers-Kiebooms *et al.*, 1993), and the views of non-pregnant individuals have increasingly been sought. A number of studies have offered scenarios in terms of risk and the consequences of a range of conditions, and have asked more hypothetical questions about individuals' anticipated use of screening, diagnostic testing and abortion. However, the point has been made that such studies cannot predict behaviour (Williamson *et al.*, 1989, Macintyre, 1995).

Numerous studies have indicated that a higher proportion of respondents approve of the availability of screening programmes than anticipate taking part in screening, diagnostic testing or abortion (Ten Kate and Tijmstra, 1989; Tymstra *et al.*, 1991; Decruyenaere *et al.*, 1992a; Evers-Kiebooms *et al.*, 1993). For example, one study of pregnant women found that 98% thought that CF carrier screening should be offered, 69% felt that they would be willing to undergo screening and 29% thought that they might terminate a pregnancy (Botkin and Alemagno, 1992).

One focus of research has been on the factors felt important in decisions - both real and hypothetical - to take part in or refuse screening programmes [see Shiloh (1996) for a review]. Empirical work has shown that a consistent proportion of pregnant women decline amniocentesis because of their opposition to abortion (Davies and Doran, 1982; Berne-Fromell, Josefson and Kjessler, 1984; Knott, Penketh and Lucas, 1986; Marteau *et al.*, 1989), or refuse cystic fibrosis carrier screening for the same reason (Mennie *et al.*, 1993b). Studies of the attitudes of the public have similarly suggested that abortion on the grounds of any foetal abnormality would be unacceptable to a proportion of respondents (Bundey, 1978; Evers-Kiebooms *et al.*, 1993). Commentators have noted that values about abortion may be firmly held, outweighing other considerations (Evers-Kiebooms *et al.*, 1993).

Some of those reporting empirical work have suggested a correlation between a view of invasive testing or abortion on the grounds of foetal abnormality as unacceptable with religious beliefs (Bundey, 1978; Knott *et al.*, 1986; Evers-Kiebooms *et al.*, 1993). Others, however, have found no such relationship (Parsons, 1990), and it has been argued that opposition to abortion on the grounds of foetal abnormality is a complex issue, with other factors besides religiosity being important (Press and Browner, 1997).

While the main reported reason for refusing diagnostic testing is the unacceptability of abortion (Marteau *et al.*, 1989), research has indicated that another important influence on decisions to decline amniocentesis is the strength of a personal desire for the child and the perceived risk of miscarriage (Marteau *et al.*, 1989), the 'wantedness' of the pregnancy (Wertz, 1992b) or 'inner motivation' (d'Amico *et al.*, 1992).

Statistics have shown that a high proportion of pregnant women take the routine screening tests that are offered (Green *et al.*, 1993a; Pryde *et al.*, 1993), in some cases despite a lack of understanding about risk or about the tests themselves (Green, Statham and Snowdon, 1993b; Pryde *et al.*, 1993; Smith *et al.*, 1994). Farrant (1985) concluded that women offered screening programmes saw them as beneficial, not only because of the possibility of terminating an affected pregnancy, but also because of the reassurance of a negative result. Reasons given for undergoing screening tests have been 'because it was offered' and for reassurance (Green *et al.*, 1993a; Roelofsen, Kamerbeek and Tymstra, 1993), and some commentators have suggested that the high figures of those taking part or anticipating taking part in screening programmes confirm Farrant's hypothesis (Evers-Kiebooms *et al.*, 1993; Roelofsen *et al.*, 1993).

Others, however, have suggested that prenatal diagnosis is perceived as more generally providing information (Green *et al.*, 1993a; Pryde *et al.*, 1993); Pryde and colleagues have pointed out that an assumption that a positive diagnostic test would be followed by abortion is now seen as unacceptable. A number of recent studies, whether of pregnant women or of others in the population (Singer, 1991; Evers-Kiebooms *et al.*, 1993) have found that a high proportion of respondents anticipate taking part in antenatal screening programmes without necessarily anticipating terminating a pregnancy (Green *et al.*, 1993a; Richards and Green, 1993). As Green and colleagues (1993a: 31) have noted, "While a strong chance of handicap was seen by the majority as an acceptable reason for abortion, a third of the sample said that they would not themselves consider it. This attitude did not appear to mean that they wished to be excluded from routine screening programmes."

Surveys examining views towards screening for genetic conditions more generally have similarly indicated an interest in finding out information; the majority of the public in

one study expressed an interest in learning their cystic fibrosis carrier status (Williamson *et al.*, 1989). Singer (1991) has suggested that such information is perceived by respondents as valuable, whether or not a 'condition' is felt to be serious, and whether or not people anticipate acting on the information.

While empirical work has indicated individual differences in motivations and values, research has also suggested that important factors in actual or anticipated decisions to participate in screening programmes, and especially in decisions about termination, are the perceived high risk of having an affected child (Faden *et al.*, 1987, Marteau *et al.*, 1991), and the perceived severity of the condition and its implications (Evers-Kiebooms *et al.*, 1993; Julien-Raynier *et al.*, 1993; Pryde *et al.*, 1993; Wertz and Fletcher, 1993). For example, the majority of respondents in one study thought that they would have amniocentesis for a 1% risk of trisomy (Julien-Raynier *et al.*, 1993). Although little has been suggested about 'absolute' limits to the conditions for which screening should be offered, studies examining the uptake or anticipated use of abortion have indicated agreement across countries about the 'relative severity' of conditions (Marteau *et al.*, 1992; Wertz, 1992b), with little interest in screening for conditions or characteristics not considered diseases (Michie *et al.*, 1995). Commentators have noted that an important factor in seeing abortion as acceptable appears to be the extent to which a disorder is perceived as disabling (Pryde *et al.*, 1993) or burdensome (Evers-Kiebooms *et al.*, 1993).

A number of studies have indicated that perceptions of risks and implications might be more important than 'objective' information in decision-making (Wertz, Sorenson and Heeren, 1984; Ekwo *et al.*, 1987; Green, 1990), and that the extent to which information or counselling resolves uncertainty or facilitates decision-making is unclear. Ekwo and colleagues found that women who accepted amniocentesis were more likely than those who did not to perceive bringing up a disabled child as burdensome. Perceptions of burden were affected by a number of factors, not only the consequences of the condition (whether a short life-expectancy, physical or mental impairment) and women's understandings of this, but also their 'susceptibility' or perceptions of being able to cope (Ekwo *et al.*, 1987). Perceptions may be influenced by both individual and cultural factors (Howard and Headings, 1991), including culturally specific meanings of illness and attitudes towards coping with a disorder. Shiloh and Berkenstadt (1992) found that counsellees' perceptions both of the severity of genetic disorders and of the risks of recurrence varied according to their background, and were influenced by personal factors such as having an affected child and the motivation for having more children.

To summarise, research examining people's use or anticipated use of screening programmes has indicated that a number of factors, individual and cultural, might shape decision-making, and that perceptions of risk and of the disorder may be important influences on their behaviour. Commentators have pointed out the difficulties in making comparisons because of differences in the way that survey questions can be interpreted (Shiloh, 1996), and because studies have examined both real and hypothetical decision-making (Wertz, 1992b; Shiloh, 1996).

Most studies of those taking part in screening programmes have examined the views and behaviour of those who do not have an affected child (Wertz, 1992b). However, many of those reporting findings have drawn attention to the fact that perceptions of being in a 'high risk' group seem to have an effect on both anxiety and decision-making (Farrant, 1980; Green *et al.*, 1993a; Julien-Raynier *et al.*, 1993; Pryde *et al.*, 1993). It has been felt that more needs to be known about the views of different cultural groups, and especially of the relatives of those affected (Pryde *et al.*, 1993, Richards, 1993).

Familiarity with the condition: the decisions and views of relatives

The views about reproductive screening programmes of those who are more familiar with a disorder might differ from those of others in the population for a number of reasons. From their experience, they might have greater knowledge of the condition and different perceptions of the consequences from others. Screening programmes might also have very different meanings for individuals or couples who know that they are at risk, and who might want to decide whether to risk having an affected child, and whether to consider means of prevention, such as prenatal diagnosis and termination (Wertz, 1992b; Pryde *et al.*, 1993; Richards and Green, 1993; Shiloh, 1996). Commentators have suggested that the reproductive decisions of family members might be influenced by a number of factors, such as guilt towards an existing affected child, the optimism needed to live with the condition, the perceived views of medical professionals (Wertz, 1992b), or attitudes within families or among communities about an appropriate course of action.

Empirical work has examined relatives' perceptions of risk (Parsons and Atkinson, 1992), and their knowledge of the condition and its inheritance (Watson *et al.*, 1991a; Denayer *et al.*, 1992); one comparative study found that relatives had a clearer understanding than others in the population about the implications of the condition (Beeson and Golbus, 1985). As outlined in Chapter 1, several studies have investigated the effect of having an affected child on decisions about family building (Woodburn, 1972; Kabak *et al.*, 1984; Evers-Kiebooms *et al.*, 1988). Commentators have noted that significant numbers of parents of children with certain conditions, such as Down's

syndrome or cystic fibrosis, refrain from further pregnancy (d'Amico *et al.*, 1992; Pryde *et al.*, 1993).

With the development of DNA tests for cystic fibrosis, a number of studies have examined the predicted use of prenatal diagnostic testing and abortion by parents of an affected child (Al-Jader *et al.*, 1990; Wertz *et al.*, 1991; Wertz *et al.*, 1992b). [Wertz (1992b) has noted that prenatal diagnosis was not the preferred approach of many parents in one study who wanted to avoid having an affected child.] Empirical work has indicated a range of views about both the appropriateness of avoiding the birth of a child affected with this disorder, and about the use of screening programmes.

More generally, some research has suggested that the willingness of couples at risk of a genetic disorder to consider testing is influenced by experience, by perceptions of whether or not a disorder is limiting in relation to family life, and by a 'world view' about the acceptability of this means of family planning, but not by factual information about the disorder, understanding of genetic risk or psychological profile (Woodman, 1989). Drawing on a number of studies, Wertz (1992b) has concluded that an important factor influencing relatives' views about their reproductive decisions was a perception of quality of life - a complex balance of the perceived lives of the parents, the child and others.

However, although one recent study has shown that most parents of a child affected with cystic fibrosis accept prenatal diagnosis in subsequent pregnancies (Lane *et al.*, 1997), in general, little is known about the use that relatives make of carrier screening and prenatal diagnosis (Marteau and Anionwu, 1996). Pilot studies of CF carrier screening have indicated some differences in response between relatives and others in the population in the uptake of carrier screening outside antenatal clinics (Super *et al.*, 1992) and in anxiety (Bekker *et al.*, 1994), but there is no evidence that families of those affected with genetic conditions are more likely than others in the population to avoid the birth of an affected child (Macintyre, 1997).

A few studies have compared the attitudes towards, and the predicted personal use of screening programmes, of relatives of those affected with CF and others in the population. Although some commentators have concluded that familiarity with a condition might have a powerful effect on the use made of screening programmes (Watson *et al.*, 1991a; Evers-Kiebooms *et al.*, 1993), Wertz (1992b) has argued that such direct comparisons are difficult to make for a number of reasons, such as the different questions asked and the differences in study populations. Reviewing a number of studies, she has concluded that family members might be more ambivalent

towards abortion on the grounds of foetal abnormality than others with less knowledge of the disorders, and that research findings suggest a complex picture.

In relation to the provision of screening programmes, most parents of an affected child have supported others' rights to terminate a pregnancy, even for the condition which affects their own child (Watson *et al.*, 1991a; Wertz, 1992b; Conway *et al.*, 1994). These findings tend to confirm the conclusions of an earlier study which found that the attitudes of mothers of children with a congenital abnormality towards abortion on the grounds of foetal abnormality were indistinguishable from those of other women, being related to attitudes towards abortion more generally (Breslau, 1987).

Although the views towards screening programmes of those affected themselves have been little sought, a few recent studies have examined the views of adults living with certain congenital conditions. One study found that adults with cystic fibrosis approved of the development and provision of carrier screening irrespective of their own experience (Conway *et al.*, 1994); in another small qualitative study, young adults with this disorder were cautious about population carrier screening, although respecting others' rights to make their own choices (Alderson, 1998).

Quantitative or qualitative methods

Most research examining attitudes towards screening programmes and reproductive decision-making has been carried out from a psychological perspective using quantitative methods (Lippman, 1992a; Shiloh, 1996). Apart from the early work of Farrant (1985) and Rothman (1986), and the more recent work of Kerr and colleagues (Kerr *et al.*, 1998a; b), there has been little qualitative work in this area.

Some of those commenting on or reporting empirical work have suggested the strengths of a qualitative approach. The topic of screening programmes is complex, involving general and personal issues, and is concerned with values. It has been pointed out that slight differences in survey questions can lead to difficulties in interpreting the findings (Shiloh, 1996), and that quantitative methods are unable to examine the process of decision-making (Beeson and Golbus, 1985), the possibility of social pressure on views (Wertz and Fletcher, 1993), or uncertainty. As Kerr and colleagues have put it, "at best (surveys) suppress ambiguity and, at worst, manufacture consensus (Kerr *et al.*, 1998b: 130). Sociologists have argued for qualitative studies, in order to discover the reasons for people's views or what is important to respondents, rather than assuming the neutrality of prenatal diagnosis (Lippman, 1992a) or reinforcing dichotomies (Stacey, 1992; Kerr *et al.*, 1998b). A qualitative approach has been seen as useful in examining societal meaning (Davison *et al.*, 1994), or differences in cultural values or views

(Richards, 1993). Lippman (1992a; b) has advocated an approach to empirical work which can investigate the possibility of social and medical pressure, or values which might otherwise be taken for granted.

Studies taking a qualitative approach have highlighted differences in values between professional and lay respondents (Farrant, 1985; Kerr *et al.*, 1998a). Qualitative research has also indicated people's ambivalence and uncertainty towards screening programmes, contradictory values or views, and some difficulties in taking decisions (Rothman, 1986; Tymstra *et al.*, 1991; Kerr *et al.*, 1998b). One study looking at decision-making found that parents offered counselling focused on the implications of risk, and that they resolved these risks into binary form. The authors have suggested that parents need to resolve uncertainty about the meaning of having an affected child, their ability to fulfil their role as parents, how to choose and how others might see their choice (Lippman-Hand and Fraser, 1979). Other qualitative work has similarly suggested the awareness of lay respondents of the role of social pressure on choices (Sjögren and Uddenberg 1987, Wertz, 1992b; Kerr *et al.*, 1998b).

Discussion

Empirical research has indicated the general acceptability of screening programmes, although suggesting little about appropriate limits. A number of studies have found that a higher proportion of respondents anticipate taking part in screening programmes than anticipate terminating a pregnancy on the grounds of abnormality, and it has been noted that attitudes towards abortion on these grounds may be changing. Quantitative work has suggested that important factors influencing the uptake of screening programmes are views about the acceptability of abortion, a (less strong) desire for the child, the perceived risk and the perceived burden of the disorder; familiarity with a condition and perceptions of being at 'high risk' may also shape decision-making. Studies taking a qualitative approach have illustrated some differences in values towards screening programmes, and have suggested internal contradictions or ambivalence.

However, while the attitudes towards screening programmes of pregnant women, other members of the public, medical professionals, and relatives of those affected, have been investigated by research, at the time of designing this study, the views of those affected with conditions which could be tested for and prevented had been little sought. There have been calls for studies examining their opinions (Kaplan, 1993; Asch, 1994). Asch has pointed out that existing empirical work has concentrated on the views of parents, and has stressed that researchers and policy makers should "look to the views of people with disabilities for their contribution" (Asch, 1994: 15). The attitudes of this group

can be seen as crucial to the debate, and their opinions, knowledge and perceptions might differ from those of others, both professional and lay.

First, those affected might have different feelings or beliefs from others about the morality of the principle of screening, or abortion on the grounds of foetal abnormality. The critique of screening programmes as discrimininatory by some disability activists contrasts with the general approval of such screening found in existing empirical work. However, a number of disabled people, especially women, have defended the rights of individuals to take reproductive decisions (Morris, 1991; Asch, 1994; Bailey, 1996).

Secondly, with concerns about the limits of conditions for which screening and 'prevention' might be felt appropriate, the grounds on which a condition might be deemed 'serious', and about who should decide such limits, it has been felt essential to find out the views of those who are affected themselves. As King (1996: 9) has argued, "Let those who actually have the condition decide. At present we rarely hear their voices; instead we hear the views of parents and doctors, speaking on their behalf". Those living with a disorder have first-hand experience of the consequences, and are the only ones able to assess their own quality of life. From their subjective perceptions and knowledge, those affected might have different views from others about whether or not it was justified or beneficial to try to avoid the birth of a child affected with the same condition or impairment as themselves.

Some disabled people have argued, from the social model of disability, that while disabled people do face problems, important difficulties can be seen as socially caused and hence addressable by social change. From this perspective, screening programmes have been seen as resting on assumptions within a 'medical model' that living with an impairment is inevitably problematic. However, it has also been noted that the social model might not reflect the experiences of all individuals living with an impairment, that little is known of the quality of life of many who would identify as disabled (Kaplan, 1993), and that this model might be inadequate for understanding the experience of living with chronic illness. While some disability activists have been critical of screening programmes, therefore, the point has been made that their views might not be shared by all disabled people (Wertz and Fletcher, 1993), or by those living with different disorders or different types of disorder.

The first four chapters of this thesis have outlined the background to the study, and the arguments for research examining the views towards screening programmes of those affected themselves. The next chapter outlines the aims and objectives of this study, and describes the process of research.

CHAPTER 5

THE RESEARCH METHODS

This chapter outlines the aims and objectives of the study and the research design, and describes the stages of carrying out the research, analysis and reporting. The chapter focuses on some of the philosophical, practical and ethical considerations which affected the design of the study, especially in relation to access to respondents and to the analysis of the data. It includes a reflexive account of my role and perspective in the research process.

The purpose of the study

Although there had been debate in the media and in medical journals about the implications of screening programmes and new developments in genetics, and although empirical work had looked at the views of various groups, the perceptions and opinions of those affected themselves had been little studied. There had been calls for research examining the views of this group.

General and personal views towards screening programmes might be shaped by a number of factors, such as the arguments expressed in the media, cultural values in families or communities or within society more generally, individual feelings about having children, and by knowledge, perceptions and experience. While individual views might differ, the interest of sociology is in what is common, the extent to which values and views, knowledge and perceptions are shared within certain groups.

Those who are living with a condition which might be 'prevented' by the use of a reproductive screening programme can be seen as in a similar position. Their views towards such screening, or their perspective, might be influenced by the fact of being affected themselves. Hence they might have different beliefs or feelings from others about the morality of the principle of screening, or the acceptability of abortion on the grounds of foetal abnormality. Also, since those living with a disorder have first hand experience of the consequences, in the sense of subjective perceptions and more objective knowledge, they might have an opinion - and perhaps a different view from others - about whether or not they would see it as beneficial or justified to try to avert the birth of a child affected with the same condition or impairment as themselves. At the same time, there might be differences within such a group. The views of younger people might differ from those of older people, and those of men from women. Those affected with certain impairments or types of disorder might see the difficulties which they experience as socially caused and not inevitable, although others - including those

living with conditions different in their implications - might have a different view. Hence the critiques of screening programmes by disability activists as both eugenics and wrong, and as resting on medical values, might not be shared by all those living with a congenital condition.

The purpose of this study was to examine the views towards screening programmes of those living with different congenital disorders. A comparative approach was felt to be useful and appropriate, since the topic of screening programmes was known to evoke a range of views. The attitudes of those affected would be considered in the context of the theoretical debate, and would be compared with those of others who had contact with and knowledge of the conditions, but who were not themselves affected. The views of young women would be sought, as young adults - or women - living with a disorder and as potential users of antenatal care.

Aims

The main aim of the study was to examine the views about reproductive screening programmes of a sample of young women living with a congenital condition for which such a programme was currently available, compared with those of professionals in the related fields of medicine and disability and those of young women in the general population. The study also aimed to understand affected women's knowledge of, and their experiences of living with, their disorder.

Objectives

1. To describe the views about screening programmes of young women affected with certain congenital conditions, those of other young women in the general population, and those of professionals in the related fields of medicine and disability.

2. To describe the perspective of 'affected young women' compared with that of other women.

3. To examine the perceptions of 'affected young women' of their experience of living with a specific condition.

4. To investigate similarities and differences in views and experiences of women living with conditions which differed in their implications.

5. To consider the impact that women's experiences of living with a specific disorder or a condition seen as 'preventable' might have on their views about reproductive screening programmes.

6. To report on the varying perspectives of all respondents in the study towards screening programmes, especially in terms of the arguments of the theoretical debate.

The study would examine the following research questions:

- what were the main concerns of young affected women?
- were there differences in views about screening within the group of 'affected young women'? If so, with what might they be associated?
- to what extent did women's views towards screening reflect the arguments of disability activists, or those within the medical literature?
- did some or all of these women describe an experience of disability or see themselves as disabled?

The research methods: methodological considerations

Semi-structured interviews were seen as the most appropriate method for examining this topic, which encompassed both personal and public issues. Interview guides, with mainly open questions, would be constructed around a number of themes, within which respondents might discuss issues which they felt were important in their own terms. At the same time, to avoid the possibility of bias, it was felt important to ensure that all interviewees were asked similar questions on key issues, as far as possible in similar terms. An inductive approach would be taken to analysis. Descriptive accounts of the views of different groups would examine the way that screening was discussed; whether in general or personal terms, the reasons given for people's views, the values and philosophies drawn on, and the arguments suggested as important.

A qualitative approach was felt to be essential because of the complexity of the topic and the exploratory nature of the study, but also in order to gain an understanding of the issues of most relevance to respondents and the strength of their feelings, the explanations offered and where there was ambivalence or inconsistency in values and views. A qualitative approach allows respondents to discuss issues in their own terms and from their own perspective (Bryman, 1988), the emphasis being on meaning or understanding. Meaning cannot be accessed directly, but may be explored by an analysis of language. As Williams and May (1996: 140-141) have noted, qualitative work, "is said to allow the respondents to construct meanings that are valid to them within their social context".

From a sociological perspective, reported subjective experience and views are regarded as being socially produced; respondents are seen as drawing on cultural knowledge and recreating it in conversation (Coffey and Atkinson, 1996). However, while qualitative sociology aims to examine social context or culture (Douglas, 1970a), or 'the socially

constructed nature of reality' (Berger and Luckmann, 1967), the objective as well as the subjective, this approach may be linked with a number of theoretical perspectives, which reflect different philosophical positions about the nature of the social world and about how this may be discovered. These different positions have different methodological implications, in particular for analysis. A naturalistic perspective suggests an inductive approach, deriving concepts from data, although this can be criticised as empiricist or atheoretical (Scambler, 1987). A structuralist position which theorises social mechanisms suggests a more deductive approach, although this position can also be criticised as presupposing facts (Lincoln and Guba, 1985).

From a hermeneutic philosophy, the shared understandings of the researcher and the researched might be used to interpret conversation to suggest meaning (Hammersley and Atkinson, 1995). However, as Schwartz and Jacobs (1979) have pointed out, there are obvious difficulties for the sociology of knowledge in examining the culture of which one is oneself a part. Because of this difficulty, a comparative approach, examining similarities and differences in views and values between groups, has been suggested as useful (Coffey and Atkinson, 1996), although the need for care in reporting the findings has also been noted (Huberman and Miles, 1994).

Reflexivity

Since findings from qualitative interviews are seen as produced in conversation, the researcher and the research setting may affect what is said and how it is said (Cornwell, 1984). The idea that people's experiences and expressed beliefs simply reflect the values of a wider culture, and that these can be 'uncovered' by the researcher, has been criticised as positivist (Hammersley, 1992). Most qualitative sociology considers interview accounts not just as reflecting beliefs or cultural values, but as constructions which are reflexively produced in interaction with a researcher (Lincoln and Guba, 1985), or which are constructed for a wider audience (Hammersley and Atkinson, 1995).

Reflexivity may be of particular relevance in certain situations or in relation to particular issues. It has often been claimed that respondents in individual interviews, where the researcher may be perceived as having a higher status, might tend to give a more 'public' or moral account (Voysey, 1975; Cornwell, 1984). Some have suggested that such normative accounts are more likely to be given in response to particular topics. For example, Voysey (1975) has argued that the prevailing ideology of family life was the only language available to respondents discussing living with a disabled child. Many have noted that health is often seen as a matter of morality or virtue (Blaxter, 1993; Williams, 1993), and Williams (1995b) has argued that research

interviews examining health issues encourage 'semi-theoretical' or professional responses.

While it has been claimed, from certain sociological perspectives, that normative accounts reflect dominant discourses or ideology (Voysey, 1975), in the debate about screening programmes, there is an array of cultural views, discourses or value-positions, all of which might be seen as moral. It has also been argued that it is not possible to tell where accounts come from. For Cornwell (1984), moral accounts about health might reflect either medical or lay discourses; she has pointed out that this difficulty of knowing what is particular to a community and what is part of belonging to wider society is a common problem for qualitative analysis.

However, Cornwell's distinction between 'public' and 'private' accounts has been seen by many as problematic; Radley and Billig (1996), among others, have argued that personal experiences might be used to make moral points. These authors have claimed, for example, that people who are interviewed because they have a particular condition might want to present themselves to a researcher in a variety of ways and for a variety of reasons; they might stress similarities with or differences from a researcher, or challenge assumptions about themselves.

While perceived differences in status between researchers and the subjects of research might have implications for the findings of a study, more general concerns have been raised about inequalities in this relationship, because of the potential for differences in power or status to be reinforced by the research process. Researchers have been advised to take care not to reinforce stereotypes or to construct participants as 'other' in reporting research, and to be attentive to the way in which a study is carried out (Dingwall, 1980).

A qualitative interview involves what Dingwall (1980) has called an unnatural relationship between the researcher and participants, involving both rapport and distance on the part of the researcher. There may be perceived differences in status between researcher and participants on the basis of class (Cornwell, 1984), gender (Finch, 1993) or disability (Oliver, 1992; Zarb, 1992). Some have argued that the lack of cultural guidelines makes for anxious interaction between physically disabled and able-bodied (Goffman, 1968; Keith, 1996), and that those who are ill or disabled might feel unequal or inhibited because of the assumption that the researcher is able-bodied (Freund, 1988; Radley and Billig, 1996). Politeness and civility on the part of respondents in an interview might indicate perceived inequality or a lack of control (Cornwell, 1984; Freund, 1988), and a number of commentators have suggested that the

reciprocity claimed by Oakley (1981) is not always achieved in practice (Zarb, 1992; Finch, 1993).

Concerns about the relative status or power of the researcher have been raised by some feminists (Fine, 1994), and particularly by some disabled people. Oliver (1992) has argued that, although qualitative interpretative methods are preferable to a positivist approach based on a 'medical model' in examining and portraying the experience of disability, nevertheless such research may still be exploitative of disabled people because of the relative powerlessness of the subjects of research. For Oliver, disability research should be emancipatory, the goals being to empower disabled people; from this view, control of the research process should be in their hands. Others have made the point that policy change is beyond the control of researchers, but that a participatory approach should be taken to empirical work. This philosophy implies research carried out in partnership with disabled people and in which the process is scrutinised, with the researcher reflecting on his/her own research practice (Zarb, 1992). From feminism, Morris (1992) has advised that a researcher should ground herself as a non-disabled person holding cultural assumptions about disability, and should see understanding as taking place in the context of an unequal relationship.

In general, commentators in the disability movement have not argued that disability research should only be carried out by disabled people. Barnes (1992) has pointed out that not only would such a condition limit the numbers of potential researchers, but that there is a range of experiences of living with an impairment; also, in his view, "having an impairment does not automatically give someone an affinity with disabled people" (1992: 121). However, he has argued that it is imperative for able-bodied researchers to interact on a regular basis with people with impairments.

In this study, the focus would be on the views of those living with a congenital condition, and most of the research interviews would be examining the attitudes of young women. The project was advertised as a studentship, and it was felt that a young researcher would share some characteristics and cultural views with this cohort. Although it has been argued in the literature that there is an affinity between women researchers and participants (Oakley, 1981; Finch, 1993), the student might be male or female. It was not felt essential for the researcher to be disabled for the reasons outlined above, and because it could be argued that a researcher identifying as disabled might distort or influence the views of all respondents in the study. However, the characteristics and the perspective of the researcher must be seen as relevant to the process of research and in reporting the results.

I came to the study from a recent academic background of sociology, with a long-standing interest in feminism and politics, but also with an interest in medicine, having spent a year at medical school after leaving school. Although female, I was aged fifty, but I came with personal experience of screening programmes, having been offered amniocentesis with my third child. I have a slight hearing impairment but do not identify as disabled. Before coming to the project, though, I had some knowledge of the issues involved from disabled colleagues, who discussed both the social model of disability and some concerns about reproductive screening programmes.

In order to increase my understanding of the needs of disabled people and to lessen possible awkwardness when interviewing women with mobility problems, from an early stage in the project I attended meetings of a 'Healthy cities' disabled women's health group, where several of the members were wheelchair users. I also took part in a workshop on disability awareness, and sat in on discussions about peer group counselling for disabled women. Although I had known people with impairments throughout my life, I found this interaction useful in learning about the sensitivities of disabled people and about the practical difficulties experienced by many, especially in relation to the availability of transport. Discussions at the disabled women's health group also covered questions such as whether accessible services for disabled people should be located in one setting or available everywhere.

It was also valuable to discuss the research project with these disabled women. Not only was I given contact the names of contacts in disability organisations, but several group members agreed to take part in the pilot study. The majority of members of the disabled women's health group at that time had mobility impairments, and at least one had been born with spina bifida, one of the conditions on which the study was focusing.

The research methods: practical considerations

At an early stage, decisions had to be taken about the number of congenital conditions that the study should examine, and which these should be. The aim was to include three, if possible. Each would be one for which a screening programme was currently offered, for which affected individuals had lived with the condition since birth, rather than a late-onset condition, and for which they could live into adulthood. If possible, the disorders should be different in their implications for the person affected.

It was felt essential for women with cystic fibrosis to be included in the study, because of the pilot population carrier screening programmes being offered at the time, and the implications of carrier screening for a condition known to be inherited. Cystic fibrosis

can be seen as a chronic illness; it is a progressive disorder, but some of those affected now live into their twenties, thirties and forties.

It was also felt important to investigate the experiences and the views of women living with a more stable impairment. In the West of Scotland, screening programmes for neural tube defects have been offered for a number of years, and it was decided to include women with spina bifida in the study. Spina bifida is associated with limitations in mobility, and I knew that some people living with this disorder identify as disabled.

It was decided to try to include women with Down's syndrome in the study. Down's syndrome is one of the major conditions for which screening programmes are offered. At the time that the study was proposed, a woman with Down's syndrome had expressed anger about the existence of the screening programme. Down's syndrome has different implications for the person affected and for others, being associated with learning difficulties (although this may also be the case for some people with spina bifida). It was acknowledged that there might be difficulties in including women with Down's syndrome because of difficulties of comprehension and communication, and it was decided to ask professionals working with people with the disorder or in the field of learning disability for their views.

This was an exploratory study, one of whose aims was to investigate the feasibility of asking 'affected young women' about their views of screening programmes. It was not known whether or not the topic might be perceived as sensitive, whether it was relevant or of interest, or whether a sufficient number of 'affected young women' might agree to take part. All three disorders are all associated with a range of severity, and the effect of limitations on women's willingness or ability to take part was not known.

The research methods: ethical considerations

In this study there were not only theoretical and practical issues affecting the design of the research, but there were a number of ethical considerations, because of the interactive method and the topic of the study. All research has ethical implications; a researcher has a responsibility not to cause harm and to protect the rights of respondents. In qualitative work, as indicated already, the flexibility of the method and the interactive nature of the interview may give rise to particular ethical difficulties. Interviews can have effects or consequences which cannot be judged. As Lee (1993) and Brannen (1988), among others, have noted, there may be particular ethical difficulties where the topic of research is a sensitive one, although there are few guidelines on the researcher's responsibility for participants' emotional well-being.

The BSA Statement of ethical practice advises that sociological research should be based, as far as possible, on informed consent, and that this implies a responsibility on the researcher to present a study in an understandable way. However, the difficulties of doing this where the topic of a study is technical or difficult to convey, or where the flexibility of the qualitative method might make the consequences of an interview difficult to predict have also been pointed out (Dingwall, 1980; Lee, 1993). On the other hand, some have argued that there may be reasons for not defining the topic of a study too closely, because this might change the behaviour of participants (Burgess, 1984) or because such research may then be unable to widen the boundaries of what is known by challenging existing definitions and traditions (Hornsby-Smith, 1993).

While there are ethical considerations for any study, it was felt that there might be particular reasons for attention to the rights and the welfare of respondents in this study. It was recognised that the topic of the study, with its focus on prenatal screening and the implication that the births of people with these conditions might be prevented, might be a sensitive issue for some, concerned with personal matters, emotions or anxieties. It might be painful for those living with a condition to focus on the implications of screening programmes, or to think about the future (Clare, 1990), and women with all three conditions might face possible difficulties and constraints in relation to having children. As discussed already, interviews with a researcher without these conditions might be perceived as awkward.

At the same time, it was felt wrong to assume either the sensitivity of the topic or the vulnerability of respondents. It has been noted that some can find a discussion of sensitive issues cathartic (Lee and Renzetti, 1993), or might prepare themselves for the possible effects of an interview (Brannen, 1988; Lee, 1993). In addition to concerns about participants' welfare, there was a concern for their rights to take part. It was felt important that the opportunity to express their views on this issue should be offered to as many women in the target population as possible.

Steps were taken to address ethical issues in designing the study. Only one interview was planned, not only because of time constraints but also because this has been seen as a helpful approach for sensitive topics; respondents know that they will not be seeing the researcher again. Although this might mean some loss of 'rapport', this was felt not to be the main aim of this study. In interviews, a matter-of-fact approach would be taken, using the interview guide, exploring issues of concern to respondents but not pursuing those which seemed sensitive or too personal. Interviews with women with disorders would close with an acknowledgement that the interview might have raised disturbing issues. Attention would be paid to the way that the study was presented, and interviews would be arranged only with those who had had time to consider the issues.

The pilot study

A pilot study of twelve semi-structured interviews was carried out with volunteers, eleven of whom were women. Several were members of the disabled women's health group, who knew me reasonably well, and who readily agreed to take part.

On the basis of these interviews, some changes were made to the interview guide. Draft questions had concentrated on participants' views about screening and new developments in genetics, but the main finding from the pilot study was that many participants preferred to talk more about their lives; not only did they seem more comfortable and fluent in discussing their own experiences, but some related their experiences and philosophies to their views about new developments. The topic of the study seemed to be perceived as important and legitimate rather than a problematic, personal one.

In the revised guide there was more emphasis on living with a condition, and questions on aspects of technology which were little understood were withdrawn. I found that a diagram was helpful to explain the inheritance of cystic fibrosis in a standardised and understandable way. Also, 'vignettes' or hypothetical personal questions were useful in encouraging discussion of general questions and consideration of moral issues.

Sampling and access

Although 'understanding' rather than representativeness is seen as the main aim of qualitative work (Mays and Pope, 1995), it has also been argued that a sample should be as representative as possible. Denzin (1970) has advised that sampling should be 'theoretically-informed', taking account of variable factors, rather than being carried out on the basis of convenience. There has recently been more attention to the question of representativeness, because of a growing interest in examining the diversity of beliefs and experience, especially those of individuals living with a disorder (Conrad, 1990). Because of this, Conrad has emphasised the need to use a variety of methods of sampling.

The aim in this study was to examine the views of young women affected with cystic fibrosis, with spina bifida or with Down's syndrome, who were aged between 18 and 25. It was felt important to sample as widely as possible, for a number of reasons. It was anticipated that the numbers of women with each of these disorders in the age-range selected was likely to be small. It was also known that, for all three conditions, there were different degrees of physical severity. In order to maximise the size of the sample and to include those with different experiences and views, the aim was to outline the study to as many of the study population as possible.

Those whose views were sought were young adults, and it was hoped that individuals could be contacted directly, rather than through their families, since the topic raises different issues for parents. However, appropriate channels for sampling were not known; for example it was not known what proportion of women with the different conditions might use statutory services or might belong to support groups. It was therefore felt that sampling should be attempted through a number of channels.

Lee (1993) has discussed the need to use a number of channels or strategies where the topic of research is a sensitive one, where numbers are small, or when researchers have no access to records. As he has pointed out, with increasing attention to the individual's rights of privacy, as well as to confidentiality and consent, the researcher is less likely to have access to lists. He has also noted that this attention to rights and welfare has led to the increasing regulation of research proposals, and possible difficulties with access, and that there are more opportunities for the research aim to be distorted or the study turned down.

The importance of the negotiation of access to research has been much discussed in the literature. The way that a study is presented might affect not only the way that the project is perceived by participants, but also the progress of the study (Burgess, 1984). It has been suggested that occasions where access is difficult or is refused can indicate the limits of acceptability of a project, or social relationships within an organisation (Burgess, 1984; Lee, 1993). However, difficulties with access have normally been discussed in relation to one situation or setting (Burgess, 1984; Hammersley and Atkinson, 1995). With the exception of Lee (1993), there has been little practical advice about sampling through a number of channels or making contact with individuals, or any discussion about the difficulties.

The aim in this study was to sample in a structured way using a number of strategies. Support groups and medical channels would be contacted, but attempts would also be made to contact individuals who did not belong to these groups. Although questions have been raised about the extent to which those living with the same condition can be regarded as a 'group', it was decided that sampling would be carried out on the basis of medical conditions; some have argued that this is essential for comparative purposes (Anderson and Bury, 1988). However, it was also recognised that some people might not see themselves as someone with a condition. Because some might define themselves as disabled, and because some disabled people have been particularly critical of antenatal screening tests, groups of disabled people would also be contacted. The process of negotiating access for the different conditions, and the difficulties of doing this, are discussed below.

Down's syndrome

While it was hoped that young women with Down's syndrome could be included in the study, nevertheless, there was concern about the feasibility of including women with learning disabilities in research, and especially about whether they could be or should be asked questions about reproductive issues and screening programmes.

At the time of the study, there had been little research with people with learning disabilities. However, the literature described a growing interest in finding out their opinions and experiences, both because of the development of community care services (Flynn, 1986; Booth, 1996), and because of the increasing emphasis on self-advocacy by disabled people (Atkinson, 1988). The literature available indicated that it was viable to include people with learning disabilities in research projects, provided that attention was paid to appropriate methods and questions, and to the allowance of adequate time (Booth and Booth, 1993), and I was encouraged by colleagues to pursue this approach.

The extent to which people with learning disabilities are able to give informed consent to research is a difficult issue. However, studies have been carried out in the UK, and the advice given by those with experience is that the most adequate possible measures should be taken to obtain informed consent, and that the rights and the privacy of respondents should be respected (Prosser, 1989).

At an early stage in the study, I approached a number of professionals working with people with Down's syndrome and in the field of learning disabilities to ask whether they would see it as right and feasible to include women with Down's syndrome in the study, as well as asking about the possibility of access. Over a period of 14 months, I contacted thirteen organisations and many other individuals. These organisations included the national support group, national and local voluntary organisations for people with learning disabilities, housing associations, special needs departments in further education, social work departments, and the health service via hospital consultants and community learning disabilities teams.

There were very wide differences in the views of professionals in the field about whether it was right to include women with Down's syndrome in the study. Some professionals, especially those at a senior level, felt strongly that there were important reasons for including them, because they should be treated as no different from other women in this respect:

I was really pleased when you had approached me initially ... and you said that having interviewed the other groups ... the next group you were wanting to tap was women

with Down's syndrome. Because I feel that women with learning disabilities should be included.

(Consultant psychiatrist)

The view of staff in the majority of organisations contacted was that women with Down's syndrome should not be excluded from the study if an appropriate and sensitive method could be found. This was seen as a matter of possible difficulties for me as a researcher rather than a reason for refusing access:

I think, you know, it would be unrealistic in talking to somebody with learning disability, not to take their sort of learning disability - mental ability - into account. ... Down's syndrome ... it affects the way you must talk to somebody. But it doesn't make the issues less significant for them. ... The issues, I think, are still important. It's just different, and probably more difficult to tap into.

(Development worker, voluntary organisation)

On the other hand, a few professionals spoke strongly against their inclusion. Some suggested that, because of difficulties of communication and understanding, the research would be meaningless, and interviews could not be compared with those of other women in the study. Some who took this view felt that it was sometimes more helpful to people with a learning disability themselves to acknowledge their differences from others. A few suggested that taking part in the study might cause harm, particularly anxiety, to the participants:

It was felt that group interviews would not be a good idea, and group discussion would not be a good idea at all, and it was also recognised that individual interviews would be quite frightening.

(Spokesperson, statutory organisation)

There were particular concerns about the topic of the study, which might introduce concepts which respondents had not encountered before, such as the fact that they had Down's syndrome or a learning disability, but especially the idea that the birth of a child with Down's syndrome might be prevented. It was also felt that questions about having children might raise false hopes or might cause distress to those who might not be allowed to have children.

Several professionals also expressed concerns about informed consent, raising questions about how the study would be presented, and about the extent to which women with Down's syndrome might feel able to refuse. Anticipated difficulties were summed up by the spokesperson from one organisation where access was turned down, who also emphasised the need to consider the implications for staff of the organisation:

Whether you should be posing these questions to people with learning difficulties is another matter ... in a way, it's a Catch 22 situation, they either don't understand the questions for you to understand that their answers are worth obtaining, or, if they do understand it well enough, then there's a need for so much support.

(Spokesperson, statutory organisation)

Those professionals who thought that it was right to include women with Down's syndrome also discussed the issue of consent. Some suggested that the study might be advertised or outlined only to women who lived independently, although most felt that carers should be informed.

These professionals also had divergent views about the need for time, and about the type of questions or about the limits to questions. Because of concerns about the sensitivity of the topic, several advised that questions should be drafted in advance, prior to gaining access. Considerations about appropriate questions are discussed in more detail in Chapter 7. After discussion, it was decided that the aim should be to ask women with Down's syndrome about aspects of their lives and their views about having children, as far as possible in the same way as with other women in the study, although avoiding any implication that the births of people with Down's syndrome could be prevented.

Some representatives of organisations concerned with learning disability questioned whether it might be more appropriate to speak to women with any learning difficulty. Several suggested that voluntary organisations or self-advocacy groups of people with learning disabilities would be preferable channels for sampling, because their philosophies of self-advocacy and making choices might lead to fewer problems with communication or consent. It was sometimes suggested that women with Down's syndrome might be more likely to think of themselves as having a learning difficulty rather than as having Down's syndrome; as the coordinator of one self-advocacy group explained, "*people are not identified in that kind of way these days*".

Several professionals raised the question of whether interviews were an appropriate method, because of perceived difficulties with communication and understanding; some also mentioned that many people with Down's syndrome did not talk very freely. It was suggested that group work might be one solution for working with people with learning difficulties, since in the presence of a facilitator and other known group members, women might feel less anxious about the presence of a stranger and differences in knowledge or perceived status might be overcome. Since there are no existing groups of young people with Down's syndrome, the members of the group would be young women with learning difficulties.

As a pilot, six volunteers from an existing women's discussion group at a voluntary organisation for people with a range of learning difficulties took part in a group discussion with the group facilitator and myself. General issues about relationships, reproduction and antenatal testing were discussed in relation to the experiences of some members of the group and their friends, and with reference to television programmes. The group situation was useful in generating discussion, although this was at times difficult to control. One participant commented that, if she were pregnant, she would want to have tests for Down's syndrome. The one woman in the group who had Down's syndrome said little.

While the group interview provided useful information, because of the nature of the interaction, I felt that this would be a difficult and perhaps inappropriate method of examining women's personal experiences or views. Some professionals also expressed reservations about this approach, because of concerns for participants and for me as a researcher:

It would be impossible to speak to groups; you would have all sorts of problems, some who don't understand. And the whole question of talking about personal issues in front of other people.

(Coordinator, voluntary organisation)

In order for this part of the study to be comparable with the rest, it was felt preferable to try to carry out individual interviews with women with Down's syndrome. I attempted to arrange interviews through a number of organisations where access had been agreed in principle; the Scottish Down's Syndrome Association (SDSA), other voluntary organisations including People First, a further education college offering courses for adults with special needs, and community learning disability teams.

The SDSA agreed to advertise the study so that parents or carers would be informed, and might outline the study or act as facilitators if necessary. At the FE college, where the philosophy was that people over the age of 18 could take their own decisions, it was agreed that the study might be outlined to the four women students with Down's syndrome by a female member of staff, who would act as a facilitator in any interviews.

However, at other organisations, although access was agreed in principle, this was qualified with the need to obtain further permission from others at a different level. Professionals responsible for granting access saw their responsibilities in different ways, and those in closer contact with women with Down's syndrome were more likely to express concerns about respondents' welfare. Several also had questions about the feasibility of carrying out interviews, and about the suitability of respondents; for example, one member of staff drew attention to the hearing impairment of a woman

whose name had been suggested. Some also raised the issue of how the study would be presented and who would do this. As one social worker pointed out, "*you see, the thing is people with learning disabilities can't decide for themselves whether to take part. Someone will have to decide for them*". Hence, while there was enthusiasm for including women with Down's syndrome, access was seldom achieved in practice.

Spina bifida

It was felt essential to try to contact women with spina bifida through a number of channels for two reasons. First, there is no specialist hospital service for adults with this condition; babies born with spina bifida or hydrocephalus are referred to a specialist paediatric clinic, but as children reach adolescence their management is taken over by GPs or local hospitals. Because there can be a range of symptoms, varying in severity, while some people with spina bifida might attend various hospitals for different symptoms, others may need little or no medical treatment, and therefore might not be contactable through a hospital.

Secondly, although a national support group does exist, I had been told that some people with spina bifida might define themselves as disabled rather than as being affected with the condition, and might have no contact either with the support group or with hospitals. It was decided, therefore, that one approach should be to try to contact young women with spina bifida through voluntary organisations of disabled people or through courses for young people with special needs.

In practice, I contacted a number of possible channels. Although I did not know whether or not young women with spina bifida might be members of the support group, I approached the Scottish Spina Bifida Association (SSBA) at an early stage. The secretary of the national association suggested that the coordinator of the Glasgow and West of Scotland branch should be contacted, and that the study might be advertised to members through the newsletter. The Association records showed that 44 women aged between 18 and 25 were members of the local branch. An advertisement was placed in the newsletter, but only one response was received; this was from an older woman who agreed to help with the pilot study.

Fortunately, through this contact with the local association, a good relationship was established between myself and the coordinator. She offered to forward a letter about the project to each of the 44 women in the branch, enclosing a compliments slip from herself, with an offer to discuss the study with anyone interested, as a way of endorsing the research. Thirteen women returned consent forms to me, and interviews were

subsequently arranged by telephone. (A copy of the introductory letter and the consent form is given in Appendix II.)

I also sent letters about the study to local voluntary associations of disabled people in the area; the Glasgow Coalition for Disabled People, SKILL - the National Bureau for Students with Disabilities, Wellbeing, and the Advocacy Project. However, there was no response to these.

Another possible channel for recruitment that had been suggested was a local college offering special needs courses. When I approached the college, it was agreed that a member of staff would outline the purpose of the study and give introductory letters to the six students there who had spina bifida. However, it proved impossible to arrange a meeting with the member of staff to discuss this. Eventually the letters were forwarded, but there was no response to them.

Snowball sampling was also attempted; I asked all women with spina bifida taking part in interviews whether they knew other young women with the disorder. Several said that they did, and some mentioned names; these included some women who had already been interviewed. A number of women took introductory letters to pass on to these people, but no consent forms were returned. I did not ask for names and addresses, in order to adhere to the previously agreed guidelines regarding privacy.

The intention was also to contact hospital consultants, as another route for making contact with young women with spina bifida. Since I had been told that women with this disorder might attend different hospitals, all interviewees were asked which hospital they attended. However, every large hospital in the area was mentioned as a possible venue for contact. One consultant was contacted, but did not reply.

I was advised that some young adults still attended the paediatric hospital, and a consultant there agreed to help with the study, although pointing out that the numbers of women would be small. Ethical approval was obtained, but this contact was not pursued. Some women taking part in interviews mentioned that they attended this hospital and it became clear that no new names could be obtained through this channel.

Thus, although access was attempted through several channels, only one was successful. However, the fact that the same people appeared to have been suggested through different routes suggested that contacts, at least in the Glasgow area, might have been saturated, and that sampling through the SSBA might in fact have reached most of the target population in Glasgow and the West of Scotland. The lack of response from associations of disabled people can be contrasted with that through the support group, although the reason for this is not known; possible factors might be the

lack of young women members or the unacceptability of sampling by medical condition through disability groups. The response rate of thirteen replies from 44 letters can be seen as low. The lack of personal contact might have been a reason for this, although nothing can be known about those who did not reply.

Cystic fibrosis

It quickly became obvious that the sampling strategy for contacting women with cystic fibrosis would have to be quite different from that adopted for women with spina bifida, because of the rarity of the condition and because of the shortened life-expectancy. In 1995, the number of women with cystic fibrosis over the age of 16 in Scotland in 1995 was 82 (UK Cystic Fibrosis Survey Report, 1995). It was decided, therefore, that the aim should be to contact all women between the ages of 18 and 25 over a reasonably wide area, such as the South of Scotland.

A specialist hospital service is available for adults with cystic fibrosis, in contrast to spina bifida. Possible channels for sampling appeared to be these hospitals or the national support group, the Cystic Fibrosis (CF) Trust; each of these had been used in other studies of adolescents with cystic fibrosis (Walters *et al.*, 1993; Blair *et al.*, 1994). I did not know, however, what proportion of young women with this condition might be under the care of the hospital or might belong to the support group.

At an early stage, I contacted a representative of the CF Trust in Scotland. The view of this coordinator was that the best way of contacting young women with cystic fibrosis would be through the large specialist hospitals, since virtually all adults were now registered at these. Although local support groups existed, she felt that they were largely aimed at families of young children, and that young people were unlikely to have much contact with these, commenting, *"they're just like you and me - they're getting on with their lives"*. I later learned that people with CF have been discouraged from meeting in groups because of the recently-discovered risk of cross-infection.

Consultants in the two specialist CF units in the South of Scotland agreed to help with the study, and they estimated that 90% of adults with cystic fibrosis now attended these clinics. The number of women aged between 18 and 25 was given as about 20 over the two clinics, and it was pointed out that the clinics covered a wider area than the South of Scotland. Ethical approval was subsequently sought and given. (A copy of the introductory letter, the summary of the study and the consent form is given in Appendix II.)

I discussed the research with the liaison sisters in each hospital, since they had regular contact with people with cystic fibrosis, and they agreed to outline the study to

individuals. The nurses' concerns were for the welfare of the women taking part; they pointed out that young people with CF had been much researched recently, and that these experiences could be stressful. They emphasised the need for a sensitive approach, and one said that she might not suggest the study to anyone that she felt might be unable to cope.

Sampling through the specialist clinics was straightforward; completed consent forms were forwarded by the clinic and interviews were arranged by telephone. Fourteen of the eighteen women approached agreed to take part; two women were not asked because it was felt 'inappropriate' and three others because they lived too far away, making a total of 23 women with CF in the target population.

From some of the early interviews, it was clear that a few women had only recently started attending the specialist centre. This suggested that some women, especially those with milder conditions, might have been missed by sampling only through the hospital, and that other methods of sampling should be attempted. The local branch of the CF Trust agreed to advertise the study in their newsletter, but there was no response to this. Snowball sampling was also attempted; all interviewees were asked whether they knew other women with cystic fibrosis, apart from through the clinic. However, unlike women with spina bifida, they all reported that they knew nobody else with the condition.

Again, therefore, although the intention was to sample through several channels, only one was successful; indirect sampling through hospital lists. From the opinions of professionals, sampling by this method is estimated to have reached 90% of the target population. The response rate was considerably higher than for spina bifida, although personal contact may only have been one factor.

Implications of difficulties with access and sampling

Because of the nature of the three conditions, and because of the need to contact as many of the study population as possible, sampling was attempted through a number of channels. However, this strategy proved unsuccessful in practice. While there were different problems with access for each disorder, these difficulties raised common methodological issues. It was often difficult to identify appropriate channels, and to make contact with an appropriate person in each setting. Lessons learned through one route did not necessarily translate to another. Also, attempts to negotiate access highlighted the different philosophies both between organisations and at different levels of the same organisation regarding the rights of the individual or the need for protection, and about the value of the research.

It has been pointed out that, for access to be successful, negotiation with gatekeepers is needed to establish potential benefits to the organisation and the trustworthiness of the researcher (Dingwall, 1980; Lee, 1993). A study might need to be approved by individuals at different levels within an organisation, all of whom need to be convinced of the usefulness of the research. As Lincoln and Guba (1985: 253) have noted, "the keys to access are in the hands of multiple gatekeepers, both formal and informal", and Dingwall (1980: 876) has spoken of a "a hierarchy of consent".

With all three conditions in this study, it was often necessary to negotiate access and obtain consent at a number of levels in an organisation. While agreement had to be obtained at a senior level, it was also necessary to obtain the consent of an 'intermediary' within the organisation who might expect to take a more personal responsibility for the study by, for example, agreeing to introduce the study or acting as a facilitator. There were sometimes wide differences between the views of these individuals about the feasibility of the study and the value of the research. Whereas more senior individuals were more likely to emphasise a philosophy of women's rights to take part, those who were in more direct contact with respondents tended to be more concerned about their welfare.

Where access was granted, I had met and spoken to these intermediaries. Many of these people commented that they saw this personal contact as essential in order to establish that the study would be carried out in a responsible and sensitive way, both during the interviews and in reporting the study. All researchers take responsibility for conducting research in an ethical way, but I did feel that I had been given a considerable degree of 'inter-personal trust' - as described by Lee (1993) - in situations and in relation to a topic acknowledged to be potentially sensitive.

These 'gatekeepers' were often not only responsible for granting access but also for outlining the study to potential respondents Lee (1993) has pointed out that, where contact is through an intermediary, the researcher may have little control over the way that a study is presented, and hence over the way that the purpose or the limits of a study are understood by respondents.

Gatekeepers in this study often expressed concern for respondents' welfare and for the feasibility of the study, and there was some scope for them to decide that some potential respondents were unsuitable or too vulnerable to be asked. A small number of women with cystic fibrosis were not asked for these reasons, and, as discussed, these concerns were often raised in relation to women with Down's syndrome. On the other hand, other individuals were sometimes mentioned as being particularly suitable, in terms

such as *"she'll give you a good interview"*, and a small number of interviewees spoke of being encouraged to take part.

In retrospect, there was some evidence to suggest that the study might have been presented in various ways, with expectations of opting-in or out, to different women, and that some women might have been more likely to hear about the study than others, because of the perceptions and concerns of gatekeepers. Also, the means by which the study was introduced might have affected the response rate, which was higher for women with CF, where the purpose of the study was outlined in person, than for women with spina bifida, who received a letter.

It might be necessary, therefore, to reflect on the extent of possible biases in the sample, which, because of the small numbers, consisted of all who had agreed to take part. While such a sample can never be statistically 'representative', in this study it was felt particularly difficult to estimate the ways in which the experiences, perceptions and views of those women with the conditions who took part might compare with those who did not. First, nothing could be known either about those who were not asked or about those who refused in each organisation, because of the need to respect privacy and confidentiality. Secondly, sampling for women with spina bifida and women with cystic fibrosis was ultimately successful through only one channel for each condition, and the effect of these different ways of drawing the sample could not be known. Thirdly, because women were contacted through a different type of organisation and in a different way for each condition, it was possible that differing philosophies and perceptions of staff within these organisations (about the value and purpose of the research, for example) might also have biased the sample. This issue will be returned to in Chapter 11.

The interviews and the respondents

Different interview guides were prepared for different groups, and are included in Appendix I. It was decided that women taking part in the study would be asked about their religious background (whether or not they attended church and if they had been brought up as a Catholic) and their educational level, and that some measure of their social class backgrounds would be ascertained.

Since I did not have access to their addresses, and hence a measure of social deprivation from post-codes, and since interviews might not necessarily take place in their homes, women were asked about their fathers', or their mothers' jobs. This question proved rather unsatisfactory in practice; some respondents described step-fathers' jobs, others those of fathers who lived away from home, and a number were unsure of the details of

their fathers' employment. However, half the sample of women with spina bifida, with cystic fibrosis and women in the general population described manual employment and half non-manual, suggesting that the social class background of each group of women was wide but roughly similar.

Brief details of the interviews and the respondents are given here, although further details are given in later chapters. All respondents taking part in this study were white. The interviews with women with congenital conditions were carried out first. Although the intention had been to arrange interviews with women with the different disorders consecutively, because of difficulties with access, all these interviews took place over the same period. Five women with Down's syndrome, fourteen women with spina bifida and fourteen women with cystic fibrosis took part in the study.

One woman with Down's syndrome replied to the SDSA advertisement, and an interview was conducted at her home, with her mother present at her request. Four women students with Down's syndrome from the FE college agreed to take part in the study. Interviews were held in a room at the college, with a member of staff acting as facilitator during three of them. All interviews with women with Down's syndrome were quite short, lasting between 10 and 20 minutes. All the respondents lived with their parents, and all had attended special needs schools.

Thirteen women with spina bifida replied to letters sent through the local support group, and another participant (aged 27) was a friend of a colleague. All interviews except two were carried out in the respondents' homes; one took place at my workplace, and another at the respondent's work. The average length of an interview was about 45 - 50 minutes, and all lasted between 30 - 60 minutes. The mothers of two of the women were present during the interviews; in one interview where the respondent had some communication difficulties her mother answered the majority of the questions.

All women with spina bifida lived in the parental home, although one was a student away from home during the term, and none had children. Within the group there was a range of social class backgrounds and also educational levels; a small number of women spoke of having some learning difficulties, whereas others reported attending courses in further or higher education. Most respondents spoke of attending a special needs secondary school, although several pointed out that this was because of mobility problems.

All respondents who had cystic fibrosis attended the two specialist clinics in the South of Scotland. All the interviews except two were carried out in people's homes; one took place at my workplace, and another in a room of a hospital where the respondent

was an in-patient. The length of interviews ranged from twenty minutes to one and a half hours. As with women with spina bifida, another family member was sometimes in the house, and the mother of one woman sat in during the interview.

Four women with cystic fibrosis were living with partners, one lived alone, and the others lived with parents, although one student was living away from home at the time of the study. One woman had a child. As in other groups, there were differences in social class backgrounds. All respondents spoke of attending mainstream school, and there was a range of educational levels, but half the sample were attending or had attended courses in higher education or had been expected to do so.

After the interviews with the women with these conditions were completed, the aim was to contact a similar number of women from the general population. Women with genetic conditions came from a range of backgrounds. Questions about education showed that, although the range of educational level was wide and a small number of respondents mentioned some learning difficulties, almost half the sample had SCE Higher grades, and several of these women were students in higher or further education. It was felt that young women on FE or HE courses with a health or special needs link might form a suitable sample for comparison, as they would share some characteristics with the other women in the study.

Two possible channels for sampling were contacted; a university nursing degree course and a local FE college offering NC child care and pre-nursing courses. Coordinators at both centres agreed that women students on the first years of these courses might be asked if they would take part in the research, and that I would outline the study in their presence.

Eight students from the degree nursing class and 21 students from the two classes at the FE college agreed to take part in the study. Interviews were carried out in a room set aside for the purpose at both settings. These interviews were normally shorter than for other women in the study, because, apart from a brief discussion, women were not asked about their own lives. One respondent in this group was living with a partner, one was lodging with friends, but all the rest were living with parents, although four students lived away home during the term. As in other groups, women described a range of religious and class backgrounds. It was clear from two of the interviews that the women were aged 17, although the rest were within the 18 - 25 category.

During the course of the study, professionals working in the fields of medicine, genetics, disability or in support groups were asked if they would agree to take part in a short interview. Twenty interviews were carried out; one with a geneticist, six with

disability professionals or campaigners, and thirteen with professionals in different fields who were contacted in the course of negotiating access. Five of these worked with people with cystic fibrosis, six with those with Down's syndrome or learning disabilities, and two with people with spina bifida. Interviews with these professionals normally took place at their place of work, and most lasted for half an hour.

All interviews except one were recorded, after checking with respondents that this was acceptable. It has been argued that, where reflexivity seems important in a study, analysis should look not only at what is said but at how it is said (Radley and Billig, 1996). It was anticipated that the issues being examined in interviews might be perceived as moral but also as sensitive. Respondents might want to convince me of their position, but at the same time, some issues might be felt to be personal or difficult to discuss. The data consisted, therefore, of 'field notes' as well as interview transcripts. Although it proved almost impossible to take notes during the interviews, the field notes were written up as soon as possible afterwards. These recorded my impressions of the context of the interview, issues which seemed awkward, those which seemed important to the respondent or forcefully expressed, and responses which seemed to me surprising. The tape was played through at the earliest opportunity to confirm or add to these impressions.

The experience of interviewing the women and professionals

As with any research, there were differences between individuals in the degree of perceived rapport, in the extent to which people wanted to talk and in what they wanted to talk about. Professionals were asked general questions about screening programmes, but I was surprised that a number also reported their personal experiences and decisions.

It has been suggested in the literature that interaction between researchers and people with an illness or an impairment might be experienced as awkward by both. I found the interviews with women with Down's syndrome awkward, perhaps because I had no experience of working with people with learning disabilities, and found it difficult to gauge their level of understanding. At the FE college, the facilitator was helpful in advising me of questions which might be attempted, and we had also discussed the limits to questions which the interview would cover. This was not the case where the mother of one woman with Down's syndrome was present at the interview, and I was concerned that some questions might have been felt inappropriate.

A few interviews with women with other disorders seemed to me inhibited, although there might have been various reasons for this, such as being a novice researcher and

being anxious not to cause offence or distress either by the study questions or by my own behaviour. A small number of women with spina bifida commented critically on the inappropriate attitudes of others towards disabled people. I was also worried about interviewing young women with cystic fibrosis because I had had no contact with young people with a short life-expectancy. However, in almost all cases, interaction seemed more relaxed as the interview progressed; I was sometimes aware that respondents were also initially anxious or cautious, but also that a number of interviewees (or their parents) took steps to minimise awkwardness. Differences between myself and participants rarely seemed to me to be intrusive during the interviews, although occasionally where women were in wheelchairs or were clearly unwell, leaving was difficult.

Many of the interviews with women in the general population seemed less awkward to me, although whether this was because of the assumed physical health of this group or because of my increased experience or confidence (as these interviews were carried out last) cannot be known. At the same time, although these women were asked about their lives in the same way as others, early conversation was often stilted, perhaps because they knew that the focus of the study was on their views about screening programmes.

Overall, I felt that interviews with all respondents were less strained than I had anticipated. Almost all participants in the study readily gave their views about screening programmes; many seemed to have given careful consideration to the issues. Similarly, contrary to expectations, abortion did not seem to be a sensitive matter. Many women appeared quite happy to discuss this, and some raised the subject themselves, a number expressing strong views on the topic. Women living with Down's syndrome, with spina bifida or with cystic fibrosis also spoke readily about their lives in general, and many respondents in the latter groups talked at some length about their approaches to living with the disorder, some of the difficulties that they experienced, and their perceptions of the quality of their lives. My impression was that many of these women wanted to convey their experiences and perceptions of living with the disorder, their knowledge and opinion about the condition more generally, and often, too, their attitudes towards screening programmes and the reasons for their views. Some women, usually those with cystic fibrosis, said that they had welcomed the opportunity to talk, and several women with both this condition and with spina bifida said that they felt that more should be known about the disorder and the implications.

It was not always possible to tell whether or not the topic was a sensitive one for respondents, or which aspects might have been sensitive. Family relationships seemed to be an awkward topic for many women in the study in all groups. A small number of women affected with spina bifida or with cystic fibrosis were somewhat distressed

when considering certain issues, in particular the deaths of friends, anxiety about their own condition and the possibility that they might not have children. However these moments passed. In response to my acknowledgement at the end of each interview that the topic might have raised difficult issues, some women with these conditions commented to the effect that 'you get used to it' or 'I'll go and have a shower and put it to the back of my mind again', indicating both a degree of distress but also that this had been expected.

This raises the question of whether women affected with these congenital conditions might have responded differently to a disabled researcher, although, as noted already, the term 'disabled' can cover many experiences. The lack of shared experience of disability might have caused awkwardness, and efforts to reduce this might have meant minimising some difficulties. On the other hand, I felt that one implication of my being able-bodied was that the concerns of many women with spina bifida or with cystic fibrosis were to educate me, and perhaps others, through reports of the research. Some respondents were forceful in their views, perhaps to challenge my perceptions, or those assumed to be generally held. It is possible that - while there might have been a lack of 'rapport' - respondents might have been less inhibited in giving their views to me than to a 'disabled researcher', who might have been expected to hold certain views, whether towards screening programmes or towards living with a condition similar to their own. These women drew on a range of discourses when discussing screening programmes, and there were some striking differences between and within groups about the importance of avoiding the birth of a disabled or affected child. For similar reasons, I felt that the age difference was not an issue because almost all participants seemed to perceive the study as an opportunity to convey their views as 'experts'.

Women in this study were not asked about their perceptions of the purpose of the study, or for their reasons for taking part. Lee (1993) has noted that increasing numbers of people are declining to take part in research, and there has been interest in respondents' reasons or perceived reasons for agreeing to do so. While Lee has proposed that those who take part might share the 'elite' values of the researcher that research is useful, Goyder (1987) has argued that those taking part in social science research are more likely to be the socially active, or those for whom the topic has salience. In a study like this one which examined controversial issues, and where participants were volunteers who had time to consider their response, one reason for taking part might have been to inform the wider world about either their philosophy - whether in relation to screening programmes or towards living with a disorder - or their knowledge, experience and perceptions. This issue will be returned to in Chapter 11.

Analysis and reporting

Qualitative analysis can be carried out in different ways and might lead to different end-products, depending on the aims of the project, on the theoretical approach, and on what it is felt that the data will support (Hammersley and Atkinson, 1995; Mason, 1996). The aim in this study was to take an inductive approach to the data. While conclusions would be based on empirical findings, the aim would be to go beyond descriptive accounts of respondents' own experience or perceptions to consider whether there were patterns in the data. Analysis would examine language and discourses, and the extent to which different philosophies were expressed, and would aim for interpretation if the data would support this. Since this was an exploratory study, and since there might be a number of cultural views or moral values, the aim was not to attempt explanation or theory. However, the study would aim to consider the impact that women's experiences of living with a specific disorder or a condition seen as 'preventable' might have on their views about screening programmes.

In this study, analysis was a long process, which did not occur in easily defined stages. Because the focus of the study was to be on the views of women with disorders, and because these interviews had been carried out before the others, early stages of analysis were carried out with these interviews alone. The interviews were transcribed, read and re-read, and the whole of each interview was coded horizontally. Broad categories for coding were mostly derived inductively from the data, although some key concepts central to the research questions were also used. The NUD.IST computer package was used for early coding and retrieval, but the transcripts were coded by hand because many responses diverged from the questions asked, and coding often 'nested' or overlapped. From a horizontal retrieval of the data, descriptive accounts were written of 'living with cystic fibrosis' and 'living with spina bifida', and of the views of women in these two groups towards new technology, looking at what was discussed or seen as important, to what extent there were differences, with some idea of the numbers of respondents holding particular views.

Coffey and Atkinson (1996) have suggested a number of strategies for moving from descriptive accounts of the data to a more interpretative, theoretically-based account, such as examining the way that language is used in narratives, trying to account for surprising findings or regarding interviews as accounts. Where reflexivity is important and interviews might be regarded as accounts, one approach is to focus on the construction of the interview itself (Silverman, 1973), although others have argued convincingly for an examination of respondents' explanations or legitimations (Radley and Billig, 1996). Coffey and Atkinson (1996) have explained that respondents might

use what Mills (1940) termed 'vocabularies of motive' to locate actions and events within a frame of reference. These authors, like Voysey (1975), have suggested that moral accounts may be expressed in terms of a comparison with others.

From early readings of the interviews, although women's experiences of living with cystic fibrosis or living with spina bifida were often very different, individual accounts were coherent. Some women spoke more in terms of narrative, others more in terms of values. Respondents usually expressed a coherent philosophy throughout the interview and sometimes in conversation beforehand or afterwards, and they often drew on these philosophies in a logical way in discussing both their lives and their views about technology. The interviews were therefore examined in a different way, as whole entities or 'vertically', in order to look at these different strategies or perspectives and the values, rhetoric or philosophies drawn on.

NUD.IST was less useful at this stage, since it is designed to assist cross-sectional categorical indexing rather than a vertical analysis (Mason, 1996). Instead, notes were written on the transcripts, and summaries were made of the values of each of these 28 respondents. Different philosophies were often linked with views about the importance of different aspects of life, and in some cases with certain use of language. A consideration of apparently unusual views in different groups helped to clarify different types of response.

At this stage more conceptual categories were developed, and interpretative accounts were written. These drew on both field notes and interviews, noted respondents' values and strategies, and considered where these seemed to be presented as different from or shared with mine. The interviews with professionals and students were analysed in a similar way, and draft chapters written of the experiences, knowledge and views of each group. At a later stage, the material about 'living with a condition' was condensed to issues which were most relevant to the topic of the study, such as perceived difficulties and quality of life, strategies and philosophies.

Decisions had to be taken about the way that the views of individuals and groups might be compared, and about how the findings should be presented. While qualitative analysis has been seen as particularly suitable for comparative studies (Coffey and Atkinson, 1996), the difficulties of carrying out a cross-case analysis have also been discussed. Huberman and Miles (1994) have argued that cross-case comparisons can only be made at high levels of abstraction, looking at generalisations which may not apply to any members of the sample.

For a number of reasons, it was decided to present the experiences and views of participants in each group separately, rather than focusing on themes or discourses. There were patterns in the data, suggesting some similarities within each 'group' of women (especially those living with spina bifida and those living with cystic fibrosis) in the issues suggested as important and in the views expressed. It was felt that the views of these women towards screening programmes, both generally and for the condition which they had themselves, could not be separated from the way that they discussed their perceptions of living with the disorder and their quality of life, and that their voices should not be fragmented. At the same time, analysis also indicated the difficulties of constructing chapters around discrete 'themes' or discourses. While rhetoric and explanations could be identified, arguments were often spoken of as being balanced against others, and women's accounts indicated ambivalence and contradictions between different value-positions. Also, while women drew on discourse, they also drew on factual knowledge and personal narrative in explaining their views. For these reasons, and because of the different ways of drawing the sample, a decision was taken to report the findings by 'group', rather than by themes.

Summary

This chapter has outlined the aims and objectives of the study, the research design and the process of carrying out the study, some of the theoretical, practical and ethical considerations affecting the decisions taken at each stage, and my impressions of the research process. The fact that contact was made with young women with spina bifida and with cystic fibrosis and interviews were carried out, has indicated the feasibility of asking women with these conditions about screening programmes, although there were difficulties with including young women with Down's syndrome. Some of the issues highlighted in this chapter, such as difficulties with access, the possibility of bias in the sample, the potentially sensitive nature of the topic, and an appropriate approach to the analysis of moral issues, have implications for a discussion of the findings, and will be returned to in Chapter 11.

The data chapters which follow outline the views of participants in different groups towards screening programmes. Some relevant details have been omitted for reasons of confidentiality and to protect respondents' anonymity; this was felt particularly important because of the small numbers in each group. The next chapter describes the views of professionals in the fields of medicine and disability about screening programmes, and the way that they discussed the implications of living with a congenital condition.

CHAPTER 6

PROFESSIONALS' VIEWS

The debate about the morality of screening programmes has been outlined in Chapter 3. While, from a 'rights' perspective, it has been argued that screening is wrong, and can be seen as eugenics or discrimination, from a 'welfare' perspective 'prevention' might be felt to be ethically right, within limits, if this can prevent harm, although benefits and harm may accrue to different individuals or groups and may be seen in different ways. Screening programmes may also be seen as beneficial in offering information and choice. However, from a view of screening programmes as beneficial, there are questions about the conditions which it might be appropriate to prevent, who should take such decisions and on what basis. Some disability activists have questioned assumptions, which might be felt to be based on 'biomedical' values, about inevitable problems for those living with an impairment, often by reference to the social model of disability.

While these theoretical views have been expressed in the media and in literature, it was felt important to find out the views of local professionals. Interviews were carried out with twenty professionals in the fields of disability, medicine and support groups; six were disability professionals who identified as disabled, five worked with people with cystic fibrosis, two in the field of spina bifida, and six worked with adults with Down's syndrome or learning disabilities. One interview was carried out with a geneticist.

Professionals were asked about the needs of the people with whom they worked and about their views towards screening programmes and new developments in genetics. This chapter illustrates the range of views among these professionals about living with a condition, about whether they would see certain conditions as serious, and about the use of screening programmes and new genetic knowledge.

Living with a condition

Professionals working with people with Down's syndrome, spina bifida or cystic fibrosis were asked about the needs of people living with these conditions. Many noted that a number of those affected might now expect to live into adulthood, and all made the point that someone living with a condition was a normal person like anyone else, with the same needs, desires, and sometimes problems, as any other young adult or young woman in today's society. As one put it, "*they're ordinary people who happen to have cystic fibrosis*". Often using similar language, all professional respondents spoke

in terms of goals for those living with disorders or impairments of having a fulfilling life, a good quality of life, or a normal life.

At the same time, they also made the point that people living with these conditions might face more difficulties than might others in living a 'normal life', because of the consequences of the condition. While some noted that the limitations associated with the conditions in this study could vary widely in their severity, there was no view that the only difficulties experienced by affected individuals might be medical ones. All professionals suggested that the experience of living with a condition might include social and psychological consequences, and that some problems, such as poverty or the way that people were treated, could be seen as socially caused. The majority pointed out that the experiences of people living with these conditions could vary widely, because of the various factors which might affect quality of life.

In discussing the needs of individuals living with a disabling condition, a number of respondents offered their perceptions of the experience of those affected, or factors which they saw as important in experience. Within this group of professionals, while all were close to those with impairments, there were different views about what the important problems of people living with these conditions might be, about the causes of such problems, and about how these might be addressed. These differences in professional views related both to the particular implications of the condition being discussed, but also to respondents' professional perspectives and their perceptions of their role.

Views about the experience of living with a condition

Medical professionals, while noting that a large part of medical care was concerned with social, emotional and psychological aspects of life, emphasised that a number of the problems facing those living with certain conditions might be seen as stemming from the condition. All discussed the beneficial nature of medical interventions or treatment, not only in prolonging life but in enhancing quality of life, and consultants pointed out that trying to alleviate physical problems was their role:

If they develop any sickness or any complications, then that needs to be sorted out. And that's what primarily we're here for, you know. To try and put right anything that goes wrong. And there's a lot that can go wrong with children with (condition). And young people with (condition).

(Consultant physician)

Professionals in the field of cystic fibrosis particularly mentioned what could be done by treatment, both now and in the future and by specialist centres, to improve the life-expectancy, health and quality of life of those affected with this disorder. One

consultant noted the costs of treatment and the difficulty of providing ideal treatment for all. All suggested the potential interference of the condition and of the treatment regime on both psychological well-being and the ability to live a normal life.

The majority of respondents, however, pointed out that 'problems' for those living with a condition should not be assumed, that it was not possible to generalise about the experiences of those affected, since all were different, and that quality of life was a matter of subjective perception:

I don't think anybody can ever put themselves in the shoes of a disabled person. That is very hard. Because it all depends, psychologically, on that individual, how they see themselves.

(Consultant physician)

Many professionals, especially those in the medical field or support groups, stressed the importance of individual attitudes, whether practical strategies or outlook, on quality of life. A number gave examples of people with a positive approach towards living with a condition or impairment, and others spoke more generally:

I think disability's like people, it affects people in so many different ways. A lot depends on your attitude to it, you know (...) and it's to do with ... people inspiring you and encouraging you, you know.

(Disability professional)

Although, as here, some disability professionals discussed quality of life and individual approaches, others did not refer to individual difference or subjective perception. One disability professional argued that disabled people did face problems, and described the experience of disability in these terms:

For people with a physical disablement, what's actually disabling us is the barriers that we encounter; by that I mean the physical barriers, the organisational barriers, the way that an organisation is structured. Or it can be attitudinal barriers, which is the big one. Because I think that's important and I think that service providers have a lot of difficulty with that.

(Disability professional)

This response reflects a 'social model' of disability, in which the important problems experienced by those with impairments may be seen, not as inevitable and stemming from the impairment, but as socially caused and hence as potentially solvable with social change. Disability professionals in this study, by reference to their personal or professional experience, discussed the way that they felt disabled people were treated, and suggested the possibility of different treatment, both in terms of rights, others' attitudes and the degree of support offered.

Professionals in various fields suggested that a number of the difficulties faced by individuals living with either Down's syndrome and spina bifida could be seen in this way. Several raised the issue of the lack of appropriate or supported employment for people with these conditions (although noting the general problem of unemployment). They stressed the importance of integration and equal opportunities for people with disabilities and especially learning disabilities; those working in this field emphasised the need for increased resources for special needs, and a number were pessimistic about the possibility of improved funding. Several also suggested that people with these conditions were often perceived and treated differently from others, whether by individuals or more generally in society. One respondent spontaneously pointed out that people were often reluctant to discuss issues of sexuality with young people with spina bifida, saying, *"there's a taboo on things like that"*. Another discussed the fact that the quality of life of young adults with Down's syndrome might be strongly affected by the way that they were treated:

Well, I mean obviously, it has got physical and mental disabilities associated with it. But there is also the disability which is imposed by society. But I think many people can have disabilities of all different sorts, but they're able to get on and have a full life. I think it's often the rest of so-called normal society that actually causes a lot of the difficulties and unhappiness, you know, of disabilities.

(Consultant psychiatrist)

Some professionals in the field of disability, in some cases drawing on their own experience, suggested that the most important problem faced by those with impairments was that of being treated as 'different', arguing that they would see the main barrier to a good quality of life as *"the stigma of being disabled"*. From a cultural approach to disability, it has been argued that those with impairments are seen as 'other' or as abnormal, rather than as part of a continuum of ways of being human. It has been further suggested that it is seen as a 'tragedy' in society to have an impairment, an assumption which can be seen as based on medical values (Shakespeare, 1995). Some disability professionals in this study did express this view. One suggested that the medical profession *"has problems with conditions it can't cure"*, and a few related their experience of medical pressure to be 'normal':

Prof: Everybody's got to be perfect. I mean, the pressure's on you, because you're not perfect. And even if you can't walk, they're trying to make you normal and making you walk.
JG: Yes. Have you found that?
Prof: Oh yes, yes I have. You know, even when I was younger, I had to get put in callipers and braces and made to walk.

(Disability professional)

However, not all disability professionals took the view that a condition was irrelevant, or that medical interventions were unwanted. Another explained a different theoretical position:

I think it (impairment) is part and parcel of who you are as a person. And I think it would be very foolish to deny that that has a big impact on you. And, as a person, you need to make sure that you get the right sort of medical support, and that's maybe the quality of support that is needed, and it's really key that people recognise that, and they don't deny it, in saying that, "Oh, my disability is caused by society".

(Disability professional)

Some disability activists have argued for the importance of including subjective experience of the physical in an analysis of disability. However, the social model acknowledges limitations (in an objective sense) in the Marxist argument that those with impairments may be seen as sharing a common social position of disadvantage and a lack of societal support for special needs. From the social model, individuals with impairments should be treated both as a normal person and yet as having special needs.

While professionals expressed very different views about the way that they saw the experiences of young people living with a disabling condition, in discussing the needs of such individuals, all used very similar language. They emphasised that someone living with a condition was a normal person, with a need to live a normal life, but also someone with a difference, who might have special needs.

Needs: normal but different

While everyone is both 'normal' and 'different', and a concern for the balance between equal rights and personhood and individual difference is a general concern for society, where people have a disorder, there are not just concerns about rights, as with any minority group, but about welfare. It might be felt right to acknowledge the condition or the difference, to consider whether limitations should be acknowledged, supported or treated, whether by medical or social interventions. At the same time, in the interests of the quality of life of the person, or from a perspective of rights, it might be felt right for difference to be ignored.

The difficult balance between the treatment of the person and of the condition has been discussed in relation to the social model, but this is also a concern for medical professionals, concerned with the welfare of the person as well as with the treatment of the condition. In discussing the experiences and the needs of those living with a condition, the responses of professionals in all fields suggested contradictions around the way that they might be or should be treated. Many also suggested dilemmas for individuals themselves.

Professionals noted possible contradictions for individuals affected with a condition in balancing their needs as a person with those stemming from the condition, or their sense of themselves (or their identity) as a normal person but yet as someone with a disorder. Several offered their view that it was important for young people to acknowledge - and often to manage or monitor - their condition and organise their lives around possible limitations, yet at the same time to *"forget about their disabilities"* and *"get on with their lives"*, and aim at a normal life. It has been noted in the literature that, for those living with a condition, social and medical (physiological) imperatives might conflict (Williams, 1993). Several respondents mentioned the need for 'strategies' or noted that people had their own ways of resolving these contradictions.

A number discussed the importance of interpersonal relationships in quality of life. Several noted the benefits for those affected with a disorder of being treated as a normal person, but they also spoke of the need for appropriate support, and some discussed the difficulties of this, especially for parents. At the same time, the responses of many suggested contradictions for society more generally. For example, one disability professional reported feeling different and excluded in situations where no allowance was made for impairment:

You know what I mean, you don't want to feel different from the rest, you want to feel included. But you're not. You're different and you're made to feel different.

(Disability professional)

The accounts of many in this group illustrated some tensions and dilemmas for professionals, and sometimes for themselves, in treating people with impairments or with the conditions in this study as ordinary people with the same rights and needs, desires and feelings, as others, and yet in acknowledging the condition. Many spoke in terms of a rhetoric of needs, choices and opportunities, but suggested some difficulties in balancing attention to rights and attention to welfare, the social and the physical, the political and the personal. There were many examples of tensions and contradictions, especially in relation to independence, employment and education. Several discussed the issue of special or integrated education, and some mentioned the general fact that giving appropriate attention to difference implied extra attention or, in some cases, unwelcome scrutiny:

The thing is <u>other</u> children are not assessed for ability at the age of five, they are not given a psychologist's report at the age of five, they're not looked at their future as if ... They're not looked at the same ... you know?

(Coordinator, voluntary organisation)

Several respondents suggested particular contradictions around questions of child-bearing. Many pointed out that women with the conditions in this study, or disabled

women, have the same feelings and possible desire for children as any other women, and that it is important that these feelings are taken seriously. At the same time some pointed out that, for some women with these conditions (although there might be assumptions about problems and in spite of individual differences), child-bearing might have negative consequences for the child and perhaps for themselves. The response of one health professional illustrated a tension between 'normality' and 'difference':

... So we do try and get contraception ... We're not saying to them, "Don't have sexual activity". They're normal people like everybody else, and we can't take that away from them, but our job is, if it's ... well, doesn't matter who it is, our job is to advise them.

(Health professional)

Reproductive decision-making can be seen as a private, personal matter, but may also be a matter for professionals and public policy. Screening programmes can be seen as a way of dealing with conditions, but they may also be seen as ways of dealing with people. In discussing the morality of 'prevention', professionals suggested similar tensions between rights and welfare, normality and difference. This issue will be returned to later in the chapter.

The meaning of 'severity'

Those professionals who worked with people affected with Down's syndrome, spina bifida and cystic fibrosis were asked whether they would see the condition as serious, and others were asked what they would see as a serious condition. Respondents were aware that the study was looking at people's views about screening programmes, and the answers of many suggested that a question about severity had been anticipated. Most indicated, whether explicitly or not, that 'serious' might be defined in different ways, or offered an explanation for their opinion.

Professionals in the field of cystic fibrosis suggested that they would see this disorder as serious. Most also noted, however, that the subjective perceptions of those affected and their families might be different from their own, some suggesting that these people's views might be affected by their 'strategies' for living with the condition. In contrast, consultants in particular focused on more objective measures of relative severity, and - while pointing out the wide variation in physical symptoms and the improvement in outlook for people with cystic fibrosis - they unhesitatingly described cystic fibrosis as serious, explaining their views by giving statistics on survival rates:

Let's face it, very few of them really live a normal life-span. However they live. So the answer is that the statistics show that it is a serious condition.

(Consultant physician)

The majority of other professionals, however, were reluctant to give a view about the seriousness of different conditions, for a variety of reasons. A number pointed out that 'severity' was a matter of subjective perception. Several interpreted a question about severity in terms of the consequences for the person affected. While, from different perspectives, possible problems were seen differently, many noted - as in earlier discussions - that the quality of life of those living with a condition might not necessarily be poor, that a number of factors might influence experience, and that the consequences could not be judged or estimated by outsiders:

Um ... I would tend to take my starting point from the people I was dealing with, and whatever kind of situation. Because I can't really know what it's like to have (disorder). You know, I'm not a bit able to know what it's like. I can only assume things, or listen to people involved.

(Coordinator, voluntary organisation)

Others suggested that the 'severity' of a disorder might be experienced by others, especially the families of those affected; this also might be a matter of perception. A number of professionals, in talking more generally, had spontaneously mentioned possible implications of certain conditions for others, especially family members. A few were careful to point out that 'serious' could be defined in many ways:

JG: Would you see Down's syndrome as a serious disorder, yourself?
Prof: As a serious disorder? We were talking about this last night ... and we were saying how much our lives have been enriched by people who have Down's syndrome. We're saying that from a position of being paid to work with people with a learning difficulty. We enjoy working with people with learning difficulties. We don't live with people with learning difficulties.

(Professional in special needs education)

While many were reluctant to give a view about severity, one respondent emphatically described Down's syndrome as serious, saying *"Yes. Very. With massive implications"*. This definite and terse answer, with a lack of explanation, suggests a possibly less acceptable view, or one that would not be discussed further. This was also suggested as a personal assessment, and, in general, most professionals concentrated on more general arguments.

One disability professional asserted, however, that Down's syndrome was not a serious condition:

JG: Would you see Down's syndrome yourself as a serious disorder?
Prof: No. It's not serious at all. It might be serious for the person themselves, is it Down's syndrome that affects your heart?
JG: It can be, I think.
Prof: So medically it can be serious for the actual person being born, Down's syndrome, but, no, actual Down's syndrome is not serious.

(Professional in the field of special needs)

Asked which conditions might be seen as serious, this respondent felt that it was not possible to suggest any condition. The firmness of this response indicates a challenge to those who might argue the 'severity' of certain conditions. The acknowledgement of possible difficulties for the affected person suggests that this is not the question at issue. Some disability activists and some sociologists have argued that disorders are constructed or defined as problematic, whether for individuals or for society. Such values (often seen as 'biomedical') are seen as implied by assumptions of the benefit of 'prevention'. In the reference to 'the person being born', the response of this professional can be seen as relating to screening programmes.

The range of views among professionals about the way that the implications of living with a condition should be seen, and about the meaning of 'severity' was further reflected in their views about screening programmes.

Views about screening programmes

All professionals were asked their views about screening programmes generally, and, where appropriate, in relation to reducing the incidence of the condition with which they worked. Several asked whether it was their personal or their professional views which were wanted, many pointing out that their organisation did not have a policy on screening programmes, as decisions were felt to be a matter of individual choice. Most professionals discussed the general question of whether or not it was right for screening programmes to be offered, and gave reasons for their view. While some argued strongly for the principle of individual choice, others gave their view of whether they would see 'prevention' as wrong, as unjustified, or as right within limits These opinions were coherent with their views about the causes of problems for those living with conditions, and the extent to which they could be addressed.

'Prevention': conflicting views

Some disability professionals criticised the principle of offering prenatal diagnosis and abortion for any condition, pointing out that screening programmes were based on value-judgements; one respondent suggested that, as with the Nazis, they indicated traits seen as unwanted, or *"what we find abhorrent"*. In the literature, disability activists, among others, have argued that the existence of such programmes can be seen as eugenics, or as discriminatory, since they call into question the rights of those with impairments to exist (Davies, 1987; Morris, 1991; Wolfensberger, 1994). This view was expressed by some disability professionals in this study, who felt that screening programmes could be seen as defining those with impairments as socially problematic:

I have a lot of problems with the idea of therapeutic abortion on the grounds of disability, because, as a disabled person, and I'm being asked my opinion. What does that say about me? The fact that I'm here, is that a problem that should be screened out?

(Disability professional)

Some have argued that screening programmes can be seen as premised on the logic of a 'medical model' which can be seen as defining both 'problems' and 'solutions' (Lippman, 1992b; Asch, 1994; Shakespeare, 1995). Not only has this model be criticised as reductionist, but from the social model of disability, the important problems of disability have been seen as not inevitable. Disability professionals in this study drew on these arguments to suggest that 'prevention' might not be felt to be justified or necessary. One respondent also noted the alternative approach of rights, rather than concerns or assumptions about welfare and responsibility:

Well, as a disabled worker myself, I think that ... disability, if you took it out of its medical context, and put it in a social context, and you say, "There is a disabled person", and, "There's someone who's going to have a disability", um, and I think ... well, not exactly, "So what?" ... but, I mean, that they've got just as much right to be here. ... So, I think, why prevent disability in the first place?

(Disability professional)

However, an opposite view of screening programmes as offering potential benefit was suggested by other professionals, especially those in the medical field. One consultant offered the view that it was right to try to reduce the incidence of certain conditions in the interests of society:

Well, I mean, it is ... the world ... as far as Western developed society is concerned, the less children they produce full stop. And the less handicapped children they produce, I suppose, even better.

(Consultant physician)

This consultant was the only person in the study to suggest that screening programmes or 'prevention' might offer societal benefit. Others in the medical field indicated that they would see screening programmes as possibly beneficial but cautioned against an over-optimistic view that certain conditions could be eradicated by this means. The responses of professionals in all fields indicated that screening programmes could be potentially helpful in avoiding suffering. However, almost all raised the question of appropriate limits, some noting the possibility of testing for IQ or behaviour:

If someone's going to have a permanent severe disability, that's obviously a major issue for ... for that person, and it's going to affect all of their lives. But then if you start screening, if you start testing babies for red hair or something like that or (inaudible) and all that kind of thing. Then you're getting something like, "Where do you draw the

line?" ... So why screen for these things? If you're not going to prevent the birth if you find it?

(Professional, voluntary organisation)

As illustrated here, several raised the question of why screening might be offered for characteristics or less 'serious' conditions. They made the point that, if a condition could not be seen as serious, then preventing the births of those affected - who might be seen as 'different' - might be seen as a drive towards perfection. One respondent expressed concerns about societal consequences:

You know, it is sort of scary, this notion that you're screening out people who have something wrong with them. And you do kind of wonder what ... what we'll be left with. And how tolerant we will be, as a society. And you kind of feel, "Well, not very", if you're going to eradicate people who are different. But then, having said that, we wouldn't ... who would avoid opportunities to ... to do away with - you know - syndromes or disabilities where people are plainly suffering? Leading short and painful lives.

(Development worker, voluntary association)

Kerr and colleagues have pointed out that the question of 'where to draw the line' was a key concern for respondents in various fields in a study examining issues concerned with genetics (Kerr *et al.*, 1998b). In their study, a number of medical professionals emphasised a distinction between 'ill health' and 'social problems' (Kerr *et al.*, 1998c), although discussions with others highlighted the difficulty of drawing such boundaries (Kerr *et al.*, 1998b). In this study, the majority of professionals noted the different moral arguments and discussed the difficulties of balancing them.

A few expressed views about whether or not they would see it as right to offer screening programmes for certain conditions. All professionals in the field of cystic fibrosis felt that it was right or acceptable for a screening programme to be available, although noting the arguments for and against, and consultants felt that screening before conception might prove to be a more acceptable, and hence more feasible, approach.

There was little discussion about whether it was right to offer screening programmes for spina bifida or Down's syndrome, whether because these are already routinely offered, or whether because their acceptability was largely not disputed; one health professional felt that these conditions might be seen as more serious than cystic fibrosis. However, an alternative view was presented by some disability professionals, who argued that there might be assumptions within the medical profession both about suffering and prognosis:

You know, when I was a child, a lot of folk with spina bifida weren't surviving. Their kind of thinking was then, "It's only for that short period, it might be a miserable life".

(Disability professional)

On the other hand, other disability professionals discussed the question of screening programmes in a more personal way, mentioning the concerns of parents and implications for them:

I've still had a good life. You know what I mean. ... So, you know. I think that the worry is more for my parents. I mean, they worried, how I was going to manage and how I was going to survive. And I think, certainly, if my mother had had the choice, she would have, probably, terminated the pregnancy. Just for the fact of going through all the hassles.

(Disability professional)

The birth of a child with a disabling condition or chronic illness has implications for others, especially family members. It has often been pointed out that screening programmes are offered in a social climate of individual responsibility (Rothman, 1985; Morris, 1991). A number of professionals, especially those in the field of disability, discussed the support which they felt might be available. While some felt that support for those with impairments was improving, others, especially some working in the field of learning disability, were less positive about service provision. From this position, one respondent argued that she would not see it as wrong for women to choose abortion on the grounds of a lack of resources, from, *"a notion of some sort of fairness - to the child and to yourself".*

Hence professionals consulted for this study offered a range of views and arguments, sometimes strongly expressed, about whether 'prevention' should be seen as wrong, as unjustified, or as possibly beneficial within certain limits. However, all suggested that they would see it as right for decisions about prevention to be a matter of choice.

Choice

Several professionals strongly emphasised the principle of choice. The argument for choice and the 'right to choose' was most firmly expressed by the geneticist, who did not suggest views about 'prevention', but rather noted individual differences in perceptions and circumstances:

I believe very strongly in the idea of free choice. Decisions that are right for one person are not right for another. And decisions that are right at one stage in life may not be right at another. So people should be allowed to make their own decisions. I think any geneticist would say the same thing.

(Geneticist: notes)

Some have argued that pragmatism may offer an ethical approach to the dilemmas of reproductive technology (Kitcher, 1996), or that reproductive decisions should be a matter for the individual, and official guidelines have been phrased in terms of autonomy (Royal College of Physicians, 1989). Professional respondents who

emphasised the importance of individual reproductive choice stressed the voluntary nature of screening programmes and most did not raise concerns about values. Asked about the possibility of others' attitudes of blame, one respondent made the point that a policy of freedom of choice and responsibility would include the idea that some might not agree with others' choices:

I would hope that, as a society, we're going to be open-minded enough to know that people have got the right to make decisions. And, once the decision's made, to take the responsibility for that. I hope that would be the case.

(Professional in special needs education)

Among those who emphasised the value of choice were those professionals who, throughout the interview, had emphasised values of independence and autonomy in discussing living with a condition. Some were disability professionals, one of whom defined both 'severity' and quality of life in terms of autonomy:

Just independence and freedom, you know. If people feel that they are free - as free as any of us are to do what we want, you know - then I think that's a good quality of life. And having choices.

(Disability professional)

While a number of disability professionals discussed difficulties of disabled people in terms of the social model, seeing difficulties as socially caused, only a minority suggested that they would see the provision of screening programmes as unnecessary or wrong. Kaplan (1993) has noted that many in the disability rights movement, while expressing concerns about screening programmes, themselves support values of rights and autonomy and defend a woman's right to take reproductive decisions. This was the view of one disability professional in this study who argued that the values on which screening was based were wrong, that the implications of conditions were portrayed negatively with screening programmes and within society, and that there was no condition for which abortion would be justified, but nevertheless felt that screening programmes should not be banned, and that it was right that choice should be offered:

... But I do ... I do worry about a kind of ... a lack of balance in this, I do worry about a kind of ... a dogmatic view which says that, "I know what is best for you people, therefore you shouldn't have the test done".

(Disability professional)

Those who emphasised autonomy suggested the right of individuals to information, making the point that it would be wrong to withhold information which was available, or that screening tests should be provided for all who wanted them. Several also suggested information as generally beneficial, as long as care was taken about how the information was given. A number noted that a desire to find out information, whether from counselling or screening programmes, was a separate matter from a decision to

prevent a birth. Support group workers noted that prospective parents could find information about an unborn child valuable or useful, perhaps in order to come to terms with a condition before the birth of the child.

The geneticist interviewed for this study was positive about the benefits of screening and counselling for families with inherited conditions, and the option to avoid the birth of an affected child. A similar view was offered by a health professional, who suggested the usefulness for those at high risk to find out information:

We certainly encourage (counselling). For a parent who has had a child with (condition), we will encourage them to go and see the genetics people. And then they'll be able to ... they'll have some idea of what the risk is like if they wanted to have more family.

(Health professional)

The response of this professional can be seen as suggesting a retraction of the outcome of screening in terms of responsibility or 'prevention'. The goal of screening programmes in terms of reducing the incidence of conditions was little articulated, the purpose of offering screening programmes and counselling generally being seen in terms of offering information and allowing individuals to take their own decisions.

Limits to choice

However, while all suggested that reproductive decisions should be a matter of choice, rather than laws, the responses of a number of professionals suggested some difficulties with the concept of 'informed choice'. They expressed concerns about the possibility of pressure on choice, but a number also questioned the adequacy and beneficial nature of information offered either by or with screening programmes.

Several expressed a concern about the provision of choice when the information provided was felt to be inadequate; as one respondent commented, *"the choice is there but the information's not as it should be"*. They stressed that screening programmes should be offered with the most complete information about the risks, the implications of tests and about the disorders, although from different perspectives. For example, many felt that more information was needed about the implications of conditions, and the degree of support which might be expected:

I think what I would like to see is the option to test. But I think that would have to be accompanied by much, much better information for the people that are being tested. ... I would want to talk to them about services, I would want to talk to them about the lack of support that's available for them, but I would also want to talk to them about what is possible, and what families have achieved, and what young people have achieved.

(Disability professional)

A number of professionals raised concerns about the test results themselves, mentioning particularly the uncertainty of some information. Some in the medical field also questioned the effectiveness and accuracy of antenatal or genetic tests in predicting either the existence or absence of some conditions, or the prognosis of those affected. Those working in the field of spina bifida noted the range of severity of neural tube defects, and pointed out the possibility of false positives and false negatives; as one argued, *"there's a lot of refining that needs to be done if you want to produce a test that is accurate, if people are going to act on it"*. In a similar way, those working in the field of cystic fibrosis referred to the complexity of genetic knowledge. Consultants discussed the limitations of DNA testing generally, and one expressed concern about the difficulty of conveying the uncertain implications of the results:

From my limited knowledge of these things - cystic fibrosis is a good example here - the relationship on an individual level between genotype and phenotype has not been proved. So you end up telling some people who ultimately are going to be really rather healthy, that they have a gene that could make them extremely ill. And that's a ... that's an endeavour that will cause a lot of unnecessary anxiety. So I think we should be careful, as a profession, what sort of information we give to people about their genes.

(Consultant physician)

Concerns about the effectiveness of tests, and the questionable benefits of information have been raised in the medical and psychological literature, and have been discussed in Chapter 1. Where health professionals raised ethical issues about offering diagnostic testing, it was sometimes unclear whether their concerns related to negative consequences for individuals or to the general question of why it might be felt right for screening programmes to be offered. As one expressed it, *"If ... if a person's not going to act on a test, and by that I mean carry out an abortion, for religious reasons, then I need some proof that the test is going to be of value"*.

Several respondents questioned whether the provision of information was always beneficial. Those who discussed the value of screening stressed that information and choice must be wanted, and that a decision to seek counselling should come from individuals themselves. A number emphasised that the role of staff (or their own role) was to respond to requests for information and not to offer advice, although some also noted the difficulty of conveying information in a neutral, objective way. In this context, some pointed out that information in itself might cause anxiety or a pressure to act on it:

... But then if people do find out, then they have the dilemma of do they want to make a choice or will they then wish they didn't know, or would they then feel pressurised into ... for whatever reason, not necessarily because they're being pressurised by someone else, but just the pressure of the situation.

(Coordinator, voluntary organisation)

Particular concerns about unwanted information and decision-making were mentioned in relation to prenatal diagnosis, some professionals suggesting, from their experience, that a number of people took part in antenatal screening programmes without an awareness of the implications. Others expressed concerns about a pressure to take part, whether from the fact that screening programmes were routinely offered, or from the fact that they were known to be available.

Pressure towards termination of a pregnancy was also suggested by some disability professionals. Several felt that the information offered could be over-negative or biased in focusing on or magnifying problems. A few further argued that the attitudes of medical professionals constituted a more active pressure on women towards termination of an affected pregnancy:

I think that when women have the test and know there's something wrong and they still decide to have this child, it's pressurised from the very beginning. And it's seen as a tragedy. And I think, instead of that, I think the pressure should be taken off women; there should be a choice. And put over as a choice. ... Because I think, even, a lot of people can put pressure on you, and very much the medical profession.

(Disability professional)

Many have argued that screening programmes can be seen as resting on assumptions about inevitable problems associated with impairment. However, one respondent suggested that 'opt-out' or routine screening programmes contained assumptions about individuals' or women's ability to care for and cope with a disabled child, since a decision to give birth to an affected child became a matter of 'opting-in'. As this respondent put it, *"It is a worrying distinction if you begin to think that there are people who can cope and people who can't. You know, there's a different type of person, and the ones who opt in are the saintly ... the remarkable, coping ones and your average person couldn't. Because there would be no truth in that".*

Hence a number of professionals in this study, although from different perspectives, suggested the potential for individuals' decisions to be influenced. For example, one medical professional, in favour of prenatal diagnosis for serious conditions, noted the cultural unacceptability of abortion, especially in some parts of the UK, compared with other cultures. Another respondent, from an opposite perspective, suggested that there were fewer problems than previously for those caring for a disabled child, but also discussed the media portrayal of disability:

Abortion is there. People can go either way. Whether programmes have been successful in portraying ... hopefully the media are portraying a more positive image of disability. So there's hope that people will maybe go the other way, And also because of the changed care and service support for people with learning disability is changing. So that parents may feel more ... are more encouraged to keep the child.

(Professional in the field of special needs)

A number of professionals, therefore, while emphasising the principle of individual choice, also indicated some discomfort with this position. They suggested concerns about the adequacy of the information but also about possible pressure on choice, and offered their own perspective or arguments about the morality of 'prevention'.

Discussion

The morality of screening programmes is a 'public issue', debated in the media and in the literature. One issue concerns policy about the provision of screening programmes and appropriate limits to conditions for which 'prevention' might be available and to autonomy. Professionals consulted for this study discussed screening programmes in these broad, theoretical terms. These respondents could be seen as an 'elite' group. While some made a distinction between their own position and that of their organisation, the concern of all professionals was with the general issue of policy. Their responses were expressed in general terms, public statements or rhetoric, discussions of right and wrong.

The views of professionals reflected the range of theoretical views in the literature about the morality of screening programmes; these views can be seen as illustrating the parameters of the debate. A few disability professionals saw 'prevention' as wrong or as epitomising discriminatory values. Others, often on the basis of personal experience, suggested that they might not see 'prevention' as justified or necessary, whether because the quality of life of those affected might be good, or because problems were perceived as not inevitable. However, while a number suggested that the experiences of those with impairments might be seen in terms of the 'social model' of disability, and difficulties socially caused, only a minority expressed disapproval of screening programmes or suggested that 'prevention' was always unjustified. Most respondents from various fields indicated that they might see the provision of screening programmes as potentially beneficial, but within certain limits. There was no view of screening programmes as offering unqualified benefit or as being cost-effective (as in a theoretical 'biomedical model'). Most considered how the different moral arguments should be balanced in an ethical way.

All professionals felt that it was right for decisions to be made by individuals, and several emphasised the principle of choice. On the other hand, several - although from different perspectives - suggested potential difficulties for individuals or with this position. They suggested concerns about the nature and the adequacy of information offered with screening programmes and suggested the possibility of pressure on choice. Several indicated the way that they felt choice should be exercised, using phrases such

as, *"I would want to say to them ... "*, or indicating arguments felt to be important. There was little discussion of concerns about individually wrong choices.

Screening programmes offering 'prevention' (while also offering choice) may be seen as suggesting a societal approach to dealing with disorders or those with disorders. While all professional respondents can be seen as close to those with disorders, they emphasised different approaches to 'prevention' of rights or welfare, coherent with their professional role or standpoint. However, few offered a prescriptive view. As discussed, the accounts of many professionals in all fields had indicated a recognition of the difficulties of balancing a concern for the person with an acknowledgement of difference in discussing the way that those living with a disorder should be seen and treated.

Professionals were asked general questions, not about their own predicted behaviour, unlike other respondents, or young women, in the study. However, a few volunteered their own experiences of being offered antenatal screening, in some cases describing the decisions they had made and their reasons. This might suggest an acknowledgement, by some, of the fact that reproductive decision-making is a personal and private matter, as well as a political and public issue, that theoretical responses alone may be felt to be inadequate for this complex issue. Where professionals readily discussed their own experience, it was usually to explain why they had refused screening tests, and to suggest other values or factors, such as religious views or a commitment to the pregnancy. These respondents did not discuss their own circumstances or qualities. The views of this group will be compared with those of others in the study.

CHAPTER 7

WOMEN WITH DOWN'S SYNDROME

One of the main conditions for which screening programmes are offered in pregnancy is Down's syndrome, and there is an emphasis on this condition in the medical literature. It was felt important, therefore, to try to include women with this condition in the study, if an appropriate method could be found.

A number of professionals, mainly in the field of learning disability, were asked whether they thought that it was feasible or right for women with Down's syndrome to be asked questions on the topic of reproductive issues. Their views, and the process of access, have been discussed in Chapter 5. It was decided that the aim should be to ask women with Down's syndrome about aspects of their lives, and their views about having children, as far as possible in the same way as with other women in the study.

The first section of this chapter outlines the way that the implications of Down's syndrome may be seen from different perspectives, and describes recent changes in policy and intervention. The second section describes the views of professionals consulted for this study and in the literature about the lives of young people with Down's syndrome and the effect of changing philosophies. The chapter presents data from interviews carried out with women with Down's syndrome, and concludes by considering whether the attempt to include them in the study was felt to have been right and worthwhile.

Background

As outlined in Chapter 1, Down's syndrome is associated with certain physical characteristics, with a degree of learning disability and, in some cases, with health problems. The quality of life of individuals with Down's syndrome may be no different from that of others (Goodey, 1991), although some may experience difficulties such as mental health problems or depression (Carr, 1994).

However, possible negative consequences of living with Down's syndrome might result from social causes such as prejudice, or a lack of support and opportunity, as well as from physical or intellectual limitations (Stratford and Lane, 1985); as Booth (1985) has noted, the extent to which impairments are a handicap depends on the degree of support received. From Goffman (1968), certain attributes might be seen as unwanted or stigmatised in society, and people with these conditions seen as an unacceptable type of person. This stereotypical view can legitimate inappropriate treatment and

discrimination. People with learning disabilities generally have traditionally had few rights, even the right to life, since learning disability has been seen as a problem which should be eradicated (Craft and Craft, 1981; Stratford, 1991).

After the Second World War, in Scandinavian countries, a concern for the equal rights of people with learning disabilities was expressed in the philosophy of 'normalisation'. This was defined as, "making available to all mentally retarded people patterns of life and conditions of everyday living which are as close as possible to the regular circumstances and ways of life of society". (Nirje, 1980: 33; cited in Emerson, 1992: 2). The concept of 'normalisation' was elaborated by Wolfensberger, in the US, drawing on the work of Goffman and on the understanding of disability as a human rights issue. Wolfensberger proposed a definition in terms of 'social role valorisation', in line with a growing interest in the way that people were perceived; "the creation, support and defence of valued social roles for people who are at risk of devaluation" (Wolfensberger, 1983: 234; cited in Emerson, 1992: 5). The goals of normalisation are prescriptive; people with learning disabilities should be valued and treated in the same way as all human beings (Wolfensberger, 1994).

While the principle of normalisation is said to be espoused by groups both 'of' and 'for' people with learning disabilities (Emerson, 1992), the concept has been criticised within sociology (as have interactionist theories of disability more generally) for its lack of explanation of why those with disabilities or learning disabilities are little valued, and for the implication that change is possible by change of attitudes alone. Some have criticised the functionalist assumptions within 'normalisation' of shared norms and values, arguing that 'culturally valued roles' can be seen as reflecting the values of the dominant or advantaged group, and masking conflicts of interests (Brown and Smith, 1989). The emphasis on the devalued status of people with learning disabilities has been seen as tending to moralism and exhortation (Dalley, 1992), and the focus on training to change attitudes as of more use to professionals than to people with learning disabilities themselves (Whitehead, 1992). The principle of normalisation has been criticised as precluding the development of a collective response to discrimination (Chappell, 1992; Brown and Smith, 1992). It has also been argued that an approach based on unconditional love, with no limits, places an unacceptable burden on carers, about whose needs normalisation is silent (Smith and Brown, 1992).

From the social model of disability, the problem of disability is located in the social structure of society, the way that society is organised, and in its processes of inclusion and exclusion (Rioux, 1992); disabled people are seen as sharing a common social position of disadvantage. From this more political analysis, people with learning disabilities have the same needs, expectations and rights as others, but may face

additional barriers, for example stigma and prejudice, but also institutionalisation, difficulties with education and employment (Clare, 1990) and economic disadvantage. Disability can result not just from being treated differently or as less valued, but also from a lack of acknowledgement of special needs for support.

A collective response to disability has also been suggested in terms of a collective identity. Whereas integration into the mainstream might imply assimilation, conformity, denial of deviance and a negative concept of disability, it has been suggested that a social model of disability might offer a potentially helpful positive identity as different, pride rather than 'passing', as with other devalued or political minority groups (Szivos, 1992a). Some have argued that an exploration of a disabled identity can offer an alternative strategy for self-adjustment (Szivos and Griffiths, 1990), as well as the possibility of making choices which are not regarded as valued in society (Whitehead, 1992).

Changing philosophies towards learning disability have emphasised social inclusion and equal rights and opportunities, but also attention to special needs. Rather than assuming needs or problems, on the basis of a medical condition for example, the focus has been on the importance of examining the views of disabled people themselves, the needs of individuals as they themselves define them, and self-advocacy (Clare, 1990), although more recently - as more generally - ideas of empowerment, self-help and self-advocacy have come to be associated with the individual.

In relation to Down's syndrome, changing philosophies based on meeting the needs of disabled people, and new knowledge about the condition have led to changes in policy and new possibilities for intervention. Over the last twenty years there has been considerable research into both clinical effects of Down's syndrome and into the psychological and educational development of people with this condition (see Lane and Stratford (eds), 1985). Neonatal surgery may correct a heart defect and other health problems may be alleviated; cosmetic surgery may also be available, although this is more controversial. Although at one time corrective surgery was withheld from babies with Down's syndrome (Silverman, 1985), there is increasing acknowledgement of the right of a person with Down's syndrome to the same health care as others (Wishart, 1993). Life expectancy for those with Down's syndrome has increased markedly in the past 25 years (Nicholson and Alberman, 1992; Wishart, 1993), and it has been suggested that this may be due, not only to improvements in health care and particularly neonatal surgery, but also to a willingness to use it because of changes in cultural attitudes (Editorial, 1990b).

There has been much recent interest in the psychological and intellectual development of people with Down's syndrome, especially of children (Buckley, 1985; Stratford, 1985; Dykens, Hodapp and Evans, 1994). Studies have suggested that the degree of learning difficulty is very variable (Carr, 1994), and that many people with this condition would be classed as having a moderate or mild degree of retardation (Booth, 1985). Empirical work indicates a wide variation in the intellectual development of people with Down's syndrome (Carr, 1994; Dykens *et al.*, 1994), although little is known about the limits for improvement (Booth, 1985; Buckley, 1985; Wishart, 1993). Recent research in educational psychology and language development has also suggested that people with Down's syndrome might learn in a different way from others (Buckley, 1985; Wishart, 1993), and this work has led to the development of support systems for young children with Down's syndrome. Although these systems are felt to need a high degree of support, often from parents (Brinkworth, 1985), many children with Down's syndrome now read at an earlier age, and it has been pointed out that new learning methods have both challenged assumptions and offered social benefits for people with the condition (Wishart, 1993).

From the mid 1970s, philosophies which focus on needs and rights have been reflected in changes in legislation in the UK. With concerns about segregation, the concept of normalisation has been seen as influential in shaping the provision of alternative services for people with learning disabilities, both by legitimating demands for improvement and by specifying aims and assessing standards of service provision (Clare, 1990; Emerson, 1992). As Chappell (1992: 35) has argued, "the goals of normalisation have become synonymous with the goals of community care". From a focus on integration, the emphasis has been on living in the community, ordinary housing, ordinary jobs, and social and leisure opportunities (Ward, 1992). An 'ordinary life' in the community has come to be seen not only as a right, and as a way of challenging stigma, but also as the most appropriate form of care (Brown and Smith, 1992). Also, although most children with Down's syndrome require special educational provision, with the increasing emphasis on integration, young children with Down's syndrome often attend local pre-school service provision, many are often able to attend mainstream primary school (Lorenz, 1995) and some attend secondary schools.

Changes in intervention, both social and medical, are seen as having improved both the life expectancy and the physical and cognitive potential for people with Down's syndrome (Stratford and Lane, 1985; Williams, 1995a). The Scottish Down's Syndrome Association has emphasised this positive view, noting, "children with Down's syndrome are healthier, more capable and more integrated into the community than ever before. With appropriate care, they grow up to be physically more lively,

socially more competent and mentally more able than was previously thought possible. ... Thankfully today the negative attitudes people once held towards disabilities are disappearing quickly." (Scottish Down's Syndrome Association 1992a: 15).

However, it has also been argued that little is known about the experiences of people living with Down's syndrome (Booth, 1985) and about the quality of their lives (Carr, 1994). As a Lancet editorial has argued, extending life expectancy "cannot be regarded as a medical triumph if the effect is merely to prolong the time-scale of suffering, handicap and dependency" (Editorial, 1990b: 888).

There has been increasing interest by social scientists in finding out the experiences and perceptions of people living with a number of conditions, and some studies have examined a number of aspects of the lives of people living with Down's syndrome (see Lane and Stratford (eds), 1985; Carr, 1994). From a different approach, there has also been a focus on the experience of disability, as noted in Chapter 2, and a number of quantitative studies have examined the equality and the opportunities available to disabled people. A focus on needs and rights has implied an interest in the transition to adulthood. From this approach, one study has concluded that young people with disabilities, including learning disabilities, might experience more difficulties than might others in achieving important goals or aspects of adult life, such as personal autonomy, productive activity, social interaction and roles within the family (Centre for Educational Research and Innovation, 1986).

Living with Down's syndrome: professionals' views

This section outlines the views of professionals consulted for this study who were working in the field of learning disability or with young people with Down's syndrome about the way that they saw the lives and the experiences of those affected. It also refers to literature reporting empirical work. All commentators have suggested that young people with Down's syndrome might face problems, but that there may be ways in which some difficulties can be addressed.

A few professionals reported that, in their experience, depression was sometimes a problem for young adults with Down's syndrome, especially for those with less severe learning difficulties. They suggested that depression might result from an awareness of being different, both in the sense of being recognisably physically different, and in the sense of frustration at the limitations in what they were able to do, and in terms of future opportunities. As one commented:

A lot of them have quite mild degrees of learning disability, so they have an awareness themselves that they are different. And I think that can be particularly painful for a young person with Down's syndrome, when they get to their late teens, early twenties.

(...) I think the other thing in particular with Down's syndrome, rather than with some of the other learning disabilities, is that Down's syndrome is the one learning disability which other people can identify from the appearance of the person. And I think that often causes a lot of distress.

(Consultant psychiatrist)

Most professionals felt that people with Down's syndrome were treated differently from other people in various ways. One - on the basis of conversations with women with Down's syndrome - reported their experiences of being *"bullied or abused in the street"*. This respondent noted their resulting fear of going out, and also their perceptions of being seen as different or unwanted, saying, *"So there is still that stigma there. They're very much aware of it"*.

Professionals working in special needs education discussed the fact that people with all learning disabilities were treated differently from others in the sense of being constantly assessed and given individual attention. One felt that many disliked special treatment:

They don't want to see themselves as being 'labelled' in any way. But on the other hand, they are different, and there are things they can't do. And it really is very frustrating for them. They hate being picked up in special transport. And they don't like the stigma of having gone to a special school.

(Professional, special needs education)

At the same time, these respondents also argued that they would see it as helpful for those who understood that they had learning difficulties to acknowledge possible limitations:

I think it's important that they recognise, themselves, their disability. That they are aware of what that means to them, and what it means for the rest of their life. So that they can come to an understanding and know where they're going in their life as well.

(Professional in the field of special needs)

While it has been argued that to be known as a person with Down's syndrome can be a negative label or 'spoiled identity' (Booth, 1985; Brown and Smith, 1989), an alternative view is that it can be psychologically beneficial for people to understand that they have the condition, as an explanation for possible difficulties (Sefton, 1995), and this was the view taken by a representative of the Down's Syndrome Association. Amongst the majority of those interviewed, it was generally held, however, that the philosophy of their organisation was to look at skills and to meet needs, rather than focusing on the condition. Some felt that most adults with Down's syndrome would be more likely to see themselves as having a 'learning disability'. This was generally felt to be an acceptable term for individuals themselves, although one suggested that it was still seen as a negative term.

Individuals who are living with an impairment or disorder may be seen as having both the same needs as others and as having special needs or requiring additional support because of the consequences of the condition. Not only might people be treated by others as both 'the same' and as 'different' (whether appropriately or not), but the contradictions between normality and difference might cause difficulties for individuals themselves. Szivos (1992b) has suggested that confusion can result from the different messages that people with learning disabilities receive from others about whether or not they are different, and that social isolation might result from not being able to talk freely about possible difference. Some empirical work has examined whether or not people with learning difficulties find it helpful to see themselves in this way. Szivos and Griffiths (1990) found that participants in group discussions did not at first readily discuss 'difference', which was felt to be a taboo subject, but that, given time, people did talk about this, and they described a variety of coping strategies.

In discussing the factors which they felt were important in the experience and the quality of life of young people with Down's syndrome or a learning disability, all professionals consulted for this study mentioned Community Care legislation and its implications. All welcomed the philosophy of integration, since it was felt that many of the problems experienced by people with learning disabilities derived from their past segregation; integration was seen as generally beneficial in its potential for education and changing social attitudes of fear or prejudice.

However, several questioned the extent to which new legislation had benefited individuals with learning disabilities themselves. Some made the point that the increasing integration of young people with Down's syndrome within education did not extend to the wider community. A number noted the difficulties for young people with learning disabilities of taking part in leisure activities, and they felt that many were socially isolated or led restricted lives, as also noted in the literature (Carr, 1994). Although some noted improvements in Adult Resource Centres, the need for opportunities for people with Down's syndrome (and other learning disabilities) for employment or for something useful to do during the day, was stressed by almost all:

I think ... young adulthood remains a major problem (...) I mean, employment's a big thing. That's a big issue for all of us, you know, everybody. Unemployment's hard for anyone, and equally for people with Down's syndrome. That would be good to change. Because there are lots of jobs that people with Down's syndrome can do. It's just - you know - that they have to be set up in a supportive way. There aren't enough opportunities for people with Down's syndrome.

(Development worker, voluntary organisation)

Although people with learning disabilities have always been in the community (Brown and Smith, 1992), with the closure of large hospitals, Community Care legislation offers

the potential for disabled adults to live independently of their families, in supported accommodation, such as sheltered housing or community homes. One view is that more accommodation is needed and that integration is limited in this respect (Brinkworth, 1985). Professionals in this study working with older adults with learning disabilities reported that an increasing number were now living independently, often with support in sheltered housing, and that this was usually successful. However, others noted that living in the community could also have disadvantages, and some suggested that the question of independent living could be a difficult issue for parents. In relation to Down's syndrome, Carr has found, from a number of studies of adults and young adults, that the majority lived at home with their families. Also, while there was a wide range of self-help skills, the parents or carers of the majority had some responsibility for supervision and for meeting everyday needs (Carr, 1988; 1994).

Professionals working in the field of learning disability emphasised a philosophy of autonomy, rights and making choices. Several discussed skills classes which offer information and opportunities to discuss the support which individuals with learning disabilities might need. A number noted, however, that many people with learning disabilities had difficulty in making decisions, because others had done this for them. Self-advocacy groups have evolved within adult education and day services as well as in independent groups of people with learning disabilities. For many professionals, leading discussion groups was part of their work, and one who was involved in organising self-advocacy groups saw this as *"a real step forward ... for some people"*.

Staff running self-advocacy groups or classes on social relationships said that inter-personal relationships and friendships were much discussed at these, and some commented that social relationships could be difficult for some people with learning disabilities. As more people with learning disabilities live in the community, sometimes independently, there are more opportunities for social and sexual relationships (Booth and Booth, 1993); adults with learning disabilities form sexual relationships, become engaged and live together, occasionally as married couples (Gath, 1988; Koller, Richardson and Katz, 1988). Some respondents in this study noted that there was an increasing acceptance by professional staff of close friendships between people with learning disabilities, that these relationships were often taken into account when deciding about accommodation, and that, in Glasgow, professional support was available for couples with learning disabilities. The majority of professionals raised the issue of close and sexual relationships for women with Down's syndrome, and the possibility of pregnancy. Several also discussed the very variable views of parents or carers and possible dilemmas for them.

Craft and Craft, writing in 1981, noted rapid changes in thinking, from an emphasis on preventing people with learning disabilities from having children, for the 'good of society', to a focus on individual rights and on positive aspects of sexuality. While it is increasingly accepted that people with learning disabilities have the same right to sexual relationships as others, it has also been argued that the principles of normalisation might be harder to achieve in relation to sexuality than in other aspects of life, because of the need for those with rights to make responsible choices (Monat-Haller, 1992). There might be problems of abuse or exploitation where there is a lack of consent to sexual activity (Roy and Roy, 1988; McKay, 1994), as well as the risk of sexually transmitted diseases (Monat-Haller, 1992).

There may also be decisions to be taken about parenthood and contraception. Although some expressed concerns about parenthood date from assumptions about the heredity of learning disability (Craft and Craft, 1981; Gath, 1988), others arise because of the difficulty of balancing the responsibilities and rights of parents against the conflicting rights of a child. In the UK, decisions to resolve such conflicts are taken by reference to the Children Act 1989, the emphasis of which is on the welfare of the child (Hayes, 1993; Gath, 1993).

Although it is rare for women with Down's syndrome to have children (Gath, 1988), evidence suggests that the number of parents with learning disabilities is increasing in the UK, and is expected to increase further (Booth and Booth, 1993). Most professionals in this study were unsure what would happen if a woman with learning disabilities became pregnant, and said that this rarely happened. Some pointed out that young women with Down's syndrome were often being counselled over a long period, leading towards sterilisation or contraceptive injections by their carers; the former was reported as still being carried out, although the latter course of action is now more common. Two respondents suggested that some young women with learning disabilities were given contraceptive injections without knowing what these were, although expressing divergent views about whether this was in the woman's best interests. Most felt that it was important for women to take their own decisions about contraception, and some thought that this was beginning to happen. However, one respondent made the point that, although a very able woman with Down's syndrome, living in the community without support, could make her own decisions about motherhood in the same way as others did, this situation was rare, and almost all women with learning disabilities had "*absolutely no choice*" in decisions about pregnancy.

While, in the past, pregnancy for women with learning disabilities was often prevented by sterilisation policies, enforced sterilisation is now widely seen as an infringement of

human rights, but there continue to be concerns about who should take decisions about contraception (Roy and Roy, 1988). In the UK, decisions about sterilisation for women with learning disabilities are made by the courts, the principle being that decisions must be taken in the best interests of the woman concerned (McKay, 1994). Some have argued that court decisions can reflect conformist or paternalistic attitudes (Rioux, 1993), but an opposite concern (mentioned by some respondents in this study) is that women with learning difficulties who seek voluntary sterilisation in their own interests might have difficulty in obtaining this (Roy and Roy, 1988).

Carers of those with learning disabilities have a duty to protect the person's welfare, but normally no right to take decisions on their behalf, or to prevent them taking reasonable risks (McKay, 1994). It has been suggested that issues of sexuality and reproduction can raise difficult and complex issues for the carers of adults with learning disabilities, whether they are relatives or service providers (Craft, 1993), because of a dilemma about how to perceive or discharge their responsibility. On the basis of empirical work, Carr (1994) has suggested that relationships with the opposite sex are often not regarded by others as serious, and tend to be discouraged by parents and carers. In another study, Shepperdson (1995) found that, although the attitudes towards sexuality of carers of young people with Down's syndrome appear to have become more permissive, these general views were often felt not to apply to respondents' own children, and that all felt that pregnancy should be prevented. He has argued that, while the issue of sexuality raises difficult issues for carers about intervention, in reality people with Down's syndrome are protected from sexual opportunities, they have little knowledge and few opportunities to have sexual relationships, or to 'take control' of this aspect of their lives.

With the increasing emphasis on education and information, in the UK, children and young people have a right to sex education under the 1986 Education Act (Craft and Stewart, 1993; McKay, 1994). Classes on social relationships, sexuality and appropriate behaviour may be offered in schools and in special needs courses as part of health and social education. Although Carr (1994) found that few young people with Down's syndrome received any sex education, nor was this regarded as appropriate for them, professionals in this study emphasised the need for education and information, several mentioned classes or discussion groups dealing with close and sexual relationships, and a number reported their involvement in organising these.

To conclude, the concern of many professionals in this study and of commentators in the literature was with the extent to which changes in interventions and policy were felt to have benefited young people with Down's syndrome. Adults are living longer and healthier lives, and most commentators have suggested the increasing social acceptance

of people with Down's syndrome (Brinkworth 1985; Stratford and Lane, 1985), in line with changing attitudes in society towards learning disability generally or to those who may be seen as not 'normal' (Inglese, 1990; Stratford, 1991). However, it has been argued that outcomes of integration for adults continue to be limited (Brinkworth, 1985); as Carr (1994: 435) has concluded, "the most 'normalised' experiences are open to only the most able and the most stable". Professionals in this study, in line with other commentators (Brinkworth 1985; Stratford and Lane, 1985; Carr, 1994) emphasised, often strongly, that in order for young people with Down's syndrome and other learning disabilities to benefit from integration and to have similar opportunities to others, the provision of appropriate and long-term support would be required. Several expressed reservations about whether such resources would be available.

While the accounts of professionals in this study and evidence from empirical work has indicated something of the lives of young people with Down's syndrome, the conclusions drawn are subjective, reflecting different perspectives or professional standpoints. There has been a growing emphasis on finding out the opinions of people with learning disabilities (Atkinson, 1988), and on learning about their perceptions and experience, and some qualitative studies have been carried out (Flynn, 1986; Booth and Booth, 1993; Shanly and Rose, 1993). In the course of asking professionals for the views about whether women with Down's syndrome might be included in the study, the researcher was referred to accounts of experience, feelings and perceptions written by people with learning disabilities and told about current research.

In this context, it was felt important to try to include women with Down's syndrome in the study if an appropriate method could be found. The next section describes the attempts which were made to carry out interviews, and presents the data. The chapter concludes by considering whether attempts to include women with Down's syndrome were felt to have been right and worthwhile.

Interviewing young women with Down's syndrome

The question of the feasibility and the ethics of interviewing women with a learning disability, especially as an untrained researcher, had been considered, particularly when questions might relate to reproductive issues. As described earlier, a number of professionals in the field of learning disability were consulted, and widely differing views were expressed about whether women with Down's syndrome could and should be included in the study. Some professionals noted general difficulties in interviewing people with a learning disability. They particularly mentioned the possibility of anxiety for respondents from the experience of an interview, the need for appropriate and possibly different questions, and the need for time to build up trust. Several also

expressed concern because of the topic of the study. Where professionals emphasised their responsibility for the welfare of participants, where there was uncertainty about harm, or philosophical or practical concerns about informed consent, access was refused by the representatives of some organisations.

Where access was agreed, there were still reservations about the nature of questions and the topics to be covered, both for practical and for ethical reasons, although there was a range of views about whether there should be more attention to the welfare and vulnerability of women with Down's syndrome than to other groups. Some warned against introducing concepts in the interview which women might not understand, or had not encountered before. Several suggested caution over the issue of pregnancy or having children, not only because this might not be understood, but because women with Down's syndrome might be receiving counselling about this issue, and hence questions might raise false hopes or might cause distress (as well as being unacceptable to carers). Some felt that to introduce the concept of abortion would be 'out of order', whereas others suggested 'playing it by ear', arguing that it would be wrong to avoid this issue, and that it would be useful to find out what was understood. However, all suggested that it would be wrong to introduce the idea of abortion for Down's syndrome; as one respondent put it, *"Abortion must be in there somewhere. ... It can't possibly be beneficial to include the idea that they might not have been born"*.

As well as suggesting ethical limits to questions, many professionals noted potential problems stemming from possible difficulties with comprehension and communication. This was suggested as mainly a difficulty for the researcher. The literature on this topic suggests that, while certain concepts, such as time or frequency may create difficulties for people with learning disabilities (Prosser, 1989), it is feasible to include people with learning disabilities in research examining concepts (Flynn, 1986; Jahoda, Markova and Cattermole, 1988; Walmsley, 1991; Shanly and Rose, 1993). Those reporting studies in this area have suggested that the ability to respond to questions is influenced by the intelligence of the respondent but also by the type of questions, which can affect the consistency of the response (Flynn, 1986; Prosser, 1989), although there are different views about whether open-ended, unstructured interview questions are useful in avoiding acquiescence or whether they inhibit response (Flynn, 1986; Shanly and Rose, 1993).

Several staff consulted offered advice. It was pointed out that some people with Down's syndrome might not be very talkative, and that the researcher should "make all the running" in an interview. Most thought that a new set of questions would be required, since they would need to be understandable, and some felt that, even then, not all women with Down's syndrome might take part. A number of professionals

suggested that some concepts such as the future and biology, and hence pregnancy, might not be understood, and that abortion was an unknown concept even for women with milder learning disabilities. Several also pointed out that the responses of people with learning disabilities might be more likely than those of others to be influenced by the way that a question was phrased. Some felt that, for findings to be meaningful, the researcher might need to understand or interpret respondents' answers; the views of people with learning disabilities were seen as reflecting the views of parents, carers or the local community more than those of others in the study for example. It was suggested that the presence of a facilitator would be useful to interpret both questions and answers, especially if the study could not be carried out over time.

Hence there was a range of views among professionals consulted about what the appropriate limits to questions should be, both for practical and ethical reasons. While most felt that simplified questions would be needed, one respondent, at odds with the majority view, emphasised that it was important not to over-protect people with learning disabilities, and argued that questions need be no different from those asked of other women:

I have found that I tended, I think, to underestimate the people that I see. But once they were given an opportunity to talk about these subjects, and felt reasonably confident that they were not going to get into trouble because of what they were going to say, that in the context of an important relationship, they have actually said that they were glad of having the opportunity to talk about these things.

(Consultant psychiatrist)

In order to include women with Down's syndrome in the study, it was decided that the best approach would be to carry out individual interviews, in the presence of a facilitator if this was preferred. The aim would be to ask women about aspects of their lives, and their views about having children, as far as possible in the same way as with other women in the study. Vignette-type questions were prepared to ask about issues of pregnancy. As with any qualitative study, interviews would be flexible, adapting to the responses of respondents. A copy of the guide is given in the Appendix (Appendix I).

A pilot interview with one young woman with Down's syndrome at a voluntary organisation was carried out by a member of staff, who knew her well, with the researcher present. This interview included questions about the difficulties that people with learning difficulties might encounter in bringing up children, and about the possibility of abortion.

Although a number of organisations were approached, access was agreed with the Scottish Down's Syndrome Association, who agreed to advertise the study, and at a college of further education, where the four young women with Down's syndrome

agreed to take part in the study. Interviews with these women were carried out at the college, with a member of staff acting as facilitator in three. An interview with the one woman who responded to the SDSA advertisement was carried out in her home; her mother sat in on the interview. The interviews were all quite short, lasting between 10 and 20 minutes

All five women with Down's syndrome lived at home with one or both parents, and all had attended special needs schooling. The facilitator at the FE college explained that two of the four students had moderate learning difficulties, and other two more severe learning difficulties.

Respondents seemed willing to answer questions about their lives. The facilitators were helpful, both in giving background information and in reinterpreting some questions to make them understandable. However, there were difficulties for the researcher, the main one being that, with no knowledge of the people, it was sometimes difficult to gauge the level of understanding. Most respondents explained when they could not answer a question, and it became clear that 'time' was a difficult concept. Rather than ask questions which might cause anxiety or which respondents would be unable to answer, there was perhaps an over-reliance on questions with a yes/no answer, which elicited few views.

All respondents said that their health was good. Asked if there were things with which they had difficulty, all replied that there were not. All women spoke of having been to special school, although only one, in response to a question, said that she had learning difficulties; when asked if this had made any difference to her life, she said that it had not. One respondent talked about herself as having Down's syndrome, in response to some encouragement from her mother. Those who were asked said that they did not find others' attitudes a problem.

All respondents described classes and interests that they enjoyed, and spoke most spontaneously and animatedly about relationships with friends and family members:

Resp: I've got three brothers.
JG: Have you?
Resp: They're all with us, where I stay. My other brother, he stays in D_. With my sister-in-law. They've got two children.
JG: That's good. Do they come and see you sometimes?
Resp: Yes. They come and see their auntie. Because I'm their auntie.

(note: Since these interviews were not coded using NUD.IST, they have no 'line numbers', and since there were only five interviewees, identifying code numbers have been removed.)

All respondents talked about their friends at a college or training centre, and the two respondents who were employed discussed this enthusiastically in terms of social relationships:

Resp: I'm going to work in the cafe in _. Make food for people.
JG: That would be good. Do you like talking to people? (this had been mentioned previously)
Resp: Yes, I like talking to people. And I get on with people. And I make friends. That's good.

M: Tell (interviewer) what you do on a Thursday.
Resp: On a Thursday I work up in Glasgow. I work in _, in the offices there. And I do quite a lot of things. Stuff like counting money.
JG: Do you like that?
Resp: It's good. I've made good friends.

The part-time job, one day a week, had been found by the respondent's mother, who emphasised the lack of work-placements and support services. This respondent described taking part in a number of activities for people with learning disabilities, which were arranged by her parents. It was difficult to tell the extent to which others took part in social activities. One respondent spoke of attending a 'girls' group', but when asked whether she saw friends outside this, replied, *"Mostly I just stay in the house"*.

All women lived at home with their parents. Asked whether they had thought about leaving home in the future, most said that they had, and one respondent discussed her desire to live independently, saying, *"I would like to look after myself. And to take care of the house"*.

The aim in interviews with young women in the study generally was to learn something not just of women's experiences, but also to find out their perceptions of their quality of life. When asked how they saw their lives, or whether there things that they liked or things that they would change, all women with Down's syndrome replied that they were happy:

JG: What do you like about your life on the whole?
Resp: I'm very happy with my life.
JG: Good for you. Is there anything in particular that you like?
Resp: (Pause) I like my whole family.

All respondents indicated that they had a happy life. Other respondents in the study were asked what they felt was necessary for a good quality of life. Women with Down's syndrome were not asked this abstract question, although they were asked about what they enjoyed. While it might be difficult for anyone to compare their life or their quality of life with that of others, nevertheless this issue might be felt important to

the debates underlying the study. The mother of one respondent, perhaps feeling at the end of the interview that a key question had been omitted, tried to remedy this:

M: You wouldn't like to talk about having ... what effect of being Down's syndrome? Do you feel different from ... if ... say you hadn't been Down's syndrome, and you were (age), do you think you would be the same, feel the same as you do now about things?
Resp: (No reply)
M: Is that too difficult, too difficult for you?
Resp: Yes.

Like other women in the study, respondents were asked at the end of the interview whether they were in a relationship, and sometimes whether they thought about getting married. While it was felt wrong to ask more intrusive questions to women with Down's syndrome than to other women, in the pilot (individual and group) interviews, questions asked by the facilitators about boy-friends and marriage had been answered very readily. A number of staff, however, had asked the researcher to note that a boy-friend might be quite a casual friend, and not to report the study in a way implying inappropriate sexuality.

Most respondents spoke of having a boy-friend, but said that they did not think about getting married. Most also said that they did not think about having children. All the women interviewed had a brother or sister with children, and they spoke about their nieces and nephews, sometimes referring to them as *"a lot of work"*.

JG: What about - you were saying you've got ... Is it nieces?
Resp: Nephews.
JG: Do you ever think about having children?
Resp: No! (Laughs)

Questions about antenatal testing were not always attempted, partly because it was often not clear how well the concept of pregnancy was understood, but also because of a reluctance to raise the issue of abortion. While the topic was introduced in three interviews by reference to a relative's pregnancy, the difficulty of asking about antenatal testing without introducing the concept of abortion can be illustrated by this response:

JG: ... Sometimes they can do tests on the babies, when they're still in their mother's tummy. And they can do tests to find out, say, if they're going to have a boy baby or a girl baby. Do you think they might have wanted to know those sort of things, or not?
Resp: Sometimes.
JG: Uhuh. Do they think it might be nice to know, before the baby arrives?
Resp: Just wait and see!
JG: Do you think you would want to know?
Resp: Just get a surprise.
JG: Uhuh. What about ... Suppose they could test to see if the baby was going to be ill, really ill. And they could do the tests when it was in its mother's tummy, before it was born. Do you think that would be a good idea, or not? To see if it was going to be ill?
Resp: It will be ill.

The possible unacceptability of this topic was illustrated by the way that these questions were firmly answered by one respondent's mother, who had otherwise encouraged her daughter to answer questions herself:

JG: What about ... Can you remember when your sister was expecting the baby?
Resp: Yes.
JG: Do you think she might have wanted to find out if it was a boy baby or a girl baby? Or anything like that.
Resp: I know that she wanted a baby.
M: She didn't mind. She didn't want to know ... to know that.
JG: No. Do you think she would have wanted to know if it was going to be a healthy baby, or have a bad chest, or anything like that?
M: No, she didn't want to know that either.

Another respondent suggested that this was a personal matter:

JG: They can do tests on some children, before they're born. When they're still in their mothers' tummies, to see if they are going to be boys or girls, the babies.
Resp: Uhuh.
JG: Do you think your sister might have wanted to know that sort of thing?
Resp: No.
JG: No, OK.
Resp: She didn't care, as long as the children were healthy. That's what she says.
JG: Uhuh, that's fine. Do you think she might have wanted to know, say, if they were going to be ill? If they could do tests for that sort of thing? Do you think she might have wanted to know that, or not?
Resp: Well, she doesn't tell me anything like that.

These responses have been included in detail to illustrate that attempts were made to broach the topic of prenatal diagnosis. However, this was an uncomfortable experience. The impression formed by the researcher was that, even if the concept of antenatal testing and abortion were understandable, this was an unacceptable topic of conversation, perhaps because it was seen as a private matter.

Discussion

Since a screening programme exists for Down's syndrome, the question for the researcher was whether it was felt right to treat women with Down's syndrome in the same way as other women with a 'preventable' condition and include them in the study, or whether the differences and difficulties of doing so might be felt more important, whether in the interests of participants or of the study. This dilemma reflects more general practical and philosophical considerations, discussed throughout this thesis, about the way that individuals with an impairment or disorder should be treated; how to balance an acknowledgement of difference and limitations with attention to the equal rights and value of the person.

Arguments against including women with Down's syndrome have been detailed already. There were concerns about possible harm to participants, because of the

difficulty of obtaining informed consent, because of the topic of the study (which might encompass questions about pregnancy and motherhood as well as about screening programmes), and because of possible difficulties with communication and comprehension. Also, because of the limits to questions which it was felt feasible or appropriate to ask, there was concern that interviews might not add anything to the study.

However, these differences and difficulties can be seen as relative; some women with Down's syndrome might be as able to be included as any other women. It was also felt that these women should have the same right to take part as other women with congenital disorders, and that it might be wrong to exclude them, to assume vulnerability for example, or a lack of understanding. At the time that the study was proposed, a woman with Down's syndrome had spoken out against the screening programme.

A decision was taken that it was important to try to include women with Down's syndrome in the study, if an appropriate method could be found. Although these women would not be asked questions about screening programmes, it was agreed that they might be asked a number of questions in a similar way to other women affected with a disorder, especially about their experience of living with Down's syndrome. At an early stage of the project, after the pilot study, it was felt that women affected with the conditions being looked at in this study might have little to say about screening programmes, or that it might be a sensitive issue, and that the focus of the study might be more on their experiences.

A further reason for including women with Down's syndrome has been suggested by Flaker (1994), who has argued that there are positive reasons for focusing on the experiences of those who might be stigmatised, and seeing such experience as legitimate, in order to widen the boundaries of what is taken to be 'normal'. From this perspective, including women with Down's syndrome might be felt to have a wider, more general benefit.

Attempts to include women with Down's syndrome in the study have been described in this chapter and in Chapter 5. Some interviews were achieved, although with some difficulty. Carrying out these interviews has shown that it is feasible to include women with learning disabilities in qualitative research, as also indicated in recent literature. Whether or not this attempt was felt to have been of benefit can be seen in different ways.

Including a consideration of Down's syndrome can be seen as helping to make the study more complete. Not only does much of the literature about antenatal screening and testing focus on this condition, but a number of respondents in the study spontaneously mentioned Down's syndrome in connection with screening programmes; they knew of people with the condition and about the screening programmes offered in pregnancy. Also, because of the decision to include Down's syndrome, background literature has usefully included reference to the implications of learning difficulties, as well as other types of impairment, and arguments about normalisation.

It was also interesting to find out the very varied views among professionals about the ethics, as well as the feasibility, of including women with Down's syndrome. These divergent views can be seen as reflecting dilemmas between theory and practice, and as highlighting questions about the boundaries of research. The limits of what might be felt acceptable in research can be seen as socially determined and as changing over time. People with learning disabilities are being included in research, they are making their views known and they are having children. Future research might examine their views about screening programmes.

A separate consideration is whether it was of benefit to the study to have interviewed women with Down's syndrome. It was educational to have learned something of the lives of the women who took part. These women, like women with spina bifida and women with cystic fibrosis, spoke about their activities, their interests, their aspirations, and relationships which they valued. They also said that they were happy. However, women with Down's syndrome did not evaluate the quality of their lives by comparison with others, nor was it possible to know whether they saw themselves as being different from other young women in any way. One respondent spoke of herself as having learning difficulties and another as having Down's syndrome, but there was no discussion of the implications of this, or what this might mean for their lives. Although direct comparison was not the purpose of the study, women affected with other disorders did discuss these issues. Some women living with spina bifida or with cystic fibrosis also referred to others in a similar situation to themselves - talking of disabled people or of 'cystics' - and it was not known whether women with Down's syndrome saw themselves as belonging to a 'group' in either of these ways. It was not really possible to gain an understanding of the views and values of women with Down's syndrome about abstract issues, nor were they asked about screening programmes.

The question of possible benefits or costs for those taking part was difficult to judge. While care was taken in interviews not to cause harm, there was doubt about the benefit for women with Down's syndrome in taking part in this study. The majority of women with other genetic conditions pointed out in interviews that they thought it important to

express their views about the morality or the importance of avoiding the birth of a child with an impairment, their perceptions of the severity of their own condition and of the quality of their lives; the topic of the study was a salient issue for them. In contrast, it was felt that the women with Down's syndrome who took part in this study might have been unaware of the existence of screening programmes and the public debate. The costs and benefits of taking part might therefore have to be seen differently for this group. It can be argued that, where people can give informed consent, it is beneficial to be offered the same opportunity as others to be included in a study examining their experiences and their views.

Hence assessing whether it was right and beneficial to try to include women with Down's syndrome in the study can be seen in a number of ways. On balance it was felt that the attempt was worthwhile. The main benefit can perhaps be seen in terms of exploring the extent to which it was feasible to do so, especially given the diversity of professionals' views. It was felt that lessons could be learned for any future research in that methods might be adapted more to participants, by allowing more time for example, in a way that was not possible in this study.

CHAPTER 8

WOMEN WITH SPINA BIFIDA

Women with spina bifida were included in the study because an antenatal screening programme for this condition is routinely offered to pregnant women in the West of Scotland. This chapter looks at the ways that women with this condition spoke about their experiences of living with the condition, their perceptions of its 'severity', and their views towards the 'prevention' of this and other conditions by the use of screening programmes in reproduction.

Background

Spina bifida is a congenital condition, caused by the failure of part of the neural tube to close before birth. It may be associated with hydrocephalus, and also with varying degrees of physical impairment, causing problems of impaired mobility and lack of sensation in the lower limbs, depending on the level and severity of the lesion. Babies born with spina bifida will have an operation to close the back, and hydrocephalus can now be treated by the insertion of a shunt. Improvements in treatments have led, since the 1960s, to a reduction in perinatal mortality for babies born with this condition, and people with spina bifida may live into adulthood. In childhood and adolescence, further operations may be necessary or useful to improve mobility, to deal with incontinence or to address curvature of the spine, although in adulthood the condition is often stable.

As with any condition, the consequences for the person living with spina bifida, whether social or psychological as well as physical, can vary. The authors of early studies considering the implications of living with spina bifida suggested that a number of difficulties, both for the families of an affected child and for the individual affected, might be addressed with improved resources (Woodburn, 1972; Anderson and Spain, 1977). More recently, with a focus on needs, it has been suggested that the needs of people with spina bifida are complex (Corbett, 1989; Tomlinson and Sugarman, 1995).

From the 1970s, there has been increasing interest in the common experience of disability of people living with certain stable impairments, such as spina bifida. As described in Chapter 2, both qualitative and quantitative studies examining the experience of disability have tended to confirm the proposition in the 'social model' of disability that important problems can be seen as socially caused, although it has also been suggested that 'disability' is not a homogenous category (Blaxter, 1983; Hirst and Baldwin, 1994).

Professionals consulted for this study expressed a range of views about the experiences and the needs of young adults living with spina bifida or a physical impairment, depending on their perspective. Those working in the field of spina bifida noted physical (and sometimes intellectual) limitations associated with the condition, and they discussed improved possibilities for those affected offered by operations and by the increasing emphasis on home care and individual management. These professionals also noted that many adults with the condition were no longer under regular medical care, a point also noted in the literature (Tomlinson and Sugarman, 1995), and they suggested the need for a coordinated approach or specialist clinic. However, like disability professionals, they stressed social causes of possible difficulties for young adults with spina bifida, referring particularly to inappropriate treatment, social isolation, and difficulties obtaining employment, and they welcomed initiatives for integration and independent living.

Living with spina bifida

Interviews were carried out with fourteen women with spina bifida. At the time of the interview, five respondents were students in higher or further education, two were working and five were unemployed. One woman who had learning difficulties attended an adult centre during the day, and another woman who had a visual impairment and some communication difficulties was hoping that part-time one-to-one learning might be arranged through the Social Work department. The interview with this respondent was conducted mostly with her mother, who interpreted questions and answers, and, on occasion, gave her own views.

The women with spina bifida who took part in the study described a range of social class and religious backgrounds and educational levels. The majority had attended special needs secondary schools - although some noted that the reason for this was because of mobility difficulties, three women had attended only mainstream schooling and two had transferred from mainstream to special needs education. Four women had taken or were taking a special needs course at a further education college, six spoke of taking other further education courses, and one woman was a student in higher education.

Asked about their fathers' (or mothers') jobs, six respondents reported non-manual and six manual employment and one woman's father was unemployed. Another woman was not asked this question because the interview was interrrupted before the end. In response to questions about church attendance and religious upbringing, seven of the fourteen women in this group described their religion; three of these attended a Catholic

church and another four reported being brought up as a Catholic. Only three women saw themselves as having no religion.

Like other women in the study, women with spina bifida were asked about their activities since leaving school, how they saw their quality of life, and in which ways living with the condition had affected their lives. The majority of respondents talked readily about a number of aspects of living with the condition, of both positive and negative experiences. Some women reported that their lives had been little affected. The majority, however, discussed some problems, or ways in which they saw their lives as different from those of others, and the extent to which they felt that these difficulties could be addressed.

Consequences of having spina bifida

Respondents were not asked about their impairment, but all mentioned some symptoms or limitations, and some women said that they had hydrocephalus as well as spina bifida. Nine women spoke about using wheelchairs all the time, three said that they used a wheelchair occasionally, one used a walking-stick, and one woman discussed having hydrocephalus but not mobility problems. Almost all mentioned the fact that they could not do some things which others could. However, while all referred to the physical condition, and to limitations, these were not generally spoken of as the cause of serious problems:

17,098: Well, there ... gradually my spine, I've got curvature of the spine, so it's like, it's actually made it sometimes quite uncomfortable sitting. ... It's like I've always had to get something that's wide enough to allow for space at the sides. But I've never ... I don't really think there's any other problems with it. As such.

There was no view that more could or should be done in the way of treatment. A number of respondents reported that their contact with doctors was confined to routine check-ups, sometimes infrequently, and spoke, instead, of managing their own condition. While several spoke of operations or their effects, again, this was often in a way which suggested that physical consequences of the condition were of little importance or low priority:

23,226: When I was thirteen, I went through a bad spell. .... (details) ... (Laughs) So I was in hospital - in and out of hospital - for about a year and a half. I was thirteen then. But apart from then, I haven't had any bother with it at all.

Most stressed that having spina bifida had not had an impact on their lives, as they had been born with it, and did not know anything different. However, two respondents spoke rather differently about the physical effects of the condition. Both these women gave more personal accounts of physical problems, which they felt were worsening, or

which could not be improved. They spoke about their anxiety about their condition, and also reported some problems of depression, of lack of self-worth and 'feeling different'.

Almost all women, however, discussed some difficulties stemming from impaired mobility, although the consequences were varied. A few spoke of depression or frustration, which was sometimes linked to their dependence on relatives. As one put it, *"I can't do anything very much for myself. I can't get out as much. I've always got to depend on my mum"*. Another noted, *"Sometimes I feel, "Crikey. My mum and dad still push me about." So sometimes that does depress me"*.

Respondents who used a wheelchair spoke about the difficulties of taking part in everyday activities, such as leisure activities, attending college or working. Many felt that difficulties with access were the main ways in which their lives had been affected, several mentioned problems with transport, and a number particularly mentioned their difficulties in finding employment. Five women were unemployed, and most of these described their frustration at being unable to obtain a job:

30,420: I would say the job situation is more ... than everything ... anything else. If you come up against anything else, you find a way to deal with it, but with the job situation, you can't deal with it. Because you're not given the chance to. So, you can't show people, "Look", you know.

Women who were at home during the day referred to a lack of social contact, speaking in strong terms of having 'nothing to do', of being 'bored out of my mind', or of doing things 'to pass the time', and a number saw their lives as restricted or constrained. Their responses suggested autonomy and self-determination as important in quality of life, as noted in other studies (Hirst and Baldwin, 1994). The right to make choices and to have equal access to opportunities might be seen as fundamental to Western society (Centre for Educational Research and Innovation, 1986), and some felt that they had a relative lack of control over their lives, compared with others:

17,236: At this time of year, it's sort of ... it really gets to me. I mean, I feel that, if you're able-bodied, you can choose whether you want to get out or not, but if the weather's not very good, then I've not got a choice.

Several women felt that difficulties with access or employment, or a relative lack of opportunities were typical of those experienced by disabled people more generally. Many suggested a common or collective experience of disability or disadvantage, as proposed in the social model of disability (Oliver, 1990).

All women in this group, whether or not they used a wheelchair, discussed a second type of problem, that of others' attitudes. Some referred to people's fear and hostility, a

few mentioned being bullied at school or in the street, and all described occasions when they had been stared at, spoken about, patronised or treated as 'not normal' or inappropriately:

40,150: I wear lipstick, I wear everything! And they still think you're a wee girl. Because you're in a wheelchair. I'm a young child. Because I'm in a wheelchair.

Keith (1996) has suggested that experiences of being talked over or treated as a child are common to people using wheelchairs. There were differences within the group, however, in the extent to which others' attitudes were seen as an important or a general problem, a number suggesting that inappropriate attitudes were only held by a minority of individuals and stemmed from ignorance. A few, on the other hand, using stronger language, suggested a more general unwillingness in society to treat disabled people in the same way as others. One woman pointed to the assumptions of employers, for example, about the abilities of disabled people as a group, saying, *"They think you haven't got it up here. But we have!"*

Societal prejudice has been suggested as one reason for preventing the births of individuals with particular conditions, but also might contribute towards problems for people living with the condition. From Goffman (1968), for those with a visible and obtrusive stigmatising condition, who might be seen as 'discredited', a negative ascribed social identity might both disrupt social interaction and affect self-concept (Scambler, 1984).

Some women reported that they found coping with others' attitudes difficult, or that they had felt this in the past. A small number referred to feelings of 'being different' or of a lack of self-worth. It has been suggested that felt stigma - which might be experienced more often than enacted stigma (Scambler and Hopkins, 1990) - might be more significant in people's lives than the physical consequences of living with a condition (Scambler, 1984). Although self-concept might be felt to be independent of the way one is treated by others (Jahoda *et al.*, 1988), some sociologists have argued, following Goffman, that people share a societal view or stereotype of those with certain conditions (Kelly, 1996). While others have criticised the theory of internalising attitudes as suggesting shared attitudes, many have drawn on the view that impairment is seen as shameful in society, that those with an impaired function are seen as inferior, and that such people may be expected to adjust to others' expectations (Zola, 1982; Davis, 1995).

To summarise, although not all women with spina bifida spoke about problems, the majority did. These difficulties were spoken of as potentially leading to frustration, depression or anger. Some studies of disabled people have noted the existence of

psychological problems, although findings vary (Hirst and Baldwin, 1994). However, the point has also been made that depression might result from social consequences of the condition and the unacceptability of expressing anger, rather than being an individual problem stemming from sickness or dependency (Zola, 1982; Finkelstein and French, 1993). Also, perceptions of dependence or self-image, as well as negative consequences of living with a condition in themselves, might not derive directly from the condition, but might be seen as resulting from social factors.

Where women with spina bifida discussed problems, they also spoke, often with feeling, of the imperative of maintaining psychological well-being, and they discussed the ways in which they felt that difficulties might be addressed.

'Solutions' or resources

The majority of respondents discussed the ways that they dealt with perceived problems, suggesting individual solutions, both practical and attitudinal. Almost all spoke about the value of having people to talk to, the need for the support of friends and families, and the importance of getting out. Many suggested taking part in 'everyday life' as important in quality of life, and, in spite of the difficulties of doing this, the majority described making efforts to take part in social life, to see friends, and to keep busy:

30,452: I mean, I feel as if I were just to stay in the house, you know, it would kind of drive you mad! You know, if you've got to do that day in and day out. You know, all the time. You've got to get out, to keep yourself mobile and sort of meet people.

In addition, many women also referred to more 'internal resources', in the sense of their own attitude. A few mentioned 'strategies' of positive thinking, comparing oneself favourably with others, or avoiding self-pity, in order to avoid depression or frustration. A number of women stressed the importance of a 'frame of mind' in the sense of self-concept. They spoke of an acceptance of physical difference or limitations, but of being the same as others and of having equal value.

Many noted that they were accepted, or seen as 'no different', by their friends, and several spontaneously pointed out that they were treated equally or in the same way as siblings within the family. The role of those in close relationships has been seen as important in the adjustment of an individual to illness and disability (Lyons, Sullivan and Ritvo 1995), and the attitudes of 'sympathetic others' as importantly affecting identity and self-concept (Goffman, 1968). Social scientists reporting empirical studies have suggested that families can act as 'stigma coaches' if they accept the stereotype of the disabled person (Schneider and Conrad, 1981; Scambler and Hopkins, 1990).

All respondents made the point that they saw themselves, and wanted to be perceived and treated by others, as a person like anyone else, a normal person or an equal person. Although a small number suggested that they found it difficult to deal with being treated inappropriately or with feeling different, the majority described the ways in which they dealt with possible problems. They spoke of ignoring others' attitudes but also of a self-concept as a normal person; as one put it, *"I have noticed a lot of people do take it to heart. But I don't. I think, "It's just a disability". You're still a normal person"*. Another suggested resisting feelings of shame:

21,211: (It's) just the kind of personality I have. I feel that if people want to say something about me, it's up to them. I know the way I am. It's not my fault.

It has been suggested that those with a stigmatising condition might need to develop strategies for the management of 'spoiled identity', whether in the sense of resisting the negative impact on self-concept or in dealing with others. While Goffman (1968) argued that people with a visible condition might manage tension in interaction by a strategy of 'covering', or minimising difference, various strategies or modes of adjustment have been suggested from empirical work (Scambler, 1984). Keith (1996) has suggested that most people with a physical disability avoid internalising others' attitudes, deal with anger, and demonstrate that they are coping psychically either by avoiding a situation or by educating others, depending on the situation and on personality.

Women reported employing various strategies to deal with actual or anticipated problems of others' attitudes. Some described an educational or explanatory approach; as one respondent put it, *"You know how, when you first meet a person, you've got to let them know that you're just the same as everybody else, just talk away to them, just like anybody else"*. Others reported challenging others' perceptions or assumptions by, for example, replying to people who assumed that they could not understand or talk, or joining in activities that they might not have been expected to, such as going dancing.

From the disability rights movement, however, personal adjustment to the problems faced by disabled people has been seen as the wrong approach; suggested solutions are in terms of social change, whether of social inclusion or increased support. A number of women with spina bifida referred to difficulties faced by all people with mobility limitations or by disabled people. Several felt that a number of the problems faced by those with mobility problems might be resolved if transport could be improved, for example, or the environment made more accessible, especially in relation to employment. They spoke of the need for general or societal solutions. A few argued for general changes in attitudes, and for disabled people to be perceived and treated in the same way as others. One respondent used the rhetoric of needs and rights:

37,267: They just treat you like you're just in a chair, and you can't do nothing for yourself, you know? But I can! I'm not like a robot that can't move, you know. I'm like anybody else, but with needs, you know. And people need to know what I need.

Only one woman reported being active in disability rights campaigns, although several said that they had been introduced to rights arguments at special needs school and one woman mentioned that she subscribed to a disability journal. Several, however, discussed having made complaints themselves about attitudes towards disabled people or about difficulties of access faced by wheelchair users. At the same time, a number of women spoke about their reluctance to join campaigs or groups of disabled people, sometimes explaining that their friends might be either disabled or able-bodied.

Many women with spina bifida spontaneously referred to themselves as a disabled person. In response to a question about whether they would see themselves as disabled, almost all gave a similar answer. 'Disabled' was spoken of as having a literal meaning as impairment, but not as a positive term:

JG: So - do you tend to see yourself as being disabled?
19,146: Um, not really. Well, I know I am disabled, but I like to just be treated the same as everybody else. Like ... the same ... things.

JG: Would you tend to see yourself as a disabled person?
37,282: No! I just treat myself like ... like mum. You know! I just treat myself ... well, I know that I am disabled, but I just treat myself like I'm normal.

All women with spina bifida pointed out that they saw themselves and wanted to be treated as a normal person, and they also noted that they were different in that there were things that they could not do. They spoke of themselves as both having limitations and as being equal, or being 'the same' and 'different', as has been noted in other studies (Pinder, 1996). Their responses made clear that this was not a contradiction, but could be a philosophy; as one woman pointed out, *"there are folk who are different"*. Goffman (1968) has argued that the fate of disabled people is to be defined as the same and different, and that a strategy of seeing oneself in this way can be seen as representing 'good adjustment'.

Discussion of 'strategies' and values

It has been noted in the literature on living with a condition that many people living with chronic illness or disability may develop strategies of coping, both with the practical consequences or with the concept of living with a condition. Such strategies, which may be both practical and attitudinal, may offer ways of adaptation to physical conditions or difficult situations (Williams, 1993), or may be ways of conceptualising the condition and its consequences (Kelleher, 1988; Bury, 1991; Radley, 1993). While the psychological literature suggests that people vary, for example in the extent to

which they see life events as under their control, and in their approaches towards dealing with or avoiding events or information (Kent, 1996), sociologists have suggested that, both in their strategies or behaviour and in talking about their 'coping strategies', people draw on cultural values to maintain an equilibrium (Bury, 1991).

In qualitative work, analysis of the way that respondents talk about their perceptions of problems and solutions might suggest different psychological approaches, but might also suggest underlying values, beliefs or philosophies. Accounts may be seen as constructs, although there are different theoretical views about the extent to which accounts reflect cultural norms. Some have argued that, when people talk about 'strategies' or coping, they might be reflecting normative or 'public' rhetoric, or presenting themselves as a certain moral or virtuous type of person with "right and proper attitudes" (Williams, 1993: 103). However, while from some perspectives, it has been argued that 'coping strategies' can only reflect a dominant discourse (Voysey, 1975), others have suggested that an analysis of strategies (and explanations) can suggest diverse cultural views (Coffey and Atkinson, 1996), or a mixture of public rhetoric and private narrative (Radley and Billig, 1996).

The interest of this study is in people's values, philosophies or perceptions, and it was felt important to look at the way that people spoke about living with a condition, both personally and more generally, in addition to being asked more directly for their views about screening programmes, in which case the issues might not necessarily be of relevance. It has been suggested that the way that people speak about dealing with illness, in the sense of balancing different aspects of their lives, might affect their attitudes in other areas of life (Radley, 1993). This might be expected to be particularly the case in considering either the morality or the justification of 'prevention', by screening programmes.

What values were suggested by this group? In addition to talking in terms of being both 'the same' and 'different', in discussing their lives, most women with spina bifida spoke of the consequences of the condition in a way that suggested these were of little importance. While it has been noted that many people speak of themselves as healthy or problem-free while at the same time discussing symptoms (Blaxter, 1993; Pollock, 1993; Williams, 1995b), a 'strategy' of perceiving the physical as of low priority has been noted in other studies of those living with a condition (Kelleher, 1988).

Possible reasons for this have been suggested. Some have argued that an approach of making light of one's afflictions or carrying on as normal is seen as the appropriate reaction, sanctioned by health professionals (Pollock, 1993). On the other hand, symptoms might be seen as part of the human condition, and it has been suggested that

there is a general cultural value in society as a whole of stoicism, or a morality attached to not being defeated by the physical (Blaxter, 1993). From medical anthropology, Kleinman (1988) has suggested that people who are defined as having a certain condition might want to ensure in interviews that they are judged as people, being reluctant to focus on only one aspect of their lives; hence individuals might present themselves in a certain way for more personal reasons. From the social model of disability, however, many of the important problems faced by people living with impairments are seen as collective and as socially, rather than biologically, caused. The social model challenges definitions - seen as resting on a 'biomedical' model - of inevitable problems, activists arguing that such assumptions may otherwise be unexamined in society.

Hence a philosophy of seeing the physical as of little consequence might reflect a number of psychological and cultural approaches or values. In a similar way, a philosophy of 'sameness and difference' might represent a number of perspectives. The concept of equal value and equal rights for minorities is fundamental to human rights. The social model includes both the philosophy of normalisation, equal treatment, integration and social inclusion, but also attention to special needs and the need for increased support. An approach of medical welfare similarly implies attention to both the person and the condition. This thesis will examine, in Chapter 11, the values expressed in other groups, and the extent to which these are felt to be different or shared, although the point has been made that analysis cannot tell where values come from (Cornwell, 1984).

In this study, as described in Chapter 5, interviews were analysed both 'horizontally', to suggest themes or concepts, but were also examined as whole entities or 'vertically', to indicate individual differences in approach, values, rhetoric or explanations. As in other studies (Pierret, 1993), it was found that, although people's experiences were varied, and strategies sometimes depended on the situation, the accounts of individual women were coherent in the ways that problems were perceived, and in the values and philosophies that were drawn on. This analysis suggested that, within the group, women expressed opposite approaches towards living with the condition, reflecting different ways in which the condition might be conceptualised. These could be seen as a more 'activist' or normalising approach, from which limitations might be seen as surmountable, or a more 'accepting' or pragmatic approach, from which limitations might be perceived as a normal aspect of the human condition or unimportant.

Activist philosophy

Some women suggested that there were, or there need be, few differences in their lives from those of non-disabled people. From this perspective, which could be seen as a more 'activist' philosophy, having the condition was perceived as not necessarily a barrier to doing the same things as others. Women with this approach spoke of taking part in the same activities, although sometimes of the difficulties of doing this. Many discussed employment; the fact that they were working, or taking part in training, or spoke of their efforts to find a job. Several emphasised their ability to be independent:

19,019: Well, I actually drive to (my work). I passed my test a couple of year ago now so I've been driving a couple of years. I'm quite independent. I get out and about!

Respondents described personal strategies or attitudes of overcoming limitations and challenging others' assumptions about their abilities, often reporting examples of having resisted unwanted help, saying, *"I can do it myself!"* Some spontaneously described actively managing or dealing with their condition, and talked positively about finding out information. Several also spoke more generally about values of autonomy, achievement, individual qualities and potential. For a few the value of independence or self-help was suggested as a philosophy:

30,230: You know, people should be pushed to their own potential. You know, let them do what they want to do. If they can do it. And if they can't do it, then they can ask for some help. You know, I think a lot of people think, you know, because you're disabled, you can't do a lot of things. But there's a lot of things that we can do.

Many women with this approach spoke about having a majority of friends who were not disabled. Zola (1982) has suggested that there is a hierarchy among disabled people, and that friends can provide identification as well as support. It has also been suggested that the attitudes of family members in relation to over-protection, as well as stigma, may be important in self-concept and identity (Scambler and Hopkins, 1990). Those who spoke in terms of an 'activist philosophy' also described a family emphasis on the value of integration, challenging pessimistic medical views, independence and self-reliance:

19,173: I think if my mum and dad hadn't pushed me from an early age ... it's helped me. If I hadn't ... like some parents don't like you going out and that, clubs and that. When I was younger, I used to go to like Brownies and that. And mixing with other kids, able-bodied kids too and that, so I've just like grown up with a mix.

Dependence and independence

Some disability activists have argued that the dependence of individuals with impairments on others need not be seen as inevitable but should be seen rather as a

matter of a lack of appropriate societal support (Oliver, 1990). There may also be assumptions within society about their inevitable dependence. From this perspective, there is a focus on independent living and on legislation towards this end, and this has been seen as having the potential to improve the lives of disabled people.

Others, however, have pointed out that a cultural value of independence is only one of conflicting moral values in society (Williams, 1993; Shakespeare, 1996). A focus on independence and personal qualities has been criticised as prioritising one cultural value over others (Zola, 1982; Williams, 1993), for implying that physical characteristics are unimportant (Corbett, 1989), or that the support of others as well as that of society are unnecessary (Zola, 1982; French, 1993b). It has been argued that a rhetoric of independence may create a pressure to achieve which is unhelpful to disabled people as a group, not only because, like others, they might want the choice of not always aiming to achieve (Zola, 1982), but because of the conflict with physiological limitations. For some, goals of independence might not be appropriate or achievable (Zola, 1982; Corbett, 1989; Williams, 1987a; 1993). As discussed earlier, other disability activists have emphasised the normality and the equal value of those who might have a diversity of limitations.

The issue of dependence may be particularly important in close relationships, since it cannot be separated from the attitudes and expectations of oneself and others towards the provision of support (Bury, 1991). Zola (1982) has argued that because of an emphasis on independence in Western culture, the giving and acceptance of nurturance is seen as unacceptable except between parents and children; disabled people may therefore feel guilty in the presence of relatives or carers. In empirical work, Williams (1993) has found that many disabled people disliked feelings of dependence, but in one study found different approaches; whereas for some dependence on family or close friends was spoken of as bound up with feelings of burden, for others who had different attitudes towards social roles, close relationships and interdependence, mobility impairments did not necessarily cause problems (Williams, 1987a).

In this study, a few women referred to their dislike of being physically dependent on other family members, although, in general, this issue was little discussed. However, some spoke more generally about limits to independence, and the need for the provision of support:

40,105: The woman on our actual (special needs) course actually said "I like to let them get on, and see how far they can go themselves". But my argument is, for people like myself, or people that are not very good on their legs, can only go so far.

Accepting philosophy

Not all respondents spoke in terms of the values of achievement or independence. Some women suggested a 'philosophy of acceptance', speaking in more fatalistic terms generally, and talking of adapting to limitations, rather than of challenging them. Dependence was discussed in a way which suggested that this did not cause problems for their own self-concept, nor was the provision of support a problem for their families. One respondent and her mother jointly discussed balancing a desire for independence and a need for support, and others quite readily discussed dependence in family relationships:

20,334: I can only go to the (leisure activity) if my dad's going, and if he's not going, I'm not going, basically. If he can't go, then that's it. Usually he can.

Women with this approach referred more to friends who were also disabled. They were also more likely to discuss leisure activities in terms of their enjoyment value, and to emphasise more generally goals of happiness, emotions and personal relationships.

The fact that some people cope with illness or disability by minimising the effect on their lives, and by challenging limitations, whereas others accept or accommodate to the condition has been noted in other studies (Radley, 1993). Kelleher (1988) has suggested that both 'normalising' and 'accepting' strategies can offer a means of control over living with a condition, the alternative being not coping. The condition may be conceptualised as manageable, as not problematic, from both perspectives. In a study examining the attitudes of relatives, Parsons (1990) also identified 'normalising' and 'pragmatic' strategies; in the latter, the person was important and the disorder or difference perceived as unimportant.

Although respondents in this study tended to emphasise one or other approach or philosophy, especially in relation to independence, these were not usually mutually exclusive. Many spoke about the need for balance, and described acceptance or a lack of control in some aspects of life, and challenging limitations, interests or possibilities in others. Several emphasised other attributes, as noted in the literature (Zola, 1982). A balance between challenging and accepting was discussed in different ways, depending on what was felt important in quality of life.

Quality of life

As well as being asked about their experiences of living with the condition, women were asked how they would see or evaluate the quality of their lives, and about what they felt was needed for a good quality of life. While many had referred to some

problems, almost all had spoken positively about their lives, and the majority saw the quality of their lives as good, one woman describing her life as "brilliant". Women with spina bifida spoke of quality of life in terms of mental and psychological well-being rather than physical health:

JG: How would you describe your quality of life?
30,458: I would say it was ... it was very good. Apart from, like, the health side. You know.

In discussing the more general issue of what was seen as important, many drew on factors which they had emphasised already. One woman who said that she had a good quality of life explained this in terms of having a good job, getting out and meeting people, while another, who saw her life as boring, suggested that it could be improved with something to do.

The responses of the majority suggested that a good quality of life or a happy life might be achieved, depending on possibilities, but also on perceptions, philosophies, or strategies. One respondent emphasised the importance of a self-concept of equal value: *"Just respect yourself. Get on with your life. Even if you are disabled"*. Another spoke of balancing acceptance with determination, *"... realising that you have to realise your own limitations ... I can't do that but I can do this"*. Some discussed a general emphasis on the positive, happiness and enjoyment of life:

17,255: I just think the right frame of mind really. ... Things like that. If there's something that you really enjoy, like maybe going to the cinema, whatever. As much as possible, just do it.

Some saw quality of life in terms of friendship and personal relationships, and a few particularly mentioned family relationships. One respondent spoke in more general, prescriptive terms about the importance of family support in quality of life

JG: What would you say is needed for a good quality of life?
30,601: A lot of back-up. From your parents. And a lot of love. Obviously that is ... you know, that is needed. And I mean, obviously everybody has their ups and downs, and everyone has times like that, and your parents have got to be there. To go through that with them. Obviously they've got to be like understanding.

Some have argued that people in interviews present themselves as a 'coping person' (Voysey, 1975; Williams, 1993), suggesting philosophies, cultural views or 'public accounts'. For example, it has been argued that a balance between accepting and challenging limitations can be a position of virtue, or a moral position (Williams 1993), as well as personally helpful. However, not all respondents with spina bifida in this study talked in terms of coping, and not all offered strategies or philosophies. Two women, when asked, suggested that they felt their quality of life was not good:

36,309: Crap.
JG: What's the main thing about it?
36: (Taps wheelchair)

This raises the question of who might take part in the study. It has been suggested earlier that some might be more likely than others to take part in this study for a number of reasons, one of which might be the degree to which participants might perceive themselves as 'coping well' with living with the condition. Questions about the representativeness of the sample, and the 'presentation of self' in interviews will be returned to in Chapter 11.

Summary

In talking about their experiences of living with spina bifida, 'solutions' to possible problems, and about their quality of life, while some women spoke in personal terms and others more generally, the responses of many indicated values. These might be seen as: that a person with a condition is both a normal human being but someone with a difference or special needs, that physical aspects life may be seen as of low priority, and either a normalising approach emphasising that limitations can be overcome, or a more pragmatic approach emphasising acceptance and that limitations may be seen as a normal part of life (although often a balance).

While strategies or solutions can suggest ways of enhancing individual quality of life, they may also indicate, more generally, the way that limitations of a condition or 'living with a condition' might be perceived, or - from more prescriptive 'solutions' or philosophies - ought to be perceived. These values might be relevant in women's opinions about screening programmes or in considering whether conditions might be seen as 'serious'.

The 'severity' of spina bifida

Women with spina bifida were asked whether they would see the condition as serious, and whether they thought that others would see it as serious. As noted earlier, 'serious' can have a number of meanings. The consequences of a condition might be experienced by those affected or by others. 'Severity' might be felt to be relative or a matter of perception. From the social model of disability, 'serious' might be felt to reflect wrong assumptions about the way that a condition or its consequences should be perceived.

The two women in this group who felt that their condition was worsening said that they might see the disorder as serious, although one pointed out that, while it was 'a serious thing to have happened', the condition had not had a serious effect on herself.

All other respondents, including those who had not spoken of their own quality of life as good, said that they would not see spina bifida as a serious condition. However, they all also felt that 'others' would. All gave the same reason for this; that 'they' would think that people with spina bifida could not do certain things:

25,564: They do, I think. If anyone had a test and they were told, "You're going to have a baby with spina bifida", they would just be in despair, I think, that they were going to have this child that ... that couldn't do anything.

While the majority suggested that there were wrong assumptions about the implications of the condition, this raises the question of who 'others' might be, and of why all thought that there was this uniform view, whether this was a view that respondents had experienced, from the media, from other individuals or from the medical profession. Although some reporting empirical work have argued that respondents share a negative or cultural world view of affected individuals (Kelly, 1996), many respondents in this study firmly asserted a different view from the assumed societal view.

Many women explained why they would not define their condition as serious. A number spoke of the consequences as 'not really' serious, often mentioning their own lack of medical problems or few limitations. In these terms or definitions, therefore, spina bifida was seen as relatively less serious than other conditions or than might have been assumed or expected. Some contrasted their own abilities or life-span with a more general idea of 'a serious disorder', or spoke of the need to challenge a pessimistic medical prognosis, or assumptions about consequences:

13,143: Not really! Probably when I was wee, they would say it was a serious disorder, and of course when I was born with it, they were like that. And they said that then to my mum, that I wouldn't live past a year, and that I was going to die ... before my first birthday. But I wouldn't say it is now.

Those women who illustrated their perceptions of the relative lack of severity of the condition by reference to their own lives or abilities made no reference to the variable physical consequences of having spina bifida, or to how representative they thought they were of people with the condition. Women who had this view often expressed a more general 'activist' philosophy, emphasising potential:

16,157: Not really because I don't ... it hasn't stopped me from doing the things that other people do. So not really.
JG: Do you think other people would see it as serious?
16: Some probably would because they would see the wheelchair and think, "She can't do certain things". Whereas I probably could.

On the other hand, as discussed already, a number of respondents, from a more 'accepting' philosophy, suggested that the condition itself and physical consequences were of little importance. One woman, describing the condition as *"just more of an*

inconvenience, basically!" did not deny limitations, but contrasted her own view with the assumed perceptions of others about their severity, saying, "*Some (employers) may see it as serious because I need help with toileting. But that's all. My legs don't worry me, my legs don't work very well.*"

These two approaches to living with the condition and reasons for refuting a view of the condition as serious were not mutually exclusive; one respondent suggested both a lack of physical problems and a lack of concern about dependence:

JG: Would you see spina bifida as a serious disorder yourself?
17,302: Not really, no. As I say, I know that I have got my limitations, that I need to rely on folk, but as I say, I wouldn't say it was exactly that serious. Not ... I mean, I've never had any problems, really. I've never been in hospital for a long time, so ... .

Answers to the question of whether women with spina bifida saw the condition as 'serious' were coherent with their views throughout the interview. The majority indicated that they did not see the disorder as serious for themselves, either because there might be assumptions about physical problems and that they could do things, or because there might be assumptions about the importance of physical aspects of life and they did not see limitations as a serious matter. In evaluating the severity of the condition, most women replied in general, rather than personal terms, but related their view to their personal experiences and perceptions.

In discussing their or others' perceptions of the severity of the condition, women did not talk about possible implications for other individuals, or suggest that the condition was seen as serious by other family members. Although women were asked a general question about the family, relationships were little discussed, and those women who talked about their families referred mainly to their relationships with their parents. Some spoke of their dislike of feeling dependent, and a small number made a passing reference, sometimes inaudible, to differences of opinion about what they were able to do. In general, however (although some noted transport difficulties), most indicated either their ability to be independent or suggested that a degree of dependence was not a problem for their families, and that needs for support - emotional or more practical - were met. In addition, almost all women spoke of being accepted, valued and treated equally within their families. A lack of discussion about others' perceptions of the condition as serious was coherent, therefore, either with a view of difficulties as surmountable, either by personal strategies and independence or by social change, or with a perspective by which difference and a condition was seen as of little importance compared with the value of the person.

It has been suggested in the literature that family relationships may be seen as a private area, too personal and difficult to discuss in a first interview (Voysey, 1975; Brannen,

1988), although Voysey as also argued that it is culturally almost impossible to talk about the rejection of a family member. Although personal perceptions of causing difficulties for others or of feeling unwanted might be something that might not be discussed in an interview, two respondents spoke in general terms about parents who 'could not cope' with a disabled child.

Advice

Women were also asked what advice they would give to relatives who were expecting a child with spina bifida. One reason for asking this question was in order to gain an impression of respondents' views about screening programmes for the condition which they had themselves. It was felt insensitive to ask about this directly. The question was not framed in terms of 'prevention', and the possibility of abortion, following a positive diagnostic test, was rarely raised.

Almost all women spoke in general terms, referring to philosophies about the right approach to bringing up a child. Some emphasised the need to treat the child like any other, for example, *"Just to love it. You know, care for it. Don't treat it any different. Because it isn't any different"*. From an 'activist' approach, some stressed the need not to over-protect the child:

19,125: I know some parents, like, molly-coddle their children if they're disabled. ... So I say, "Just let your children go", sort of thing. Let them get out and that, let them join sports clubs, you know.

Although a few made the point that possible difficulties had to be accepted, the responses of the majority of women did not indicate possible difficulties, either for the parents or the child. They did not suggest concerns about the responsibility of parents for bringing a child with spina bifida into the world. Rather, they indicated that there might be assumptions about suffering, that parents might worry unnecessarily, stressing that physical consequences were not major problems. As one pointed out, *"Well, if it's the same as myself, I would say to them, "It's only the legs". There's nothing else wrong with them, you know."*

However, while most spoke in general terms, another type of answer was given by four other women in this group who reported having been asked for advice, whose relatives had been pregnant, or who had considered the question of having children themselves. These answers were personal rather than hypothetical, more hesitant and ambivalent, and suggested concern. One woman wondered whether the mother might be able to cope. Others expressed some anxiety about what the child might be like, mentioning their concern about risk, especially in relation to their siblings:

JG: Suppose relatives of yours knew that they were expecting a child with spina bifida, what kind of things would you be saying to them?
21,249: I don't know. My big brother's just actually ... she's got a wee boy just now and he's only ... just turning (age), and that's what I ... I was worried, you know. Like, in case he would be like me.

One woman spoke positively about her view that relatives would not regard the condition as a reason for termination, because of herself. Another, however, described very different reactions of family members towards testing. She also reported her own difficulties when asked for advice about the implications:

25,491: They knew that their baby was going to be born with spina bifida, and she was wondering what was going to be wrong with it. And my mum went to speak to her first, because, I was like ... I didn't know what I was going to say. (...) And she said that she felt really encouraged because ... they saw that I just, kind of, just got on with things and didn't really let it hold me back, and I think it just gave them a wee bit of hope. Although, I think, their baby's got, sort of, more disability than I have.

These responses indicate responsibility, although it is unclear whether the concern is about the implications for the affected child or the parents. The mother of one respondent noted that she had been asked for advice about termination, and - while pointing out that this would have to be a personal decision - she stressed the concern of parents about the child, and also, like the respondent above, pointed out the variation in severity.

New reproductive technology, screening programmes and 'prevention'

The aim of this study was to find out respondents' views about new developments in technology, particularly screening programmes. It was felt that the views of women with 'preventable' conditions might be shaped by a number of factors. Their experiences of living with a condition and their perceptions of the consequences might influence their views about whether this condition should be prevented. They might also have different views from others in the population more generally about the morality of 'prevention', or the importance of 'choice'. At the same time, women's views might be influenced by their feelings about having children themselves.

All women in the study were asked if they thought about having children, and how they felt about new developments in reproductive technology, such as abortion, and sex selection. They were also asked their general views about abortion on the grounds of foetal abnormality. Women were asked about their knowledge of the conditions in this study, whether they felt that it was right for screening programmes for these conditions to be offered, and whether they might consider undergoing screening programmes themselves.

Motherhood

None of the women with spina bifida had a child. Two spoke of being in a relationship although none was living with a partner. Several women said that they might think about having children in the future, and a few had discussed different aspects of this, including the risk of spina bifida, with a doctor; one reported being told that if she took folic acid, her chances of having a 'normal child' were high. However, several women replied that they were unsure about whether they would want children, and a few said that they probably or definitely would not have children. They were not asked for reasons for their view, but some gave explanations. One woman spoke of her concern about the risk of inheritance, saying that she would worry about 'how a child might turn out'. Two women suggested that they felt they might be unable to care or would not 'feel able to cope' with bringing up a child. One woman was not asked this question.

Crow (1996), a disabled woman who has argued that the social model of disability is inadequate for examining the experience of disability since it ignores the physical implications of impairment, has suggested that many disabled people are ambivalent about having children. Although numbers in this study were small, the responses of women with spina bifida tended to confirm this view. Some women indicated that questions about family building or about interventions in reproduction were of little relevance or interest, and, for some, these issues provoked little discussion.

Abortion

The views of a number of women with spina bifida towards abortion were rather different from those of women in other groups. Concerns that abortion might be a sensitive issue were unfounded; most seemed to have assumed that this issue would be raised, and discussed their views on abortion quite readily. The majority of respondents spoke of themselves as being 'against abortion', or of abortion as something that 'I don't agree with'. A few referred to their religious beliefs or upbringing, but several spoke in terms of a 'right to life' or 'the chance of life', or saw abortion as murder:

21,432: I think it's like killing somebody. It's like you're consenting to it if you know what I mean. It's like you're killing somebody, and I just don't agree with it at all.

However, almost all women made a distinction between their own moral view that abortion was wrong or was something that they 'would never do', and a view that the option of abortion should be available for others:

17,435: I mean, I've always said that, because of my upbringing, it's like, in a way, I would always say that I was against abortion, but I would always just say that it's like ... I wouldn't ever ... I've been brought up to think that it's not right to let the pro-lifers and things like that, it's not quite right to actually force people to have children.

As in other groups, a view that abortion was personally unacceptable did not necessarily mean that women felt committed to bringing up a child; a few mentioned the option of adoption in certain circumstances. Also, although the majority of women with spina bifida said that they were against abortion, four women did not take this view, and some said that, in certain circumstances, or for certain conditions, they might consider this themselves.

Women were asked their views about the use of abortion in some cultures to prevent the birth of a child of the unwanted sex and about the possible use of genetic tests to provide information about the physical characteristics of an unborn child. The views of women with spina bifida were similar to those of other women, in that sex selection was spoken of, often in strong terms, as unacceptable. The provision of information about physical characteristics was widely seen as unnecessary, the responses of many suggesting that this was 'not relevant', or questioning why the information was being given. However, this was not seen as wrong, and there was little discussion about the nature of such information. The implication of abortion on the grounds of physical characteristics was raised by some women, who saw this as very wrong, but also as unlikely:

JG: Should they be able to tell people or not?
19,462: Well, they should be able to tell people, but I don't really think people would really mind, as long as their baby was healthy, I don't think they'd really bother about the colour of the eyes or their hair and that.

Abortion on the grounds of foetal abnormality

The majority of women indicated that they did not approve of abortion, and possible reasons for which abortion might be felt acceptable were rarely mentioned. The question of abortion on the grounds of a condition or foetal abnormality was only referred to by a small number of women, whose responses were sometimes hesitant:

19,404: (Abortion) should be available if people are really wanting it, maybe if the woman's life's at risk, so I think that's an option too. Maybe to ... or maybe for definite ... or if they really know for definite, the child's going to be sort of damaged and all that. But you don't really know if a child's going to have a disability.

When asked directly for their views about abortion on the grounds of foetal abnormality, almost all said that, as with social abortion, they thought that the option of abortion on these grounds of a condition should be available. However, the majority of women suggested that this was something that they would not consider themselves.

Some made the point that they would not see abortion on the grounds of foetal abnormality as a different issue from social abortion, since any abortion would be murder:

23,446: Well I look upon it ... a baby's a baby and it's a human being. I wouldn't kill any baby, whether it was disabled or not.

Some respondents, especially those who spoke in terms of a personal philosophy of 'acceptance' in their approach to living with the condition (although not those who most emphasised discrimination against disabled people), suggested that they would see abortion on the grounds of foetal abnormality as less acceptable than abortion on some other grounds. Women who held this view spoke of abortion on the grounds of foetal abnormality as wrong, used stronger language, and spoke of the equal rights to life of those with a disability:

23,501: There is people that if they discover they're having a spina bifida baby or a Down's syndrome baby, do abort their baby, which I think is really terrible. I think no matter what disability, or however it's developed, it should still be allowed to be born.

As illustrated here, those who took this view spoke about the wrong choices and wrong values of individuals. They did not suggest that it was wrong or discriminatory for screening programmes to be offered, or that such programmes could be seen as evidence of societal prejudice. Unlike disability activists, women with this view discussed the morality of individual action; abortion on the grounds of foetal abnormality was seen as a search for perfection, not in society, but in individuals:

31,867: I mean, so what if your kid's going to have cystic fibrosis? Or spina bifida? So you're going to have to cope with that. Like, you aren't having a kid, like, to be a perfect baby. You're having a baby to be a part of the family. I mean you <u>should</u> have been having a baby to be part of the family! (Laughs)

As with abortion generally, some made a distinction between what others might do and what they might do themselves. It has often been noted that accounts can have a moral function, and that a moral position may be established by contrasting one's own choices with those of others (Voysey, 1975) or using 'contrastive rhetoric' (Coffey and Anderson, 1996). In such accounts, "the speaker and his or her practices are legitimated or justified by means of comparisons with what goes on elsewhere, what has been done in the past, or what others do. The contrasts are constructed so as to provide the hearer with the opportunity to recognise which state of affairs is to be preferred" (Coffey and Atkinson, 1996: 104). Some respondents referred to the principle of choice and of not judging others, but indicated nevertheless a moral view about how this choice should be exercised:

31,798: I mean, some people do feel as if they can't cope with somebody that's going to have ... all these things. Which I totally disagree with, but I think that's individual choice. I mean, I wouldn't sit and say, "Well, you shouldn't have done that". You know, I would never say to somebody, "You shouldn't have an abortion, that's really wrong". Even though myself, I do think it's wrong. (Laughs) ... I mean, I would accept it's the person's choice. But then, your choice isn't my choice.

Those who took this view that parents should accept all children equally did not discuss the circumstances or qualities of parents or the implications of different conditions. Several noted that, since their views about selective termination applied to any condition, information about the unborn child in pregnancy was unnecessary (although some might find it helpful). As one woman, asked about screening and diagnostic testing, said, *"they should be born anyway"*. Nor was there any discussion about the ability to cope, rather, from this position, *"you have to get on with it"*.

As discussed, almost all women felt that the option of abortion on the grounds of foetal abnormality should be available, and some women, often those who spoke more of agency and independence in their own lives, tended to stress the value of choice, rather than morality and the right to life. It has been suggested by sociologists and disability activists, however, that choice is offered in a social context of prejudice (Morris, 1991) or medical values (Lippman, 1992b). Some women who supported choice nevertheless expressed their own views about the way that impairment or disabled people should be seen:

JG: Do you think other people should be allowed to have abortions for these kind of conditions if they want to?
13,279: If they're really against having it, yes. But I don't see why they should be. Because they are just like everyone else.

Another respondent noted that selective abortion might be felt to be a necessary course of action by some people or in some circumstances, saying, *"If the person feels that they just can't cope with ... the father or whatever and the disabled child, because of the disability, then it's better getting an abortion, rather than having a child, and the marriage maybe breaking up"*. At the same time, she suggested a contrast with her own different priorities or perceptions, emphasising both the lack of importance of physical abilities, equal rights and value, and using the term 'disability':

30,641: I've got my views on abortion and things like that. I don't agree with that. Like if the baby has spina bifida and obviously it is a big thing, spina bifida and other disabilities. But I say, "Why shouldn't they have, like, a chance to live, when it's got as - you know - as well a chance to survive as anybody else?" You know, it is a human being after all, even if it can't do things that other people can.

Other women, several of whom thought that they would not themselves have an abortion (on any grounds) emphasised others' right to choose, saying simply, *"I mean it's their choice. Whether they want to have (an abortion) or not"*. This type of brief response was given by respondents in all groups who emphasised the principle of choice. From this perspective, there was no discussion of whether 'prevention' might be seen as right or wrong, suggesting that others' might have their own perceptions or their own values; it suggests a non-judgmental approach to others' choices.

The majority of women with spina bifida indicated that they would not consider abortion on the grounds of any abnormality themselves. However, a few women, who supported choice or who anticipated more personal difficulties of having a disabled child, spoke about dilemmas and decisions for a parent. Termination of a pregnancy was spoken of as a serious step, but as preferable to bringing a child into the world who would be ill, or that people might have difficulty in caring for. From this perspective, information from screening and diagnostic tests was spoken of positively as helpful, even necessary:

JG: How do you feel about these kind of tests being available?
40,896: I think they're rather good. Because ... because it lets you know. It would let you find out for yourself. And it gives you so long ... I think it's so many weeks it gives you, to think on it, and that sort of thing.
JG: Mm. So, if they found out that it was, you know, people might be offered an abortion. How would you feel about that?
40: I think it's terrible to ... to have an abortion. But I think if it has to be, it has to be.

At the same time, most expressed some concern about the difficulty of taking a decision, and about possible pressure to take a decision quickly. Women spoke about the need for full information about the implications of different conditions, and one questioned the adequacy of the information offered with screening programmes:

19,428: I think it depends on the parent ... I think the parents should have a view too. But I think they should, maybe when they go and get advice, they should get a lot of advice before ... what they heard before, maybe visit like counsellors.

Personal predicted use of antenatal screening and diagnostic testing

Women were asked what information they might want to have in pregnancy about an unborn child. Several said that they did not think about having children, and a number of others said that, since they would not terminate a pregnancy on the grounds of any condition, information would be unnecessary:

30,834: Nothing really. I mean, as long as it ... I mean, obviously, I do want a healthy child, but if it is disabled ... I'm not saying I would love it any less, probably I might love it even more, you know, because of the disability.

Some, particularly those who had emphasised fate, nature, and a more 'accepting' philosophy, argued that there were reasons for not having tests. They mentioned concerns about the accuracy of the information, both the possibility of mistakes, and the fact that testing might not indicate the severity of the disorder. Reluctance to spoil the surprise of a new baby was also mentioned, but particularly the risk of amniocentesis, the possibility of harm:

17,551: I think really I would just leave it. Because, as I say, I've got reservations about a lot of the different things like that. I don't know whether it's the amniocentesis test but it can actually harm your baby, and I think if there was something wrong with it

I would rather just find out, rather than actually the baby being harmed. Before it's actually been born.

Although a number of women with spina bifida suggested that there would be no reason to have any tests in pregnancy, others, often those who had spoken of choice and the value of information in their lives, suggested that they might find information in pregnancy helpful. Most explained that information could be useful in preparing for the birth of an affected child. One respondent implied that she might consider her ability to care; hence finding out information could be seen as responsible:

16,350: Well, I'd like to know if it's got a disability and that. And whether I'd be able to handle it. Like, when it is born.

In talking generally and more personally about screening programmes, the majority of women did not talk about different conditions, coherent with their views that abortion was unacceptable or that the condition was not an issue. However, a few women suggested that they would see the implications of conditions as important.

Antenatal screening programmes for Down's syndrome and spina bifida

The views of most women towards the provision of screening programmes for spina bifida and Down's syndrome reflected their arguments and philosophies more generally. An antenatal screening programme is routinely offered for these conditions, and women suggested that they thought it was acceptable for this to be available. However, the implications of these conditions were little discussed, the majority implying that their general and personal views about the unacceptability of 'prevention' applied to any disorder. From this perspective, one respondent, reporting being asked for advice by acquaintances who had had a positive diagnostic test for neural tube defects, did not refer to the consequences of the condition, but advised a fatalistic philosophy:

31,584: I says (to her), "Well," - this was at the time - "what difference does it make, X?" I says, "If that's how your baby's going to be born", I says, "then that's obviously what was meant for you". And I says, "And if something happens in the birth", I says, "and the baby dies, then it was meant".

The majority of women suggested that they would not themselves consider abortion on the grounds of any abnormality. For example, one respondent spoke of the lack of importance of spina bifida in terms suggesting the equal value of the disabled person rather than the relative severity of the condition:

JG: If you were pregnant, would you think about having these sort of tests?
13,270: No, because I wouldn't bother if it had spina bifida. Because they're just like everybody else, they've just got a disability.

However, a few women who spoke generally of supporting choice, or who considered taking part in screening programmes themselves, discussed the implications of spina bifida. Most indicated that they would not terminate a pregnancy on the grounds of this condition, because the condition was not seen as serious, partly perhaps because of their familiarity with the condition; as one woman put it, *"you know what you're letting yourself in for with spina bifida"*. As noted earlier, all respondents felt that others in the population would see the condition as more serious than they did themselves. One woman argued for improved information to reflect the possibilities for people with spina bifida:

19,438: Just as soon as they (prospective parents) see spina bifida, they don't realise ... maybe they don't really know what the doctors are saying to them. I think doctors maybe feel like they are to blame too. Like they could say, "They can't do this for themselves ... ", but they should say, "They have difficulty but however they ... they're able to have a quality of life too."

This small group of women also discussed the different implications of Down's syndrome. Those women who discussed knowing people with Down's syndrome, whether through special schools or whether they were relatives or acquaintances, spoke in different ways. A few spoke very positively of friends, in terms of personal characteristics as *"brilliant"* or as *"just like everybody else but just got Down's syndrome"*.

On the other hand, a small number of women, while discussing the increasing integration of people with Down's syndrome, also spoke about their need for support and the implications for carers. Two women who took a more 'activist' approach to living with their own condition made a distinction between spina bifida and Down's syndrome:

19,497: They can help themselves more now. A lot of them are more independent now too. So a lot of people are seeing this. But I actually think, like Down's syndrome, it's like different because a lot depends on what ... when I'm older. Who's going to, sort of look after the kids, too, I think that's it with the Down's syndrome. I think everything's completely different.

Cystic fibrosis carrier screening

Women were asked about their knowledge of cystic fibrosis and its inherited nature, and a diagram was used to explain the pattern of inheritance. It was explained that carrier screening for this condition could be offered both during and before pregnancy, and women were asked whether they might think about having such tests and whether they thought that carrier screening should be offered.

Within this group of women there was a wide range of knowledge about cystic fibrosis. Several women said that they had very little or no knowledge of this disorder, and some of these people felt that they knew too little about the disorder to answer questions about screening. Several other women, on the other hand, had known children with cystic fibrosis through their attendance at a paediatric clinic or at school. Almost all said that these people had died, and this was spoken of as 'hard to deal with'. Questions about cystic fibrosis carrier screening were mostly answered by this group of women who had some first-hand knowledge of cystic fibrosis.

All those who were asked thought that it was right for cystic fibrosis carrier screening to be offered, whether before or during pregnancy. A number of respondents thought that they might find antenatal screening or diagnostic testing useful to prepare for the birth of an affected child. The responses of some suggested that information about cystic fibrosis might be welcome in pregnancy, sometimes in contrast to their views about information about spina bifida or Down's syndrome. There might be a number of reasons for this view: cystic fibrosis could be a less familiar condition; it might be felt more important to find out about a chronic illness than an impairment; non-invasive carrier screening can give information about risk, which might be useful for others.

Most women indicated that, as for any condition, they would not terminate a pregnancy. Another respondent thought that she might be less likely to consider a termination for cystic fibrosis than for some other conditions such as Down's syndrome; she felt that the condition was one that she "*would just learn to cope with*".

However, one respondent had an opposite view. She had indicated that she saw no reason to prevent the birth of a child with spina bifida, saying, *"I mean, it's just like any other kid except they're in a wheelchair. I mean, we need a bit extra hospital problems but there won't be ... there'll never be anything drastic"*. She spoke in very different terms about cystic fibrosis, talking of the condition as *"a very serious thing to happen"*, and of a friend who had died as "*very very ill*". She felt strongly that population carrier screening would be useful, so that carriers could avoid having affected children, but also felt that a concern for the suffering of a child with this condition would over-ride her reluctance to have an abortion:

JG: You know, how do you feel about abortion?
20,999: Oh I don't know, I don't really like that idea, but if the baby was - you know - ill or there was no chance ... that it was going to be healthy, then I wouldn't want it ... I wouldn't want it in this world.

All those who discussed cystic fibrosis carrier screening felt that screening before pregnancy was preferable to antenatal screening, and several thought that they would

have such tests if they were offered. However, although one respondent expressed concerns about a logical outcome that people might stop having children, in general, there was no view that it might be wrong to prevent the birth of an affected child by this means. The response of one woman who spoke in strong moral terms about antenatal screening as a search for perfection suggested that carrier screening before pregnancy was seen in a different way:

31,790: Like, (name): she's been tested and she's found out that she's a carrier. And what her mum said was, she'll have to think really hard before she goes and has children. Not saying that she wouldn't have them, but she'd really need to know the pros and cons before she goes ahead.

Discussion

For a number of women with spina bifida, questions about intervention in reproduction provoked little discussion. Almost all felt that it was acceptable for screening programmes and for the option of 'prevention' to be available. However, only a minority of these women discussed reasons why screening programmes might be felt to be beneficial, such as the welfare of affected individuals, difficulties in caring for a disabled child, or the provision of reproductive choice.

Where women raised concerns about screening programmes, these related to those offered in pregnancy. The majority of respondents indicated that they would not have an abortion on the grounds of any foetal abnormality. For many, this position was linked with a general view about the unacceptability of abortion. The responses of some suggested some disapproval of the values of those who might terminate a pregnancy on the grounds of foetal abnormality. They spoke about equal rights to be born, and several used the term 'disabled people' in this context. Unlike disability activists, however, these women did not argue that the provision of screening programmes was wrong or indicated evidence of societal prejudice. Their concerns about the morality of action and about 'selection' related to wrong individual values of a lack of acceptance.

The concerns of many of this group about people's attitudes towards having a disabled child, rather than concerns about welfare, were coherent with their opinions about the unacceptability of abortion, but also with a general view of the lack of importance of physical limitations. The implications of different conditions, for those affected or for others, were only discussed by a minority of women who indicated less strong views about abortion on the grounds of foetal abnormality. Their responses suggested that cystic fibrosis was seen as more serious for the affected individual than spina bifida, whereas the consequences of Down's syndrome of dependence were seen as more

serious than those of spina bifida for the parent or carer. However, numbers were very small.

Women were asked these questions because it was felt that their views might be shaped by their knowledge and experience of living with spina bifida or their experience of disability. Almost all women suggested, often on the basis of their own experience and their own perceptions, that spina bifida was a condition which it was not necessary to prevent, since having the condition did not preclude a happy life or a good quality of life. All also felt that others in the population would have an over-negative view of the condition, especially about the limitations of those affected. Their views can be seen as challenging assumptions about inevitable problems or suffering for the affected person and perhaps also about difficulties for others, although a few women offered a more personal and hesitant view.

In discussing living with spina bifida or with a physical impairment, most women discussed some problems. The majority of those mentioned, such as difficulties with access, a lack of opportunities, others' attitudes, were seen as not inevitable, but as potentially addressable with social change. Many women also saw such problems as being common to all individuals with a a mobility impairment or to disabled people as a group. Several women referred to themselves as disabled (although disliking the negative connotations of 'difference' in the term), and suggested some experience of socially constructed disability, as proposed by the 'social model'. Although only a small number reported being involved with campaigns, their accounts suggested a familiarity with the arguments of disability activists.

At the same time, although many spoke of socially-created difficulties, women also discussed more personal 'solutions' or strategies. There were differences within the group in women's approaches towards living with the condition and perceiving their impairment, and these different philosophies seemed to be linked with their approaches towards screening programmes. Those women who suggested a more 'activist' approach to life tended to support the principle of choice, and some indicated that they would see the dependence of an affected child as important. Other women, from a more 'accepting' philosophy, stressing equal value, tended to emphasise unconditional acceptance of an affected child.

Views towards screening programmes might be shaped by a number of factors, both personal and cultural. The views of women with spina bifida can be understood within their social context of their lives. Disability activists have been critical of screening programmes, arguing that 'prevention' is both discriminatory and unnecessary - being based on a 'medical model' of assumed problems - and have claimed that these values

underlie many of the problems faced by disabled people. Women's views about the unacceptability of abortion on the grounds of foetal abnormality might be seen as reflecting this rhetoric, feelings about the value of others like themselves or their perceptions of their own experience.

However, women did not give reasons for their views. The majority of these women (eleven of fourteen respondents) reported having a religious belief, and eight spoke of being brought up as a Catholic; the unacceptability of any abortion expressed by many of these women might be seen as associated with these values. Some commentators on empirical work have suggested a link between a view of abortion on the grounds of foetal abnormality as unacceptable and religious beliefs (Faden *et al.*, 1987; Evers-Kiebooms *et al.*, 1993), although others have found little evidence of such a link (Parsons, 1990) or have argued that this is a complex matter (Press and Browner, 1997).

At the same time, a view of abortion as personally unacceptable might reflect cultural values in the West of Scotland, or might be shared by women of a similar age. As noted earlier, research has suggested that abortion on the grounds of abnormality has been seen as less acceptable recently among young people (Birth Control Trust, 1997). In order to consider similarities and differences within and between groups, the following chapter outlines the views of women with cystic fibrosis towards screening programmes and the way that these women spoke about their experience.

CHAPTER 9

WOMEN WITH CYSTIC FIBROSIS

Women with cystic fibrosis were included in the study because of the recent development and trials of carrier screening programmes for this condition, and the debate about whether it was right to offer such screening to the population. This chapter looks at the way that women with this condition spoke about their experiences of living with the condition, their perceptions of its 'severity', and their views towards the 'prevention' of cystic fibrosis and other conditions by screening programmes in reproduction.

Background

Cystic fibrosis can be seen as a chronic illness. It is an inherited condition which causes mucus to be produced both within the lungs, causing a progressive lung disease and a proneness to chronic chest infections, and within the pancreas, causing an enzyme deficiency. Although the condition is always present from birth, there is considerable variation in symptoms and in the severity of the disorder between individuals, and a person with the condition may have periods of ill-health alternating with periods of stability. Cystic fibrosis was once fatal in infancy or in childhood, but increasing knowledge about the condition has led to improved diagnosis and treatment, and to a dramatic improvement in prognosis and survival. People with this condition can now live into adulthood.

As outlined earlier, a number of studies have examined the characteristics and the experiences of adolescents and young adults living with cystic fibrosis. Empirical work has suggested the increasing autonomy of many people with the condition (Shepherd *et al.*, 1990; Walters *et al.*, 1993). A recently published study in the UK of the experiences of adults has indicated that the perceived impact of the disorder varies with its severity, but that respondents have a positive attitude towards living with the condition (The Gallup Organisation, 1996).

Professionals in the field of cystic fibrosis consulted for this study all felt that they would see the disorder as a serious condition, pointing out as evidence the short life-span and the potential interference of both the condition and the treatment regime on the lives of those affected. However, they also noted the benefits of new techniques for management of the condition, the resources available at specialist centres, and the improvements in both prognosis and life-expectancy (although some cautioned against over-optimism in relation to early treatment by gene therapy).

Living with cystic fibrosis

Interviews were carried out with fourteen women with cystic fibrosis. In response to questions about their father's (or mother's) job, seven women reported non-manual employment and seven manual. Only two women in this group said that they attended church regularly; others described themselves as having no religion, and none had had a Catholic upbringing. In response to questions about educational level, some women explained that they had had to abandon their education because of ill-health. However, half the sample described attending or having attended courses in higher education, or said that they had been expected to do so.

Women were asked what they had done since school, about the ways in which they saw the condition as affecting their lives and about their quality of life. Like women with spina bifida, many respondents talked at some length about their lives, and discussed various aspects, such as jobs, relationships and hobbies. While the responses of several suggested similarities to the lives of others, women also pointed out the ways in which they saw their lives as different because of the consequences of cystic fibrosis.

Consequences of having cystic fibrosis

All reported having attended mainstream school, although a few said that they had increasing health problems during secondary school. Two women were currently in full-time higher or further education and one part-time, five were working full-time, two part-time, and two others, who reported being unable to get work to suit their state of health, were doing some voluntary work. Two women said that they were too unwell to work.

Several women discussed the nature of cystic fibrosis, its unpredictability, and the variation in symptoms, and all described the ways in which they were affected themselves. There are few visible symptoms of the disorder, and most respondents did not look ill, although some had a cough. All felt that the condition had an impact on their lives, although their perceptions of important consequences and their reported experiences and histories varied widely. Women described a range of symptoms and physical limitations, from one who felt that the condition had almost no impact on her life, since she took no medication and had no chest problems, to another who was unable to leave the house.

All women with cystic fibrosis had been contacted through a specialist clinic and, in talking about their lives, all discussed - sometimes at length - their contact with hospitals and medical staff. Although women were ambivalent about attending clinics, all referred positively to the specialist knowledge of consultants, using such phrases as,

"they really know their stuff", and to the information that was available. They also discussed recent improvements in the treatment of cystic fibrosis. The accounts of most women indicated a considerable knowledge of the disorder and its effects; several mentioned seeing others affected with CF at hospitals and clinics.

Almost all women - by reference to symptoms, their level of medication, or to the frequency of their contact with the hospital - compared the 'severity' of their condition with that of others with CF, and several felt that they had a milder version of the disorder than many. A number compared their present state of health with that at an earlier age and described a stage at which their condition had worsened. Many also compared what they felt able or unable to do with people who did not have cystic fibrosis. The 'relative severity' of their condition was often discussed, and seemed to be an important issue.

All respondents described the extent to which they felt able to take part in the same activities as other people without cystic fibrosis, because of the limitations of the condition or because of having treatment. Two women, who reported being more seriously ill at a young age, referred to losing contact with friends who did not have CF, saying they had *"missed out on a lot"*. A number of women referred to their ability to work.

However, employment was not always spoken of as a straightforward matter of being able or unable to work. Some discussed anticipated or actual difficulties of getting work when employers knew that they had CF, and a few spoke more in terms of a lack of opportunities:

22,003: Well, when I left school, I did a YTS for two years, I did (vocation). But when I left that, I went to the Job-Centre, and they says, "Forget it. Go on the sick."

Although the majority of women spoke of consequences as related to the severity or progress of the condition, some spoke of certain difficulties as more socially caused. A number of women referred to ways in which they thought that people with cystic fibrosis were perceived by others. Several pointed out that they looked no different from anyone else; as one explained, *"I don't look not well; I just look like any other person"*. At the same time, some felt that people who were known to have CF were seen and treated differently from others.

While some spoke of certain symptoms as intrusive or embarrassing in themselves, several made the point that 'the cough', for example, might 'give away' a diagnosis of cystic fibrosis. A number of women reported being worried about others' reactions, because of the way that they thought the condition itself would be seen:

34,127: I don't know, I mean, I've got CF, and I know I shouldn't be so embarrassed about it. But you know, when you say CF, people automatically think, "Oh my goodness, cystic fibrosis". But, you know, people know I'm quite healthy and fit so ... so I don't know.

Cystic fibrosis is a progressive condition, and those affected have a shortened life-span. Some women reported that, where people knew that they had the disorder, they were treated differently from others in the sense of being pitied or over-protected (or "molly-coddled"). There was no mention of societal attitudes of prejudice - as noted in another study (The Gallup Organisation, 1996) - suggesting that the condition was not seen as stigmatising. However, others' attitudes were not mentioned as a problem by all. Those who reported being more seriously affected did not speak about others' perceptions of the condition, or others' treatment of themselves as problematic; as one woman said, she did not *"bother about anybody else's attitudes"*. They talked instead of their need for support.

In discussing their perceptions of others' attitudes, several women noted that other people were 'scared' of cystic fibrosis. A number pointed out the possible effect on others of knowing someone with a life-threatening condition:

26,235: ... Because it's ... for other people, you know, they're kind of frightened as well, because they know that I've lost a lot of people, so maybe they think it's going to happen to me. It happens to everybody but they tend to think me, sooner than later, because of the situation.

The majority of respondents referred in some way to consequences for close friends and family of their having cystic fibrosis. While most women reported being treated 'normally' by friends and family (like women with spina bifida), or being encouraged and supported, a number also mentioned possible effects on close relationships. Some described difficulties of telling close friends that they had the condition, and a few noted that boyfriends might want to consider whether to become involved; one pointed out that some *"walk away ... if they can't cope with it"*. As Bury (1988) has noted, there may be different implications for those in close relationships with people with a chronic illness or a disabling condition, because of the future, or dependence.

Parents and siblings are in a different situation from friends in that they do not have a choice about the relationship. Professionals in this study suggested that there could be strain in family relationships, both between the affected sibling and others, and between parents, where a child had cystic fibrosis (or spina bifida). Most women, like others in the study, did not discuss family relationships, although a small number referred to some tension with siblings. Some respondents with cystic fibrosis described the practical involvement of family members, usually their mothers, in their care, often when they were younger, and a few spoke about their (own) current need for support.

Several reported the concern, worry or anxiety of their parents, and a few spoke in stronger terms about the effect on the whole family, or about grief or distress.

A number of women suggested generally that living with cystic fibrosis could cause emotional and psychological difficulties for those affected themselves. A few spoke more personally, reporting the depressing nature of long-term treatment, of being in hospital for long periods, and of feeling unwell. Others talked about their anxiety about their condition worsening, a lack of direction in life, or a general depression about the concept of the condition and the future, sometimes reinforced by seeing others more seriously ill at the clinic. Two women discussed the difficulty of dealing with the deaths of others with cystic fibrosis who they were close to, friends or a sibling. These issues have been suggested as important for people living with a terminal illness by others reporting empirical work (Fulton, Madden and Minichiello, 1996). Although Fulton and colleagues have suggested that emotional issues might be little discussed in interviews, in this study a number of women with cystic fibrosis did talk about such difficulties.

However, not all women discussed problems; many spoke positively about their lives. They also considered the means by which and the extent to which difficulties might be addressed. All respondents spoke of the contact with the specialist centres as useful, because of the knowledge and expertise of the medical staff. They also spoke positively about new treatments and techniques for management of the condition, although some were more optimistic than others about a future cure by gene therapy.

Most also mentioned their own strategies or approaches towards living with the condition. Like women with spina bifida, a number discussed the importance of maintaining psychological well-being, and several also spoke about looking after their health. Practical strategies for the management of the condition were often spoken of together with attitudinal strategies, a number suggesting that "a good mental state" or a positive frame of mind could affect one's physical condition.

Some respondents also said that it was useful to be able to talk about feelings of fear, grief, or anxiety, although several also noted that it could be difficult to find an appropriate person to talk to. One pointed out that she would not discuss worries with a doctor, saying, *"I just ... I never ... you feel as though you should never ask a doctor, you know, "How am I really?" You know?"* Women said that they did not want to worry or distress others; as one pointed out generally, *"some people find death hard to talk about"*. While a few spoke of talking about worries openly within the family, the majority suggested that they tried to minimise problems, or suggested emotional independence:

15,254: I tend to basically deal with it myself. Do you know what I mean? I mean, my friends, my close friends know. But I still, sort of, deal with it myself. I'm quite, sort of, independent!

As with women with spina bifida, a vertical analysis of the interviews suggested that, within this group, women had different approaches towards living with the condition. These might be seen as 'activist' and 'accepting' strategies.

Activist approach

The majority of women with cystic fibrosis spoke of an approach to living with the condition which could be seen as 'activist' or 'normalising'. They emphasised that having the condition did not prevent them being able to take part in the same activities as others, or a 'normal life', and many described their social life, their hobbies, their goals or plans for the future, and their jobs. As one said, *"I mean, OK I've got an illness, but it's not that bad that it stops me going out and doing whatever. I can still get out and about, which I'm grateful for, do you know what I mean?"*

Women with this approach spoke in terms of preserving a normal way of life for as long as possible, a strategy noted in other studies (Bluebond-Langner, 1996). Several suggested that they had very full lives, and some spoke of the need to *"work round"* the condition, or *"organise your day"*, in order to both deal with the condition and take part in everyday activities. They described a general approach of dealing with the condition and forgetting about it, getting on with life as a person. Several spoke of 'trying not to let it interfere' or the need to *"put it to the back of my mind"* as helpful in minimising the impact of the concept of the condition:

26,1598: You learn to cope with it. If you be normal, if you act as if everything's normal, then you'll get on all right. Because, believe me, I don't go to my bed at ten o'clock every night.

The accounts of a few women suggested that an approach of getting on with life, living as normal a life as possible, minimising the effects, was seen as the best philosophy, not only for themselves in avoiding self-pity, but as generally seen as preferred or right. As one woman put it, by means of a contrast:

28,261: She (best friend) is like that, "See the difference between the way you handle things and the way P handles things". Nobody would know - apart from her - that I had anything wrong with me.

Women with a more 'activist' approach often compared their physical condition with that of a person without CF. As one reported, " ... *So basically ()% of my lungs is functioning correctly. So I says, "So how does that compare ..." I always say, "How does that compare with somebody that's normal?"* A number of respondents with this

perspective also spoke about the measures that they took to challenge limitations or to control the progress of the disorder; they referred to their knowledge of new treatments, and described trying to keep as fit as possible. Several mentioned the encouragement of family and friends:

34,431: X is very encouraging. She goes to (exercise) with me and things. So really, I mean, she's ... you know ... confirms the view that she doesn't think I'm badly affected by it, and ... you know.

At the same time, some also reported some ambivalence about information and hospital contact and a dislike of being a 'patient', and the accounts of some indicated some anxiety about the progress of the condition.

Acceptance approach

Other women, especially those who had described a deterioration in their condition, spoke in more pragmatic terms about accepting the limitations of the condition, or mentioned the short life-span. These women spoke more of welcoming the support that they received from friends and relatives, and from hospital staff. One woman, by means of a comparison, suggested an approach of acceptance as personally helpful:

18,673: But T just totally denies that there's anything wrong with him. For him it works. Because ... I think T's a different type from me. He doesn't get (infections). And when he does, T seems to be able to cope. And fight. I can't. I've tried it. I've tried coping with it.

Several women suggested a more fatalistic philosophy of acceptance, rather than always worrying, saying, for example, *"I just get on with everything And I don't really let it get me down"*, or *"I just make the best of what I can"*. However, within this latter view, some women suggested a more positive philosophy of acceptance, saying that having a disorder with a short life-span need not be seen as important, and need not preclude a good quality of life.

While some women spoke more of a normalising 'strategy', and others more of acceptance, like women with spina bifida, a number spoke in terms of a balance. Professionals in this field spoke approvingly of an approach of living as normal a life as possible, keeping CF as a part of life, but they also pointed out that to ignore the condition could be seen as 'denial' They noted the different approaches of 'denial' and 'acceptance' of different people with CF, as ways of coming to terms with the condition.

Although not all respondents offered 'strategies', it was clear that, because of the progressive nature of the condition, different philosophies might be employed, or felt more appropriate, as the condition deteriorated, as noted in other studies (Bluebond-

Langner, 1996). Kübler-Ross (1970) has proposed a series of psychological stages of acceptance of a terminal condition.

Those women who mentioned the actual or anticipated attitudes of others as a problem described a variety of ways of dealing with this. Several said that they told almost no-one that they had cystic fibrosis, apart from immediate family or very close friends, or they described concealing symptoms, because of the way that they felt they might be seen:

24,082: Nobody knows. I don't ... I don't really tell anybody. You know. Because I feel they will judge me on that, and it colours their opinion of me. I'd probably be more likely to tell people once they've got to know me. And then I can let them be surprised, when they know I'm just normal.

A number of women referred to strategies of managing information or 'passing' as normal, a mode of adjustment suggested by Goffman (1968) as useful both in maintaining self-concept and in avoiding being treated in a stereotypical way. However, since cystic fibrosis is a progressive condition, which can only be hidden in the early stages, women spoke of telling others when this was felt necessary and according to the closeness of the relationship. Several noted that it was difficult to explain the implications of the disorder, and said that they did not give *"full-blown explanations"*, because, as one woman put it, *"When you start to explain it to them, you can see ... a lot of ... sort of ... face changes and that"*.

Many, however, felt that not enough was known about the implications of CF, and a few women said that, in some circumstances, they made a point of 'educating' others about the condition, or correcting wrong assumptions:

26,123: If I met people, I didn't want to tell them what was wrong with me, and just ... things like that. And then I thought to myself, "No - people should know, because then it might make them more aware", you know. Everybody that ... I've met a lot of people through my boy-friend, and they all know about it now, and they ... they just look on me as being a normal person. Because I am a normal person.

The 'strategies' described by women in this study were similar to 'typologies of adjustment' - 'secret', 'pragmatic', and an educational approach emphasising equal rights - which have been suggested in a study of people with epilepsy (Schneider and Conrad, 1981). However, while different approaches were described for dealing with others' perceptions, or with being both 'the same and different', all included the idea that women wanted to be seen as a person like anyone else, rather than as defined by their condition, as discussed by Kleinman (1988).

In talking about dealing with possible difficulties, women did not suggest the need for changes in the way that people with cystic fibrosis (or other conditions) were treated in society. The majority, when asked, said that they did not see themselves as disabled.

Many of the findings of this study confirm those suggested by Alderson (1998), who interviewed ten young men and women with cystic fibrosis about their lives and about their views of screening programmes. Alderson found that respondents had a positive approach towards living with the condition, although noting that this attitude might be required in order to take part in the study. Most suggested the need for improved knowledge within the population about cystic fibrosis. Several also felt that improved benefits and opportunities for those affected would be helpful.

Quality of life

Respondents in this study were asked how they would define quality of life - what they saw as needed - and also how they would evaluate their own quality of life. As when they were talking about living with the condition, a number of women with cystic fibrosis suggested a link between the physical and the psychological.

The majority saw the quality of their lives as related to the severity and the progress of the disorder. They suggested, in talking both personally and more generally, that a good quality of life was achievable until the condition deteriorated, saying for example, *"I think it's not too bad. When you're not well at all, it's not very good. The thing is with CF, you're in the house, really, you don't get out"*, or *"As long as I can still walk and do things myself, then I'm happy"*. There was a range of assessments of personal quality of life, from one woman who replied, *"not too great the now"*, to others who reported being less seriously affected and having a 'normal life' or a 'good life', although this was qualified in many cases with, *"at the moment"*.

Asked what they would see as important for a good quality of life, several women with cystic fibrosis, unlike other women in the study, spoke of the importance of health. One said, *"a healthy body"*, and another replied:

18,561: Well I would say, 'good health for everybody'. But until you don't have good health, that's not a factor. You know, a lot of people, they would say, 'money' or 'happiness', but I would say, 'good health', always. It would always come top priority. If I could scrub something in life, this is what I would scrub.

Others, however, suggested a philosophy of focusing on or pursuing happiness rather than 'normality' or health. One woman spoke of *"getting rid of things that don't make you happy"*, especially in terms of relationships. As mentioned earlier, some women

felt that having cystic fibrosis or a condition that shortened life need not prevent happiness or a good quality of life:

15,310: I think, life-wise, I prefer to live a happy short life than a miserable long life, I think. Do you know what I mean? I think you should just take out of life what you can, you know, you've got to put something extra into things as well.

While a few suggested that a good quality of life in terms of happiness or relationships might be independent of the condition, another respondent resisted the idea of evaluating quality of life:

38,338: (Pause) I suppose it's fine. Just like everybody else's really. I don't really think of it as a quality of your life, you know. Everybody's got to go at one point, I mean, whether you've got cancer or cystic fibrosis or anything.

A reluctance to evaluate quality of life in terms of happiness or health, but just 'life', can be seen as a fatalistic or 'acceptance' philosophy. This may also be seen as relevant to the study, in that the important issue may be seen in terms of a valued life, rather than happiness or health.

Summary

As discussed in the previous chapter, 'strategies' relating to the way a condition and its consequences are conceptualised or perceived might have implications for the individual, for example in enhancing quality of life, but they might also suggest more general philosophies. Strategies might vary with the person, but might draw on more general cultural values. An analysis of strategies and of the way that people discuss these can suggest more general values or philosophies.

Most women with cystic fibrosis spoke in narrative terms, and they described very different experiences of living with the condition. A vertical analysis suggested different approaches of normalising or accepting, often depending on the stage of the disorder. The responses of the majority suggested that health was seen as important, quality of life being linked with the physical condition. The fact that many women spontaneously described 'strategies' for dealing with the physical effects of the condition or the concept suggests that having the disorder was perceived as important. These may be seen as 'medical' values.

Like some women with spina bifida, several women made the point that they saw themselves and wanted to be seen by others as 'normal'. However, few emphasised rights, equal value or societal prejudice, and the majority did not speak of themselves as disabled. The social model of disability may be less appropriate for reflecting the experiences of those living with chronic illness.

A number of concerns about being seen as 'different' related to the way that women thought that cystic fibrosis was perceived. Almost all respondents spontaneously discussed others' assumptions about the condition, the relative severity of their own condition and of the disorder more generally. Since women knew that the study was looking at reproductive issues, the topics that were raised when women spoke about their lives might have been important for themselves but might also be relevant to screening programmes.

New reproductive technology, screening programmes and 'prevention'

Motherhood

Like other women in the study, women with cystic fibrosis were asked whether they thought about having children. Men with cystic fibrosis are infertile (Dodge, 1995) but affected women can have children. One woman in this group had a child, and others spoke about hoping to have children in the future. Other women said that they did not think about having children; although they were not asked for reasons, a few noted concerns that they might not be able to care for a child or that child-bearing could endanger their own health.

Professionals in this field noted that an increasing number of women with cystic fibrosis were having children, but they also stressed that child-bearing could be risky for women with a low lung-function. Consultants referred to literature suggesting that pregnancy might exacerbate both lung and nutritional problems, but was well tolerated where a mother had mild disease (Edenborough *et al.*, 1995), and they pointed out that they would have a view about the advisability of pregnancy for different women under their care. They also noted that the partner of a woman with CF would be advised to be screened for carrier status, since, if he were a carrier, children would have a 50% chance of being affected.

Those women who thought that they might have children said that they would discuss this with consultants, and several said that they had done so. All saw carrier screening of a partner as acceptable, and often as sensible. Some women reported that they had been advised to have children *"sooner rather than later"*, and a few spoke of considering pregnancy in spite of medical advice and an awareness of the risks. Some said that they had discussed the possibility of having children with other family members; one woman referred directly to the fact that others might have the responsibility for caring for or bringing up a child, saying that consultants *"want to know who'd be there to look after the bairn"*.

Abortion

Abortion was spoken of as option which should be available. Only one woman in this group spoke of abortion as wrong because it was terminating a life. Like other women in the study, some respondents thought that they would not have an abortion themselves, talking of this as something which they would not want to go through. This response was typical of several:

35,434: Uh, I'm not really for or against abortion. I would rather not do it, it would be a last resort, abortion. But I'm not against it, really.

In common with other women in the study, all felt that abortion on the grounds of the sex of the child was wrong. Women were asked what they felt about the possible provision of information in pregnancy about physical characteristics, such as fair hair or blue eyes, but a number had spontaneously raised this issue in discussing tests for foetal sex. Although some spoke about not spoiling the surprise of a new baby, a number of women expressed concerns about the provision of certain information in itself. Several women broadened this issue to discuss the limits of acceptable testing, and some suggested more general social consequences. The dangers of *"genetic engineering"*, trying to get *"that kind of society"*, *"a super-race"*, or *"the Aryan race"* were mentioned by several women, who argued for diversity, suggesting not only that these tests were unnecessary, but also that they should not be done.

Abortion on the grounds of foetal abnormality

Asked their views about abortion on the grounds of foetal abnormality, all thought that it was appropriate for this to be available, within limits. Only two respondents expressed concerns. While one saw all abortion as wrong, the other used the term 'disabled', suggesting that abortion on the grounds of foetal abnormality could be seen as discriminatory:

22,359: I think some abortions are wanted, you know. But the likes of ... you know, just aborting it because it's disabled or something, well I think that's like trying to get rid of all disabled people and I don't think that's right. Maybe in severe ... severe cases.

Like women with spina bifida, these respondents suggested individuals' wrong choices or values, not that screening programmes were wrong in themselves. In contrast to women with spina bifida, however, both pointed out that there might be conditions for which abortion might be felt to be the right course of action or justified; as one put it, *"Very very ... really strong circumstances ... that's different"*.

Most women suggested that they would see it as generally right for individuals to take decisions about abortion on the basis of the perceptions of their ability to care for the child. As one suggested, *"a lot of people wouldn't be good parents, if they were faced with something that they couldn't cope with personally"*, or another, *"I mean, somebody else might not have the attitude that they could handle it. So I guess it's their choice"*. At the same time, however, some pointed out that there could be assumptions about the implications of different conditions, that public perceptions might be inaccurate, or that people could be frightened of what they did not know about.

Screening programmes

All respondents saw the provision of screening programmes generally as acceptable and the majority felt that they were potentially helpful for individuals; as one woman put it, *"they're only offered really for your benefit"*. In relation to antenatal screening and diagnosis, while some pointed out that information might be useful in itself, the responses of many women with cystic fibrosis implied that benefits might include the option of terminating the pregnancy:

18,827: I think if it happened, you'd rather know ... you would think, "Well, I'll have to look after it." It gives you the option as a person as well ... Because it affects your life, not just its own, and I think if you were pregnant, you would want to know what's going to happen.

Asked what they themselves might want to know about in an unborn child, a small number of women felt that they would want no information, since they would not terminate a pregnancy. The majority, however, thought that they would want to know about the 'health' of a baby. Several referred to perceived difficulties in caring because of their own health, and some felt that they might want to avoid giving birth to a child with certain conditions.

Antenatal screening programmes for Down's syndrome and spina bifida

In line with their general views, all women thought that it was right for antenatal screening programmes for Down's syndrome and spina bifida to be available. Most referred to the implications for the carer, the perceived ability to care being spoken of as an ethical or responsible reason for deciding about abortion:

39,621: Everybody's different. Some people would be able to cope, with having somebody with spina bifida or Down's syndrome. Other people just know that they wouldn't be able to handle it. I don't really see the point in ... you know, some people say, "Oh, you can't do this and you can't do that". I think it's up to them as individuals. You know.

A few women considered the severity of these conditions for the affected person. One respondent who felt that it would be wrong to terminate a pregnancy on account of almost any condition also argued that, in her view, people with both spina bifida and Down's syndrome were able to 'have a life', and that 'prevention' would not be justified.

However, in contrast, a number of women in this group, in discussing whether screening programmes should be offered for different conditions, suggested that they would see spina bifida and Down's syndrome as 'more serious' than cystic fibrosis:

34,595: I mean, I can't say that that should be available and not CF, you know. But it's just because I don't see CF as so serious, but that's really through personal experience, you know. Um, I don't know. I mean ... I suppose spina bifida comes with varying degrees of severity as well, doesn't it, really?

Although it was not always clear whether 'severity' related to the implications for the carer or for the affected person, two respondents contrasted their perceptions of the quality of life of people with cystic fibrosis with that of people with either Down's syndrome or spina bifida. One suggested that someone with a visibly different condition such as Down's syndrome would experience a poorer quality of life:

27,1041: You know, the difference between maybe having a baby with Down's syndrome and cystic fibrosis is the fact that one's going to have a better quality of life. Because as soon as you're born with a physical appearance of disability, you know that it's not ... not to be normal then that's ... that's a big barrier you've got to face. And I mean, the worst you're going to get with people with cystic fibrosis is chest infections.

The other spoke in very similar terms about the relative severity and untreatable nature of spina bifida, again implying few difficulties for individuals affected with cystic fibrosis:

28,811: That (spina bifida) is more of a disability than CF is. Do you know? That can't be treated with a tablet. That won't make your back straighten out and your ... do you know what I mean?

Although tests for the two conditions were largely discussed together, a number of women particularly referred to Down's syndrome, and to perceived difficulties for the carer. Some suggested that there was a lack of knowledge of this condition, leading to fear. One woman saw a pressure from others in society to have a termination for Down's syndrome, saying, "*Everybody was saying, 'Oh, oh, she's some girl! Imagine coping with that!' you know. 'Oh', some folk were saying, 'she should have had an abortion'.*" Pressure was seen as coming from other individuals, rather than from medical professionals or from assumptions of benefit in routine screening programmes, as suggested by social scientists (Farrant, 1985; Macintyre, 1997).

On the other hand, a number of other women suggested that there might, in fact, be particular difficulties for a carer, that support might not be available, and that particular qualities might be required. As one put it, *"I don't know how many people would be able to do that"*. Some women expressed uncertainty about their own ability to care for any child because of their own ill-health, and five of the fourteen women in this group, sometimes spontaneously, suggested that they might have difficulty in caring for a child with Down's syndrome:

15,417: ... If they tell you something really bad, like Down's syndrome or something. Because I don't know, because, I mean, with my health, I couldn't have a child with Down's syndrome. I couldnae cope with it. ... Because it's so demanding, it's so demanding, they need all your time, and I, obviously I couldn't give them all my time, so ... just ... I don't think I could.

Studies have suggested that an important factor in people's views about termination is the perceived burden of the child (Evers-Kiebooms *et al.*, 1993), and that perceptions of burden relate not just to the implications of a condition but to perceptions of individual susceptibility or the ability to cope (Ekwo *et al.*, 1987). Whereas a child with Down's syndrome was spoken of by a number of women as a burden on the parent or carer, many women spoke in a different way about the implications of having a child with cystic fibrosis. Some contrasted their perceived ability to care for a child with Down's syndrome or spina bifida, with caring for a child with cystic fibrosis:

JG: What sort of things would you like to know about, yourself? About a baby before it was born?
28,892: Just that it didn't have, um, the Down's syndrome or spina bifida. Um, if I found out that it had CF, that wouldn't really bother me, because I know I could handle it. You know what I mean, it wouldn't cause me a problem at all.

Although women were not asked directly how they would feel about having a child with cystic fibrosis, almost all women discussed this issue. They spoke about their perceptions of the severity of the condition, the importance of preventing the births of those affected, and by what means, and whether they felt that it was right for population carrier screening to be offered.

Cystic fibrosis carrier screening

The debate about whether it is felt right to offer a screening programme for cystic fibrosis, where the principle of screening is accepted, has been outlined earlier in the thesis. Some, including those in the medical profession (Scully, 1985; Alper, 1996), have questioned the appropriateness of preventing the births of those affected with cystic fibrosis, because of the improved prognosis and the hope of further improvements in treatment (Kabak *et al.*, 1984; Evers-Kiebooms *et al.*, 1988). In one study of the views of obstetricians, 13% did not recommend termination for cystic

fibrosis at any stage of pregnancy (Green, 1995). Consultants in this study felt that it was right to try to reduce the incidence of the condition, but suggested that screening before conception might offer a more acceptable means of prevention.

The implications of screening for an inherited genetic condition are more complex than for conditions such as spina bifida and Down's syndrome. Where carrier couples can be identified, the birth of an affected child can be prevented by other means than abortion, generally by avoiding conception. There are implications for reproductive decision-making for relatives, who might be at increased risk. Professionals in the field explained that, in Scotland, carrier screening and counselling is available for partners and relatives of people with cystic fibrosis, although one noted that not all relatives choose to be screened, and that some conceive children without being screened, only testing the children when they are born.

At the time of the study, a pilot programme of population carrier screening was being routinely offered at antenatal clinics in Edinburgh as part of trials to evaluate the feasibility and acceptability of offering CF carrier screening, either in pregnancy or on an opt-in basis to individuals and couples before conception. There is debate about whether carrier screening for cystic fibrosis, or other less familiar genetic conditions, should be introduced on a routine basis to the public, rather than only to those with a known family history. Particular concerns are that those with less knowledge might be more anxious (Richards, 1989) or more likely to terminate a pregnancy than families at risk (Marteau, 1996).

As a way of introducing the issue of screening, and because it was felt insensitive to ask women directly how they would feel about having a child with cystic fibrosis, women were asked what advice they would give to relatives who were expecting a child with this condition. Rather than being a sensitive issue, many respondents said that they had considered the question of whether they would see it as right to have an affected child, and several had strong views on the topic. The majority knew about the possibility of population carrier screening. Most also knew of, and several discussed, the availability of genetic counselling and carrier screening for partners and relatives.

Although the question did not suggest abortion, almost all mentioned that this was a possibility, and they suggested that they would see abortion for cystic fibrosis as inappropriate. This response was typical of many:

34,462: I don't think I'd counsel them to have it ... well, say I was an independent person, or a doctor, I wouldn't counsel them to abort it or anything like that.

In discussing 'advice', almost all respondents noted similar positive and negative factors. Several mentioned the potential of new treatments. All also referred to their own quality of life, which was often perceived as reasonably good, but they also pointed out that they were less seriously affected than many with the condition. However, while some emphasised the positive, others spoke in a more negative or pessimistic way. There were widely different views within the group about the 'severity' of cystic fibrosis and the appropriateness of preventing its occurrence, and about the provision of carrier screening.

The severity of cystic fibrosis

Women were asked whether they would see cystic fibrosis as a serious disorder and whether they thought that it was seen as serious by others. Almost all felt that they would see cystic fibrosis as a serious condition, and they explained this view in terms of the shortened life-expectancy, using similar factual or objective language to that used by consultants:

39,505: (seriously) Mm. Yes. ... I mean, there isn't any doubt that it is ... definitely life-threatening.

Several women mentioned that their opinion was based on their knowledge of other people with the disorder, saying, for example, "*when you've buried nineteen people, that is serious - in the space of three years*", or "*Yes I do. Having been involved with other people with cystic fibrosis*", or "*well, I didnae until a few years back. Then I saw quite a few CFs, and they were dying*".

Most women did not refer to their own condition in stating that they would see the condition as serious. The responses of several suggested an alternative viewpoint focusing on other aspects of life or the present, or avoiding a consideration of severity; one respondent expressed this overtly:

29,955: Yes, I would. Because it is. I certainly would. At the end of the day. I would say it's a serious disorder, but I would say I try to let it affect my life ... as minimum as possible.

A number of women expressed considerable ambivalence about the relative severity of CF. Their responses suggested balancing both subjective perceptions and objective assessments, and a knowledge of the very variable symptoms:

JG: Would you see it yourself as a serious disorder?
27,851: No, not at all. Not ... I do, no, I do see it as a serious disorder because I think ... I think there's different, you know, there's like different severities of it or whatever. I think I'm quite a mild case. I really do.

While this response relates to the relative severity of the condition for the affected person in medical terms, a few women suggested different ways of defining severity. One felt that she would not see the condition as serious at all, since people with CF were very similar to others both in what they were able to do and in appearance.

In discussing the severity of the condition, several women pointed out that the implications for others, such as family members or close friends, were different from those for themselves. They also noted that others' perceptions might differ from their own:

38,358: R seems to see it as serious. One day I might not be there for him and things like that, which ... but I don't tend to see things like that. I mean, he could go away from me, or something stupid. He sees things differently from me.

While some suggested that close friends and family members might see the condition as more serious than they might themselves, others felt that partners or close friends *"don't see it as a problem"*. The responses of some indicated that this was a shared view, but one woman expressed an opposite concern that family and close friends underestimated the severity of CF:

24,585: They act as if, "Don't mention it," you know. "There you go again, feeling sorry for yourself". Sometimes I get a wee bit annoyed with them. Because they don't ... you know ... they don't see it as being anything. I think if anything was to go wrong it would be a rude awakening for them, because I don't think they really ... Obviously, they do know all about it and they know about the future, and its causes, but I don't think they really think about it.

The relative severity of cystic fibrosis

In talking about cystic fibrosis, all respondents suggested, not just that those not affected themselves would have a different perception of the condition from their own, but also that others might have more generally wrong views about the effects of the disorder. Many suggested that most people had little knowledge of cystic fibrosis. Almost all contrasted their own opinion of the 'severity' of CF with their understanding of the assumptions of others. Often depending on their own views, some suggested that the public perception was of too serious a condition, others that the condition was not seen as serious enough.

Although the majority of women with CF felt that they themselves would see the condition as serious, several respondents said that they thought others would have an even more negative view. They felt that this perception stemmed from a lack of knowledge of the variations in symptoms or of the recent improvements in life-expectancy:

27,646: A lot of people are ignorant of a lot of things. Like, I mean, that girl, you know, who said, 'Oh, you must be really unhealthy'. And I says, 'No, I'm not', you know. Because she knew a guy, who was really thin and really ill and ... I don't know if he's dead, now, or whatever.

In general, those women who saw public perceptions of cystic fibrosis as too negative did not suggest that these reflected a medical view. Several suggested, like some professionals consulted, that these perceptions might derive from the representation of the condition in the media where the disorder was portrayed as affecting children, rather than adults, and there was a focus on the short life-expectancy.

Other women in the group, in contrast, felt that many people did not appreciate the serious nature of the condition. Some made the point that, since they did not look ill, friends or colleagues would underestimate the severity of the condition, and also did not understand the variation in prognosis:

32,149: When I explain to them what it is, they don't know what it is at all. I explain to them what it is and they are quite amazed at how serious it is. Unfortunately some people do have something wrong with them.

From this perspective, one woman expressed concern that television programmes focused on medical advances, and minimised - or could not convey - the effect on a family:

39,447: It's totally different seeing something on the telly, like, for five minutes, and saying, "Oh this is what it's like". But it's a different reality living with it. Like in your house. It really is. It just dominates everything. It really does. You know. (...) Somebody's living with that every day, as a family, like. And it's totally different. I mean, I don't think people even realise how much input there is into it. Like, seeing it from videos and all the rest of it. I don't think they realise. It's a full-time job. You know, it really is.
JG: Uhuh. Is that from the family?
39: Uhuh. Everybody. It affects everybody. It does. It really does.

While a number of women expressed ambivalence themselves about the 'severity' of the condition, and indicated the difficulty of conveying the implications of the condition to others, the majority also suggested that the public perception was muddled or wrong. Women did not suggest that there was a stereotypical or 'world' view of the condition, as has been suggested in some studies of other conditions (Kelly, 1996), but that there was a range of views in the population:

26,1018: Well, some people exaggerate it ... to an extent just ... As my mum says, "You're going to die when you're twelve", and ... But some people don't see it serious enough.

Women in this group expressed divergent views about the relative severity of cystic fibrosis. A vertical analysis of the interviews showed that these opinions were

consistent, and were coherent with respondents' positive or negative approaches to 'advice', and their views about whether they saw it as generally right or important for the condition to be prevented.

Cystic fibrosis is a serious condition which should be prevented

Some women suggested, without qualification, that they would see cystic fibrosis as a serious disorder, stressing the short life-expectancy; as one explained, *"it's a very very serious illness, there's so many of them out there waiting for transplants"*. Asked for their 'advice' to relatives expecting a child with CF, the responses of women with this view were more negative, although they pointed out the variation in prognosis, and they did not advocate abortion. One replied, *"If they knew? (Pause) I don't know what I'd say. What could you say?"* Another was pessimistic about the potential for treatment, saying, *"I think people tend to think now that with medical advances, etcetera etcetera, that things are getting better. But I don't know that it always works that way, you know."*

Women who stressed the severity of the condition were often those who, in talking about living with the condition, had spoken of their own or others' psychological difficulties in living with the condition, or or who reported being distressed by the deaths of others with CF that they were close to. A few of these women also spoke in strong terms about the consequences, whether practical, economic, or emotional, for other family members, especially parents. Two women said that caring for a child with CF was a 'full-time job', and one referred to the grief, fear and worry of families:

26,1114: Many a time I've seen my mum and dad come up to the hospital when I've been ill and you can see the pain in their eyes, you know, just through seeing me the way I am. (...) Because, I think, from my point of view, to watch a child in hospital suffering and being really ill must be terrible.

These women indicated that they would see it as right to avoid having a child with CF. Although not all gave reasons for this view, one respondent stressed that she would see it as wrong knowingly to conceive a child with cystic fibrosis, because of the consequences for the affected person, explaining, *"I wouldn't put another person through what I've been through, and people have been through a lot worse than me, and it's not so much the physical side, it's the mental side"*.

From this perspective, some suggested that they would see it as important, and a responsible course of action for those at increased risk, to find out their carrier status. One woman said that her parents would talk to relatives who were thinking of having children about cystic fibrosis. Another, asked about advice to relatives, suggested that, with carrier screening before conception, relatives need not be in the situation of finding

out in pregnancy that they were expecting a child affected with CF, commenting, *"Do they want to take that chance? Because I'm one of the healthier ones."*

Women were asked if they knew whether family members, such as brothers or sisters, knew of their carrier status. A number of women who spoke about the importance of preventing the disorder, or who saw screening as sensible, indicated that family views about the severity of the condition were similar to their own. Several volunteered the information that relatives, and sometimes the partners of these relatives, had been screened. One discussed the decision of close family members to have no more children:

39,489: They've decided that they're not going to have any more. Because they feel that they were lucky enough to get away with that. Without sort of risking it again, if you know what I mean.

Women with this view about the severity of cystic fibrosis spoke positively of population carrier screening, and were in favour of this being offered to everyone; one woman felt that it was relatively common to be a carrier for CF. While noting that a screening programme was and should be voluntary, they emphasised that they would see screening as important and beneficial:

33,259 (notes): I think it should be entirely up to yourself. If people want the tests - it's up to them. They've checked you over ... it's safer in the long run. (...) If you're thinking of having bairns, I would say "Get the screening done". So you ken your chances then.

There is debate about whether population carrier screening for CF should more appropriately be offered during pregnancy or before conception. While screening before conception is felt to maximise the principle of autonomy (Raeburn, 1994; Clarke, 1995), it has been argued that screening in pregnancy is both more relevant for individuals and more effective in reducing the incidence of the condition (Wald, 1991; Brock, 1995). Although some women who saw prevention as important felt that they might not consider an abortion themselves, they did not question the availability of screening programmes offered in pregnancy. However, one suggested that those with a positive result might face a dilemma.

It was generally felt that screening before conception would be preferable, although some questioned the usefulness of this approach in terms of prevention. One woman pointed out that individuals would be unlikely to choose a partner on the basis of carrier status:

39,745: I don't really see how it would really work. Because the only way it would be any use to you was if two carriers were going to have children. And, you cannae very well ask somebody, "Are you a CF carrier? Can I see your identity card?" kind of thing! (Laughs)

A number of possible negative implications for carriers, such as stigma, more negative perceptions of health, anxiety or an unwelcome sense of responsibility about conceiving an affected child, have been suggested in the literature (Richards, 1989; Marteau, 1992). Some of those who saw it as important to avoid the birth of an affected child pointed out that, with voluntary screening before conception, not everyone in the population might choose to be screened for CF carrier status; one respondent considered whether screening and information should perhaps be compulsory. They also made the point that not all couples screened before conception might avoid pregnancy, especially if they did not appreciate the severity of the condition:

39,427: I think at the end of the day they're still going to have children anyway. I mean, I don't really see it as ... like putting people off, if you know what I mean.

Cystic fibrosis is less serious than many conditions

Although some women with cystic fibrosis felt strongly that the disorder was a serious condition which should be 'prevented', others took an opposite view. These were women who saw the condition as less serious than others in its effects, or those who suggested - often by reference to their own lives - that the quality of life of those affected could be good, whether because of the relative lack of severity or whether because of an approach to life emphasising happiness and relationships rather than health. These women did not talk about difficulties for other family members.

The views of these respondents towards the severity of the condition were similar to those of the majority of women with spina bifida. Asked about their 'advice' to relatives, women with this positive view, often using similar terms, did not suggest a responsibility to the unborn child. Rather, they referred to their own experiences, perceptions and philosophies for living with the condition. For example, *"I'd tell them not to dwell on it, I mean, it's only part of them"*, *"I'd just tell them about, you know, doing the treatment right, and the diet"*, or, *"I certainly wouldn't make it into a big drama for them, because I don't feel that it is"*.

Unlike spina bifida, there are new possibilities for treatment for cystic fibrosis, both current medication and the potential for treatment by gene therapy in the future, and some women, in addition to other arguments, were optimistic about progress:

15,286: I mean, I wouldn't put them off having it! I ... definitely ... I would never put anyone off having it. I mean, I'd tell them that, in the early days, you know, in my own life you know, from having so much wrong with me - and there's that many new treatments that are starting for it.

In discussing the provision of a screening programme, women who saw the condition as less serious spoke against prenatal diagnosis in particularly strong terms. One described

the termination of a wanted pregnancy on the grounds of cystic fibrosis as *"ridiculous"*, and others saw this as inappropriate or unjustifiable, especially when the prognosis was felt to be improving:

28,694: I don't think it's fair to have an abortion if it's CF. I really don't. Because it's something that ... it can be treated. Do you know what I mean? It's not something that ... they're just going to be really ill, really poorly, and then they're just going to pass away.

Women with this view stressed that those having a positive diagnostic test in pregnancy should be offered adequate information about the implications, saying, for example, *"Well, I don't think you should just abort it because it's CF, you know. Find out about it first"*.

Screening before conception was spoken of as generally preferable to antenatal screening programmes; in the words of one respondent, *"that seems to be the best way forward"*. However, some women who saw the condition as less serious expressed ambivalence about whether they would see 'prevention' by any means as important or right; pre-conception carrier screening was suggested as interfering with nature or fate to an extent:

27,993: Well, I think it's good but again, I mean, if it puts people off having a kid, you know, I mean, I don't think it's right. You know?

Most women who had reservations about the importance or necessity of 'prevention' did not spontaneously describe the views or actions of members of their families. Asked their views about carrier screening for relatives, the majority described this as sensible, although without suggesting what actions relatives might take; as one woman put it, *"at least then they would know what to expect"*. Several women thought that their siblings would not want to be screened until a pregnancy was planned.

Asking whether relatives knew of their carrier status often felt awkward to the researcher. There might have been a number of reasons for this. Child-bearing and reproductive decision-making might be seen as a private matter, concerned with emotions, rather than being a public matter of rational or scrutinised decisions. Pregnancy is not always a matter of decision-making, it might be accidental, and there could be doubts about paternity (Macintyre and Sooman, 1991). Not only might one's carrier status be felt to be a personal matter, but there might be a number of reasons not to want to know. Also, there might be different views within the family about the severity of the condition or the importance of prevention.

While these might be issues for relatives, there might be additional reasons why respondents might not want to discuss or consider the views or decisions of family

members. This might be to cast doubt on their perception of the condition as not serious or to call into question their perceived value within the family or their self-concept. Some women said that they had no idea whether their brothers or sisters, or their respective partners, had been screened, or whether they would want to be.

However, a few women who suggested that they did not see the disorder as serious indicated that this was also a family view. Some referred to younger siblings conceived after their own condition had been diagnosed. One woman emphasised that her siblings thought they would wait until a pregnancy was confirmed before finding out their carrier status. Some suggested that their brothers or sisters might not see it as a problem to have a child with the same condition as themselves, because of their familiarity with the condition:

28,674: I know my sister .... She would have ... if she had ... if my sister X had a child and it had CF, she would half have a clue what it's about. Because of me having it. And so it would be easier. It would be easier than it has been with my mum and dad, because they didn't know anything about it, whereas X has grown up with it. ... But I think it would be a better idea beforehand.

Antenatal screening programmes can be seen as offering information and choice, and the majority of women with this view generally supported the provision of screening. Asked about the possibility of population carrier screening, a few suggested generally that decisions should be a matter for the individual, using similar non-committal terms to respondents in other groups who supported the principle of choice:

JG: How do you feel about that sort of thing?
35,387: Um. I can understand that, yes. I realise that it's a big responsibility to bring up somebody with CF. So - you would have to think about it seriously.

However, most women with cystic fibrosis offered their opinion about whether or not they would see 'prevention' as justified. Where women saw it as less important to prevent the births of affected children or were ambivalent about the 'severity' of the condition, their responses indicated a tension between this position and a view that it was generally right for screening programmes offering information and choice to be offered:

27,1057: It depends if it was going to be of any benefit to ... I mean, everything's got to be so ... You know, it's all individual. You know, I mean, that's why I can never make decisions about anything, and I can never judge anyone else because I always think, "Well, that's making them happy", or, "That's OK for them, but I wouldn't do it", or "God, I'd love to do that", you know. I mean, you can't say. I think it ... it can be a good thing if, you know, you're not sure or, you know, just to give you a bit more insight into things.

Several women who saw the condition as less serious than others expressed some reservations about whether they would see a screening programme for cystic fibrosis as necessary:

34,541: Well, if people are so ... so worried about it then they should have access to it I suppose. I suppose ... you know ... well, I feel that a reduced incidence of CF in the population would be good. Maybe. But then there wouldn't be so much research into it I suppose.

A small number of women took a stronger view, questioning whether it was right or beneficial for CF population carrier screening to be offered. Some raised concerns about the benefits of screening for individuals at low risk; one woman stressed the rarity of the disorder, feeling that many people would be put through the worry of screening for nothing. Several felt that carrier screening should be restricted to family members, making the point that others might have little knowledge or understanding of the condition or have an over-negative view. Routine screening has been seen as implying that prevention is both right and beneficial (Farrant, 1985; Macintyre, 1997). One respondent argued that offering information would create a pressure to act on it; another noted that prospective parents increasingly wanted information about an unborn child, but questioned whether it was necessarily right to offer this:

15,532: (firmly) See that's ... like carrier screening, and that. It could put a lot of people off, I think, having children. Because some people, I think, they just don't understand. They hear the words and that's that. So I think, for the whole population, I don't think it's a good idea, it would send people into shock, and that would be the end of it, do you know what I mean. I think that ... for people that have got CF or anything like that, I think it's all right, not for the nation. Because there's some ignorant folk, do you know what I mean. Some ignorant, ignorant people.

As illustrated here, some women used stronger language in suggesting that they would see 'prevention' as unjustified. One reported her anger at learning of someone who wished she had terminated her pregnancy when she found out that her child had CF:

27,1002: ... And that really, that infuriates me, you know, because ... I mean, if she was to meet any of us, I mean, we're all, I mean, every one I know is over twenty, you know, and we're all ... Well, OK, people have died, it's a fact of life. People die from other things at a young age, and it really ... it really annoys me that people can be so narrow-minded.

This respondent suggested disapproval of the values of individuals who might terminate a pregnancy on the grounds of cystic fibrosis, a condition for which she felt 'prevention' was unjustified. She also questioned the provision of the means to 'prevent' cystic fibrosis, asking whose definition of 'serious' should prevail, and raising concerns about a drive to perfection.

Several of those who raised concerns about offering CF population carrier screening or about preventing the births of those affected with this condition questioned the provision of information about physical characteristics. Some also suggested possible discrimination in employment, or described concealing the fact that they had cystic fibrosis.

Their concern was with appropriate limits of conditions for which a screening programme should be offered. Although one woman said that she could see why people might want to ban screening programmes, since her own parents had not been offered such a choice, in general, there was little view that concerns about the provision of screening programmes extended to other conditions such as Down's syndrome or spina bifida.

Women who saw cystic fibrosis as a less serious condition suggested that they would not themselves prevent the birth of child affected with this disorder, for different reasons, although screening of a partner was seen as useful in offering information. One felt that it was "not right" to avoid conceiving a child on the "slim chance" of a child being affected. Another felt that a child could have a *"good enough life"*, as she had herself. A third, more personally, suggested that a concern for the well-being of an affected child might be outweighed by a desire for the child:

22,338: I was saying to my mum, "Well if I was pregnant, and I knew it was going to be CF for definite", I says, "I would still have it, why not?". But she says, "But, you know, you've got to think about it seriously", sort of thing. But, I mean, I know ... what to expect, and things. ... I know it's unfair, bringing up someone with CF, because I know what I go through. I would still like to do it.

Like women with spina bifida, several women suggested their familiarity with the condition as a factor in feeling more able to care for an affected child. Some also noted the support of family members or a partner, and their responsibility in bringing up a child. One woman reported her partner's view that, *"it didn't matter whether a child had CF, because he'd read up as much information as he could about it"*. The findings of this group tend to confirm Richards' (1989) suggestion that, where people suspect a high risk, the birth of an affected child might not only be less of a surprise, but the greater knowledge of the condition might be reflected in family support.

Within the group as a whole, however, there were divergent views about the appropriate use of screening programmes. Women's accounts of the reported views and actions of relatives indicated a similar wide range of views, The findings of this study suggest that decisions about whether to prevent the birth of an affected child might be a potentially difficult issue for relatives.

Although most respondents discussed CF carrier screening in hypothetical terms, a small group of women talked about planning a pregnancy. They reported attending counselling clinics with their partners, who had been screened for carrier status. Although all partners had tested negative, most said that they would not want to have a child with CF or an abortion, and one woman spoke of carrier screening as giving *"peace of mind"*. Another said that she and her partner had discussed what they might have done if the test had been positive, and she thought that they might have decided against having children.

There are some similarities between the findings from this group and a study examining the reproductive careers of a small cohort of HIV positive men and women. Green (1994a) has concluded that there was a gulf between respondents' hypothetical and actual decisions about pregnancy, with younger respondents (those under 25) expressing the strongest and most hypothetical views about what they would do. She has suggested that both public and medical opinion is hostile towards pregnancy for those with this condition, both because of the risk of passing on the condition and because of the short life-expectancy of the affected parent, and has noted that some who are HIV positive become pregnant 'accidentally', i.e. without asking medical advice.

Discussion

Women with cystic fibrosis suggested generally that they would see the provision of screening programmes as beneficial. A number suggested reasons for this view in terms of allowing individuals to take their own decisions about giving birth to a disabled child. They also suggested that they might see it as right to try to avoid the birth of an affected child in certain circumstances, whether in the interests of the parents or the individual affected. There was little suggestion that such 'prevention' or abortion on the grounds of any foetal abnormality was wrong, or that those affected with a disorder had equal rights to life. However, while the provision of screening programmes for Down's syndrome and spina bifida was suggested as unproblematic, women expressed stronger views about the morality or importance of offering a screening programme for cystic fibrosis.

Within the group there were divergent views about whether cystic fibrosis was a condition serious enough to be prevented, although almost all saw abortion as inappropriate. Those who saw it as important to prevent the birth of an affected child emphasised the importance of - and individuals' responsibility for - avoiding suffering. They saw population carrier screening as beneficial, although expressing concern about others' lack of knowledge about the severity of the condition. In contrast, those who saw the condition as less serious, or who were optimistic about progress, questioned

both the justification for and the morality of 'preventing' cystic fibrosis. They expressed particular reservations about the value of offering routine screening outside families, because of concerns that people might not question 'prevention' as the logical outcome. Some of these women suggested that the existence of a screening programme in reproduction for a condition felt not to be serious - such as cystic fibrosis - could be seen as selection, or as a drive within society towards perfection. They raised questions about the limits to conditions for which it might be right for a screening programme to be offered, and - unlike other women in the study - expressed concerns about the provision of genetic information about physical characteristics.

At the time of carrying out this study, there was debate about whether or not population carrier screening for cystic fibrosis should be routinely offered; currently no decision has been taken. Widely different views have been expressed in the literature, and screening policies vary between Glasgow and Edinburgh. The findings of this study confirm the views of commentators that the question of the relative severity of cystic fibrosis is a difficult issue (Green, 1995). Women with cystic fibrosis might have perceived the study as an opportunity to offer their perceptions of the condition; almost all offered the view that not enough was known about cystic fibrosis, or that others' understanding of the disorder might differ from their own.

The results of this study confirm those of another small qualitative study, which also found a range of views among young adults affected with cystic fibrosis about the relative severity of the disorder and whether or not they might consider preventing the birth of an affected child themselves, and caution about the appropriateness of offering prenatal population screening (Alderson, 1998). As in this study, respondents respected others' right to make reproductive decisions but argued that choices should be informed ones.

Women's views about screening programmes generally or for cystic fibrosis in particular might be influenced by their own perceptions of living with the disorder. Like women with spina bifida, women with cystic fibrosis described their approach to living with or conceptualising the condition, and 'activist' and 'accepting' strategies could be distinguished. To an extent these general philosophical approaches to life could be seen as linked with views about screening programmes; an 'activist' approach with support for individual choice and valuing independence, and an 'accepting' approach with a view of any child as equally valued. However, strategies were suggested as at least partly pragmatic, as appropriate at certain stages of the disorder.

Women described very varied experiences of living with cystic fibrosis, and gave a range of assessments of their quality of life. Although they spoke of a personal

approach to life as important, most saw the quality of life of people with cystic fibrosis as related to the progress of the disorder. Similarly, in assessing the 'severity' of the condition, while women referred to their subjective experience, the majority also referred to their more objective knowledge of the condition itself and future progress in treatment, or to consequences for others affected. Many also mentioned their perceptions of the effects on others. The very varied views about 'severity' expressed might be seen as reflecting a tension between these different perspectives.

In discussing living with cystic fibrosis, although a few women mentioned a lack of opportunities or some concerns about the way that they might be perceived, they did not suggest that the main problems they experienced were socially caused, deriving from the way that they were treated, or common to those living with cystic fibrosis or a chronic illness. These women did not describe themselves as disabled, and some refuted this definition of themselves. The social model of disability may be inadequate for understanding the experience of chronic illness; women with cystic fibrosis did not suggest that physical consequences of the disorder were unimportant or of low priority. In a similar way, in talking about screening programmes, most women focused on the consequences of disorders for those affected or for others; their concerns were mostly about welfare.

In considering their own anticipated use of screening programmes, most of these women suggested a pragmatic approach; they indicated that their reproductive decisions might depend on a number of factors. Few expressed a view of abortion on the grounds of foetal abnormality as always unacceptable. While women gave their opinions about whether or not they saw it as right to try to avoid having a child affected with cystic fibrosis, several mentioned more personal factors. Familiarity with the disorder was sometimes suggested as a reason for seeing it as unnecessary to avoid having a child with CF; some also spoke about their desire for a child or about their own ability, or that of others, to cope with children. Several anticipated difficulties in caring for a child affected with certain disorders.

Again, the views towards screening programmes of women with cystic fibrosis can be seen in the context of their lives. A number of these women spoke about considering having children, and their focus on the consequences of conditions for others might be seen as reflecting the perspective of a potential mother. They also described discussing decisions about motherhood with other family members and with medical professionals. The accounts of these women and those of medical staff - in discussing carrier screening of a partner for example - indicated that pregnancy for women with CF was seen as scrutinised by medical professionals to an extent, often in connection with implications for women's own health. The outlook of these women more generally, and

their perceptions of cystic fibrosis, can also be seen as informed by medical knowledge and as reflecting medical rhetoric; women described frequent and regular contact with hospitals, and their reliance on and respect for medical professionals.

The less strong views against abortion expressed by these women, compared with women with spina bifida, might relate to the fact that cystic fibrosis might be avoided by other means, although the fact that few women in this group reported having strong religious views might also be relevant. At the same time, some women did express concerns about eugenics, and about whether it was right to offer a screening programme for conditions felt not to be serious. The educational level of this group was higher than that of other women in the study; they might have had greater access than other respondents to theoretical debates in the media. In order to further examine similarities and differences in views, the next chapter outlines the ways that reproductive screening programmes were discussed by young women in the general population.

CHAPTER 10

WOMEN IN THE GENERAL POPULATION

One aim of the research was to examine similarities and differences between the views of women affected with 'preventable' conditions and those of young women of a similar age in the general population. The study was outlined to women in the first year of a degree nursing course, and in the first (NC) year of a pre-nursing course and a child-care course at a FE college. It was felt that these women would form a comparable sample to the disabled sample in having similar levels of education, and in having some familiarity with antenatal screening programmes and with the conditions being screened for, if only from their studies.

Twenty-nine students, who, like other women in the study, were volunteers, took part in interviews. Eight of these women were students on the degree course and twenty-one on an FE course; of this latter group, seven had passed some SCE 'Highers' and the others had passed 'Standard' grades. Asked about their fathers' (or mothers') job, three said their fathers were unemployed, twelve described manual jobs and thirteen non-manual, and the father of one woman was retired. In response to questions about religion, eleven women spoke of having a religious belief. Three of these attended a Catholic church, and four other women said that they had been brought up as a Catholic. The other fourteen women described themselves as having no religion.

This chapter looks at the way that these young women, as representative of women in the general population, discussed issues of motherhood, reproductive technology and screening programmes, especially in relation to Down's syndrome, spina bifida and cystic fibrosis.

Quality of life

Respondents were asked similar questions to other women in the study about their activities since leaving school and about their quality of life. In discussing what they felt was needed for a good quality of life, most - like other women - referred to happiness and good relationships, and many suggested the importance of having interests in life or a job. Very few mentioned health, but some spoke about money, and 'getting by' financially.

However, unlike the majority of women with spina bifida or cystic fibrosis, who readily evaluated their quality of life, often with reference to explanations or philosophies, a number of respondents in this group were initially uncertain about how to describe their

own quality of life, suggesting perhaps that this was a question that they did not often consider. As one put it, *"Don't know, I'm quite happy with it."* On consideration, most concluded that they would see their quality of life as 'all right' or 'good', or said that they felt reasonably happy. Several qualified this by some reference to quality of life as relative by, for example, comparing their lives with those of others, saying that they felt they were *"quite lucky compared to some"* or *'"there's a lot of people better off, but there's a lot of people worse off"*, or by explaining that good and bad days, or aspects of life, were in balance.

Motherhood

Women were asked whether they thought about having children. Five women had a child, and almost all said that they hoped to have children in the future. The majority also discussed their desire to have a career, or a job that they enjoyed. When asked about combining work and motherhood, most thought that they would manage to do both, perhaps by relying on relatives, taking career-breaks or working part-time. However, those women who had children described some difficulties with finding appropriate child-care, and several noted that looking after children could be tiring and time-consuming. One woman felt that it was impossible to combine having a career with motherhood, because of the commitment needed to fulfil one's responsibilities:

1,182: You know, when you have children, they are like your responsibility and they're important. And I don't think you should take it lightly, I think you should really want them and have all the time in the world to look after them. And I wouldn't have that if I want to have my career.

She reported that her partner held a similar high standard of parenting, saying, *"He's the very same, he thinks you've got to be a really good parent, you've got to be a certain type of person"*.

Although other women did not take the view that having a job would preclude having children, several spoke about the responsibility of parenting or the qualities or resources felt to be required. Many felt they were too young at the present time to either want children or feel able to care for them, saying that they valued their freedom or independence, or that they were not 'settled' with a partner or an income.

Abortion

Like other women in the study, women spoke of abortion as an *"option that should be available"* in certain circumstances. Like others, many made a distinction between their general view that abortion should be legally permitted, and their personal belief that this was something that they *"just don't agree with"*, or would not consider themselves.

Several contrasted their own values or anticipated behaviour with that of others. As one woman put it, *"I wouldn't have an abortion, no way. I think, though, that there's some situations where I can see folk, why they do. But not me."*

In this group, theoretical issues were often interpreted in individual, personal terms, respondents often referring to their feelings or to the course of action which they anticipated taking themselves and their reasons. Most felt that their views about whether they would regard abortion as wrong or as acceptable might depend on whether there was felt to be an adequate reason:

JG: How do you feel about abortion on the whole?
18,191: Well, it depends. On the person, on the circumstances. I know ... I couldnae. Well, there's ... there's extremes ... you know, like, you think, "I couldnae kill anything, good grief, I couldnae do anything like that", but it depends ... on the circumstances.

A number of women gave their opinion of what they would see as a sufficiently good reason for abortion, whether for themselves or others. There was a range of views. While some felt generally that abortion on social grounds should be restricted (some noting the possibility of adoption), the majority stressed that others might have their own perceptions of benefit. As one argued, *"It is their decision. If they think it's better off in the long run for that baby, not to have it ... I don't really have anything against abortion"*. This response may be seen as in line with a recent general emphasis on individual rights, freedom and difference, personal values or moralities, but also on a 'right to choose' abortion. Several noted peoples' different circumstances, and they indicated a non-judgmental approach towards others' choices, focusing rather on their right to decide:

12,263: I mean, everybody's an individual, so it depends on how ... what's happened in their situation. Whether they want to get rid of the child or not.

Several women spontaneously suggested that, while they would see abortion as generally unacceptable, they would regard certain foetal abnormalities as offering an ethical reason to have an abortion. As one argued, " ... *but I think there are certain exceptions, where there's something wrong with the baby, or whether the chances are it could damage the mother's life*". Another expressed a similar view, saying, "... *if there's a ... a good reason for it, fair enough, but anything else I don't like ... If there's a good reason, like you're too young or you've been raped or the baby's just going to die, fair enough*". The views of these women confirm findings from other studies which indicate that foetal abnormality is felt to be an acceptable reason for abortion (Faden *et al.*, 1987), as noted by disability activists (Finger, 1984).

The responses of these women suggested certain societal or cultural limits on the acceptability of abortion, however. All saw abortion on the basis of foetal sex as

wrong, although a small number referred to the fact that there were different views in other cultures. However, in discussing this issue, a number of women noted that they saw nothing wrong with finding out the sex of a baby, and several suggested that they might like to have this information themselves.

Asked their views about the possible provision of genetic information about the physical characteristics of an unborn baby, a number of women (nine of twenty-nine), like some women with spina bifida, also felt that this was acceptable and unproblematic. However, others saw this as wrong, and all suggested that such tests were unnecessary, questioning, *"why do they want to know?"*. Some felt that a desire on the part of parents to have a child with certain wanted characteristics or of a particular sex was the wrong reason for having children, and several suggested that they would not want such information themselves. There was little discussion of this issue in more general, conceptual terms, however, such as that certain characteristics might be constructed either as preferable or as problems, or that there might be wider social consequences of offering such tests or having such information, or of taking decisions on these grounds.

Several respondents made the point that they would see testing for physical characteristics as unnecessary or wrong in the sense of interfering with nature. They felt that there were reasons for not having tests, such as the surprise of a new baby, and a few spoke about the risk of causing harm to the child by unnecessary invasive testing or *"interference"*. Many women spontaneously contrasted information about the sex of a baby or about physical characteristics, which was spoken of as not needed, with information about 'health', which was discussed in a different way, as something which one might *"need to know"*, or as a reason for having tests:

14,209: It sounds a bit kind of unnecessary, that kind of thing. I don't think you really need to know, do you? (Laughs) You know, it would be different if it concerned the baby's health or something. You know, curly hair and blue eyes! It doesnae really matter!
JG: What kinds of things do you feel as though you would like to find out about, about a baby before it was born?
14: Um. More or less what you do find out. Just if it was healthy, weight, things like that.

Personal predicted use of antenatal screening programmes

This response was typical of many. As an introduction to discussing prenatal diagnosis, women were asked what they might want to find out about an unborn child. The majority of respondents used similar language, and often a similar contrast, to suggest the benefits and importance of knowing whether a baby was 'healthy'. However, there seemed to be various meanings of the term. Two women said that this meant *'heart*

beating', indicating a lack of interest in abnormality. A number of respondents suggested that they might want to know about possible 'problems', 'damage' or 'disability'. Many kept the meaning of 'healthy' quite vague:

JG: What sort of health things do you think you might want to find out about?
16,221: Well just how healthy the baby was, if there was going to be anything wrong. So you can prepare yourself ... for after the birth.

The majority of women had a good knowledge of antenatal screening and diagnostic tests, and a number spontaneously mentioned the fact that screening tests and scans were routinely offered in pregnancy. Across a wide educational and class range, many women said that the information offered by *"the routine tests"*, or *"just the screening programmes"* would be what they would want to know about in an unborn child.

Those women who had been pregnant said that they had undergone the screening tests and scans which were available, and the majority felt that they would take the tests which were offered. Women were not asked why they had or would want the routine tests, although several offered reasons in general terms such as, *"so you know ... what's ahead of you. ... Because it is a big thing"*. Although it has been suggested that women might find antenatal screening tests useful for the reassurance of a negative result, and that their goals might be different from those of medical professionals of finding out about abnormality (Farrant, 1985), in this study, women spoke of learning about 'health' together with 'problems'. Diagnostic tests were spoken of as something that women would only consider *"if the baby was at any risk"* or *"if there was a high risk in my family"*, or to get a more accurate result after a positive screening test:

10,158: I don't know whether I would have had the amniocentesis. Just ... for them to be certain, for them to tell me for sure ... what the risk was instead of having maybe a kind of ... just a ... an inaccurate test, although it is a small chance. I'd rather know for certain.

When asked if they would thought they would take part in screening programmes, a number of women spontaneously said that they thought they would follow medical advice. Their responses indicated that tests which were recommended might be assumed to be beneficial:

15,186: Right. I think if they advised me to go for it, I would go. But I think if I wasn't at any risk, or any way of losing the baby, I wouldn't go. But if they were to say, "I think you should go for the test", I would go for it. I think I would anyway! (Laughs)

There is some evidence that women are more inclined to have tests that are seen as being supported by the institution where they are offered (Press and Browner, 1997). Although the psychology literature suggests that people differ in their approaches to information, and that deferring to 'experts' can be seen as self-determination (see

Shiloh, 1996), some social scientists have argued that it might be difficult for pregnant women to resist the advice of medical 'experts' (Clarke, 1991; Richards, 1993). The views of these women can be contrasted with those of other women in the study, most of whom did not suggest following medical opinion. However, while several women in this group suggested that they might follow medical advice about having tests, a number indicated that they would take their own decisions about how to act on the information, often giving reasons for their views.

Abortion on the grounds of foetal abnormality

In line with their views that foetal abnormality might be an ethical grounds for having an abortion, all saw it as right for screening programmes and the option of abortion on these grounds to be available. They pointed out that there were circumstances in which they might see it as right to prevent the birth of an affected child by this means, because of the perceived consequences, either for the child or for others, especially parents.

Consequences for affected individuals

Many women suggested that abortion on the grounds of foetal abnormality might be seen as right for humanitarian reasons if the affected individual might be expected to suffer. They mentioned a short life-expectancy or a poor quality of life as factors which might outweigh either moral considerations about abortion or a more personal desire for a child:

3,440: ... But for something like severe brain damage I mean ... you're unable to physically do much but you're also unable to do mentally very much, and therefore it is not much of a quality of life really.
JG: Right, I see what you mean. So that's the kind of thing that you might want to find out about.
3: Yeah. Yeah ... not so much for my own benefit but I don't think it would be fair to ... to bring someone in to the world knowing that they're going to be living ... I mean, it could be - you know - twenty years or so just with no quality of life whatsoever.

Several offered criteria for assuming a that it might be in a child's best interests not to be born; they mentioned severe brain damage, severe pain or complete dependence. Particular disorders were rarely mentioned, and not those preventable by screening programmes, such as those being examined in this study. Some were later to make the point that the quality of life of individuals living with certain conditions might depend on the way that they were treated, both in society generally, and by individuals. Asked what they would see as a 'serious' condition, answers were sometimes hesitant, respondents drawing on their knowledge or their experience of working with disabled children and adults to indicate those who *"weren't having a life":*

9,245: Things like ... if you couldn't move. Or couldn't like ... if you were totally paralysed, you couldn't see ... things like that. Just that you couldn't live a ... not a normal life, but ... you know. I worked at this place in R_.. And it was for - you know - children with more severe ... special needs, sort of thing, just ... because some of them were going to die, like, maybe in the next week, and ... and you felt so ... and you had to see like babies that couldn't move, disabled, and I don't know if I could put somebody through all that. I think it just depends how severe it is.

Where women suggested that they might see abortion on the ground of foetal abnormality as appropriate in the interests of the affected person, they did not suggest that there was an individual 'right to health', which has been suggested as a current value (Hubbard and Wald, 1993), but of responsibility for the child's well-being. Many suggested this responsibility in personal terms, using language such as it being 'not fair' or 'cruel' to *"put somebody through that"*. Several spontaneously suggested that they might consider abortion themselves where a child might be expected to suffer; this was suggested as an ethical position:

6,313: Eh ... I mean, I don't really ... agree with (abortion) for just likes of ... sort of ... coming in off the street, saying, "I want an abortion". But, I think that if they have got a disorder, and if the person's not going to have a very good quality of life, or if it's going to have no quality of life at all, then ... I mean, you're going put them in less pain by destroying it before ... it actually grows and gets like ... I know there's a certain stage that I wouldn't want to ... destroy it.

Their responses indicated that they would see it as wrong to give birth to a child who might suffer, when such suffering might be preventable; allowing such a child to be born was suggested almost as causing suffering, rather than being fate. These findings confirm the view of Rhodes (1998) that, in considering genetic testing, people's considerations are in terms of 'responsibilities to one another'. Rothman (1985) has pointed out that women are seen within society as responsible for the well-being of a child.

Consequences for women

A number of respondents suggested that severe problems for a child affected with certain conditions could not be seen separately from the effect on other family members, particularly the mother, a point noted by Wertz (1992b) in discussing the views of affected families. As one woman put it, talking about giving birth to a child who would have a short life-expectancy, *"for a few years of a life it would be hard for the person, but for the mother as well"*.

The majority of women in this group discussed their perceptions of the implications for women of having a disabled child. Although not all suggested negative consequences, many did, drawing on their work experience or on their knowledge of other families to suggest both practical problems and possible strain in caring for a severely disabled or

dependent child. As one noted, *"Eh ... one of my mother's friends, she's got a mentally and physically handicapped daughter. And it takes such a ... it takes such a lot of her time, well, all of her time she takes up"*. Several felt that they would see caring for a child with certain disorders as *"really hard"*.

In discussing consequences for women or for families, many indicated the support which they thought might or might not be available, whether from other individuals - especially a partner - or from services. Several expressed the view that, while there were more resources for children with special needs, there was little support or respite for parents or carers. It has often been noted that screening programmes are offered in a climate of increased individual responsibility (Rothman, 1985; Morris, 1991), and the comments of these women indicated that they saw the responsibility for care as resting with individuals and often with women. Care was spoken of as a *"full-time job"* or *"24 hours a day"*, or as lasting for a life-time; as one woman put it, *"You'd ... kind of give up the rest of your ... your life, you've got to care for it you know"*.

Perceived consequences for other family members and possible difficulties of caring for a child affected with certain conditions or limitations were less discussed by women in other groups. There might have been a number of reasons for this. First, women in the general population discussed the question of 'prevention' from the standpoint or perspective of a mother, and perceived consequences for the woman or the parent were seen as the most relevant or salient issue. Most women in other groups, on the other hand, emphasised the standpoint of the affected person, and the study had been presented to them in this way. Women affected with spina bifida and cystic fibrosis might also have wanted to challenge possible assumptions or public perceptions about consequences for parents, or to suggest that negative consequences should not be seen as inevitable (although these conditions were not suggested as problematic for others by women in this group). On the other hand, the possible negative effect on others might have been a sensitive issue for those affected, and might not have been discussed for this reason.

Many women in this group suggested that they might see 'prevention' or abortion on the grounds of foetal abnormality as right or ethical, where women might feel unable to care for the child. As in other empirical work, the perceived 'burden' of the child was felt to be an important factor in seeing abortion as acceptable (Faden *et al.*, 1987; Evers-Kiebooms *et al.*, 1993). However, a number interpreted perceived problems of 'coping' quite widely, in some cases as meaning wanting the child. One noted, *"some people have difficulty coping with a child who is not perfect"*, and, as another put it, *"some would not be comfortable with a disabled child"*. A few expressed a tolerance of a

wide range of reasons, values or priorities underlying a decision to terminate a pregnancy on these grounds:

24,458: I don't know if I'd get that done. But I can see why other people would. I mean, say you work, and you were a woman and you had a career! You wouldn't want to have this extra burden.

Ekwo and colleagues have made the point that perceptions of burden might depend on a number of factors, including personal susceptibility (Ekwo *et al.*, 1987). Women in this group noted that people's capacity to love and care might depend on the effects of the disorder. Many also made the point that individuals might vary in their personalities, qualities, resources and circumstances, and hence in their perceptions of what they might feel able to cope with or 'handle'. The expression *"some can't cope"* was used by many:

13,267: I mean, some people couldn't cope. I mean, some people just could not cope with an illness like that. And if they couldn't cope, then ... I mean, it wouldn't be fair to the child, letting parents have this child and then denying it - putting it into some kind of institution or not loving it or whatever. I think it should be offered to them, yes.

As illustrated here, several also suggested that the feelings of parents might be reflected in their treatment of the affected child. Although a few noted the possibility of adoption, a number of women argued, sometimes strongly, that if a child was not wanted, and might not be loved and cared for, then it might be in its best interests not to be born. They felt that they would see abortion on the grounds of foetal abnormality as right or beneficial for this reason.

A number of women offered a prescriptive view that, although taking part in screening programmes was voluntary, they would see it as right or responsible to find out about any disorder affecting an unborn child. As one put it, *"it's kind of sensible in that people will know that they're going to have to live with the condition"*. Some went further, suggesting that women ought to consider the implications of having an affected child and whether they might feel able to care for such a child before the child was born.

Personal predicted use

In discussing prenatal diagnosis, the majority of respondents indicated whether they might consider abortion on the grounds of foetal abnormality themselves. A number suggested that, while they might take part in screening programmes, they would not consider abortion. This difference between the predicted uptake of testing and of abortion has been noted in empirical work, both quantitative and qualitative (Evers-Kiebooms *et al.*, 1993; Richards and Green, 1993). A few women indicated that they

would see abortion for any reason as personally unacceptable. Others suggested that they would not terminate a pregnancy on the grounds of any condition, saying that this *"would make no difference"*. Many used similar language, pointing out that the child was a wanted child or *"my baby"*:

23,234: I wouldn't have that (abortion on the grounds of foetal abnormality). I don't agree with that sort of thing. I don't care what the baby's like. I'd be, 'No, no, it doesn't matter! I'll just get it.' It's still my baby.

While the individual desire for a child (as well as the unacceptability of abortion) has been suggested as a strong influence on decisions, which can outweigh perceived risk (Marteau *et al.*, 1989; Shiloh, 1996), the language used by some women suggested that a disregard for the condition might also seen as a moral matter of personal values. A few women, sometimes by means of a contrast with their own anticipated actions, suggested that people who would terminate a wanted pregnancy on the grounds of a disorder had the wrong attitudes towards motherhood or parenting; a lack of commitment rather than an unconditional love and acceptance:

3,514: ... People who feel that they have to know everything about it, and have to have all the details and decide, you know ... if everything is as they want it. But I think if you really ... if you really want children, then you're going to ... you're going to have it no matter what, and really that doesn't matter to you. I mean if you're that picky about small things then you're obviously not a ... a determined parent! You're not ... committed to it. As some people who don't have the tests because they're not bothered because they're going to love it anyway, would be.

Like some women affected with conditions themselves, these women expressed some disapproval of the values of individuals who might prefer some children to others or want a perfect child. They did not suggest that attitudes of prejudice were general values in society, or that screening programmes offering 'prevention' might be seen as implying that those with conditions were unwanted, nor did they refer to the equal rights of those with conditions to be born. Their responses suggested that the rhetoric suggesting eugenics, expressed by some disability activists, has been absorbed in the general population, although translated or interpreted into more personal terms as a matter of individual rejection. This critique can be seen as reflecting that of Rothman (1985) that children may be seen as commodities, in terms of their characteristics, although Rothman argued that such values could be seen as socially constructed and as inherent in the provision of screening programmes.

In discussing moral issues, people might suggest an 'ideal' position which might not necessarily predict their behaviour. At the same time, various approaches or actions might be felt to be moral or right in relation to screening programmes. A number of women in this group made the point that they could not predict how they would feel if they had a positive diagnostic test, or what they might do. Several, often with some

hesitancy, said that, although they did not agree with abortion, they might consider this. They pointed out that any decision 'would depend' on circumstances or support, or that they would have to be *"in that situation"*.

Whereas some were reticent about expressing this view, a few women more readily suggested that they might consider abortion on the grounds of foetal abnormality. They felt that they might find it difficult to care for a child that was *"really disabled"* themselves; some mentioned their circumstances, whereas others anticipated strain and felt that they might lack the qualities required to cope. One woman saw herself as *"too emotional a person"* to care properly for a child with severe disabilities, and another expressed concerns about her patience and her ability to love a disabled child. As noted already, many had argued more generally that they would see it as right to avoid giving birth to a child for whom one might feel unable to care. In discussing 'commodification', Rothman (1985) has pointed out that material circumstances may constrain choice, but also that women are held responsible for the well-being of a child and that the fear of being unable to love a child is common to pregnant women. Those women who discussed perceived difficulties did not speak of poverty, but they talked about the need for both emotional and practical support. The responses of some suggested a responsibility to themselves as well as to a child.

On the other hand, other women felt that they would feel able to care, in a practical sense, for a disabled child, so that abortion would not be justified. While some referred to their own personality, a few suggested that they might feel particularly able to bring up a child with certain disorders. Familiarity with a condition has been suggested in other studies as an important factor affecting uptake of screening (Marteau and Anionwu, 1996).

Hence, in discussing abortion on the grounds of foetal abnormality both generally and personally, the majority of respondents suggested balancing concerns about the morality of abortion with concerns about the perceived consequences. Almost all thought that there were reasons or circumstances in which 'prevention' on these grounds might be felt to be right or responsible. Their responses indicated few absolute moral values, but that whether 'prevention' was right or wrong might be felt to be a relative and an individual matter.

Choice

Screening programmes may be seen as providing information and as enhancing autonomy or reproductive choice. As illustrated already, a number of women, in contrast to many respondents in other groups, strongly supported the principle of

choice. They argued that individuals should be able to take their own decisions about whether or not to terminate a pregnancy where abnormality was found; as one woman put it, *"I think it's good that you can have the option"*. For a few, the right to choose was paramount:

1,324: If the parents decide that they don't want the child then that is up to them. I think they should have a choice. No matter what the child is like, they should still have a choice. If the child's going to have three heads, the parents should still have a choice to have it.

In a similar way, the information offered by screening programmes was seen as valuable or beneficial in itself, and in order for people to consider whether or not to prevent the birth of an affected child. Some argued that such information should be available, using the expression *"the right to know"*.

Many have expressed concerns about appropriate limits to autonomy, and it can also be argued that broader or more difficult questions about the morality of screening programmes may be avoided by a recourse to an individual approach and consideration only of implications. On the other hand, it has been suggested that individual choice and pragmatism might offer an ethical approach to the difficult moral issues posed by screening programmes (Kitcher, 1996). Choices may constrained by a cultural consensus of what is acceptable, since decisions must be legitimated within society, or by a sense of inter-personal responsibility (British Medical Association, 1998).

However, the rationale for this study is that there can be different cultural values about what is morally 'acceptable'. Also, decisions about welfare depend on perceptions of costs and benefits, and there might be different views about whether 'prevention' is felt to be justified. Sociologists and disability activists have argued that decisions cannot be seen as separate from the social context in which such choices are offered, and that choices may be constrained by material factors, by powerful cultural norms about disabled people (Finger, 1984), or about appropriate behaviour (Tymstra, 1991). It has also been suggested that screening programmes routinely offered in pregnancy imply benefit to mother and child (Lippman, 1991a; Macintyre, 1997), but may be seen as resting on unexamined 'biomedical' values or assumptions, such as that living with a condition is inevitably problematic (Lippman, 1992b; Asch, 1994; Bailey, 1996).

The majority of respondents in this group felt that they would take part in screening programmes which were offered in pregnancy. Since one issue in the debate concerns the conditions for which 'prevention' is felt appropriate, for what reasons and by whom, there was interest in the way that women spoke about the conditions being examined in this study. Many suggested that they would see the consequences of conditions as important factors in considerations about possible abortion. A number of women also

spoke about the fact that some tests were routine as an important factor in whether they might have these themselves. How did they view screening and diagnostic testing for Down's syndrome, spina bifida and cystic fibrosis?

Antenatal screening programmes for Down's syndrome and spina bifida

Women were asked about their knowledge of people with spina bifida and Down's syndrome, and whether they would see these conditions as serious. They were also asked their views about antenatal screening and diagnostic tests for these conditions, and whether they thought they might have such tests themselves.

Knowledge and perceptions

Only four women in this group spoke of having any knowledge of people with spina bifida, so there was little discussion of whether this was seen as a serious condition. Two had met people through their work with special needs, and described these people as severely handicapped, one spoke of a neighbour's daughter who had died, and someone else spoke about a good friend of her own age, who, she reported, used a wheelchair but took part in most activities.

All women, however, had some contact with people with Down's syndrome. A few spoke about people that they knew well, but all had met people with the condition either through the FE college, where there were courses for adults with special needs, or through placements at day centres.

Women spoke about people with Down's syndrome in various ways, often depending on how well they knew them. A few made the point that people with this condition were individuals, and some described the characteristics and personalities of people that they knew, and the activities in which they took part. One woman spoke of a relative as having friends and a good social life and as, *"absolutely brilliant ... she's got a great sense of humour"*.

A number of respondents, especially those attending the FE college, noted the increasing opportunities for people with Down's syndrome and other learning difficulties to take part in the same activities as others. Several spontaneously suggested that people with Down's syndrome were increasingly accepted, and they spoke of integration as beneficial for people with learning disabilities and for people without.

Many emphasised in their accounts that individuals with Down's syndrome were people like everyone else. As one put it, *"I mean, they're kind of doing the same things as*

myself, they're learning, they're in the canteen with us, they're doing the same things just like every other person", and another, *"I'd see them probably as just the same as the rest of us. Got a life just like the rest of us"*. A number of women emphasised the rhetoric of normalisation, but often in a personal way, making the point that they would treat a person with Down's syndrome equally or in the same way as they would any other person:

28,235: I just treat them normally, and the kids in here as well. I mean, I don't ... talk to them any different, I don't talk down to them.

However, the responses of several indicated that people with Down's syndrome were not always seen and treated in the same way as others, although sometimes this was not acknowledged by interviewees. The adults at the college were often referred to as a group, and sometimes as children. Some described staring at them or laughing at them. A few referred to the unpredictable behaviour of some, and one woman spoke of her fear of people with Down's syndrome.

A number of respondents described people - especially children - with Down's syndrome as positively different; as having *"special qualities"*, or as being *"lovely children ... lovely natured"*. As one woman expressed it, *"They're really really special, so different from ... you know, normal kids. They're ... they're just lovely. They're ... they're innocent"*. At the same time, all noted that people with Down's syndrome had learning difficulties, and a greater need for support.

A number of women suggested that they would see people with Down's syndrome as 'the same' but also as 'different'. One woman made this point overtly, saying, *"To me they're just like the rest of us, they're just that wee bit 'different', if you like - in quotes!"*. Another suggested this more indirectly:

15,122: You know, they're just ... like everybody else. They talk away to us, quite the thing. You know, one of the wee girls - not a 'wee girl', she's probably older than me! - talks away to you quite the thing.

Asked whether they would see the condition as serious, the responses of many women suggested several meanings to the term, in the sense of for whom the condition might be serious. Some indicated a possible meaning of 'serious' as 'a problem' - perhaps for society - in making the point that people with Down's syndrome were 'no different'. At the same time, women spoke about their perceptions of possible consequences of the disorder both for those affected and for others. These meanings were often used together:

25,376: It depends how bad it is, right enough. Because they're just ... like any other people, just that ... they're slower and ... like they're more hyper-active and that, I'd just see them like normal people.

17,075: I don't think it's serious as in fatal. I think, if they're happy enough, then I don't think they should treat them any differently really, I don't see them as much different. They're still people.

Many suggested that they knew little about the implications of the condition for affected individuals, or *"what they've to go through"*, and were reluctant to suggest whether the condition might be experienced as 'serious', although often noting that the degree of learning disability could be variable. Others, however, indicated their perceptions of the quality of life of people with Down's syndrome.

A number of respondents suggested that people with Down's syndrome appeared to be happy. Some felt that integration and improved opportunities were improving the quality of life for people with learning disabilities more generally. Several, in some cases referring to their knowledge of people with Down's syndrome, made the point that there could be assumptions about inevitable problems for individuals, or that they might be as likely as other people to have a good quality of life:

18,078: I think they're probably still got the same ... quality of life as what I've got, because ... you know, it's just ... I think they're ... I mean, it's like ... probably their mothers and fathers have got good jobs and ... they're still getting the same as what I've got. But it's ... it's just whenever I see them, they look quite happy.

On the other hand, a few respondents felt that some people with Down's syndrome might not have a good quality of life, especially because of the way that they might be seen or treated by others. They suggested that, because they were visibly different, or because of their learning disabilities or behaviour, they might be stared at or bullied, or others might be frightened of them. A small number of women, also referring to their knowledge, suggested that some people with Down's syndrome could be lonely or unhappy living in the community.

Most women, however, made a clear distinction between their perceptions of the quality of life of people with Down's syndrome and their acceptance within society, and their dependence, or need for care and support. Asked whether they would see the condition as serious, many focused on the implications for parents; as one women replied, *"If I had to look after them, yes"*. A number of women, referring to their experience, questioned the availability of support services or alternative care for the families of people with learning difficulties. Women suggested 'severity' in terms of the perceived difficulties, often of strain, for mothers:

25,265: They've got some special qualities an' all. So they wouldnae be any different from other people, but ... it ... I don't really think of them. I think of their mothers, how it must be hard to cope.
JG: What do you think are the problems that their mothers might have?
25: Don't know really. But if it's really bad, they might not be able to feed themselves, or ... they might be like that or they might ... have to dress them or ... whatever, help

them with the toilet. Just do everything for them, because they might not be able to do anything theirself.

On the other hand, other women, also drawing on their knowledge of families or their experience, made the point that people might make assumptions about problems for parents, or might be unduly frightened of the condition. They suggested that, because of their familiarity with people with Down's syndrome, they would not see the consequences as so serious as might be thought, or as others might:

12,098: Well, I suppose Down's syndrome is serious ... but not to me! Not to me. But as folk say, if you're kind of afraid of it, it's more serious - if you look at it that way that can be serious. But if you're around it every day, you wouldn't see it as that serious. (...) It's not easy. I mean, even X, that's S's mum, I mean, she even says that. It's great to have just a wee hand from time to time. But ... but he's able to get it. So.

While these women spoke of the need for support, they also spoke positively about mothers' and families' ability to cope or to manage; one described a family that she knew where the child with Down's syndrome - now an adult - was not the youngest.

Screening programmes for Down's syndrome and spina bifida

Women were asked their views about screening and diagnostic tests for spina bifida and Down's syndrome, and whether they might consider having such tests themselves. As noted already, most thought they would want to have 'the routine tests' and the responses of the majority indicated that they knew which disorders these tests could identify. Several also gave their views about whether they might consider abortion, usually referring to the implications of Down's syndrome.

A few women pointed out that neither Down's syndrome nor spina bifida was a life-threatening condition, and one argued that people with both these disorders - in contrast to some other conditions - were able to "*have a life*". Several felt that Down's syndrome was less serious than other conditions for the person affected, so that they might see abortion as inappropriate for this reason:

19,168: I don't think I'd really have an abortion for that. But if that baby was ... was sort of, it was going to be in pain or something, you know, it was just going to die, in sort of a week or so. But if it was a Down's syndrome baby, they seem really really happy.

In general, however, women considered the question of whether or not they might feel able to care for a child with Down's syndrome. A number felt they would terminate a pregnancy for this condition. Some who thought that they might have difficulties in coping with any disabled child spontaneously suggested Down's syndrome as an example of a condition with anticipated problems:

4,287: Because there's all ... quite severe mental handicap ... like Down's syndrome ... I suppose their quality of life is affected, but if ... it's ... well, the best quality of life it could have, but if it really really just can't do anything, I don't think I could cope with it personally. So anything like that ... I just don't ... it wouldn't be fair to the child, because I don't think that I would react well to that. So.

On the other hand, other women, talking about either their familiarity with the condition or their own approach or capabilities, felt that they might feel able to care for a child with Down's syndrome, although often stressing the need for support:

18,181: Well I mean, I think ... I know that I could give somebody a good ... a good life. You know, I would spend time with them. I've got a lot of time for them. I know it's a serious responsibility, because, as I said to you, I've never seen people with Down's syndrome before. But I feel that I've got a lot of time for them.

A relatively high proportion of women in this group suggested that they would not terminate a pregnancy on the grounds of Down's syndrome. This contrasts with statistics for the West of Scotland which suggest that the rate of pregnancies diagnosed and the rate terminated are very similar (Crossley *et al.*, 1994), or national statistics indicating a rate of termination of 92% (Alberman *et al.*, 1995). This difference between the reported unacceptability of abortion on the grounds of foetal abnormality and statistics of uptake has been noted by others (Richard and Green, 1993). As discussed earlier, one study has indicated an increasing general disapproval of abortion on these grounds, especially in respondents aged under 25 (Birth Control Trust, 1997).

In line with their personal and more general views, women saw it as right for screening programmes for these conditions to be offered. Although one woman made the general comment about the severity of the condition that, *"I wouldn't see Down's syndrome as a condition that you 'could not have that child' because of it"*, all respondents made the point that people could vary in their circumstances and in their perceptions of their ability to care.

While women drew on a cultural or professional rhetoric of 'normalisation' in talking about the way that people with Down's syndrome (or disabled people more generally) should be treated, in discussing screening for this condition, women suggested that considerations of practical consequences might, and perhaps should, take precedence. The value or rhetoric of choice was suggested as tying in with individual concerns about the ability to care or to cope, and values of responsibility.

Women's views about screening and diagnostic testing for Down's syndrome suggested considerable knowledge both of the condition and of the routine tests offered in pregnancy. The majority felt that they would probably take part in 'routine tests', although their reasons might have been because of the opt-out basis on which the tests

are offered, or because taking part might have been felt important or necessary because of the perceived consequences of the condition itself. How did these views compare with their views about a screening programme for cystic fibrosis?

Cystic fibrosis carrier screening

Knowledge and perceptions

Some women had encountered cystic fibrosis on their course, and had some theoretical knowledge of the disorder, while a small number had had some contact with individuals with the condition. Respondents therefore may have had a greater knowledge of cystic fibrosis than many in the general population.

Asked whether they would see the condition as serious, some women said that they were unable to answer this question without further information. Others offered their perceptions of the severity, although often noting that they had little knowledge of people with the condition, prefacing their views with comments such as *"from what I've read up on it"*, or *"as far as I know"*. A number of empirical studies have suggested a lack of knowledge about cystic fibrosis in the general population (Williamson *et al.*, 1989; Cobb *et al.*, 1991; Watson *et al.*, 1991a).

Most of those who had some knowledge of the disorder made the point that the consequences of the condition could be variable. Some were careful to point out that their evaluation of 'severity' related to only one person; one noting, *"From what I've ... it wasn't a major problem, you know, for the wee boy. But it could be for some people"*, and another, speaking of someone who was dying in young adulthood, *"Well, I mean, in his case it seems serious"*.

In discussing severity, women did not speak of people with cystic fibrosis as 'different', nor suggest that they should be treated in the same way as others; they talked about possible consequences for those affected and for their families. Some felt that they would see the condition as very serious; others that it was less serious in its implications than other disorders.

Those who felt that they would see cystic fibrosis as serious referred to the short life-expectancy. One woman, who had a relative with the condition, used similar 'objective' language to that used by women with this condition and by professionals in this study, saying, *"Mm-hm. Aye. Because, I mean, it's not as though they get better. It does kill. So, aye, it is. Yes."* Another suggested the implications of a short life-span for others, in concluding, *"It is serious. Aye. Because nobody knows when they're*

going to die." One respondent felt that cystic fibrosis was *"a very nasty disease"*, for both the family and those affected.

Several considered the implications for the quality of life of people living with cystic fibrosis. However, there was a range of views, and considerable uncertainty, about what possible consequences might be, and about the way these might be perceived. One felt that the condition could be experienced as serious, suggesting it as *"very unpleasant for the sufferer"*. Women spoke about the poor physical health of people with cystic fibrosis, and about their inability to join in the activities of their peers:

11,284: As far as I know, yes, it can be. Because we've covered a wee bit of it, in the course. And we've seen, like, slides and stuff of kids with it. And some of them can be ... yes, well, their lungs can be kind of twisted and stuff. It can make life really uncomfortable for them. Specially when they're in nursery school and they can't do what other kids are doing.

On the other hand, although many were uncertain about the prognosis, several suggested that they might see cystic fibrosis as less serious in its implications for the affected person than other conditions. One woman felt that it might be *"annoying"*. Two described the disorder as *"controllable"* or *"treatable"*, and others mentioned improvements in both treatment and prognosis. A number suggested that the condition might not prevent those affected leading a relatively 'normal life', or doing the same things as others. One woman suggested that, *"it would be serious but ... um, I think the child would still have a life"*, and another explained:

3,354: I mean you can still go out, and go out with friends, you can still go to ... a park or a funfair or whatever, and enjoy yourself with your friends and family with ... I mean it's not a life-stopping ... I mean it doesn't ... it doesn't prevent any kind of quality of life whatsoever. Like perhaps some sort of severe things, like perhaps severe cerebral palsy or severe brain damage might well do.

Several respondents who saw the condition as less serious than others made the point that people with cystic fibrosis can *"do normal things"*. A few pointed out the lack of learning disability, one saying, *"I mean, they're quite normal. You know, the learning with them is quite normal"*. These comments might be seen as suggesting fewer negative consequences for others; in talking about the implications for parents, one noted only that, *"you'd need a lot of hard work bringing up the child; it might be a bit frustrating at times"*.

Screening programmes for cystic fibrosis

The inheritance of cystic fibrosis and the risk of being a carrier and having an affected child was described by the researcher, with a diagram. Women were told about the carrier screening tests which were being offered on a pilot basis in some antenatal

clinics and which might be available for individuals and couples before conception. They were asked whether they would see these as useful and whether they might consider having CF carrier screening themselves. In contrast to the way that many spoke about testing for Down's syndrome, most women did not readily suggest their anticipated personal use of these tests, and discussion was more often in general terms of whether or not prevention was appropriate or whether it was right for carrier screening to be offered.

One respondent was shocked to learn that antenatal testing could be offered for cystic fibrosis, indicating that she would see the abortion of a wanted child with this condition as unjustified, saying, " ... *An abortion? And it was your child? Oh! No! For the ... for what I know of cystic fibrosis, it's not that serious".* A number of others suggested, sometimes spontaneously, that they might not have an abortion for this condition, saying that they would see cystic fibrosis as "*not that bad*" compared with other disorders. Those who expressed this view included eight respondents who said that they would consider termination for Down's syndrome or spina bifida, one explaining, in relation to cystic fibrosis, *"I wouldn't actually class that as handicapped".* Another suggested that the risk of an invasive test would be a factor if a child might be expected to have a 'normal childhood':

25,508: Well if it was going to cause any harm to the baby, well no I wouldn't. But if like ... the child was coming up that it wouldn't have like a normal childhood, then I would. But if like, you know, I thought maybe there was a chance of miscarriage or anything like that, then no. I would probably just have to wait and see, just wait and see how the child came out.

In talking about decision-making, women's responses suggested weighing up costs and benefits or resolving uncertainty, as proposed in the literature (Lippman-Hand and Fraser, 1979; Shiloh, 1996). Expressions such as 'normal' and 'handicapped' indicated that important factors in decisions might be seen in terms of the potential of the child and in perceptions of the consequences for carers.

In discussing whether they might take part in screening programmes for Down's syndrome and spina bifida, most women talked in terms of their ability to cope or care. In considering the same question for cystic fibrosis, respondents spoke more about the implications for the affected person and the responsibility to avoid giving birth to a child who might suffer. However, a few expressed considerable hesitation about the justification for 'prevention', by whatever means, if prognosis was thought to be improving:

22,431: I don't know if I would ... like to bring a child into the world, if it was not going to live that long. And you don't know how long it's going to live ... how long a life. ...

(Pause) I mean, they can ... if they're trying to make their life longer then - fair enough. Then I would ... (Pause) They can live a fairly long life and that.

Another respondent, however, who had emphasised the severity of the condition because of the effects on others, was initially emphatic that she would want carrier screening in pregnancy:

JG: Is that the kind of thing you think you might ...
23,322: I would take it.
JG: You would take it, OK. What ...
23: Because ... oh, this is a hard one, because I don't know if I could go through - like - having that child ... with me for twenty years and then it dying. It would be really hard. (Pause) ... Ach, it would, it would really hit me. Oh yeah. I don't think I'd be ... if I knew there was a test for cystic fibrosis and things. And I found out that my child could have it, I think ... I don't know, I don't really agree with abortion, but I don't know what I would do! Just pure panic, that's what I would do!

This response suggests that an important reason for taking part in CF carrier screening might be for the reassurance of a negative result, as suggested by Farrant (1985). Several women noted that finding out in pregnancy that a child was affected might present a dilemma about how to act on the information. Those women who said that they would see cystic fibrosis as a serious condition thought that screening before conception rather than during pregnancy might be more acceptable.

Cystic fibrosis carrier screening before conception was little discussed by the majority of women, although many, when asked, thought that they might take tests which were offered, describing these as *"helpful"* or as giving *"peace of mind"*, and taking part as *"sensible"*. A few women discussed the implications of screening for recessive conditions more generally, pointing out that they would see it as 'better' to find out about inherited conditions before pregnancy rather than having to consider abortion. They were aware of the implications that a couple who found that they were both carriers might want to consider whether to have children together. One felt that screening before pregnancy might be 'useful', saying, *"it wouldn't do any harm just finding out what ... it would be interesting to find out what kind of genes you've got"*.

Others, however, felt that they might not want such information. One respondent felt that a positive result might *"start off a big network of problems"*, and there was some ambivalence about whether screening was beneficial overall:

10,371: Aye. I think so, uhuh, I think it would help. I don't know. It would help so you knew whether you were carrying the gene or not, but I don't know whether it would be a good thing that if you were a carrier, that if you really wanted children, that you don't know what you'd do after that. But, um, I think it would help, uhuh ... definitely it would, uhuh it would.

The majority of women saw it as right for cystic carrier screening (during or before pregnancy) to be available; this was seen as *"a good thing"*. Several felt that they would see it as important to find out about any health risk to a child; as one woman put it, *"It's something you would want to know, I think. You'd want to know if there was something that would affect your children"*. The language used by almost all respondents suggested reasons in terms of the value of information rather than in terms of the importance of 'prevention'.

However, the responses of several women indicated some uncertainty about why or in whose interests screening was offered. Where women saw the condition as less serious, they expressed some hesitancy about whether arguments for reproductive choice might be appropriate, especially where the means of prevention was abortion:

13,390: Really, compared to some illnesses you can have, it's not ... during their childhood, it's not really that bad. I mean, there is, obviously, your child's going to die, maybe young. Which I know that some people maybe couldn't cope with. And therefore they wouldn't want to have ... that kind of thing happening really, so ... I don't know, I mean, it's a good ... I mean, obviously, it's good that they offer that kind of choice. Um. Just in case certain people would ... you know ... would like to have their baby aborted.

Hence although most women felt that it was right or acceptable for cystic fibrosis carrier screening to be offered, and that information could be useful, women did not readily suggest their own personal use of such tests, as they did in relation to screening for Down's syndrome and spina bifida. One reason for the different response may have been that women were told that carrier screening was being offered on a trial basis rather than routinely. However, women suggested difficulties in deciding the right course of action or weighing up costs and benefits when the main criterion for seeing 'prevention' as beneficial was felt to be the 'suffering' of the affected person. The general difficulty of using quality of life as a justification for taking decisions about prevention has been discussed by others reporting qualitative work (Kerr *et al.*, 1998b). Although many women felt that they had a lack of knowledge of the effects of cystic fibrosis, diverse views were expressed within the group about the relative severity of the condition, similar to those expressed by women affected themselves.

Discussion

Although there were differences within this group, there were similarities in the way that screening programmes were discussed, in the issues which were spoken of as important, and in values suggested by these views. Respondents generally approved of the provision of screening programmes, as has been noted in other studies of the views of women in the general population (Farrant, 1985; Wertz and Fletcher, 1993). They suggested that they would see screening programmes as beneficial because they offered

both information and the possibility of avoiding the birth of a child affected with certain conditions. Most women strongly supported the principle of choice, and felt that it was right for individuals to take their own decisions about family building.

These respondents suggested, often spontaneously, that that they would see it as morally right to avoid giving birth to an affected child in order to prevent suffering. Some mentioned the poor quality of life of those affected with certain conditions, but women also discussed possible difficulties for others, especially mothers, in caring for a child with a disabling condition, and they saw the consequences for the affected person and for others as inter-related. The majority suggested that they would see abortion on the grounds of foetal abnormality as an ethical or responsible course of action in certain circumstances or for certain disorders.

One issue in the debate about screening programmes concerns the limits to conditions for which the provision of screening and 'prevention' might be felt to be appropriate, on what grounds, and who should take such decisions. These women did not offer general or prescriptive views about limits, coherent perhaps with their emphasis on individual choice and on people's differing perceptions or circumstances. Most discussed the complex issues raised by screening programmes in a sophisticated way, as noted by others reporting lay views in this area (Kerr *et al.*, 1998a). Women's reflections on their own anticipated use of screening programmes indicated factors felt important and possible dilemmas, and it was clear that the perceived implications of conditions, for those affected and others, were felt important. A number of women mentioned difficulties for parents in caring for a child with Down's syndrome, but the ambivalence in many women's accounts indicated some uncertainty about how to take decisions in the case of cystic fibrosis. The responses of many suggested that, in spite of some theoretical knowledge, many had little practical understanding of the consequences of either spina bifida or cystic fibrosis.

Almost all these women emphasised values of welfare and responsibility. There was no discussion of the rights of those with disorders to be born. In discussions about Down's syndrome, for example, several respondents made a clear distinction between the way that people living with this disorder should be treated, and decisions about whether it might be felt right to try to avoid the birth of an affected child. Only a small number of women suggested some disapproval of those who could not accept any child; the majority spoke more about the responsibility of giving birth to child affected with a disorder. Where a woman might feel unable to love and care for a disabled child, although adoption was mentioned, many saw it as right to consider avoiding the birth of the child.

Almost all these women discussed screening programmes in a personal way, considering their own anticipated uptake of screening programmes and abortion. Most felt that they might take part in screening programmes offered in pregnancy; some saw themselves as following medical advice, but many suggested that they saw the information offered as useful. However, they indicated that decisions about abortion might be a separate issue. While several felt that they might not terminate a pregnancy on the grounds of foetal abnormality, others suggested that their decisions might depend on their circumstances or on the condition that was diagnosed.

These respondents discussed the provision of screening programmes from the standpoint of a potential parent or mother, in contrast to the perspective of many other women in the study. They considered how to be a good mother, in both accepting and loving an affected child, and in caring for the child and considering its well-being. The accounts of some suggested tension between these aims. It has been suggested that one consequence of offering routine screening programmes, especially in a climate of individual responsibility, is that it might be assumed that only certain women might be able to cope with a child affected with the disorder in question. A number of women in this group said that they thought they might find it difficult to care for a severely disabled child.

The responses of these women, as representative of women in the general population, indicated that their social class and religious backgrounds and their educational level were broadly similar to those of 'affected young women' in the study. However, their views suggested some differences in the meaning of screening programmes which can be seen in the context of their lives. These women were all students on nursing or child-care courses, and they suggested that they saw themselves as having a job or a career, unlike many women living with a congenital condition. Almost all also anticipated having children. Their focus on anticipated difficulties of caring might be seen against a background of a future envisaged as combining motherhood and working.

The readiness to accept medical advice suggested by many might be seen as reflecting a respect for medical knowledge (perhaps related to their courses in 'care' professions), but also women's acknowledged lack of understanding about the implications of certain disorders and their age. At the same time, their views might be seen as reflecting a general view of antenatal care and screening programmes as broadly beneficial. A focus on parental responsibility which might be seen as 'medical' can also be seen as an accepted value in modern society.

The final chapter of this thesis draws together the findings from all the data chapters to consider the conclusions which can be drawn about the views towards reproductive screening programmes of young women for which such screening is currently offered, compared with the views of others in the study.

CHAPTER 11

DISCUSSION

The preceding chapters have described the views about screening programmes of professionals in the fields of medicine and disability, women with spina bifida, women with cystic fibrosis and other women in the population. They have also outlined the way that women with either of these two conditions spoke about their lives. This chapter draws together the findings from these chapters to address the aims of the study and the research questions.

The chapter first reports on the attitudes of all respondents in the study towards screening programmes, looking at the way that the rhetorics of the debate were used, which were seen as important and by whom. A range of views was expressed about whether it was felt right or wrong to try to avoid the birth of an affected child, although many suggested the need for a balance, and the accounts of several respondents indicated ambivalence when talking more personally. All respondents emphasised the principle of choice, however, and the implications of this are considered.

Next, the chapter compares the views of young women living with spina bifida or with cystic fibrosis with those of professionals and other women in the population. (The attempts to include women with Down's syndrome in the study have been outlined in Chapters 5 and 7, and the views of this group are not included here.) All lay respondents, unlike professionals, tended to focus on individual decision-making, rather than on policy issues. However, there were a number of differences between 'affected women' and other women in the study in the way that screening programmes were discussed. Women living with congenital disorders described their own experiences of living with the condition; almost all felt that others in the population had inadequate knowledge or wrong perceptions of the consequences, and might take reproductive decisions in a different way from the way that they might themselves. In talking about screening programmes, they were more likely than other women to refer to issues of rights and values.

While there were similarities, there were also differences in views and values within the group of 'affected young women'. Women drew on different but coherent philosophies in talking about living with a congenital condition and in discussing screening programmes. There were also differences between women living with spina bifida and those living with cystic fibrosis in the way that screening programmes were discussed and in the issues suggested as important. The chapter considers a number of differences between the two conditions in their implications for those affected. The extent to which

women's attitudes towards screening programmes might be associated with their different experiences - such as perceptions of disability - and with their social context is considered, as well as with other differences between the groups. The chapter concludes by considering the limitations of the study, and also implications for policy and for further research.

The views expressed by all respondents in the study towards screening programmes

Since the study aimed to examine the views of both lay respondents and professionals, and professionals with a background in both medicine and disability, a range of attitudes towards screening programmes had been anticipated. Divergent views were expressed about whether or not it was felt right or wrong to avoid the birth of individuals affected with a disorder. Only one disability activist argued that the existence of any reproductive screening programme could be seen to indicate that those with impairments had less value in society or fewer rights to be born. However, a number of respondents, especially professionals, expressed concerns about the eugenic implications within screening programmes, especially where a condition was felt not to be serious.

On the other hand many respondents - including professionals in the field of health - felt that screening programmes could offer benefit. One medical professional put the case that it could be seen as generally beneficial to avoid illness and impairment in society. Others emphasised individual benefit, seeing it as right to offer a screening programme, or to make use of screening to avoid the birth of an affected child, if this would avoid severe difficulties for those caring for a child, or if the child might be expected to suffer. In expressing this view, a number of respondents discussed the need to balance concerns to avoid suffering with concerns about eugenics; they felt that there were limits to conditions for which such 'prevention' would be appropriate, raising the question of 'where to draw the line'. Most, however, were reluctant to specify conditions which might be defined as 'serious', and the responses of many indicated that the severity of a condition was felt to be a matter of perception or perspective. In a similar way, the fact that the quality of life of someone living with a disorder might be conditional on the way that they were treated, by others or in society more generally, was well understood. Respondents in this study did not see physical limitations as determining experience, as in a hypothetical 'medical model'.

The majority of those who were living with an impairment themselves drew on their subjective experience and perceptions to consider whether they would see it as right or beneficial to avoid the births of others like themselves. Many felt that they had a

reasonable quality of life, or suggested generally that assumptions about suffering might not be justified, although some discussed difficulties for others affected or for other family members. A number additionally expressed the view that, while they did experience problems, these should not be seen as inevitable but as socially caused. Disability activists have been critical of screening programmes as premised on medical values (as well as being discriminatory); from this view, social rather than medical interventions have been seen as a more appropriate response to difficulties associated with impairment.

Whereas professionals interviewed for this study tended to focus on issues concerned with the provision of screening programmes or policy, most lay respondents considered the morality of individual action. Women interpreted the conflicting discourses of 'avoiding eugenics' and 'avoiding harm' in individual terms. Hence many of those who saw it as wrong to avoid the birth of a disabled child did not suggest that the provision of screening programmes was wrong or evidence of societal prejudice, but spoke rather of an individual lack of acceptance of a disabled or affected child. Others who saw screening programmes as beneficial discussed the responsibility of individuals to avoid the suffering of a child, or noted possible difficulties for parents, or mothers, in caring for a disabled or affected child.

Most respondents discussed screening programmes both generally and personally; several drew on their own anticipated behaviour to make more general points. The accounts of a number indicated contradictory views, and some ambivalence about the best or right course of action, especially when talking more personally. For example, some women affected with spina bifida or with cystic fibrosis who questioned whether it was right or justified to avoid the birth of an affected child were more hesitant when talking about the implications for their relatives. At the same time, others who emphasised the importance of avoiding suffering were uneasy with abortion as the means of such 'prevention', and many expressed uncertainty about how to take a decision when the effects of a disorder were unfamiliar. Several suggested that the provision of screening programmes might cause dilemmas for individuals.

It was striking, however, that all respondents in the study, regardless of their own views about whether or not it was right to avert the births of individuals affected with certain or any conditions, and in spite of some concerns about anxiety or about the adequacy of information, emphasised a rhetoric of choice. All felt that decisions about using reproductive screening programmes should be a matter for the prospective parents. This position is coherent with a current focus on individual rights and responsibility, and with a post-modern or 'liberal' tolerance of a diversity of values. These findings of a dominant discourse of individual choice confirm those of a recent qualitative study in

which both professional and lay respondents stressed the value of autonomy (Kerr *et al.*, 1998b; c).

It has been proposed in the literature that such a pragmatic approach might offer an ethical approach to screening programmes when there are conflicting moral views about 'prevention' (Kitcher, 1996). It has also been seen as right for decisions to be a matter for the individual, because personal circumstances and perceptions might differ and because there is a 'right to choose'. (Here the 'right to choose', at one time a feminist discourse, has not only become more generally accepted, but there has been a shift in meaning from a choice about abortion to a choice about trying to avoid having an affected child.) In contrast to this positive view of 'individual reproductive choice', sociologists - from different perspectives - have drawn attention to difficulties with the concept, arguing that 'choice' can create dilemmas when values conflict, that an unchallenged rhetoric of autonomy can mask material constraints on decisions, and that 'choice' should be seen, not as a positive option, but as an obligation. Respondents in this study did not suggest that 'choice' was seen as a boon, but rather that personal decision-making, unconstrained by law, was felt to be the right approach to a difficult issue. However, some of those who initially advocated individual choice expressed some hesitancy after further discussion.

A prerequisite of offering choice is that decisions should be informed ones, and many respondents emphasised the importance of providing adequate information with screening programmes. Information was generally seen as beneficial, but it was also recognised that it could cause anxiety and might be unwanted, and that offering screening tests could create pressure. Where there was doubt about whether information was necessary, or about what beneficial action might be taken, some respondents questioned why tests were offered.

One implication of the principle of autonomy is that people might take decisions with which others might not agree. Some respondents made this general point, and the accounts of many suggested a reluctance to judge others' decisions or values. At the same time, there were felt to be limits to 'conditions' or characteristics which it was felt acceptable to avoid. Participants in this study did not advocate constraints on choice; the question was often felt to be whether or not it was right for certain options to be available.

A second implication of offering choice is that individuals might be assumed to take responsibility - or might be held responsible - for the consequences of their actions. The accounts of a number of respondents indicated that it was seen as important to try to anticipate the likely effects of reproductive decisions. However, there was no view

that actions might have social consequences; outcomes were seen in terms of individual costs and benefits. Many mentioned responsibility for the welfare of others, or 'interpersonal responsibility', as noted by Rhodes (1998) but it was also suggested as right for women to consider consequences for themselves; not to consider one's capacity for coping with a disabled or affected child was seen by some as irresponsible or wrong.

At the same time, responsibility for the welfare of others or oneself was often spoken of as having to be weighed against other ethical considerations or values, such as beliefs about the morality of the means of 'prevention' or concerns about prejudice, but also more personal desires and considerations. It was recognised that to try to avoid the birth of an affected child, or not to do so, could be seen as right, but also as blameworthy, for a number of reasons. The hesitancy and internal contradictions in some accounts can be seen as reflecting the contradictory ethical imperatives regarding 'prevention', as well as tensions between general and personal views, or consideration for the welfare both of the affected person and of others. A number of respondents who spoke more personally suggested a complexity of feelings and opinions about screening programmes, and almost all offered explanations for their anticipated behaviour or their views.

Hence, while there was a theoretical tolerance of others' decisions, trying to avoid the birth of an affected child was seen as a moral issue, and most respondents were concerned to convey their own ethical stance. The point has been made that respondents in research interviews want to be seen as a moral person (Voysey, 1975; Pinder, 1995). A number of respondents in this study expressed concern about the way that their decisions might be perceived by others (as selfish, for example), indicating an awareness of others' disapproval or social pressure. Others however, expressed more prescriptive views about the right course of action, or indicated issues of importance to themselves.

To summarise, the rhetorics identified in the debate about screening programmes - the contradictory ethical values of rights and welfare, as well as a critique of a 'welfare' position as 'medical', and a rhetoric of autonomy - were drawn on by respondents in this study. The responses of many suggested that the conflicting arguments of rights and welfare have been translated into individual terms, coherent with a generally accepted discourse of individual choice and responsibility. Perhaps for this reason, there were felt to be limits on the screening programmes that it was right or beneficial to offer.

While a horizontal analysis has indicated the ways that the discourses identified in the literature were used, and which were suggested as important, this was a comparative study. A vertical analysis of the data showed that the accounts of individuals were coherent and consistent in the discourses drawn on and in the way that contradictory values and general or personal concerns were balanced. There were also patterns in the data, in that differences and similarities both within groups and between groups could be identified.

The views of 'affected young women' about reproductive screening programmes, compared with those of professionals in the related fields of medicine and disability and those of other women in the general population.

Professionals in the fields of medicine and disability offered a range of views towards screening programmes which reflected the parameters of the debate about screening policy. Several raised the question of an appropriate balance between concerns about eugenics and concerns for welfare. While supporting autonomy, a number of professionals - from different perspectives - noted possible influences on choice, and some questioned the adequacy of the information on which people might base decisions.

Like many professionals, women with spina bifida or with cystic fibrosis drew on their own knowledge, experience and perceptions to indicate the arguments about screening programmes which they felt were important. Even more than professionals with a background in medicine or disability, those affected themselves can be seen as 'experts', in terms of their feelings about screening programmes, but also their familiarity with the condition; these women also raised questions about the existing knowledge and perceptions about their condition among the general public. Unlike professionals, however, most affected young women focused on individual decision making or choices, as did other women in the study.

Other women, as representative of women in the general population, saw the provision of screening programmes as beneficial, whether or not they anticipated taking part themselves; these women strongly supported the principle of choice. In this group, general issues were often interpreted in more personal terms, and dilemmas resolved in terms of perceptions of welfare and a mother's responsibility. Often by reference to their own anticipated use of screening programmes, women suggested that they might see it as ethical and appropriate to prevent the birth of a child affected with certain disorders, where a child might be expected to suffer or where a woman might feel unable to care for or cope with a child.

Like other women in the study, young women affected with spina bifida or with cystic fibrosis supported others' rights to take their own decisions about the use of screening programmes. Several of these women also discussed whether or not they might consider using a screening programme themselves if they thought of having children, and some felt that they might take medical advice. There were a number of similarities between 'affected' young women and others; the social class background of all groups of women was similar (although wide), 'affected' and 'unaffected' young women reported a range of educational and religious backgrounds, and all 'unaffected' women had some theoretical knowledge of the conditions being examined in this study.

However, while there were similarities between 'affected young women' and others, there were also differences between the two groups in the way that screening programmes were discussed. The majority of women with spina bifida or with cystic fibrosis focused on the question of whether or not they would see it as justified to avoid the birth of a child affected with the same condition as themselves. Most spoke in general, rather than personal, terms. They reported on their own experience of living with the disorder, including their perceptions of their quality of life, and they offered their views about the 'severity' of the condition. Inspection of the data showed the wide differences in knowledge and perceptions between individuals affected themselves and others. Almost all 'affected' women also spontaneously suggested that there were misconceptions and a lack of knowledge about their disorder among the public, and many felt that improved information should be offered with screening programmes. Thus the views of the 'affected women' in this study echoed those of disabled women in the literature; that women offered screening should be offered a 'real' or informed choice (Saxton, 1984; Morris, 1991; Asch, 1994).

In discussing their lives, almost all these women spontaneously referred to their approach towards living with a congenital disorder. They mentioned practical strategies for dealing with the disorder or its effects, but women also described attitudinal strategies or personal philosophies. Such strategies - of trying to lead as normal a life as possible, or of focusing on the person rather than on the condition - have been noted in other studies, and have been seen as ways of perceiving a disorder or limitations as manageable. From a view that those affected with certain disorders may be seen - by others and by themselves - as a negatively different type of person, such strategies have also been explained as ways of conceptualising difference, resolving tension between normality and difference or managing 'spoiled' identity (Goffman, 1968). This study was not exploring questions of identity or self-concept. However, in talking about their lives, a number of 'affected young women' spoke of a

sense of being both 'the same' and 'different'; a 'normal person' yet acknowledging the implications of being affected with the disorder for their lives and for themselves.

While the accounts of these women indicated a heightened awareness of the concept of 'difference', most did not use the language of normality and difference in relation to screening programmes. Nor did they discuss the tension inherent in such programmes between 'avoiding the condition' and 'avoiding the person'. However, in response to questions about screening programmes, a number of 'affected young women' referred to the rights and the value of the affected individual, as well as to issues of responsibility and welfare. Some raised concerns about value-judgements in the provision of screening programmes, whereas others spoke more about individuals' lack of acceptance. Other women in the study, however, rarely raised these issues.

The responses of 'affected young women' indicated that they were aware of arguments both about the valued lives of those affected with conditions which might be 'prevented' by the use of screening programmes, and about the importance of finding out the subjective perceptions of those living with the disorder, rather than making assumptions about 'problems'. Several suggested that having an affected child did not or should not matter; that difficulties might not be inevitable or that definitions of 'normality' might be widened. The accounts of many indicated an aversion to the idea of avoiding the births of others like themselves, and some expressed the view that they might see such decisions as ill-informed or unaccepting. On the other hand, many of these women described experiencing difficulties themselves. The responses of several indicated some ambivalence or contradictions in their views about screening; women were loath to condone either the provision or the use of screening programmes, yet at the same time they were unwilling to minimise the difficulties they were experiencing. The general views expressed by these women can be seen as indications of a dilemma or a reluctance to offer 'advice'. Several thought that they might not have children themselves.

The more complex way in which screening programmes were discussed, and the differences from the views of other women, suggests that the perspective of women living with spina bifida or with cystic fibrosis can be seen as shaped by their being affected with a 'preventable' condition in several ways; overtly in that these women offered opinions informed by their familiarity with the disorder - their subjective perceptions and their more objective knowledge - and in their greater sensitivity to issues of rights and values. Most suggested that others in the population might take reproductive decisions in a different way from the way that they might themselves, indicating that they saw their experience of being affected with such a disorder as having an impact on their views or their perspective.

Were there differences in views within the group of 'affected young women'? If so, with what might these differences be associated?

While there were similarities in the ways that 'affected young women' discussed screening programmes, differences in views and values could also be identified. There were differences between women with spina bifida and women with cystic fibrosis in the issues suggested as important (which will be discussed below), but there were other differences within the group.

Women with either of the two conditions described their 'strategy' for living with a congenital disorder, and two opposite philosophical approaches could be identified; an 'activist' or normalising approach, and an 'accepting' approach. While such strategies might be pragmatic, reflecting an appropriate approach to individual limitations, the philosophies expressed were coherent, to an extent, with women's views towards screening programmes. Those women who described a more 'activist' approach, emphasising potential, overcoming limitations and taking part in the same activities as 'able-bodied' people, tended to stress the importance of the provision of parental choice; some also indicated that the dependence of the affected person might be a concern. Other women with a more 'accepting' philosophy, by which the person is important and limitations do not matter - those who emphasised happiness, relationships and a valued role within families - tended to suggest a more fatalistic approach towards having children and the acceptance of any child. Having an appropriate approach towards living with a disorder has itself been seen as a moral matter (Pierret, 1993). Radley (1993) has argued that personal philosophies towards living with a condition may be reflected in approaches towards other aspects of life, and this study has tended to confirm this view.

There were also differences between women with spina bifida and women with cystic fibrosis in the way that screening programmes were discussed and in the issues suggested as important. A number of women with spina bifida did not discuss screening programmes at length; the focus of interviews with many of these women was on their experience of living with the disorder or with an impairment. These women did not question the provision of screening programmes, whether for neural tube defects or more generally, but few offered arguments in their support. The majority saw abortion as unacceptable, and most also suggested that impairment did not provide grounds for abortion (although the few women who discussed having children themselves were more ambivalent). A view of abortion or abortion on the grounds of foetal abnormality as wrong precludes discussion of the consequences of a disorder, whether for the affected person or for others, and a number of women made the point that their general views applied to any condition. At the same time, almost all also

suggested, often drawing on their own experiences and perceptions, that they would not see spina bifida as a serious condition which it was necessary to prevent, whether because they could do most things which others could do, or because they saw limitations as unimportant.

Like women with spina bifida, women with cystic fibrosis did not question the provision of reproductive screening programmes in general; several discussed the possibility of undergoing screening themselves in certain circumstances or to find out about certain conditions. Their responses suggested, however, that population carrier screening for cystic fibrosis was a salient issue. Almost all rejected the idea of terminating a pregnancy on the grounds of cystic fibrosis. However, there were divergent views - and some ambivalence - within the group about whether or not cystic fibrosis was a condition serious enough for 'prevention' (by any means) to be justified, and consequently about whether or not carrier screening should be generally available. Those who saw it as right and responsible to avoid the birth of an affected child emphasised the shortened life-span, psychological difficulties for those affected and the effects on other family members, and they expressed concern that others in the population might underestimate the severity of the condition. Other women who saw the disorder as less serious were optimistic about treatments, they drew attention to their own good or happy life, and their concerns were about others' over-negative perceptions. Some women with this latter view raised more general concerns about appropriate limits to screening and about the provision of genetic information about characteristics, and they suggested that offering diagnostic tests in reproduction for conditions not felt to be serious could be seen as a drive within society towards perfection.

There may be a number of reasons for differences in views between women affected with spina bifida and those affected with cystic fibrosis. Although the incidence of both conditions might be reduced by a screening programme, there are differences in the screening programmes themselves. There are also differences between the two conditions in their implications for those affected, as illustrated in the table below.

	Spina bifida	Cystic fibrosis
Screening programme	Offered during pregnancy Means of prevention is abortion Routinely offered	Can identify carriers Implications for population Means of prevention need not be abortion Not routinely offered
Physical limitations associated with the disorder	Stable impairment Mobility limitations	Progressive chronic illness Increasing periods of ill-health Shortened life-expectancy
Important consequences for those affected	May be social rather than medical, e.g. social isolation, fewer opportunities for employment	May be medical rather than social
Implications for social interaction	Limitations normally visible. Those affected may be seen as 'discredited'.	Limitations may not be visible, and might be concealed where the condition is less severe. Those affected may be seen as 'discreditable'.
Engagement with the concerns of disability activists	High	Low
Contact with medical professionals	Limited	High

The women affected with spina bifida or with cystic fibrosis interviewed for this study described very different experiences of living with their impairment or disorder. They perceived the main problems and the causes of difficulties in opposite ways, and they reported different degrees of contact with medical professionals.

In talking about their lives, almost all women with spina bifida described some difficulties of being unable to take part in the same activities as others, or a lack of equal opportunities, and some mentioned others' attitudes; problems which most saw as not inevitable but as having the potential to be lessened or solved by social change or indirectly through campaigns. Women with spina bifida saw themselves as disabled (although disliking negative connotations of difference in the term), and they described the experience of disability in similar terms to the arguments of the social model. From the social model of disability, people with impairments can be seen as disadvantaged by society, whether because of material inequalities, or from unchallenged assumptions about those with impairments or about the medical or individual causes of problems. Almost all women with spina bifida stressed that they saw the physical effects of their condition as of low priority, and as minor in themselves, although in many cases having serious effects on their lives and their life chances. These women saw a good quality of life as achievable, although - unlike disability activists - they spoke about personal strategies and interpersonal relationships, as well as changes in society, as important.

The majority of women with spina bifida described having little contact with doctors and hospitals.

The views and values of women with spina bifida towards screening programmes can be seen as coherent with the way that they spoke about their experiences of living with the disorder. In discussing giving birth to a child affected with spina bifida, a number of women used the phrase, 'it's just a disability'. This might suggest that impairment does not give grounds for 'prevention', whether because difficulties should not be seen as inevitable or because difference should not matter, and a number suggested that their views might apply to any condition. Hence the views of women with spina bifida towards screening programmes can be seen as reflecting some of the arguments of disability activists. Some concern about eugenics were expressed, although in personal terms, and the social causes of problems of disabled people were emphasised. However, women did not condemn screening programmes; they supported the principle of choice. Many also mentioned the role of perceptions or philosophies in their own views, and their references to the particular implications of spina bifida indicated some acknowledgement that the condition being tested for might be relevant in decision-making.

The women with cystic fibrosis taking part in this study described very different experiences of living with the condition. Almost all referred to the relative severity of their symptoms, whether by comparison with others in the population, others affected with CF or their earlier state of health. These women did not suggest that the important problems that they - or others with cystic fibrosis - experienced were socially caused. Rather, most saw the quality of life of those affected as dependent on the progress of the disorder, and some - unlike other women in the study - stressed the general importance of health in quality of life. Women spoke about the importance of trying to maintain their own health, and the accounts of many demonstrated considerable knowledge of the disorder and its effects, improvements in treatment and current progress in research. The views of this group suggested a more 'medical' perspective. People living with cystic fibrosis are highly dependent throughout their lives on medical care. The women in this study described having regular contact with doctors and hospitals; they spoke positively of the expertise of consultants and they described discussing treatments, and sometimes pregnancy, with medical professionals. When asked, most said that they did not see themselves as disabled, and some strongly refuted this definition of themselves.

As with women with spina bifida, the views of women with cystic fibrosis towards screening programmes can be seen as coherent with the way that they spoke about their lives. In talking about avoiding the birth of an affected child, women discussed their

perceptions of the effects of various disorders for the person affected and for others. The general issues raised by this group, such as appropriate limits to screening or the relative severity of cystic fibrosis, reflected current concerns in the medical literature, and different medical perspectives. The ambivalence about the 'severity' of cystic fibrosis can be seen, to an extent, as a tension between the objective knowledge of the disorder and subjective perceptions of the consequences. Many women saw the condition as serious while suggesting that they had a good or happy life themselves. While women with cystic fibrosis did not see themselves as disabled, some did express concern about the potential for eugenics in screening programmes, although only in relation to conditions felt not to be serious.

Thus, while both spina bifida and cystic fibrosis are congenital conditions, the way that women described their lives and their perceptions of the main causes of difficulties suggested that the two conditions differ widely in their implications for the affected individual, and that the experience of living with a physical or mobility impairment is perceived very differently from the experience of living with chronic illness. Women with spina bifida described an experience of disability by which important difficulties were common to those living with impairments and socially caused; women with cystic fibrosis did not hold this view. The different issues highlighted by women with each condition in discussing their lives were reflected in their concerns - or lack of concerns - about a child being affected with specific, or with any, disorders and in their views about screening programmes more generally.

The impact that women's experiences of living with a specific disorder or a condition seen as 'preventable' might have on their views about reproductive screening programmes

Views about screening programmes might be shaped by a number of factors, both individual and cultural, and those of 'young affected women' must be considered against the background of their lives. Their views might be seen as shaped by their being affected with a condition for which such screening is offered in a number of ways. As noted already, the responses of this group indicated a greater sensitivity than suggested by other women to issues of rights and values. 'Affected' women also drew on their own experience and knowledge of their disorder to consider whether or not they would see it as right to try to avoid the births of others like themselves.

While there were similarities in perspective, however, there were also differences in views and values within the group. Some differences in women's attitudes towards having an affected child were coherent with opposite philosophical approaches towards 'difference' or limitations. Other differences between women with spina bifida and

women with cystic fibrosis in the issues about screening programmes suggested as important can be seen as associated with the way that women with each of these two conditions described their experience of living with a specific disorder or type of disorder.

However, in talking about their lives and in reporting on their experience, these women drew on either 'disability' or 'medical' rhetoric. From a sociological perspective, interview accounts are considered as constructions, as reflecting social context or cultural values. Women's 'experience' cannot viewed separately from the values of those with whom they have most contact, or the rhetoric of powerful groups which might resonate most strongly with their concerns or offer most hope for improvements in their lives. The accounts of 'affected women' were coherent in the discourses drawn on in discussing both their lives and their views about screening programmes. At the same time, many also spoke in narrative terms, drawing on their perceptions and their knowledge as well as on values and rhetoric.

The point has been made that it is not possible to report on the origins of the views of individuals or groups (Cornwell, 1984). There might have been other factors influencing the way that women with each of these conditions spoke about screening programmes. As illustrated in the table above, the screening programmes for the two conditions differ in their implications. Also, while the social class background of each group of women in this study appeared to be similar, there were other characteristics which were strongly represented among the groups of women with one or the other condition.

The majority of women affected with spina bifida, unlike other women in the study, saw themselves as having a religious belief, and a number reported having had a Catholic upbringing. The religious values of these women - which might also be held by their families - might be associated with the expressed view of many of abortion (on any grounds) as unacceptable. Also, several women with spina bifida felt that they might not consider having children themselves, and for these women screening programmes were seen as having little relevance. The general views expressed might have reflected a lack of personal interest.

A higher proportion of women with cystic fibrosis reported considering having children. Women with this condition tended to discuss screening programmes in more personal terms, and several said that they had thought about their own ability, or that of their partners or relatives, to care for a disabled or affected child. The concern of a number of these women about the consequences of conditions, or the effect on others, can be seen as reflecting the perspective of a potential mother, as well as a 'medical'

perspective. In addition, half the sample of women in this group described having attended courses in Higher Education or having been expected to do so. The concerns about policy and about eugenics expressed by these women might have reflected a greater access than that available to other women to theoretical debates in the media, and a range of discourses.

As well as these individual factors which might have influenced women's views, the sample for each group was drawn from different contact organisations - a support group for women with spina bifida and specialist clinics for women with cystic fibrosis; these ways of drawing the sample might have affected the way that the study was presented and perceived, and whether women decided to take part. Also, the views of women with spina bifida or with cystic fibrosis were expressed in the context of an individual interview, and there is a view that disabled subjects of research might be inhibited in the presence of a researcher perceived as of higher status, especially if he or she is seen as non-disabled. My impression was that women affected with each of these conditions perceived the purpose of the study as an opportunity to make their perceptions and their philosophies more widely known (and to educate me); nevertheless it is possible that the interview might have been experienced differently by women with each condition, thus shaping their views. Also, as in any qualitative research, reporting on respondents' views is necessarily subjective and selective.

Limitations of the study

Discussion about differences within and between groups, and about possible reasons for different views, raises the main limitation of this study, the question of who might have taken part. This is important in reflecting on the extent to which making comparisons between groups is felt to be valid, or findings generalised to other areas. It has been argued that it is philosophically problematic to generalise from the particular in qualitative work (Hammersley, 1992; Coffey and Atkinson, 1996), and the additional difficulties of cross-case analysis have been mentioned earlier. In this study, it was felt that, although true 'representativeness' would have been impossible to achieve, and might have had little meaning, there were reasons for considering the possibility of bias in the sample, especially when the number of women affected with spina bifida and cystic fibrosis who volunteered to take part was small.

Because of the way that sampling was carried out, nothing was known about those who did not take part. As discussed earlier, the topic of screening programmes was known to evoke strong feelings, and it was felt that those with definite views, about abortion on the grounds of foetal abnormality for example, might have been more likely than others to volunteer. If this were the case, general rather than personal issues might have

been felt more important, and the study perceived as less sensitive. In addition, it was felt that those women who saw themselves as 'coping well' with living with the condition might have been more likely to volunteer or to have been encouraged to take part by gatekeepers. As with any study examining the views of people living with a disorder, those with a milder condition or who were more physically fit might also have been more likely to volunteer, and conversely those for whom the study might have been sensitive or difficult, less likely.

It was felt that considerations about interviewing only those less severely affected were relevant to both conditions. In the case of cystic fibrosis the response rate was high, suggesting that the sample consisted of the majority of affected women in the age range living in the South of Scotland. However, at the moment, people with cystic fibrosis who are aged between 18 and 25 are mostly those with a less serious condition, and two women from this target population were not asked to take part in the study. In the case of spina bifida, where the response rate was lower (13 women from 44 invited by letter), there were reasons for thinking that volunteers might have been less severely affected than others with the disorder. For example, although it had been pointed out that a number of women with spina bifida might need a facilitator to take part in an interview, only one respondent was not able to speak for herself and half the sample had attended further or higher education.

Conclusion

Many have argued that it is important for the voices of those affected with conditions for which screening programmes are offered to be heard in the debate about screening, and for their views and experience to be included in research (Asch, 1994; King, 1996; Shakespeare, 1999). It has been suggested that their views and perspective might differ from those of others. In examining the views of women with two such conditions - spina bifida and cystic fibrosis - this study has addressed a gap in the existing empirical work and the theoretical literature.

Compared with other respondents, professional and lay, women living with spina bifida or with cystic fibrosis discussed screening programmes in a complex way, in both general and personal terms, and with reference to arguments about both rights and welfare. Most focused on the question of whether they would see it as right or justified to avoid the birth of a child affected with the same condition as themselves. The main concern of these women was to convey their perceptions of living with the condition; they reported on their own experience and quality of life, and several also offered their more objective knowledge about the disorder and its effects. Almost all felt that there

were misconceptions about the disorder, and they advised that improved information should be offered with screening programmes.

At the same time, these women also discussed their 'strategy' for living with a congenital disorder, and a sense of being 'the same' yet 'different', and they drew on the same philosophical approaches towards 'difference' or limitations in considering the implications of having an affected child. In addition, in discussing screening programmes, women living with a condition for which a screening programme was offered were more likely than other women to raise issues of the rights and the value of affected individuals, although concerns about eugenics were often interpreted in individual terms.

While there were similarities in the perspective of 'affected women', there were also differences within the group, both in 'strategies' for conceptualising limitations, and in the issues about screening programmes suggested as important by women living with spina bifida and by women living with cystic fibrosis. These different views were coherent with the way that women with each condition spoke about their lives. Women with spina bifida - who saw themselves as disabled - indicated generally, as have disability activists, that they might not see it as necessary to avoid the birth of a disabled child. Hence 'affected women' drew on some of the critiques of disability activists, but in different ways and to different extents, and their more general concerns were often spoken of as modified by, or in tension with, more personal considerations and perceptions.

The differences between these women and other women in the study in knowledge, perceptions, values and views indicated that being affected with a condition for which a screening programme is offered does have an impact on perspective; women themselves saw this as the case. Yet the differences within the group suggested that views about screening programmes might also be associated with philosophical approaches towards difference, with the experience of living with a specific disorder or type of disorder, with relevant dominant discourses towards living with a disorder or towards screening programmes, as well as with social context more generally.

While there were differences between the views and perspective of 'affected women' and others in the study in relation to screening programmes, there were also similarities. Like all other respondents in the study, women living with spina bifida or with cystic fibrosis emphasised individual difference and the principle of reproductive choice. Although expressing some concerns about limits to conditions for which screening programmes should be offered, or about the information provided, they saw it as right for decisions about screening to be a matter for individuals.

Carrying out this study has suggested implications for further research. First, the numbers of affected young women interviewed for this study were small, and it was not possible to ask women with Down's syndrome about screening programmes. It would be useful to examine the extent to which the findings of this research were replicated in a larger study investigating the views of people with various conditions for which a screening programme can be offered. It is also possible that different views and experiences might be suggested by men.

Secondly, a number of women in this study referred to the implications of screening programmes for their relatives, especially siblings. They discussed the way that they thought siblings might see their risk of having an affected child, but some also suggested whether or not they thought that relatives might take steps to avoid the birth of a child with the disorder which they had themselves. Others seemed to avoid this issue. The existence of screening programmes might cause particular conflicts or dilemmas for relatives, although alternatively family members might be more certain than others in their views. There have been few studies of the views of siblings about screening programmes, or of their anticipated or actual use of them. Such studies could be of general benefit in understanding how the consequences of the disorder and 'risk' are seen by this group. However, this might be a sensitive and personal issue for relatives, without the mitigating effect of the political aspect of the current study.

The findings of this study do not imply major recommendations for policy. Questions about whether or not screening programmes should be offered, and for which conditions, are already a matter of national and international debate. However, since almost all affected young women felt that not enough was known about their disorder, it seems right to recommend (it is recommended?) that more adequate information about the condition be offered with screening programmes. Information technology and improved means of communication might offer many possibilities for making such information available.

At a more personal level, my hope is that the findings from this study can contribute towards a continuing and broadening public debate about the implications of reproductive screening programmes. New developments in technology and new genetic knowledge have drawn attention to important questions, whose implications need to be widely discussed. There is a need for views which are informed by experience, and for more nuanced discussion. I hope that dissemination of the findings from this study can make a contribution towards this debate.

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* *These references have not been read*

APPENDIX I

Interview guides

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INTERVIEW GUIDE: WOMEN WITH DOWN'S SYNDROME

Introduction: explanation

I am going round talking to a lot of young women, the same sort of age as yourself. I want to talk to young women first about their lives. Then I would be interested in some of their views (on issues to do with getting married and having children). But it doesn't matter if you haven't thought about some of these issues before - there are no right answers to the questions.

Each interview will be confidential.. That is, anything that you say in the interview will be kept in confidence, and your name will not be revealed in any material that is published about the study.

First, can you tell me a bit about your life up to now - for example, where you went to school, and what you've done since then.

(Follow up interests, college, work.)

What do you like doing?
Anything you don't like?
What do you hope to do in the future?

Do you have friends at college/work?
Who are your friends?
What do you do?

What do you do in your spare time?
What do you like doing?
Anything you don't like?

What about school?
Was that a special school?
Do you still see friends from school?

What about your family?
Do you live at home?
Who with?
Do you have brothers and sisters? Older/younger?

What about your health?
Do you keep quite well?
Do you have to go to the doctors or the hospital?

Do you do any sports/exercise?

Suppose you had to describe your life as a whole:
how would you describe it?

What's good about it?
Anything you would like to change?

Would you describe/see yourself as disabled?
Do you belong to any groups?

When you were at special school, did people say that you had a learning difficulty?

Would you say, yourself, that you had a learning difficulty?
Or Down's syndrome?

If yes (disabled / learning difficulty / Down's syndrome):
You were saying that you have ..... Can you tell me how this affects your life?

Does it make a difference?
If so, what difference does it make?

Does it ever stop you doing things that you would like to do?

I am quite deaf, and sometimes people get cross about this.
Have you ever found anything like that?

Is there anything you would like to change about the way you are treated?

Do you have a boy-friend?

Do you ever think about getting married?
Have you spoken about this to your boy-friend? (What does he say?)
Have you spoken abut this to your mum and dad? (What do they say)

Some people say that people with learning difficulties shouldn't get married. What do you think about this?

Do you ever think about leaving home?
Have you spoken about this?
What do people say?

Do you ever think about having children?
What do you think about that?
What problems or difficulties might there be?
If people with learning difficulties have children, who might help them?

Do you know anyone who has spina bifida?
Do you know anyone who has cystic fibrosis?

Even if you're not thinking of having children, some of these questions are about having babies.

Is there anyone you know who's got young children?
brother/sister/friend

Can you remember when they were pregnant?
Do you know what that means?
The baby is still in its mother's tummy - before it's born.

These days, people can do tests on the baby while it's still in its mother's tummy.
Sometimes they can test to see if it's going to be a boy or a girl.
Do you think your (brother etc) might have wanted to know that?

Do you think, if you were going to have children, you would want to know that?

They are saying in the newspapers that maybe one day, in the future, there could be tests on unborn babies, to see whether they will be tall or small, or have curly hair or have blue eyes.
Would you want to know that?

Suppose they could do tests to find out if the baby would be terribly ill.
Would you want to know that?

Suppose that, in the future, they could do another sort of test.
Not on babies, but on people.
Suppose they could do tests on you.
To find out if you might be very ill later on - when you were older.
Would you want to know if you might get an illness later on in life?

Is there anything else you feel we haven't talked about?

Finally, some questions about things not covered in the interview:

Do you go to church regularly? Were you brought up a Catholic?
Can you tell me what your father's job is? (or mother's or partner's)
Are you in any kind of relationship?

INTERVIEW GUIDE: WOMEN WITH SPINA BIFIDA

Introduction: explanation

I shall be talking to young women, some of them, like yourself with certain disorders, such as spina bifida, others who do not have this.
I'm interested in your views about some of the new developments in antenatal care and the new genetics. It doesn't matter if you haven't thought about some of these issues before; there aren't any right answers to these questions.

This interview will be confidential; that is, anything that you say in the interview will be kept in confidence, and your name will not be revealed in any material that is published about the study.

1. First, can you tell me a bit about your life up to now - for example, where you went to school, and what you've done since then.

schooling, special or mainstream, education level
family - parents - brothers and sisters
friends, relationships
working/student/not working
any particular interests

2. You have spina bifida. Can you tell me how this affects your life?

What are the main disadvantages, do you find?
physical limitations?
good days, bad days?
attitudes?
discrimination?
unable to do things that would like to do?

Are there advantages?

Do you tend to tell people about your disability?
reactions?
Have people's attitudes or reactions ever made you feel uncomfortable?
what do you think could be done about this?

3. Would you describe yourself as disabled?
Would you tend to describe yourself as someone who has spina bifida?

Do you belong to any organisations or support groups?
for disabled? for spina bifida?
If yes, what do you find useful about them?
Are you involved in campaigns?
If no, do you know of any groups?
views about disability rights campaigns

4. What sort of things do you find help you to cope with your life?

5. Thinking about your life as a whole, how would you describe your quality of life?

What would you say is needed to achieve a good quality of life?

6. Do other people see your condition as a serious disorder, would you say?
What do you think?

What would you describe as a serious/less serious disorder?

7. Suppose that relatives of yours knew that they were expecting a child with spina bifida.
What would you say to them, what advice would you give?

8. How would you describe your own health?
good days/bad days?
Do you take any particular measures/steps to look after your health?

Some people that I have spoken to have told me that they do have problems with not feeling well, or with physical discomfort. or pain Is this ever a problem for you?

Some people feel that there is extra pressure these days to be fit and healthy. Do you think that there is or not? If so, from where?

Have you had much contact with hospitals and doctors?
How helpful have they been? Has your GP been helpful?
pressure to be 'normal"?

9. Do you think about having children?
Have you any children?

Have you spoken to doctors about anything to do with reproduction, or about sexuality?
helpful/not helpful
others been helpful?
particular problems being a woman with spina bifida?

Have you had any genetic counselling?
Has anyone in your family had this?

Even if you're not thinking of having children, I'd still be interested in what you think about new scientific developments to do with reproduction.

10. Nowadays scientists can do a whole range of things that were seen as science fiction in the past. For example, test tube babies ... where the egg is fertilised outside the mother and then the baby put back inside the mother.
Some people see this as helpful for women who can't have children, or who have a serious genetic condition.
Other people see it as interfering with nature.

How do you feel about this?
Might you think about this yourself?

Infertility treatment is expensive, and not available everywhere on the NHS.
Do you think that it should be available for everybody?

Some women agree to donating some of their eggs so that other women can have babies (or for research)
Can you imagine (donating some of your own eggs, or) using eggs from someone else?

Sometimes there is a shortage of donated eggs. One suggestion has been that eggs from foetuses, from abortions, might be used.
What would your reaction be to this?

11. This study is looking at some conditions that are inherited. Scientists think that spina bifida may be partly inherited.
Does anyone else in your family have spina bifida?

Some other conditions that are thought to be partly inherited are cystic fibrosis and Down's syndrome.
Do you know anyone who has cystic fibrosis or Down's syndrome?

12. Cystic fibrosis is a disorder which is genetically inherited.
It's quite a rare disease. A child with cystic fibrosis can be born to parents who are both carriers for the disease. People who are carriers often don't know this, because they are not ill themselves. Sometimes they find out that they are carriers when they have a child affected by cystic fibrosis.

Diagram showing 1 in 4 chance of a child of carrier parents being affected with CF.

It is quite common for people to be carriers for cystic fibrosis. It only matters if they have children with someone else who is also a carrier.

In some parts of the country, tests have been offered for people to find out whether they are carriers for cystic fibrosis. This screening test is quite simple and harmless.

Do you think you would be interested in finding out, yourself, if you were a carrier for CF?
reasons? serious/not serious, anxiety, risk

At the moment, there are some trial programmes, offering these screening tests to couples at antenatal clinics.

If you were pregnant, would you want yourself and your partner tested, do you think?

Do you think that this kind of screening should be available to everyone?
Or should it be just for those with a family history of cystic fibrosis?
Should people be able to pay for this test?

13. If a couple are both carriers for cystic fibrosis, then their unborn baby could be tested to see whether it has cystic fibrosis.
This would be done by a diagnostic test at the antenatal clinic.

These kinds of diagnostic tests have been used with older pregnant women, for some time, to test babies for spina bifida and Down's syndrome.
(Do you know anyone who has had this kind of diagnostic test?)

These days, though, all pregnant women are offered a screening test to see whether their pregnancy might be at a high risk of either Down's syndrome or spina bifida. The test itself is quite harmless, a blood test.

If you were pregnant, would you think about having this yourself?

If no, how do you feel about this screening test being offered to all pregnant women?

abortion, choice, risks of tests, information provided.

If there is a high risk, a woman will be offered a more accurate diagnostic test. And if the unborn baby is found to have either Down's syndrome or spina bifida, then the parents would be offered an abortion.

(depending on previous replies)
Is this something you would think about yourself?

Do you think abortion for these conditions should be available?

14. Some diagnostic tests can tell the sex of an unborn baby.
In some countries people can choose not to have a baby, if it's the sex that they don't want.

Do you think people should be allowed to choose the sex of a baby?

15. They are saying in the newspapers that scientists will soon be able to find out more and more about unborn babies, for example, whether a baby will be tall or bald, or have blue eyes.
Would you want to know this sort of information?

Should people be allowed to have this information if it is available?

Is there anything that you might want to find out about yourself, (about an unborn child)?

medical/non-medical, serious/non-serious
treatment available?
late onset, preconditions?
knowledge, ignorance, fate
anxiety

Where would you draw the line?

16. Some people think that prenatal screening and diagnostic tests should be banned. At the other extreme, in China for example, genetic tests for married couples are compulsory. In this country, most people are offered antenatal screening and diagnosis, but they have the choice - they have the right to refuse.

Where would you stand on this?

Now that these tests are available - say for Down's syndrome - suppose that a lot of people decide to have the tests and decide not to have a baby with Down's syndrome.
Do you think that this could change our attitudes towards people who decide to have a baby with Down's syndrome?

Or towards people who decide not to have the tests done?

17. All newborn children are tested at the moment to see whether they have certain conditions. In the future, scientists might be able to find out more about the heredity of newborn children. For example, whether they are carriers for genetic disorders or whether they might develop a disease - say a heart condition - later in life.
Would you want to know this kind of information about a child?

Might there be advantages? Might there be disadvantages?

18. How would you feel about having this kind of information about yourself?
Say in terms of your risks of developing disease later in life?

Some people think we shall all soon know all about our own heredity and have 'DNA identity cards'. This might tell us if we were carriers for a genetic disorder. Or it might tell us if we had a tendency to develop something like a heart condition later in life.

What would your reaction to this be?
Do you think it could be helpful? Unhelpful?

19. Is there anything else you feel we haven't talked about?

20. Finally, some questions about things not covered in the interview:
Can you tell me at what stage you left school? Did you take Highers? Standard grades?
Do you go to church regularly? Were you brought up a Catholic?
Can you tell me what your father's job is? (or mother's or partner's)
Are you in any kind of relationship?

INTERVIEW GUIDE: WOMEN WITH CYSTIC FIBROSIS

Introduction: explanation

I shall be talking to young women, some of them, like yourself with certain disorders, such as cystic fibrosis, others who do not have this.
I'm interested in your views about some of the new developments in antenatal care and the new genetics. It doesn't matter if you haven't thought about some of these issues before; there aren't any right answers to these questions.

This interview will be confidential; that is, anything that you say in the interview will be kept in confidence, and your name will not be revealed in any material that is published about the study.

1. First, can you tell me a bit about your life up to now - for example, where you went to school, and what you've done since then.

schooling, special or mainstream, education level
family - parents - brothers and sisters
friends, relationships
working/student/not working
any particular interests

2. You have cystic fibrosis. Can you tell me how this affects your life?

What are the main disadvantages, do you find?

infections
never feel well, breathless
good days, bad days?
discrimination
others' attitudes
unable to do things that would like to do

Are there advantages?

Do you tend to tell people that you have CF? If no, why not?

reactions? attitudes?

Have people's attitudes or reactions ever made you feel uncomfortable?

what do you think could be done about this?

3. How would you describe your own health?

good days/bad days?

Do you take any particular measures/steps to look after your health?

Some people that I have spoken to have told me that they do have problems with not feeling well, or with physical discomfort. or pain Is this ever a problem for you?

Some people feel that there is extra pressure these days to be fit and healthy. Do you think that there is or not? If so, from where?

Can you tell me a bit about your treatment at the moment?
- medicines
- physiotherapy
- hospitals

How do you fit this in with your life?

Have there been any changes in your health recently?
- new problems
- new treatment

How long have you been going to (specialist clinic)?
- before this? few problems/somewhere else
- paediatric/adult hospitals
- health when younger - physiotherapy? - family involved

Have you had much contact with hospitals and doctors?
- spells in hospital?
- how often for check-ups?

How helpful have they been?
- information about CF

What about your GP?

4. Do you think about having children?
Have you any children?

Have you spoken to doctors about anything to do with reproduction, or about sexuality?
- helpful/not helpful
- others been helpful?
- particular problems being a woman with CF?

5. Would you describe yourself as disabled?

Do you belong to any organisations or support groups?
- for CF? CF Trust? Local support groups?
- for disabled people?

If yes, what do you find useful about them?
Are you involved in campaigns?
If no, do you know of any groups?
- views about disability rights campaigns

6. What sort of things do you find help you to cope with your life?

7. Thinking about your life as a whole, how would you describe your quality of life?

What would you say is needed to achieve a good quality of life?

8. Do other people see your condition as a serious disorder, would you say?
What do you think?
What would you describe as a serious/less serious disorder?

9. Suppose that relatives of yours knew that they were expecting a child with CF. What would you say to them, what advice would you give?

Even if you're not thinking of having children, I'd still be interested in what you think about new scientific developments to do with reproduction.

10. Nowadays scientists can do a whole range of things that were seen as science fiction in the past. For example, test tube babies ... where the egg is fertilised outside the mother and then the baby put back inside the mother.
Some people see this as helpful for women who can't have children, or who have a serious genetic condition.
Other people see it as interfering with nature.

How do you feel about this?
Might you think about this yourself?

Infertility treatment is expensive, and not available everywhere on the NHS.
Do you think that it should be available for everybody?

Some women agree to donating some of their eggs so that other women can have babies (or for research)
Can you imagine (donating some of your own eggs, or) using eggs from someone else?

Sometimes there is a shortage of donated eggs. One suggestion has been that eggs from foetuses, from abortions, might be used.
What would your reaction be to this?

11. This study is looking at some conditions that are inherited. As you probably know, cystic fibrosis is a condition that is inherited.
Does anyone else in your family have cystic fibrosis?

You probably know quite a lot about the inheritance of CF?
Do you know how it is inherited?

Diagram showing 1 in 4 chance of a child of carrier parents being affected with CF.

It is quite common for people to be carriers for cystic fibrosis. It only matters if they have children with someone else who is also a carrier.

People can find out whether they are carriers for cystic fibrosis. This screening test is quite simple and harmless.

If you were thinking of having children, your partner would be advised to have a carrier screening test for CF.
How would you feel about this?

Have any of your relatives had this screening test?
Or genetic counselling?

Do you think this kind of screening should be available to everyone? Or should it be just for those with a family history of cystic fibrosis?
Should people be able to pay for this test?

Screening can be offered at GP's surgeries, or else at antenatal clinics.
Where would you say is the best place to offer this test?

If a couple are both carriers for cystic fibrosis, then their unborn baby could be tested to see whether it has cystic fibrosis.
This would be done by a diagnostic test at the antenatal clinic.

(depending on previous replies)
Would you think about having a baby tested?

If the baby was found to have CF, you would be offered an abortion.
How would you feel about this?

12. This study is looking at some conditions that are inherited.
Some other conditions that are thought to be partly inherited are spina bifida or Down's syndrome.
Do you know anyone with spina bifida or Down's syndrome?

13. Diagnostic tests have been used with older pregnant women, for some time, to test babies for spina bifida and Down's syndrome.
(Do you know anyone who has had this kind of diagnostic test?)

These days, though, all pregnant women are offered a screening test to see whether their pregnancy might be at a high risk of either Down's syndrome or spina bifida. The test itself is quite harmless, a blood test.

If you were pregnant, would you think about having this yourself?

If no, how do you feel about this screening test being offered to all pregnant women?
abortion, choice, risks of tests, information provided.

If there is a high risk, a woman will be offered a more accurate diagnostic test. And if the unborn baby is found to have either Down's syndrome or spina bifida, then the parents would be offered an abortion.

(depending on previous replies)
Is this something you would think about yourself?

Do you think abortion for these conditions should be available?

<u>Questions 14-20 as for women with spina bifida</u>

INTERVIEW GUIDE: WOMEN IN THE GENERAL POPULATION

Introduction: explanation

I'm talking to young women, the same age as yourself, about their lives and about issues to do with having children. Some of these women have congenital conditions such as cystic fibrosis, spina bifida and Down's syndrome.

Although I've contacted you through (college), this interview has nothing to do with the course that you're doing. I'm interested in your views, as an individual, about some of the new developments in antenatal care and the new genetics. It doesn't matter if you haven't thought about some of these issues before; there aren't any right answers to these questions.

This interview will be confidential; that is, anything that you say in the interview will be kept in confidence, and your name will not be revealed in any material that is published about the study.

1. First, can you tell me a bit about your life up to now, for example, what you've done since school.

schooling, special or mainstream, education level
family - parents - brothers and sisters
friends, relationships
any particular interests

What do you hope to do after the course ?

2. One of the questions I'm asking is about people's quality of life.
How would you describe your quality of life?

What would you say is needed to achieve a good quality of life?

3. How would you describe your health?
Do you take any particular measures/steps to look after your health?

Some people feel that there is extra pressure these days to be fit and healthy. Do you think that there is or not? If so, from where?

4. This study is looking at people's views about having children.

Do you think about having children?
Have you any children?

When to have children?
Combining work and motherhood?

Even if you're not thinking of having children, I'd still be interested in what you think about new scientific developments to do with reproduction.

5. Nowadays scientists can do a whole range of things that were seen as science fiction in the past. For example, test-tube babies ... where the egg is fertilised outside the mother and then the baby put back inside the mother.
Some people see this as helpful for women who can't have children, or who have a serious genetic condition.
Other people see it as interfering with nature.

How do you feel about this?
Might you think about this yourself?

Infertility treatment is expensive, and not available everywhere on the NHS. Do you think it should be available for everybody?

Some women agree to donating some of their eggs so that other women have can have babies (or for research)
Can you imagine donating some of your own eggs, or using eggs from someone else?
Sometimes there is a shortage of donated eggs. One suggestion has been that eggs from foetuses, from abortions, might be used.
What would your reaction be to this?

6. This study is looking at the views of disabled women.
Do you know anyone who would see themselves as disabled?
meaning of 'disabled'

7. As you know, I'm speaking to women who have certain inborn conditions, such as spina bifida and Down's syndrome. Do you know anyone who has any of these conditions?

If yes, for each condition:
What's your impression of (condition)?
Would you describe this as a serious condition?

What would you see as a serious disorder?

8. Thinking about Down's syndrome or spina bifida ...
These days all pregnant women are offered a screening test to see whether their pregnancy might be at a high risk of Down's syndrome or spina bifida. The test itself is quite harmless, a blood test.
If you were pregnant, would you think about having this yourself?

If no, how do you feel about this screening test being offered to all pregnant women?

If there is a high risk, a woman will be offered a more accurate diagnostic test. And if the unborn baby is found to have either Down's syndrome or spina bifida, then the parents would be offered an abortion.

Do you think abortion for these conditions should be available?

(depending on previous replies)
Is this something you might think about yourself?

9. Some diagnostic tests can tell the sex of an unborn baby.
In some countries people can choose not to have a baby, if it's the sex that they don't want.
Do you think people should be allowed to choose the sex of a baby?

10. They are saying in the newspapers that scientists will soon be able to find out more and more about unborn babies, for example, whether a baby will be tall or bald, or have blue eyes.
Would you want to know this sort of information?

Should people be allowed to have this information if it is available?

Is there anything that you might want to find out about yourself, (about an unborn child)?

medical/non-medical, serious/non-serious
treatment available?
late onset, preconditions?
knowledge, ignorance, fate
anxiety

Where would you draw the line?

11. Some people think that prenatal screening and diagnostic tests should be banned. At the other extreme, in China for example, genetic tests for married couples are compulsory. In this country, most people are offered antenatal screening and diagnosis, but they have the choice - they have the right to refuse.

Where would you stand on this?

Now that these tests are available - say for Down's syndrome - suppose that a lot of people decide to have the tests and decide not to have a baby with Down's syndrome.
Do you think that this could change our attitudes towards people who decide to have a baby with Down's syndrome?

Or towards people who decide not to have the tests done?

12. Cystic fibrosis is a disorder which is genetically inherited.
What do you know about cystic fibrosis?
Do you know anyone who has cystic fibrosis?

If yes:
What's your impression of the disorder?
Would you describe this as a serious condition?

If no: It's a lung condition.
People with cystic fibrosis can live a fairly normal life, with some restrictions.
Some women with cystic fibrosis can have children.
But people with cystic fibrosis die quite young, in their twenties and thirties.

Cystic fibrosis is quite a rare disease. A child with cystic fibrosis can be born to parents who are both carriers for the disease. People who are carriers often don't know this, because they are not ill themselves. Sometimes they find out that they are carriers when they have a child affected by cystic fibrosis.

Diagram showing 1 in 4 chance of a child of carrier parents being affected with CF.

It's quite common for people to be carriers for cystic fibrosis. It only matters if they have children with someone else who is also a carrier.

In some parts of the country, tests have been offered for people to find out whether they are carriers for cystic fibrosis. This screening test is quite simple and harmless.

Do you think you would be interested in finding out, yourself, if you were a carrier for CF?

reasons? serious/not serious, anxiety, risk

At the moment, there are some trial programmes, offering these screening tests to couples at antenatal clinics.

If you were pregnant, would you want yourself and your partner tested, do you think?

Do you think that this kind of screening should be available to everyone?
Or should it be just for those with a family history of cystic fibrosis?
Should people be able to pay for this test?

13. All newborn children are tested at the moment to see whether they have certain conditions. In the future, scientists might be able to find out more about the heredity of newborn children. For example, whether they are carriers for genetic disorders or whether they might develop a disease - say a heart condition - later in life.
Would you want to know this kind of information about a child?

Might there be advantages? Might there be disadvantages?

14. How would you feel about having this kind of information about yourself?
Say in terms of your risks of developing the disease later in life?

Some people think we shall all soon know all about our own heredity and have 'DNA identity cards'. This might tell us if we were carriers for a genetic disorder. Or it might tell us if we had a tendency to develop something like a heart condition later in life.

What would your reaction to this be?
Do you think it could be helpful? Unhelpful?

15. Is there anything that you feel your family is particularly prone to?
Or inherited conditions?

16. Is there anything else you feel we haven't talked about?

17. Finally, some questions about things not covered in the interview:
Can you tell me at what stage you left school? Did you take Highers? Standard grades?
Do you go to church regularly? Were you brought up a Catholic?
Can you tell me what your father's job is? (or mother's or partner's)
Are you in any kind of relationship?

INTERVIEW GUIDE: PROFESSIONALS WORKING IN THE FIELD OF DOWN'S SYNDROME AND LEARNING DISABILITY

1. You work as a ..... with people who have Down's syndrome/learning disabilities. Can you tell me briefly what this involves?

2. I have been carrying out interviews with young women with other inborn disorders - cystic fibrosis and spina bifida - looking at their lives and their experiences of living with the disorder, and at their views about some issues to do with motherhood.

Do you think it is possible to include/involve young women with Down's syndrome/learning disabilities in this kind of research?
Can it be done?
Should it be done?
Will results be meaningful?

If yes, what would you see as an appropriate approach?
Groups, individuals?

What particular problems do you see?
Consent, confidentiality, unreliable answers.

3. What would you say are the particular needs of young people with Down's syndrome/learning disabilities?

What are the main ways that this affects their lives?

4. What is the reality of having children for women with women with Down's syndrome or a learning disability?

5. Do people with Down's syndrome understand that they have this condition? Do you think it would be helpful if they did?

Do they normally see themselves as having learning disabilities?

6. Do you think that to see themselves as disabled would be helpful for people with Down's syndrome?

Some people see those with disabilities as being 'disabled by society'.
Do you feel that this is a helpful approach?

7. What do you think would improve the quality of life for people with Down's syndrome?

8. How do you see future developments for people with Down's syndrome or a learning disability?

9. Antenatal screening for Down's syndrome and spina bifida is now provided routinely as part of antenatal care.
Genetic screening of couples is offered at antenatal clinics, with the possibility of abortion if the foetus is affected with CF.

Do you think there is an argument for either banning this kind of antenatal testing or for making it compulsory?
Or should there be a choice?

Do you think that the numbers of people with Down's syndrome will be reduced by antenatal screening and diagnosis?

10. Would you see Down's syndrome as a serious disorder?
What would you see as serious?

11. Thinking about new developments in genetics, some people think that one day we shall all have a 'DNA identity card'.
Do you think that this would be a positive advance?

12. Do you think there should be any limits on DNA testing?

INTERVIEW GUIDE: PROFESSIONALS WORKING IN OTHER FIELDS

1. You work as a ..... (with people who have cystic fibrosis / spina bifida).
Can you tell me briefly what this involves?

2. I have been carrying out interviews with young women with other inborn disorders - cystic fibrosis and spina bifida - looking at their lives and their experiences of living with the disorder, and at their views about some issues to do with motherhood.

What would you say are the particular needs of young disabled people / people with cystic fibrosis / spina bifida?

What are the main ways that this affects their lives?

3. What is the reality of having children for women with women with cystic fibrosis / spina bifida?

4. (Where appropriate) How do you see future developments in relation to the treatment of people with cystic fibrosis / spina bifida?

5. What do you think would improve the quality of life for people with cystic fibrosis? spina bifida?

6. Many of the people that I have spoken to have said that they would not see CF/SB as a serious disorder. Would you?
What would you see as serious?

7. I have asked people whether they would see themselves as disabled, and have had a wide range of replies.
Do you think that to see themselves as disabled would be helpful for people with cystic fibrosis / spina bifida?

Some people see those with disabilities as being 'disabled by society'.
Do you feel that this is a helpful approach?

8. To what extent do you think that people understand the genetics of their condition?

9. Antenatal screening for Down's syndrome and spina bifida is now provided routinely as part of antenatal care.
Genetic screening of couples is offered at antenatal clinics, with the possibility of abortion if the foetus is affected with CF.

Do you think there is an argument for either banning this kind of antenatal testing or for making it compulsory?
Or should there be a choice?

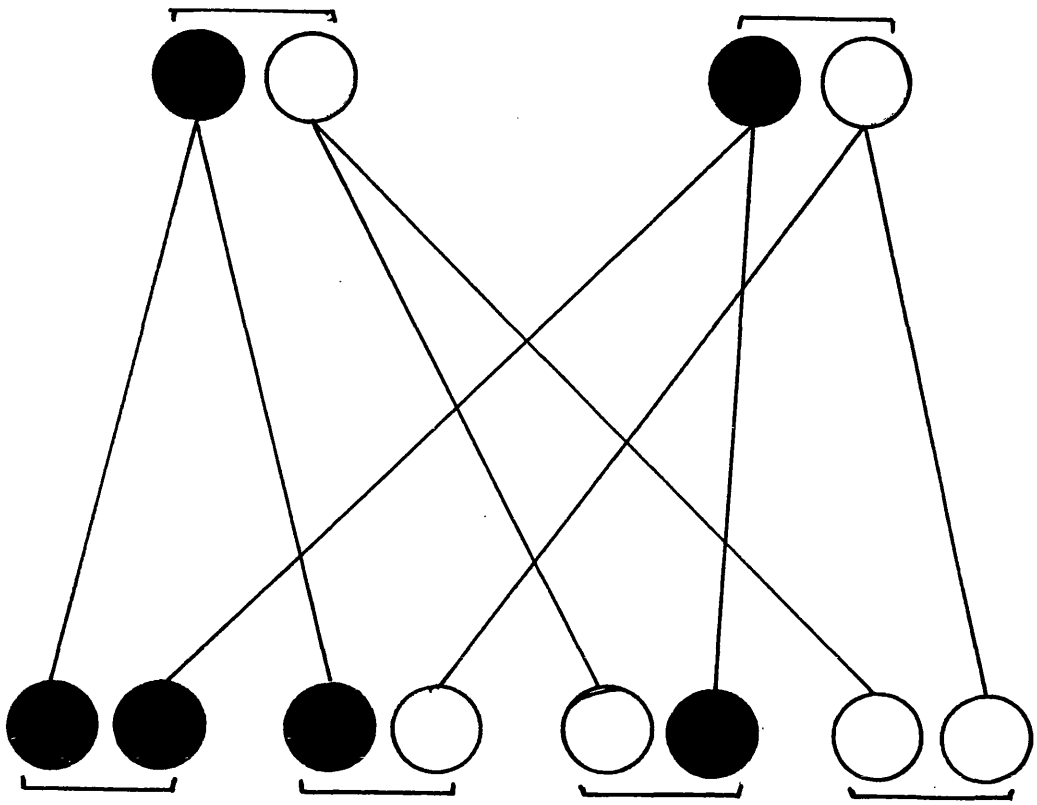
(Where appropriate) Do you think that the numbers of people with (condition) will be reduced by antenatal screening and diagnosis?

10. Some people think that one day we shall all have a 'DNA identity card'.
Do you think that this would be a positive advance?

11. Do you think there should be any limits on DNA testing?

THE INHERITANCE OF CYSTIC FIBROSIS

Both parents are carriers of cystic fibrosis.
They are not ill in any way.



1 in 4 of their children could have 2 affected genes.
This child would have cystic fibrosis.

2 in 4 of their children could have one affected gene.
These children would be carriers of cystic fibrosis.

1 in 4 of their children could have no affected genes.
This child would not have cystic fibrosis,
and would not be a carrier either.

APPENDIX II

Letters introducing the study and consent forms

(a) forwarded to women with spina bifida via the SSBA

(b) given to women with cystic fibrosis by clinic staff

UNIVERSITY
of
GLASGOW

date

name,
address,

Dear ,

I am trying to contact young women aged between 18 and 25 who have spina bifida or hydrocephalus and who might be willing to help with a small research project.

I am carrying out a research study which is funded by the Medical Research Council. The aim is to find out the views of young women, some of whom have genetically-inherited conditions, towards new developments in medicine, particularly in antenatal care.

Some of these new developments mean that it is now possible to test babies, before birth, for certain inherited conditions such as cystic fibrosis, as well as for conditions such as spina bifida and Down's syndrome. This study is looking at the views of young women towards some of these new tests.

Disabled women have often not been asked their views on these issues; this study will therefore try to change this by concentrating on women with disabilities. I am also interested in your own experience of living with a disability.

Even if you have never thought about some of these issues before, I am still interested in your views. It is not necessary to have detailed knowledge of the tests, nor do you need to be married or in a long-term relationship.

I should particularly like to include young women with spina bifida and hydrocephalus in my study, so I hope you will agree to take part. It will involve one interview, either in your own home, or in a place which is convenient for you, such as my work place at the Department of Public Health. The interview will take about half an hour, and would be completely confidential.

If you would be willing to take part in the study, or if you would like further details, please let me know by writing to me at the Department of Public Health (address below) or by phone (ext. 2254 or via the department secretary, Mrs Adrienne Girvan).

Yours sincerely,

Jane Gow

DEPARTMENT OF PUBLIC HEALTH
University of Glasgow, 2 Lilybank Gardens, Glasgow G12 8RZ
Telephone: 0141-339 8855 *Ext* *Fax:* 0141-330 5018

Consent form

I would be willing to take part in the Medical Research Council study.

I understand that it will involve one interview, which will be confidential.

Signed ..... Date .....

NAME .....(please print)

ADDRESS.....

.....

.....

Tel:.....

I can be contacted at .....

.....

Please return to:
Jane Gow, Department of Public Health, University of Glasgow,
2 Lilybank Gardens, Glasgow G12 8RZ

UNIVERSITY
of
GLASGOW

date

name,
address,

Dear ,

I am writing to ask if you would be willing to take part in a small research study, which is funded by the Medical Research Council. I'm interested in talking to young women aged 18-25, who have a genetically-inherited condition such as cystic fibrosis, about their lives and families, and about scientific developments which might affect their future.

I should be very grateful if you would be willing to take part in the study. It would involve one interview, either in your own home, or in a convenient place, such as my work place at the Department of Public Health. The interview would take about half an hour, and would be confidential.

If you would be willing to take part in the study, would you complete the attached consent form and post it (free of charge) back to me in the envelope provided. If you would like to discuss the study further, you could contact me at the Department of Public Health, telephone 0141-339-8855 (either ask for extension 2254 or the department secretary, Mrs Adrienne Girvan).

I look forward to hearing from you.

Yours sincerely,

Jane Gow

DEPARTMENT OF PUBLIC HEALTH
University of Glasgow, 2 Lilybank Gardens, Glasgow G12 8RZ
Telephone: 0141–339 8855 *Ext* *Fax:* 0141–330 5018

Lay views of the new genetics: a comparative study of the views of the young female general population with a sample of young disabled women

We invite you to participate in a study to investigate the views of young women towards antenatal care and new developments in genetics. The research project is funded by the Medical Research Council. Its aim is to look at the views of two groups of young women, those who have a genetically-inherited condition, and those who do not.

We should particularly like to include young women with cystic fibrosis in the study, as many of the new treatments, as well as new screening methods, relate to cystic fibrosis.

If you agree to participate in the study, it will involve one interview, which will last about half to three-quarters of an hour. This could take place either in your home, or in a place which is convenient for you, such as the researcher's workplace at the Department of Public Health. The interview will cover topics such as quality of life, your experience and understanding of disability, your knowledge and views about cystic fibrosis and new methods of treatment, and your knowledge and views about antenatal care and new developments in genetics. Detailed knowledge of these topics is not needed.

The interview will be completely confidential. Any views that you express or any information that you give will be kept in confidence, and your identity will not be revealed in any material from the study which is published.

Please note the following points:

Your participation in this study may not be of direct benefit to you, but could help in the development of treatment for the benefit of future patients.

If you do not wish to participate in this study, or wish to withdraw at any time, your care will in no way be affected.

If you wish to take part in the study your GP will be informed.

This research is particularly looking at the views of young women who are not pregnant; if you are pregnant, you should therefore not take part.

If you have any questions, or if you would like to discuss any aspect of the study further, do not hesitate to contact the researcher, Jane Gow, at the Department of Public Health, University of Glasgow, 2 Lilybank Gardens, Glasgow G12 8RZ, (telephone 0141-339-8855 extension 2254 or via the department secretary, Mrs Adrienne Girvan).

Form of consent

Title of project

Lay views of the new genetics: a comparative study of the views of the young female general population with a sample of young disabled women.

By signing this form you give your consent to your participation in the project whose title is above. You should have been given a complete explanation of the project to your satisfaction and have been given the opportunity to ask questions. You should have been given a summary of the research project to read and to keep. Even though you have agreed to take part in the research, you may withdraw this consent at any time without the need to explain why.

Consent:

I, ...(PRINT)

of .....

.....

.....(address)

give my consent to the research above, the nature and purpose of which have been described to me

by.....

Signature..... Date.....

I can be contacted at .....

.....

Telephone:.....