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Exploring the short term psychological impact of Mayer-
Rokitansky-Kuster-Hauser (MRKH) syndrome for young women;
an interpretational phenomenological analysis approach

and clinical research portfolio

Volume 1

(Volume 2 bound separately)

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July 2012

Mental Health and Wellbeing

University of Glasgow

Submitted in part fulfilment of the requirements for the degree of Doctorate in
Clinical Psychology (D.Clin.Psy)

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Acknowledgements

Firstly I would like to offer my sincerest thanks to the women who took time to participate in this project. I am extremely grateful for your open and valuable insights into your experiences.

Thank you to Professor Andrew Jahoda for your guidance and support throughout the completion of this thesis. Your research advice and encouragement has been greatly appreciated. Thank you to Dr Rebecca Crawford for introducing me to this area of research, for sharing your expertise in working with women with MRKH, and for your persistence in identifying potential participants. To Dr Ruth McGowan, thank you for allowing me to link in with your study and for aiding the recruitment process.

Thank you to my fellow trainees; you have been a tremendous support over the past few years. It has been great to know I was not alone through the various challenges we have been set.

I would also like to warmly thank my friends and family, you have been a wonderful source of support and distraction throughout. Thanks to Craig and June for always being there for me and for bringing Mia into my life, her smile has never failed to lift my spirits. A particular thank you to my parents for your unconditional love and belief in me, and for listening to my many stress filled rants. Finally, a huge thank you to Robert for always knowing what to say when I need you most, and for arranging many surprises and glasses of wine to help me relax! I am so excited about now having the time for us to make our new house our home and to plan our wedding!

CHAPTER 1: SYSTEMATIC REVIEW

Women's lived experience of infertility: a systematic qualitative review

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Prepared in accordance with guidelines for submission to *Health Psychology Review* (see appendix 1.1)

Submitted in part fulfilment of the requirements for the degree of Doctorate in Clinical Psychology (D.Clin.Psy)

Abstract

Background: Infertility can affect women's well-being, self-esteem, relationships and sexual satisfaction. Explanations for these effects are, however, speculative as few studies have examined women's own reports of their experiences of infertility.

Aims: The review therefore aimed to systematically examine qualitative studies of women's lived experiences of infertility within modern Western cultures.

Methods: Meta-ethnography was used to select and synthesise the studies. Articles published between 2000 and 2011 were searched for in EMBASE, MEDLINE, CINAHL and PsychINFO. Reference lists and journals of selected articles were also hand searched. Eight studies were identified for inclusion. Quality was assessed using a rating scale based on Walsh and Downe's (2006) quality rating framework.

Results: Six themes were identified; feeling different, compromised female identity, maintaining hope, sensitivity of professional support, developing a new life, and affected intimate relationships. Together these suggest that Western cultural expectations remain focused on traditional family patterns and roles. Infertility becomes a central focus for these women, with hope of pregnancy being persistent beyond an active decision to concentrate on other areas of life.

Conclusions: The need for professionals to consider the emotional impact of infertility and the potential benefits of social support are identified. Further research focusing on infertility in different contexts is advised.

Key words: Qualitative systematic review, lived experience, infertility, cultural expectations

Introduction

Around 8-10% of couples are thought to experience difficulties with fertility (Boivin, Bunting, Collins & Nygren, 2007). Despite advances in medical treatment, success of interventions for infertility remains limited, with around 30-40% of couples remaining involuntarily childless after medical treatment (Pinborg, Hougaard, Nyboe Andersen, Molbo & Schmidt, 2009; U.S. Department of Health and Human Services, 2004). A wide range of negative emotions, such as stress, anxiety and depression are commonly reported as psychological responses to infertility (Demyttenaere et al., 1998; Verhaak et al., 2007; Volgsten, Skoog Svanberg, Ekselius, Lundkvist & Sundstrom Poromaa, 2008). Literature has shown that infertility and its treatment can affect self-esteem, relationships, sexual satisfaction and psychological well-being (Bergart, 2000). Women are generally thought to be more affected by infertility than men, reporting similar levels of psychological distress to those who have physical illnesses such as cancer (Chen, Chang, Tsai & Juang, 2004; Domar, Zuttermeister & Friedman, 1993; McCarthy, 2008).

Becoming a parent is seen as a natural part of life by the majority of people (Johansson & Berg, 2005). Indeed, Carter and McGoldrick's (1989) traditional family lifecycle model makes the assumption that individuals go through eight life stages, including parenting young children and adolescents, children leaving the family home, and becoming grandparents. Infertility has therefore been conceptualised as a life crisis where an individual's assumptions about their lives and their world views can be challenged, leading to a sense of loss (Diamond, Kezur, Meyers, Schaf & Weinshel, 1999; Greil, 1997).

Infertility, however, is thought to encompass *recurrent* losses of future dreams, of parenthood, and of control over one's body (Glazer & Cooper, 1988). In keeping with Carter and McGoldrick's model, these losses are thought to become particularly pertinent at certain life stages. Wirtberg, Müller, Hogström, Tronstad and Lalos (2007) therefore proposed that,

although infertility may cause individuals to adjust their expectations and world views, crisis models are not sufficient to explain individuals' experiences of infertility as the recurring nature of associated stressful events means that adaptation and resolution are difficult.

Becker and Nachtigall (1994) proposed that pronatalist ideologies in Western cultures, which link social worth to having children and consider biological parenthood to be optimal, have led to childlessness being considered abnormal. This is in keeping with Miall (1989) who reported that those who experience infertility may be stigmatised. Fisher (1992) suggests that women may feel under more pressure to conform to societal ideals than men, which may also help to explain why they have been found to experience higher levels of distress about infertility.

These explanations, however, are somewhat speculative. Most of the research on infertility focuses on quantitative methods and on diagnostic criteria for psychiatric indicators of distress rather than exploring individuals' lived experiences (Greil, 1997, McQuillan, Greil, White & Jacob, 2003). The aim of qualitative research is to 'provide an in-depth understanding of people's experiences, perspectives and histories in the context of their personal circumstances and settings' (Spencer, Richie, Lewis & Dillon, 2003, p.3). Such an approach offers the possibility of providing insight into the lived experience of infertility and for the delivery of effective infertility care (Dyer, Abrahams, Hoffman & van der Spuy, 2002). Qualitative studies have considerable potential to provide insight into such lived experience, however there has been no proper review to determine, as a whole, what the data shows. Further, qualitative studies adopt a range of methods and also differ in quality, therefore some may be more helpful in providing insights into people's experiences than others.

This study aims to systematically review qualitative research on the experience of infertility, synthesising the data whilst taking account of methodological quality. Drawing out

themes from qualitative studies could offer theoretical insights *and* help develop more sensitive supports or interventions, based on an understanding of individual's experiences. Examining people's own experiences of being infertile is also in line with broader initiatives to gather service-user perspectives to inform health care and service development (e.g. Scottish Executive's, 2006, *Delivering for Health*).

Given the different psychological consequences for men and women indicated in the literature, this study aims to review only women's experiences. Only those who have not had a biological child through the use of reproductive technologies are included, as retrospective accounts may have been affected by having become biological mothers. Having had a child after infertility treatment may result in women feeling less different from others, hence their experiences may differ from those who remain infertile. Further, only studies from Western cultures are included to help maintain a homogeneous sample and to help elucidate the link between women's views of infertility and Western cultural notions of female identity. Such cultural beliefs, however, are historical and, as Ulrich and Weatherall (2000) have noted, there may now be a changing pattern of child bearing as more women are working and delay having children until they have developed their careers. For this reason the review focused on papers published from 2000 onwards.

Objective

The review aimed to facilitate a greater understanding of women's experiences of infertility in the current social context by identifying central themes of importance to emerge from qualitative studies, and exploring any discrepancies that were found. The review also aimed to assess the methodological quality of the reviewed studies and consider the implications of this for the interpretation of findings.

Research question

What is the lived experience of women who are infertile?

Method

Search strategy

Qualitative research can be difficult to identify using electronic databases and bibliographic database indexing systems' thesaurus terms are of limited value for qualitative research (Barroso et al, 2003; Evans, 2002; Shaw et al, 2004). However, it remains good practice to complete a robust search, with transparency being vital (Walsh & Downe, 2005). Barbour and Barbour (2003) recommend that, due to the iterative nature of qualitative research, literature searches should be broad and inclusive. A broad search strategy using free text was therefore employed to encompass the diversity of possible themes.

The EBSCO host was used to search CINAHL and PsychINFO databases, and OVID was used to search EMBASE and MEDLINE databases. All searches were completed on the 23rd September 2011 using the following terms:

1. Qualitative OR grounded theory OR interpretative phenomenological OR narrative
OR thematic analysis OR social constructionis\$ OR phenomenolog\$ OR experience\$
OR content analysis OR focus group\$ OR grounded OR interview\$ OR ethnograph\$
2. Infertility OR involuntary childlessness OR female infertility OR infertile
3. 1. AND 2.

Searches were limited to studies using human participants, and to those published in English.

Studies yielded in the electronic search were compared to the inclusion and exclusion criteria in a three step process; comparing against study title, abstract and full text. Eight

studies were identified in the search (see Figure 1 for flowchart of search results and Table 1 for study details).

In order to ensure the sensitivity of the search, reference lists from all included articles were manually reviewed. This is suggested by Walsh and Downe (2005) who note that electronic searches may not identify all relevant qualitative studies. Four further articles were identified by their title in this search, however all were discounted after reviewing their abstracts. The journals which published all selected articles were also hand searched using the same three step process, with no additional articles being identified.

Inclusion and exclusion criteria

Inclusion criteria

- Human participants
- Female participants
- Participants aged 16 and over
- Participants live in Western cultures
- Participants have undergone treatment for infertility
- Participants remain biologically childless
- Uses qualitative research methodology
- Published in the English language
- Published between 1st January 2000 and 23rd September 2011
- Published in a peer reviewed journal
- Aimed to explore some aspect(s) of women's lived experiences of being infertile

Exclusion criteria

- Primarily quantitative in design, including only elements of qualitative methodology

- Unpublished, case study or book chapter
- Focuses on the experiences of male participants only
- If both male and female participants are included, it is not possible to separate female participant data
- Participants have become biological mothers
- Participants have secondary infertility, and have previously had children
- Focuses purely on women experiencing infertility as part of a wider medical condition

Figure 1. Flowchart of Search Results

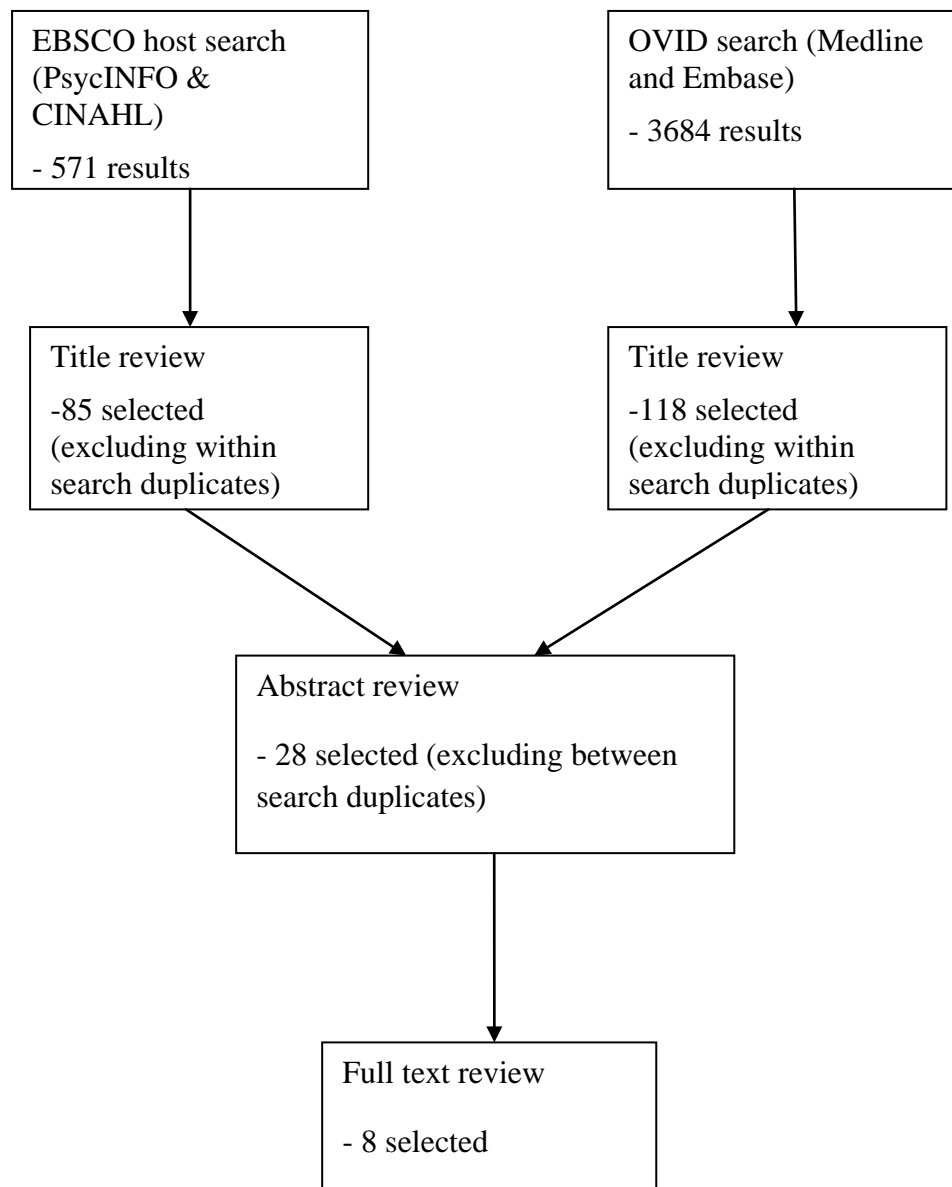


Table 1. Selected study details and quality ratings

| Study (Year) | Country | Method | Participants | Themes |
|-----------------|-----------|-------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Bergart (2000) | America | Grounded theory | <ul style="list-style-type: none"> - 10 women - 32 to 45 years old -After unsuccessful IVF treatment (ended at least 6 months earlier after at least 3 years treatment) | <ul style="list-style-type: none"> Entering the role of an 'infertility' patient Physical impact of treatment Emotional impact of treatment Reactions to extended, unsuccessful treatment The impact of pregnancy loss The impact on other areas of life Value of emotional support from other infertile women Stance of the physician Reaching limits Decision to stop treatment |
| Gonzalez (2000) | America | Thematic analysis | <ul style="list-style-type: none"> - 25 women - 20 to 40 years old -All had sought fertility treatment | <ul style="list-style-type: none"> Failure to fulfil a prescribed societal norm Assault on personal identity Mourning Transformation Restitution |
| Peters (2003) | Australia | Thematic analysis | <ul style="list-style-type: none"> - 6 women - mid 30s to early 40s -Received IVF for 2 to 3 years - Some participants had adopted or fostered | <ul style="list-style-type: none"> Keeping secrets Why Me? Trying different avenues Getting it wrong Being let down |
| Johansson and | Sweden | Phenomenology | <ul style="list-style-type: none"> - 8 women | <ul style="list-style-type: none"> Childlessness is a central issue in life |

| | | | | |
|------------------------------------------------|----------|------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Berg (2005) | | | <ul style="list-style-type: none"> - 34 to 41 years - 2 years after end of IVF treatment - Infertile for at least 7 years - One participant fostered and awaiting adoption | <p>IVF is an important and positive part of life Contact with other people is not an important issue Hope of achieving pregnancy still exists Attempts to identify other central issues in life</p> |
| Peddie, van Teijlingen and Bhattacharya (2005) | Scotland | Thematic analysis | <ul style="list-style-type: none"> - 25 women - Mean(SD) age 37 years(4.5) - 3 to 24 months after end of IVF treatment. - Some had adopted | <p>Difficulty with acceptance of infertility Stress associated with IVF Unrealistic expectations of treatment Pressure from media and society Insufficient information specific to the individual Social and professional opportunity costs Physical and emotional pressure exerted on a couple's relationship Information provision and communication skills during final consultation Lack of continued support from the IVF unit</p> |
| Wirtberg et al. (2007) | Sweden | 'Qualitative approach' | <ul style="list-style-type: none"> - 14 women - 48 to 60 years old - 20 years after end of infertility treatment | <p>Looking back at time as infertility patient Other means of parenting Long term effects of infertility and childlessness Effects on sexuality Development of non-parent life style Life as a caring activity And some did not turn the corner The silent story</p> |

| | | | | |
|--------------------------------------------------------|---------|------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| McCarthy (2008) | America | Phenomenology | <ul style="list-style-type: none"> - 22 women - 33 to 48 years old - Average 3.9 years after end of treatment - Some had adopted | Revisioning self in life's context Revisioning the world in life's context Experiencing isolation: A sister set apart Permanent presence Choosing to go on Creating a different kind of life |
| Volgsten, Skoog Svanberg and Olsson (2010) | Sweden | Content analysis | <ul style="list-style-type: none"> - 10 women (and 9 men, not included in current review) - 35 to 43 years - At least 3 years after IVF treatment - Some in process of adopting | Putting up a shield Late realization of the need of professional support Affected partner relationship Frustrated at ending of IVF Unanswered questions after ending IVF Feeling excluded and lacking understanding Loss of future life goals |

Quality of data

Assessing the quality of qualitative research has been debated for many years (Holloway and Wheeler, 1996; Perakyla, 1997; Popay & Rogers 1998). One area of controversy is how best to appraise the quality of qualitative research (Spencer et al., 2003). For example, Dixon-Woods, Shaw, Agarwal & Smith. (2004) note that it is difficult to identify what may be considered as a fatal methodological flaw in qualitative studies due to differences in qualitative research designs.

In order to develop a practical guide for assessing the quality of qualitative studies which is firmly grounded in qualitative research, Walsh and Downe (2006) reviewed eight previous frameworks for qualitative research, pulled them together and eliminated non-essential criteria. The quality of studies included in the current review were assessed using a scoring system based on Walsh and Downe's resulting framework which includes essential criteria covering the following stages; scope and purpose, design, sampling strategy, analysis, interpretation, reflexivity, ethical dimensions, and relevance and transferability (see Appendix 1.2 for scoring criteria). Each study was compared to a total of 29 criteria, with each being marked as present or absent to determine a profile of strengths and weaknesses. Criteria were marked as absent if they were not met or if it was not possible to determine from the information given. As it is difficult to define fundamental flaws in qualitative research, the framework was used to indicate the quality of studies rather than to exclude those failing to meet a predetermined threshold. All papers were second rated by an independent reviewer and any disagreements were resolved through discussion.

The scope and purpose of all the included studies was described well, with all researchers making a clear statement of their focus, rationale and research aims. All studies were contextualised by existing literature.

Studies generally had good designs that were appropriate to the research questions and all authors provided a rationale for their method of data collection. Samples and sampling methods were described for all studies and, with the exception of Gonzalez (2000) and Peters (2003) a justification was provided. In contrast, the level of background information about participants, in terms of age, fertility treatment and whether they had adopted children, varied across studies.

All studies provided an appropriate analytic approach, however specific quality criterion for analysis varied across studies. Half of the studies involved a second researcher in data analysis (Gonzalez, 2000, Peddie et al., 2005, Volgsten et al, 2010; Wirtberg et al., 2007). A strength of Gonzalez' (2000) study was that participants were also involved in the analysis. Only Gonzalez (2000) and McCarthy (2008) discussed having reached a point of data saturation.

Study contexts were described, however only Peddie et al. (2005) and Volgsten et al. (2010) explicitly accounted for the socio-cultural setting and for infertility being a sensitive issue to discuss in their interpretations. In contrast, all researchers considered the overall context of remaining infertile despite having had infertility treatment in their interpretations. All of the researchers used interview data to support their interpretations, however there were no audit trails of the processes that led to their interpretations. Only McCarthy (2008) and Peddie et al. (2005) demonstrated researcher reflexivity in their study report.

Explicit reference to ethical dimensions also varied across studies. A weakness of Gonzalez' (2000) study was that there was no indication of whether ethical approval had been granted. Whereas most (with the exception of Bergart, 2000; McCarthy, 2008; Wirtberg et al., 2007) made reference to confidentiality and anonymity, only Gonzalez (2000), McCarthy (2008) and Volgsten et al.(2010) discussed the process of participant consent.

Finally, evidence of relevance and transferability of findings was provided in all studies and findings were discussed in the context of previous theory or literature.

Acknowledgement of study limitations was generally provided, with only Bergart (2000), Gonzalez (2000) and Johansson and Berg (2005) failing to discuss such weaknesses. Three studies gave no suggestions for further research directions (Bergart, 2000; McCarthy, 2008; Volgsten et al. 2010).

Method of synthesis

The term meta-synthesis is commonly used to describe a number of methods developed to synthesise qualitative research (Barnett-Page & Thomas, 2009). These methods are interpretative in nature, aiming to further develop understanding and explanations of phenomena, generating new insights and understanding (Walsh & Downe, 2005).

Meta-ethnography (Noblit & Hare, 1988), one type of meta-synthesis, was used in the current review. This involves a seven stage process proposed by Noblit and Hare (1988) and described further by Atkins et al. (2008). The seven stages are outlined in Table 2. Meta-ethnography was selected as it allows the synthesis of studies employing a range of qualitative methods (Ring, Ritchie, Mandara & Jepson, 2011). Themes were compared across studies in order to determine the most dominant factors. This allowed for the most prominent themes to be explored in order to develop a holistic understanding of women's experiences. Participants' views and beliefs as elicited from quotations in the published studies, and the original author's interpretations of these, were synthesised. This led to the current author's interpretation of the women's experiences as a whole.

Table 2. Stages of meta-ethnography

| Stages | Description |
|------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------|
| 1) Getting started | Determine research question |
| 2) Deciding what is relevant to the initial interest | Defining the focus of the synthesis Locating relevant studies Making decisions about inclusion Quality assessment |
| 3) Reading the studies | Become familiar with the content and detail Begin to extract ‘metaphors’ or emerging themes |
| 4) Determining how studies are related | Create a list of themes and metaphors Juxtaposition of above Determine how themes are related Reduce themes to relevant categories |
| 5) Translating studies into one another | Arrange papers chronologically Compare paper 1 with paper 2, and the synthesis of these papers with paper 3 and so on |
| 6) Synthesising translations | Third order interpretation leading to a line of argument synthesis |
| 7) Expressing the synthesis | Presentation of results Publication of findings |

Results

As recommended by Noblit and Hare (1988), study themes were listed in chronological order to determine relationships between study findings and to compare identified themes. Study themes are outlined in Table 1. This process elicited the following six key factors in women's experience of infertility: (i) feeling different, (ii) compromised female identity, (iii) maintaining hope, (iv) sensitivity of professional support, (v) developing a new life, and (vi) affected intimate relationships. Highlighting the most prominent elements identified in the synthesis, each theme will be discussed in turn. Quotations from study participants appear in italics, quotations from study authors do not.

Feeling different

Women in all studies reported a sense of feeling different. The participants described a general feeling that they did not fit in with their peers, attributing this to not sharing experiences with them. One participant said:

'I hated ... not blending with everybody and being the couple that everyone felt sorry for ... It's like a sorority that I'm never going to be a part of' (McCarthy, 2008)

This quotation highlights women's feelings of being an outsider. It also suggests a desire to feel part of social group and a sense of loss that this is not the case.

Women also experienced difficulties being around others who had children or were pregnant. Gonzalez (2000) for example, notes that women in his study reported feelings of envy towards other women. Some reported that such difficulties resulted in them withdrawing from social situations, as in the following quotation.

'I didn't feel sociable, but deep down I missed the social interaction. I suppose I was afraid that someone would appear with a baby or to say that they were pregnant, and I, just couldn't cope with that.' (Peddie et al., 2005)

It seems that it was sometimes easier for women to miss out on social situations than to manage their feelings of being different and the pain of seeing others with children.

The ongoing impact of feeling different from peers was demonstrated in Wirtberg et al.'s (2007) study, which showed that 'the experience of isolation had persisted for half of the women and re-emerged when their peer group was reaching the stage of grandparenthood'. Feelings of social isolation therefore seem not only to be long lasting, but also to be particularly pertinent at certain life stages. One lady said:

'I found myself making excuses for not visiting, or taking a trip to the bathroom when the latest photos of grandchildren were passed around. I really had to get a grip on myself and break that; suddenly it was like turning the clock back twenty years.'

(Wirtberg et al., 2007)

Peers becoming grandparents echoed the women's previous experience of their social group becoming mothers. Social avoidance again became a coping strategy in this later life stage.

Feeling different from others also resulted in women feeling stigmatised or anticipating that they would be. Peters (2003) stated that 'participants acknowledge an attached stigma to aspects of infertility and assisted reproductive treatment'. He goes on to report that some women 'feared the cultural and religious implications of being infertile and seeking assisted reproductive treatment. Others wished to avoid being the target of gossip.' Women therefore seemed to be aware that their situation differed from social expectations and, as a result, often opted not to discuss their infertility with others, fearing a negative outcome. One participant said:

“It was a hidden burden. I kept it a secret. I was ashamed. I didn’t want anyone to know.” (Gonzalez, 2000)

Further, Peters (2003) stated that ‘the general public remained relatively uninformed, and therefore ill equipped to deal with issues surrounding it’. This factor was reported to further influence the sense of isolation these women experienced. Participants talked about friends and family not understanding and being insensitive or hurtful. Such experiences are apparent in the quote below.

‘Some [friendships] didn’t survive ... the infertility ... One in particular was very, very painful, because it ... started ending shortly after I lost the baby, and this had been a ten year relationship ... [S]he got pregnant about three months after I had lost the baby ... But then when she got pregnant, it became, ‘Oh, I can’t believe ... how I’m feeling. I have to go through this shit. Men don’t know how lucky they are’. And ... I said ... I can’t listen. I can’t listen to your complaint about being pregnant. I-I can’t. I just can’t’. (Bergart, 2000)

A final concern about being different from others was that of being alone in old age. McCarthy (2008) reported that ‘The lack of a family introduces an ambiguity about what to expect as one ages and contributed to a concern about being alone’. Women reported having concerns about who would look after them in later life, continuing the family, and passing on inheritance and family heirlooms.

Feeling different was described by women in all studies, including those where some participants had adopted children, suggesting that becoming a parent through this means did not overcome feelings of difference.

Maintaining hope

Women in all studies reported that they maintained a hopeful stance through infertility treatment and beyond. They explained that their menstrual cycle brought a monthly pattern of hope for pregnancy followed by despair at menstruation. Johansson and Berg (2005) described how this was true during IVF treatment, where ‘The fertilized egg is experienced as a child and when menstruation occurs it feels like a miscarriage’. Explaining this experience, one woman reported:

‘You’re so on the edge; you’re trying to get pregnant. Every month it’s a roller coaster of emotions waiting, waiting, waiting, - and then the big disappointment’
(Bergart, 2000)

Medical staff appeared to play a role in maintaining this hope. For example, Bergart (2000) found ‘The doctors of seven of the ten women maintained a hopeful stance, not indicating that treatment might fail’ and stated ‘Several women saw this stance as a “carrot”, feeling that their doctors were “dangling” hope in front of them.’ One woman in Peters (2003) study reported:

‘The staff in the clinic were sort of too positive I think. They sort of made you feel like this is really going to happen. They weren’t negative at all. They made you feel real sure of yourself. You’d hit rock bottom when it wasn’t successful’

This woman’s belief that staff were too positive echoes that of others who felt they should be given all information, including the negative, to ensure they are fully informed. The optimism of medical staff influenced the women’s beliefs about treatment success which in turn had an impact on their feelings when treatment was not successful; experiencing it as a letdown or

failure. Staff optimism and lack of accurate information therefore contribute to maintaining hope and to feeling a sense of despair at menstruation.

Women also, however, reported that they held onto the possibility of pregnancy many years after treatment finished, emphasising that as long as they menstruated there was still a chance they could have a child. One participant stated:

‘I do not think one wants to hear that it is definite before one reaches menopause, then one has to accept it. (Johansson and Berg, 2000)

This quotation illustrates that hope remains until menopause makes it explicit that the women cannot become pregnant. This was evident too in women followed up longer term, with Wirtberg et al. (2007) reporting ‘Still some of the women, who thought they had worked through the issue, surprised themselves when they reached menopause and reacted with depression and grief.’ Hope therefore, although perhaps not consciously so, seems to remain present for these women, who finally grieve their fertility when they reach menopause.

Johansson & Berg (2000) reported that ‘Hope is experienced as immensely important, being the driving force that enables life to continue’. On the other hand Volgsten et al. (2010) note that ‘it was difficult not knowing how to handle grief’. Hope therefore seems to be employed as a coping mechanism which allows these women to avoid fully experiencing the loss of their fertility. Although useful in the short term, this delayed the grief reaction and maintained a focus on fertility throughout these women’s lives.

Infertility as a central life focus indeed came across in many of the studies. As Peters (2003) reports ‘participants had a major change in their focus to life with everything now revolving around getting pregnant.’ One woman stated:

'[Y]our whole life becomes like your menstrual cycle ... [E]verything else about your life is very peripheral, and ... that's sort of like how you measure things.'

(Bergart, 2000)

Not only does maintained hope of becoming a mother become a focus, it therefore also seems to detract attention from other areas. As infertility 'takes up a large part of their existence' (Johansson & Berg, 2005), the women do not have the same resources for other areas as they may otherwise have had. One participant stated:

'I just felt like my life was in limbo for so long ... I was stuck in a job I didn't like. I was doing this treatment ... I put the rest of my life in limbo and did this. And this is what I did for three and a half years – went to doctors, had treatment, waited for my period ... It's ... almost like the freeze frame [in a] video or something.'

(Bergart, 2000)

Treatment therefore became a central aspect of life whereas other areas became less important, frozen to be resumed once pregnancy is achieved. Again this indicates a hope that pregnancy *will* be achieved.

When pregnancy is not achieved, however, women continue to focus on the hope of future pregnancy. Wirtberg et al. (2007) reported 'Another felt very strongly that the desire for a child still dominated her life and she would never get over it. To live a life of trying to conceive a child had become her dominant state, and now when her biological clock had finally 'tollled', she was at loss at how to find meaning for herself in life'. Again, focusing on the hope of pregnancy had detracted from reconsidering her life without children, which was forced upon her at menopause. In this study of the longer term experience of infertility Wirtberg et al. (2007) reported that 'The interviewer found herself often being transported

back in time, listening to a story that was a very living and dynamic part of the present, everyday life of these women.’ Although many years after treatment had finished, infertility remained a central focus which impacted their daily lives.

Compromised female identity

The majority of studies discussed an altered sense of self, largely in terms of a compromised female identity (Bergart, 2000; Gonzalez, 2000; McCarthy, 2008; Peters, 2003; Volgsten, et al. 2010; Wirtberg et al., 2007). Participants felt that a woman’s role was to become a mother and that they therefore should be able to have a child. Gonzalez (2000) reported that ‘This failure to fulfil a societal norm was described as an assault on their personal identity, often in terms of threats to self-concept and body image. One lady described this as:

‘It was almost as if you cannot run with your legs – you have two legs but you cannot run with them’ (Gonzalez, 2000)

This alludes to a perceived sense of failure of their bodies. As women they felt they should be designed to reproduce, however they were unable to do so. This often affected their self-esteem and ‘Many women described a sense of personal failure and worthlessness’ (Bergart, 2000). They felt that they were not complete as women, and were therefore less worthy than others. One woman reported:

‘there was something wrong with me I wasn’t a proper woman, I didn’t understand why my husband wanted to stay with me ... because I couldn’t give him what he wanted ... I felt worthless ... I thought I couldn’t do anything’
(Volgsten et al., 2010)

It is apparent that a compromised sense of self as women in turn affected beliefs about their worth to others. The women appeared to place a strong emphasis on child bearing in their judgement of self-worth, seeing this as a defining characteristic of a woman. This is echoed in the following quotation:

‘It’s like you thought you were this solid chocolate bunny and you’re not. You’re the hollow chocolate bunny, which is the less expensive version, not quite as good and not what everybody really wanted at Easter.’ (McCarthy, 2008)

This reveals an expectation that one will be able to have children and the challenge to one’s identity when you realise this is not so. Not only did this cause a readjustment of the women’s sense of self, but also led to a compromised one, which saw the self as inferior to others, unworthy of affection and as incomplete.

Sensitivity of professional support

Women in half of the studies reported that they were not wholly satisfied with the support provided for them by professionals during infertility treatment (Bergart, 2000; Peddie et al. 2005; Peters, 2003; Volgsten, et al., 2010). This was largely in terms of “mechanical” and impersonal aspects of the treatment, which tended to make them feel like machines or body parts rather than people’ (Bergart, 2000). One woman described her experience as:

‘[S]ometimes you’re shuffled in and out so quickly it doesn’t seem that they realize how important this is. This isn’t a bunion you’re having removed. This is something that is affecting your life, day in and day out. And you want more personalized care. You don’t wanna feel like you’re ... that 3:15-in-the-after-noon-appointment, whatever-your-name-is.’ (Bergart, 2000)

This illustrates that women felt they were not treated as individuals and that the sensitive nature of the treatment was not taken into account.

A particularly salient time for many of the women was the ending of treatment. Bergart (2000) reported that ‘Four of the ten women spoke positively of having doctors who helped them and their husbands to look at low probabilities of success, or to set limits of their treatment’ (Bergart, 2000). The time given to discuss the likelihood of success and the ending of treatment, however, was rarely reported. The majority of women’s experiences were captured by Volgsten et al. (2010) statement that ‘Treatments were described as being too forced and the ending often being abrupt.’ Peddie et al. (2005) also explained that ‘Respondents reported a sense of being ‘unprepared’ for the major step of ‘decision-making’ and felt that an additional appointment/s to discuss the effects of ending treatment would have been beneficial.’ Women expressed a desire for more time to be taken to discuss ending treatment and the impact that this may have on them.

Support in the decision making process was described as lacking and ‘The need for continued support from the Unit some time after ending treatment appeared to be universal’ (Peddie et al., 2005). Many women had no further contact with the service after they decided to end treatment and professionals made no acknowledgement of what the women had been through, or the lasting impact this may have. One woman explained:

‘[W]hen I went in there for the final conference, [the doctor was] just sitting there talking to me about the medical aspects of it, but never once said, ‘Well, how are you? ... How do you feel about this?’ ... And I was just floored by it, because ... [she] had been so good throughout everything ... [so] supportive, but then right at the end it was like, as far as any psychological support ... nothing ... I was just so hurt by [her way of] speaking to me.’ (Bergart, 2000)

This suggests that women felt they were let down by those they had placed their trust in and that there is a need for a more holistic approach to treatment, taking into account the emotional aspects as well as the medical. Women in some studies felt that additional input in the form of counselling may be beneficial:

‘To me communication is everything, and as for making the final decision to end treatment, enlisting the help of a counsellor at this stage would be a more sensitive way of going about it.’ (Peddie et al, 2005)

A failure to take adequate account of individual and emotional factors left women feeling unsupported and on their own, particularly at the end of treatment.

The above reports differ from those described by Johansson and Berg (2005) who state that: ‘The women are pleased and proud of being able to participate in IVF treatments. The treatment has given them a stronger feeling of self-esteem.’ The lead researcher and interviewer for this study, however, was involved in the care and treatment of infertile couples. Hence, participants may have felt less able to describe negative aspects of their IVF experience due to social desirability.

The studies also came from a range of Western countries and cover a range of healthcare systems, including public services in Scotland and Sweden, mixed public and private systems in Australia and private care in America. Further, despite some women having access to publicly funded treatment they opted for private care. Women’s experiences are likely to have been influenced by the particular healthcare systems they used. For example, in Peddie et al.’s (2000) study, they noted that two thirds of people opt for private fertility treatment and that financial restraints influenced the women’s decision to end treatment. In such instances it may be that endings felt abrupt as no further input could be

offered when the couple no longer had resources to pay for this. Participant's more positive experiences reported in Johansson & Berg's (2005) Swedish study may reflect their being more scope to offer support beyond active treatment in a public setting. Given the variations in sources of funding both within and between studies, however, it was difficult to fully interpret the impact of healthcare systems on the women's experience of care.

Developing a new life

Contrary to the theme of maintaining hope, which suggests that women hold onto the chance of pregnancy throughout their adult lives and that this becomes a central life focus, studies also found the women attempted to develop an alternative life to that of parenthood (Bergart, 2000; Gonzalez, 2000; Johansson & Berg, 2005; McCarthy, 2008; Peddie et al., 2005; Wirtberg et al., 2007). As McCarthy (2008) reports 'The women described very active awareness of taking stock, letting go, and doing whatever it took to create a new space to move on into.' Women made a decision to move on to a life that does not involve having children and tried to identify a different life focus. One woman explained:

'I am always going to regret that we can't have a child, but you have to accept what you've got rather than what you haven't. You have to make alternative plans and they don't include children.' (Peddie et al., 2005)

This suggests that, although a child is still desired, a point of acceptance was reached when other life opportunities could be realised. The alternative plans made varied, however Wirtberg et al. (2007) reported that 'The most-told story about making meaning in life and coming to terms with childlessness was that of caring for somebody – most often a child'. Again this contrasts the notion of avoiding situations in which there are children discussed

under the *feeling different* theme, and Wirtberg et al. (2007) note this is a shift in the women's behaviour.

'The loss of parenthood, of not having a family including a child, was described as a different life situation; but at the same time, a life situation with more freedom' (Volgsten et al. 2010). This notion of seeing a life without children as being a different life that created further opportunities was indeed identified in other studies. For example, a woman in Johansson and Berg's (2005) study reported:

'One can see how very positive it is to not have children. One can travel, I can go out in my car and drive wherever I want and whenever I like.' (Johansson and Berg, 2005)

Women identified that there were more options for them in terms of work, travel and hobbies as they were less restricted without children. Identifying such lifestyle opportunities and finding other ways to fulfil a caring role appeared to allow the women to move beyond focusing on their loss and towards developing a new life. As Wirtberg et al. (2007) report 'through consciously trying to focus on one's good fortune rather than one's misfortune, several had found a sense of meaning.' These women therefore appeared to develop meaningful lives after an active decision to focus on areas other than becoming a parent.

Despite this, it was not found that developing a new life resulted in full acceptance of infertility. For example, Gonzalez reported 'Despite achievement of a realistic perspective, the inability to conceive and bear a child continued to be a significant factor in the definition of self.' (Gonzalez, 2000). One participant describes this saying:

'Even though that old nagging thought is still back there; it is not the same. So, I don't think it's resolved, but I think it's different.'

Affected intimate relationships

Four studies reported women experiencing difficult relationships with partners or sexual problems (Bergart, 2000; Peddie et al., 2005; Volgsten et al., 2010; Wirtberg et al., 2007). Volgsten et al. (2010) stated ‘Some men and women described how the relationship with their partner had been strengthened in a critical situation. Other men and women experienced strain in the relationship and temporary separations after ending IVF’. The studies gave various reasons for the strain on couples, emphasising financial pressures, poor communication, self-perceptions about being inadequate as women, and the competing demands of treatment and other aspects of life. Again relationship pressures may partly be influenced by the health care system the couple are using and the related financial pressure this brings. One woman in Peddie et al.’s (2005) study explained the financial strain of treatment:

‘not that we grudged the money; I mean you can’t put a price on a child, but it added to the stress of it all.’ (Peddie et al., 2005)

Studies also reported a lack of lust and enjoyment in sexual life.

‘[O]ur sexual relationship really got strained ... [We] didn’t make love for a year, we had sex. We had this copulation where I was making a baby, and he was indulging me. (Bergart, 2000)

It seems that being infertile and going through infertility treatment changed sexual experiences for these women, with the hope for pregnancy being dominant. One woman explained that ‘*the enjoyment just ebbed away*’ (Wirtberg et al., 2007), suggesting that additional pressure took the pleasure away from sexual relationships. Wirtberg et al. (2007) found that ‘as many as nine reported that their sexual life and sexual desire was lost forever’

suggesting that this was a problem for couples long beyond infertility treatment. The women in this study also felt they would have benefited from counselling in this area.

Discussion

This review synthesised recent qualitative data about women's experiences of infertility in order to facilitate greater understanding of such experiences in a Western context. Women reported that being unable to have children made them feel different to their peers and led to a sense of isolation. Withdrawal from social contexts where there was a risk of being judged by others, or of being in the company of those who highlight a sense of difference, has been proposed by Goffman (1963) to be a means of coping with stigma. Women also reported an altered sense of self, feeling that they were not complete or adequate as women. Infertility as a challenge to women's core female identity has also been identified in previous literature (Cousineau and Domar, 2007). The above themes led to additional strain on couples, with intimacy becoming less enjoyable and solely focused on achieving pregnancy. Again this is in keeping with literature which has found infertility to affect relationships and sexual satisfaction (Bergart, 2000).

The expectation that women will become mothers is reflected in Carter and McGoldrick's (1989) family lifecycle model. The women's experience of stigma suggests that cultural expectations and Western pronatalist ideologies remain focused on traditional family patterns, despite many families no longer following the stages set out in Carter and McGoldrick's model (Ulrich & Weatherall, 2000). Further, the women's feelings of being different became particularly pertinent at certain life stages suggesting that social norms continue to follow set patterns of childbearing. The family lifecycle model therefore appears to remain useful in understanding the experiences of infertile women.

Maintaining hope throughout treatment and beyond was described by many women, with menstruation bringing disappointment. This supports Glazer and Cooper (1988) who proposed that infertility is associated with recurrent losses rather than representing a single life crisis. As hope for pregnancy persisted throughout most of their adult lives, the desire to become a mother became a central focus. This seemed to have an adverse effect on other areas of life which were effectively put on hold. Contrary to Eugster & Vingerhoets (1999) and Weaver (1997), who proposed that life grief is a central factor for infertile women, these women's accounts suggested that losses were responded to by focusing attention on hope for a positive future outcome, allowing grief to be delayed.

Although women described active attempts to find meaning in other areas of life, maintaining hope for pregnancy and delaying grieving for their fertility appeared to prevent full acceptance of their infertility. It also seems that some women felt they were not offered the opportunity, or did not feel comfortable enough, to discuss the emotional impact of infertility and its treatment with professionals. More sensitive care and a more holistic ending to treatment may have provided greater opportunities to process their emotional reactions.

Diversity in the quality of included studies may, however, have impacted on findings. As only half of the studies employed second raters in the analysis stage, study interpretations may be affected by researcher's own assumptions and values. Some differences between studies therefore may reflect individual interpretations rather than participants reporting different experiences. Further, only two studies mentioned achieving data saturation, therefore some themes may have been more dominant in the synthesis had larger sample sizes been recruited and data saturation reached. The context in which the data was collected must also be considered, yet was rarely mentioned in study reports. Study findings may therefore be a product of the healthcare systems and specific contexts they took place in. For example,

the women's experiences may have been affected by how well services are set up to offer emotional support and whether such support is available once active treatment, and potentially payment for this, have ceased.

Limitations

There are a number of limitations to the current review which should be addressed. Firstly, word limit requirements of journals may have meant that some methodological information was not provided despite it being considered in the research project. Profiles of strengths and weaknesses may not therefore accurately reflect the research project itself, and rather reflect quality of the written report within such restraints (Sandelowski and Barroso, 2002).

Secondly, there may be a sampling bias in these studies as women may have been more motivated to engage in the research if they had particularly strong feelings about their experience of infertility. All women included had sought infertility treatment and may differ from those who do not pursue this route. The voices of those unable to pay for infertility treatment in countries where this is required were also missing. In order to ensure a certain level of quality, only published studies were included in the current review. However, there may be a publication bias meaning other relevant studies may have been excluded. Finally, controversy exists over whether the integration of studies using a diverse range of methodological approaches is appropriate. While a number of strong themes did emerge from the studies, there were a number of subtleties that were difficult to capture.

The samples of women included in the studies tended to be from across a wide age range. Although this is beneficial as it allows for accounts of those initially coming to terms with their infertility and those looking back over their life experiences, specific experiences

associated with different stages may have been missed. Future studies should therefore aim to examine the experience of infertility at distinct life stages and take account of the particular health conditions that may be the cause of the women's infertility.

Implications

The current review suggests that care should be taken to consider the individual and emotional aspects of infertility as well as the medical. Staff need to remain alert to the sensitive nature of infertility in their consultations with women, and training for staff on the psychosocial impact to infertility, as well as skills development in normalising, active listening and containment, would help them to respond appropriately. Time should be given to assess and discuss how the women are coping and the wider impact of infertility on their lives. Follow-up appointments should also be offered after the end of treatment to provide women with support regarding the longer-term impact of their infertility. Given the challenge that feelings of difference brings to women's sense of self, the provision of social support from other infertile women may be beneficial.

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CHAPTER 2: MAJOR RESEARCH PROJECT

Exploring the short term psychological impact of Mayer-Rokitansky-Kuster-Hauser (MRKH) syndrome for young women; an interpretational phenomenological analysis approach

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Prepared in accordance with guidelines for submission to Psychology & Health (*see appendix 2.1*)

Submitted in part fulfilment of the requirements for the degree of Doctorate in Clinical Psychology (D.Clin.Psy)

Lay summary

Mayer-Rokitansky-Kuster-Hauser (MRKH) syndrome is a medical condition where women are unable to have periods, sexual intercourse, or to have a child. Previous research has shown that MRKH has an emotional, as well as physical, impact. The unique experience of MRKH for young women as they become adults, however, has not been investigated. The aim of the current study was to explore young women's experiences of having MRKH in terms of social and emotional challenges, and the impact of MRKH on their identities. Five women aged 18 to 22 years old were interviewed and asked about their experiences of having MRKH. The results of the interviews found four main themes which explained their experience of MRKH: hindering independence, a sensitivity to difference, managing intimacy and managing threat to identity. These themes showed how difficult it is for young women to discuss their condition with others and that having MRKH affects their confidence and female identity. This study offers new insights into the significant social impact of MRKH as young women become adults and highlights the need for professionals to address this. Running support groups for women of similar ages is also advised.

Abstract

Background: MRKH is a congenital condition which renders women unable to menstruate, carry a child, or have sexual intercourse (Edmonds, 2000). However, there is a scarcity of research regarding the psychological impact of the condition. Diagnosis is made in mid to late adolescence, a time where social relationships and identity are salient, hence there may be specific challenges at this point and through the transition to adulthood.

Aims: The study aimed to explore the impact of MRKH on young women.

Methods: Five women (aged 18 to 22) diagnosed within the past five years took part in a non-directive semi-structured interview. Transcripts were analysed using Interpretative Phenomenological Analysis.

Results: Four themes were identified; hindering independence, a sensitivity to difference, managing intimacy and managing threat to identity. The personal and sensitive nature of MRKH had a significant social impact for the young women's developing identity, autonomy, and negotiation of peer and sexual relationships. Fear of being stigmatised caused them to manage their presentation to others in order to minimise the impact of their diagnosis.

Conclusions: The study offered a unique insight into the social challenges of MRKH in the transition to adulthood. Implications for services and future research directions are indicated.

Key words: MRKH; late adolescence; lived experience; identity; social relationships

Introduction

Mayer-Rokitansky-Kuster-Hauser (MRKH) syndrome is a congenital condition where, although women have normally functioning ovaries, the vagina and uterus do not develop (Edmonds, 2000). The condition, which occurs in around 1 in 5000 women, causes them to be unable to menstruate, participate in sexual intercourse and carry children (Bean, Mazur & Robinson, 2009; Edmonds 2003). Women typically present to services when menstruation has not begun despite normal development of secondary sexual characteristics (Aittomaki, Eroila & Kajanoja 2001; Mungadi, Ahmad, Yunusa, Agwu & Ismail, 2010).

Historically treatment and research regarding MRKH have largely focused on physical aspects and on medical procedures to create a neovagina (Bean et al., 2009). Holt and Slade (2003) found women's experience of such services to be that there was a scarcity of accessible and appropriate information, and that treatment felt impersonal. There is now, however, increasing recognition of the need to support women with the psychological impact of MRKH (Edmonds, 2003, Hughes, Nihoul-Fekete & Thomas, 2007). In keeping with this, Brain et al. (2010) proposed that care should be provided by a multidisciplinary team, involving psychologists as well as medical professionals. The development of more holistic services requires a deeper understanding of the experiences of women with MRKH to ensure that services target areas of importance and provide sensitive support to service users.

In a review of research studies, Bean et al (2009) reported a range of reactions to a diagnosis of MRKH including depression, shock, fear of partner rejection, and feeling different. Women who were diagnosed with MRKH ten years previously indicated scores for mental health disorders that fell between normal and psychiatric ranges (Heller-Boersma, Schmidt & Edmonds, 2009a). The inability to carry a child has been suggested to be one of the most distressing effects of MRKH, and therefore may have a strong impact on women's wellbeing (Bean et al., 2009; Kimberley, Hutson, Southwell & Grover, 2011). Carrying a

child is typically thought to be a key life role for women and the inability to do so has been found to have the potential to challenge their self-esteem and gender identity, and to lead to jealousy and a sense of isolation around peers who have children (Holt & Slade, 2003, Bean et al., 2009).

Although a cognitive behavioural model for MRKH has been developed by Heller-Boersma, Schmidt & Edmonds (2007, 2009b) this was developed on the basis of a quantitative study comparing women with and without MRKH on a number of questionnaire measures of psychological distress and on past quantitative research. Without a qualitative exploration into their experiences, there is limited insight into the unique challenges that MRKH brings, hence the essence of women's experiences may not be captured.

A recent review of qualitative studies investigating women's experiences of infertility identified the following themes from the literature: feeling different, compromised female identity, maintaining hope, sensitivity of professional support, developing a new life and affected intimate relationships (Espie, this volume). However, a downfall of this literature was that the study samples represented a broad age range. Therefore identified themes were likely to reflect only the commonalities across the age span. This prevented the accounts of the particular experiences of women at different life stages from emerging. Further, a need to explore the distinct experiences of specific populations where infertility is an issue was indicated.

In the only qualitative study of women's experiences of MRKH, Holt and Slade (2003) found that the role of time came out as a theme for women diagnosed between two and 22 years previously. It is unsurprising that women's views will change as they adjust or adapt to the condition, but this highlights that one theoretical model is unlikely to explain women's experiences at different stages of their lives. There is therefore a need to study the specific experiences of MRKH at different time points in order to understand the subtleties of its

interactions with various life stages. Given the unfolding nature of key developmental tasks of different life stages, the aspects of MRKH which cause the most distress may change and fluctuate over time.

Diagnosis of MRKH typically occurs in mid to late adolescence, (Aittomaki et al. 2001; Mungadi et al. 2010) hence initial adjustment will likely take place during this transitional stage. This is an important life stage for identity formation and is associated with physical, cognitive and social changes, (Brinthaup & Lipka, 2002; Brown, 2004; Erikson, 1968; Sebastian, Burnett & Blakemore, 2008) therefore MRKH may pose particular challenges in this context.

Perceived gender role inadequacy in the transition to adulthood has been shown to be related to depression and anxiety in the short term, and to predict poor adult adjustment in the longer term (Aubé & Koestner, 1992; O’Heron & Orlofsky, 1990). Further, growing independence from one’s parents and increasing focus on social and sexual relationships during this life stage leads to an emphasis on social acceptance, with much time and effort being dedicated to building peer relationships (Engels & van den Eijnden, 2007; Parker, Rubin, Price & DeRosier, 1995). The challenge that MRKH poses to one’s sense of self, gender identity, and social and sexual relationships may therefore be particularly problematic during the transition to adulthood. Indeed, Heller-Boersma (2006) found that distress is greatest immediately after diagnosis of MRKH and Bean et al. (2009) suggested that this time period is one which may benefit most from psychological intervention. These young women are being asked not only to cope with the transition to adulthood but also to cope with and adjust to a diagnosis of MRKH. It is therefore pertinent to understand the experience of MRKH at this particular stage in order to inform the development of sensitive and timely care, and to help prevent longer term adjustment difficulties.

The current study set out to investigate young women's experiences shortly after being diagnosed with MRKH in order to understand their experiences at this critical stage of development. A qualitative approach, using Interpretative Phenomenological Analysis (IPA, Smith, 1996) to analyse data, was employed in order to explore the young women's lived experiences. IPA deals with significant and emotional life changing experiences and has been found to be an appropriate method for exploring health and illness, sexuality, psychological distress, life transitions and identity (Smith & Osborne, 2003; Smith & Osborne 2008).

Aims

The study aimed to explore young women's experiences of MRKH in order to address the following broad research questions;

- What are young women's experiences of having a diagnosis of MRKH?
- What are the social and emotional challenges faced by young women with a diagnosis of MRKH?
- How do individuals manage a diagnosis of MRKH in relation to their developing identities?

Method

Participants

Five females with MRKH participated, aged 18 to 22 years old. This is within the recommended sample size for Doctorate level research studies employing IPA and enabled data saturation whilst allowing for a detailed interpretative account of each participant's experiences (Smith, Flowers & Larkin, 2009; Smith & Osborne, 2008). Participant information is shown in Table 1, using pseudo names to maintain anonymity. Time since diagnosis was determined by the participants' own reports of when they were diagnosed rather than being based on a formal diagnosis per say. This was due to variable delays

between professionals indicating that the young women had MRKH and having this confirmed by a MRI scan. The participants' perception of the time when they were diagnosed was thought to best represent when it first became salient for them.

The Scottish Index of Multiple Deprivation (SIMD) was used to determine the socio-economic status of participants. The SIMD provides a ranked list of postcodes in Scotland from the most (rank 1) to least (rank 6505) deprived area based on indicators from seven domains: income, employment, health, education, skills and training, housing, and geographic access and crime. None of the participants' post codes were within the 25% most deprived areas in Scotland (rank 1627 and under) and three fell within the 25% least deprived areas (ranked 4879 and above) indicating that study participants were of a middle to high socioeconomic status.

Table 1: Participant information

| Participant pseudo name | Age | Time since diagnosis | SIMD rank |
|--------------------------------|------------|-----------------------------|------------------|
| Susan | 22 | 5 years | 5955 |
| Catherine | 22 | 5 years | 5329 |
| Katy | 22 | 4 ½ years | 5493 |
| Louise | 18 | 1 ½ years | 1687 |
| Fiona | 18 | 2 years | 3946 |

Inclusion and Exclusion Criteria

Inclusion and exclusion criteria were developed with the aim of maintaining a homogeneous sample. Women were invited to participate if they were aged 16 or over and had a formal diagnosis of MRKH at least six months prior to being interviewed. Participants

who were over 25 years old, who had been diagnosed more than five years previously, or who did not speak English as their first language were excluded.

Recruitment

Participants were recruited through a multi-centre genetics study taking place throughout Scotland; *Spectrum of Clinical and Genetic Abnormalities in Women with Congenital Abnormalities of the Female Reproductive Tract*. Potential participants for the genetics study were identified through the Scottish Disorders of Sex Development (SDSD) database; a central register set up to contain a core dataset of patients seen by the SDSD multidisciplinary team.

Women were contacted by their named clinician or local gynaecologist and given an information sheet (Appendix 2.2) about the overall study before being invited to a clinic to discuss any questions or concerns, and to provide written consent (Appendix 2.3). As part of the consent process, potential participants were asked to agree to be interviewed for this study.

Women who met the current study's inclusion and exclusion criteria were then contacted to arrange a suitable time and place to meet to carry out the interview. When there had been a delay of more than two weeks between consenting for the study and being contacted to arrange the interview, a letter and stamped addressed envelope were sent requesting that the women return a reply slip indicating they still wished to participate (Appendix 2.4). Women who had consented to the study within the past two weeks were telephoned to arrange a suitable time and location to meet.

Considerable efforts were made to achieve the final sample size. Problems were identified with the SDSD dataset due to the fact that the number of people recorded on the dataset with a diagnosis of MRKH was below the expected level given the known prevalence

rate of the condition. It also became apparent that some women who had been seen by services were not registered on the database. Further, the contact details available for those on the dataset were at times found to be incorrect. Gynaecology clinic lists were therefore hand searched in order to identify potential participants and regular contact was made with professionals working as part of the SDSD multidisciplinary team throughout the recruitment period to ensure all women who met the study criteria were identified.

Six women initially provided consent, and five agreed to participate at the time of the interviews. One woman did not reply to a letter or a telephone message asking her to contact the researcher to arrange a suitable time to meet, therefore no further contact was made. Another three women were identified who met the study criteria, one from the SDSD database and two from clinic lists. Unfortunately they did not respond to invitations to attend a clinic to discuss the wider study.

Semi-Structured Interview

Non-directive, semi-structured interviews were used to explore the young women's experiences of MRKH. Care was taken at the start of each interview to make it clear to the young women that the researcher was interested in their views as experts on their own lives. A topic guide was used to help structure the dialogue, while the interviewer remained alert to different topics raised by the participants themselves and encouraged the exploration of these.

Considerable thought went into the development of the topic guide, taking guidance from a Clinical Psychologist working in the field, and drawing on past research concerning adolescence, infertility and MRKH. The resulting guide covered the experience of, and feelings in relation to, initial diagnosis, the effect of MRKH on their life in general, experience of talking to peers and family members, sense of identity, experience of intimacy, and concerns for the future (See Appendix 2.5 for topic guide). Additional probe questions

were used to explore participant's views (e.g. Can you tell me more about that? How did that make you feel?).

Due to the sensitive nature of the topics covered, care was taken to ensure that participants felt at ease and that a rapport was developed before discussing the most personal areas. Care was taken to try to avoid asking leading questions.

The interviewer summarised key points made by the participants to check her understanding. These summaries also provided the opportunity for the participants to correct misconceptions and opened up topics for further discussion. At the end of the interviews time was taken to check how the participant was feeling and to ask if there was anything else they would like to discuss. Interviews lasted between 32 and 46 minutes (mean 37 minutes).

Procedure

Interviews were digitally recorded and were carried out in a NHS clinic which was convenient for each participant to travel to. Travel expenses to reimburse the cost of public transport to attend the interview were offered to all participants. When they attended the interview the young women signed a further consent form agreeing to the interview being voice recorded and to quotations being used in published reports (Appendix 2.6). They were also given the opportunity to ask any questions prior to the interview commencing.

Researcher reflexivity

Although IPA aims to generate understanding through participants' perspectives of phenomena, there is an acknowledgement that the researcher brings their own pre-existing beliefs to the process and plays an active role in interpreting the data (Reid, Flowers & Larkin, 2005). Therefore, care was taken to acknowledge the researcher's own experiences

and beliefs, and to consider how these may influence the interpretation of participants' experiences.

The researcher had observed a Clinical Psychologist working therapeutically with women with MRKH. She had also worked clinically with other populations who experience complications with fertility and this had provided insight into the typical challenges faced, and potential reactions to these. As the researcher was at a life stage where she was beginning to think about when she would like to start her own family, she also remained aware of her personal feelings about what being able to carry a child meant to her.

Data Analysis

Interviews were transcribed verbatim by the researcher. Each participant was allocated a pseudo name and other potentially identifying information was anonymised. Transcripts were analysed using IPA, as described by Smith and Osborne (2008). This involved repeated and careful reading of the transcripts whilst listening to the digital recordings. Points of significance were noted line by line. The researcher then re-read the transcripts and noted recurring themes. Emerging themes for each interview were compared and integrated, noting similarities and discrepancies between them. Themes were clustered together and their relationship to each other considered in order to generate super ordinate themes. Capturing the participants' personal experiences of MRKH as experts was the aim of this process. The transition to adulthood provided the main context for the interpretation of the participants' accounts. See Appendix 2.7 for a sample extract of an analysed interview.

In order to ensure a rigorous and transparent process, a reflective journal was kept to record the key decisions made and the reasoning behind these. Identified themes were discussed in research meetings as a means of audit, and two transcripts were analysed independently by a research supervisor to further verify the reliability of the analyses (Elliott,

Fischer & Rennie, 1999; Reid et al., 2005). Ultimately, themes were linked back to quotations from the interviews to ensure they continued to represent the essence of the women's experiences.

Ethical approval

Ethical approval was awarded by the relevant NHS ethics committee, as part of the study *Spectrum of Clinical and Genetic Abnormalities in Women with Congenital Abnormalities of the Female Reproductive Tract* (Appendix 2.8).

Results

Four themes concerning the experience of MRKH in the transition to adulthood emerged from the interviews: (i) hindering independence, (ii) a sensitivity to difference, (iii) managing intimacy and (iv) managing threat to identity; dealing with a disrupted developmental process. See Appendix 2.9 for a table of themes and their main dimensions.

Quotations from the interviews have been selected to help illustrate each theme. For clarity, participant quotations are presented in italics, with pauses in their speech indicated by a series of three dots. Author responses are written in parentheses.

Hindering independence

The women described their diagnosis as being intensely sensitive and personal to them. The enormity of discussing MRKH with others came across throughout the interviews, with the young women expressing feelings of it being a social taboo and a desire to keep their diagnosis private.

‘I just, kind of a private thing that I just didn’t think people would talk about, it’s not really a thing that a lot of people talk about. (Okay) So I just didn’t really want to talk about it myself.’ Fiona

Despite the deeply personal nature of MRKH, and the young women being at a stage when one typically becomes less dependent on one’s parents, participants described their mothers being heavily involved around the time of their diagnosis. Some felt that their mothers took the lead in consultations with professionals; deciding on when to initially attend the GP and asking questions throughout the process. This resulted in participants feeling they had not been at the centre of their care. Susan chose to attend appointments by herself. However, as shown below, she was irritated by her mother’s questioning after appointments. Here she portrays a desire to manage the process on her own, with her mother’s questions being perceived as an intrusion on her privacy.

‘but my mum, you know, every time I go for an appointment she’s always like ‘what’s happened, what’s happened?’, and I’m like ‘it’s got nothing to do with you mum, just leave it.’’ Susan

In addition, Katy felt that the professionals she saw in relation to her diagnosis did not recognise her growing autonomy, challenging her perception of herself as an independent adult. In the example below she shows her frustration about this.

‘I just didn’t like him, (be)cause I asked him questions and he ignored me and ... he just talked to my mum, so I was like ‘I am an adult’’ Katy

Many participants described their mothers’ strong emotional reactions to their diagnosis, with one mother being said to have had a panic attack. Some believed that these

intense reactions were due to their mothers being so involved in the process of diagnosis that they felt it was happening to them first hand. It may also be that, as mothers themselves, they reflected back on their experiences and of what their daughters may miss out on through not being able to carry a child. Catherine also felt that receiving a diagnosis of MRKH caused her mother to be protective of her, talking in terms of a mother-child relationship, rather than a more adult relationship.

'I think (Catherine laughs) it affected my mum probably more. She was quite, like, distressed but that's kind of, you know, finding out there is something wrong with your child I suppose.' Catherine

Another way in which the participants' growing independence appeared to be hindered was in relation to disclosure of their diagnosis to others, with over half of the participants stating that their mothers had told other family members that they had MRKH. In two instances, the participant had not told anyone apart from their mother about their diagnosis, hence the only way others knew was through being told by their mother. Louise felt that she had no power over who knew, with the words 'everybody' and 'absolutely' emphasising a real sense of feeling this had been spiralling out of her control.

'My mum, I didn't really want everybody in my family knowing but my mum told absolutely everybody (Louise laughs).' Louise

MRKH seemed to complicate the ongoing negotiation of the mother-child relationship to a point where it was difficult for the young women to remain in control of their lives. Their wishes to keep their diagnosis private due to the personal nature of MRKH were undermined by their mothers' own beliefs about who should be told, or perhaps their mothers' own need to discuss the diagnosis with others due to their emotional reactions. Below, Fiona describes

such opposing beliefs. Here, her own wishes were overridden by her mother's, resulting in not only her privacy, but also her autonomy as an adult, being compromised.

'I kind of ... I didn't really want them to know (be)cause it's, it was like, my, private kind of thing, but if, my mum said 'well it would be better if they knew', (be)cause it would, like, I'd be awkward in the future trying to tell them, but at the moment I just, didn't really want anybody to know, just kind of wanted to deal with it first and then tell them.' Fiona

A sensitivity to difference

The interviews revealed feelings of being different to others and the situations where such feelings come to the fore. Some participants felt that they could not take part in certain conversations with their peers due to their different experiences, such as discussing menstruation. All women also mentioned being around children or those who were pregnant as being difficult for them, provoking a sense of loss of normality and anticipated future loss. Below, Fiona reflects on being around others with their children, and how adopting her children would compare to being a birth mother.

'it just like makes me feel 'right, I can't have that kind of bond'. But, I know it's going to be my child in the end, but it's not actually really mine. (Mm hmm) So I think that's just, kind of ... daunting to think that (yeah) but, it's just, kind of, hard to, kind of, think 'aw ... they're not actually really going to be my ... children.'

Fiona

The young women's feelings of being different from others and concerns about the future impact of such differences caused them to feel down and hurt. Louise provides an example of this, describing that she experienced a range of emotions which fluctuated daily.

'But it's, it started playing about wi(th) my mind so I was like down one day, and I'd be happy the next, and then just really depressed and, I don't know, it just really took a lot from me so it did. It, like, it was bad, it was really bad.' Louise

Such feelings of being different and the subsequent impact on the young women's emotions were in contrast to reports that, because they were not at the life stage where they were thinking about having children, they were not currently particularly affected by MRKH. Rather, they described their present priorities as being those more typically associated with the transition to adulthood, such as developing their careers.

'but at the moment it doesn't affect us really at all, because ... it's just, it's, it's not (Catherine laughs) having a uterus doesn't really affect your day to day life? (Yeah) And you know, we're kind of, you know, early twenties, we're not really up to much which requires a uterus at the moment.' Catherine

Most participants described active attempts to try not to think about their diagnosis which appeared to be a means of allowing them to focus on more current priorities and those which were salient for their peers. Fiona's description below shows her effort to do so, with her attempting to think about anything but her diagnosis.

'I've always kind of just pushed it to the back of my mind, so (mm hmm) it's not really ... the thing I'm always thinking about, it's the last thing I'm thinking about, so I'm, like, concentrating on other things. (mm hmm) So its, I don't think it's really affecting me right now, (be)cause I'm always just pushing it to the back of my mind.' Fiona

It seems that, despite wishing to maintain a focus on current priorities and avoid thinking about their diagnosis, certain social contexts triggered feelings of difference for these young women, leading to them thinking ahead to future difficulties. Being sensitive to difference, they reported using avoidance to help minimise their exposure to such situations. In the example below, Katy shows that the hurt and feelings she experienced in response to feelings of difference was significant enough for her to change her planned career path.

'I mean, I always like, kind eh, just felt quite different and as an outsider and like ... why am I different, why am I the one that can't? And just ... kind eh, a wee bit gutted, but (Katy inhales) so I just took myself out of the situation rather than sorting the situation if that makes sense.' Katy

For Catherine, feeling different from others became a viewpoint from which she interpreted her life. Her reflective comment below show she has a tendency to view herself as categorically different, and that such differences are seen as the cause of the difficulties she experiences. This highlights how pervasive feelings of difference can be at a life stage in which peer acceptance is strongly sought.

'Emm ... I don't know, it's kind of an ir', irrational response to a situation where you are singled out to assume that if you weren't different then, like everything else in your life would be different in some way, or better in some way, but ... I mean you know, there's, there's been moments where you've, I've felt like that because you know, you're down and you're feeling 'oh why has the world chosen me, why?'(Catherine laughs)' Catherine

In addition to participants feeling they did not fit in with others who could carry children, they also felt that those without MRKH were unable to understand their situation.

Although positive relationships with friends and family were described, the participants felt that only those who had MRKH could truly relate to their experiences, as Susan describes below.

'I mean my friends were very supportive but, so that was helpful, but I think because none of my friends obviously have MRKH, eugh, MRKH, it's a bit different, you know, you could talk about it but only to a certain extent, you don't get how they feel about having it as well kind of thing' Susan

Some of the women indicated that they would like to meet or talk to others with MRKH who they may be able to relate to more. Participants felt, however, that previous experiences of talking to women with MRKH on an internet forum had been of limited value as this had tended to be with older women, as shown by Susan below. They expressed a wish to meet or talk to others with MRKH who were of a similar age to them.

'I think ... emm, the websites weren't actually that handy, because at the time I was 17 or 18 and all the people that had been diagnosed were 30s, 40s, they'd adopted, they'd, you know, had surrogate children et cetera, and it wasn't, I didn't feel particularly close to them because of the age gap.' Susan

Managing intimacy

Participants expressed an overall sense that MRKH caused difficulties in partner relationships. Those who were in, or who had experience of, such relationships reported that it was difficult to negotiate when to tell their partners about their diagnosis. They seemed torn between waiting until further into the relationship to tell partners they had MRKH and telling them from the outset.

'it's kind of, (be)cause it's obviously quite personal you know, and if you are only with them for, you know, a month or so because it doesn't work out but, you don't want to tell them too quickly, you don't want to leave it too long either so I think that's a thing I've always had trouble with.' Susan

As Susan describes above, on one hand participants wanted potential partners to know about their diagnosis of MRKH prior to them becoming invested in the relationship, however they also felt that they would rather not share such personal information if it was not to become a long term relationship. Those without experience of such relationships also discussed concerns for the future.

Worries about potentially negative reactions from partners appeared to drive concerns about disclosure. Participants feared that prospective or current partners would want to end their relationships if they knew they had MRKH. In some instances, this was stated despite having found partners to be understanding and supportive. Some participants felt that men may not want to be in a relationship with them because they could not have children. Others, such as Louise, felt that relationships would not last beyond disclosure due to the more current potential difficulties with sexual relationships:

'When I say, 'oh', to a guy, 'oh look I've got this problem', I've, I feel as if they are just going to be like that 'oof no thanks, don't want to go through it with you', do you know what I mean?' Louise

Motivation to tell partners sooner partly related to wishing to avoid awkward situations. Susan, for example, explained that when partners did not know she had MRKH she felt unsure how to respond when they asked about her use of contraception, and embarrassed about needing to use lubrication to participate in sexual intercourse.

Difficulties with negotiating intimacy resulted in two of the participants avoiding becoming involved in romantic and sexual relationships for long enough for this to become an issue. The quotation below shows Katy's active attempts to escape this difficult conversation by not allowing relationships to progress to a more serious level.

'That's something I've always done by pushing people away, so it's just part of, my nature, and I wanted to avoid telling them. So I just pushed them away.' Katy

Two participants had been friends with their current partners prior to entering a more intimate relationship, hence the partner either already knew they had MRKH or the women felt able to trust them from the outset of their relationship. As shown below, Catherine talked about being conscious that this made it easier for her to talk to her partner about having MRKH.

'Well I mean, I wasn't nervous (be)cause I knew him (mm hmm) so that was different. If it had been like a new relationship which I hadn't known him as a friend or anything before it would have been different' Catherine

Likewise, Katy described how her current situation has allowed her to avoid full discussions about MRKH. Again, a sense of relief is apparent in relation to this.

'We've discussed minimal things but he doesn't know, we're not into any discussions about physical things until a wee bit later on in the relationship, because of issues at his end and mine, so ... there's ... it, I've worked out quite well here that I don't need to tell him for a wee bit yet (laughs)' Katy

Catherine described feeling more confident when entering intimate relationships than other participants. For her, having had previous sexual experiences where partners had not been able to tell she had MRKH allowed her to feel more comfortable about future intimacy. As she states below, knowing that others would not necessarily be aware of physical differences allowed her to feel more in control of who she told and when.

'(be)cause of when I found out, (be)cause I was like, you know, I'd had boyfriends and (inhales) and you know, I had already kind of like, you know, had, not, not sex with guys but um, I'd, you know, I'd done stuff with guys before I found out. So I hadn't seen, I hadn't, I think maybe if I'd found out earlier I would have like kind of avoided going into those sort of situations more? Because, I thought, I would think that I was way different from other guys, (be)cause I'd already kind of seen other guys and stuff before I found out I thought well obviously they didn't, you know, they didn't notice (Catherine laughs). So I guess I was more confident because of that.' Catherine

Managing threat to identity; dealing with a disrupted developmental process

Participants spoke about MRKH having affected their developing sense of self in terms of their gender identity and self-esteem. They described a compromised female identity, feeling they were not complete as women because they were unable to participate in tasks typically associated with being a woman. For Louise, not having periods caused her to feel only part female as she had not completed the steps she saw as being involved in becoming a woman. Indeed, people's gender identity usually becomes more consolidated following such experiences.

'basically what I mean was, in a way, like a girl goes through the stages of puberty, period, growth in the, the chest, and all that, and when I didn't have my

period I was kind of like, when I got told that I wouldn't never have a period, I was li(ke), I felt as if, that's the stages a women goes through, like every month, having a period and getting the sore stomach and the cramp and, I just felt as if I'm never going to have that, so I part of thought that ... part woman, part of my woman side had gone. ' Louise

Child bearing was talked about as a fundamental role of a woman, hence the inability to carry a child was seen to be the most distressing aspect of their condition and posed a particular challenge to the young women's core female identity. As indicated by Susan below, they felt that, by being unable to fulfil this central role, they were by definition incomplete as females.

'I think I feel a lot more ... kind of, boyish, if that makes sense, (right) like I'm not completely feminine since, (mm hmm) I suppose ... when you go down to the basics the whole point of being a female is that you provide, you know, have children and put the population up and stuff. So it does make me feel slightly less feminine' Susan

Susan also explained how she responded to these challenges to her female identity:

'I think ... I kind of try harder to be girly sometimes, so I'll wear a lot of makeup (okay) and skirts and stuff, whereas before I didn't bother. Then when I got the diagnosis I was like well I'll just make sure everyone knows I'm a girl (Susan laughs).' Susan

This was in contrast to descriptions of herself as being a tomboy growing up, indicating that she had previously not been as concerned about appearing typically female. Hence, her

diagnosis of MRKH seemed to challenge her sense of self as female, leading to her being more sensitive about her identity as a woman and more concerned about how others saw her. She therefore made more active attempts to present herself as being typically female to others.

The majority of participants also described feeling less confident than they had been prior to their diagnosis, both in general and in relation to developing intimate relationships. Participants appeared to feel less worthy of such relationships and believed that partners would be at a disadvantage to be with them rather than another woman. Louise's lowered self-esteem and concerns about withholding typical experiences from her partner resulted in her ending their relationship.

'But I think it was because it did knock confidence off me I was just like 'no, I want you eh have someone better, I want you be able to have a sex life, I don't want you to be able t(o), I don't want to be, like, not go through the stages that every man goes through, I don't want eh take that away from you.' Louise

In light of their lowered self-esteem, it was implied in the interviews that participants felt self-conscious about their diagnosis. They talked of thinking carefully about who to discuss having MRKH with and were concerned about people's reactions to this. As Catherine demonstrates, other people's perceptions of them were particularly salient and fear of potential negative responses prohibited them from talking about their diagnosis due to the effect this may have on them personally. This is in keeping with young adults being more conscious of, and more effected by, other people's perceptions and suggested that the women saw MRKH as a potentially stigmatising condition.

'see I think that's why I don't tell people if I don't need to because I don't find it's an issue for me but people make things issues and then it can become an issue for you, it is kind of like a feedback thing.' Catherine

With concerns about what their peers think of them and lowered self-esteem, participants were worried that others would 'find out' about them having MRKH. Some felt that others would be able to detect that they had MRKH without being told. In the following quotation Katy shows that she was concerned about how she looks to others, believing that they would notice differences in her interactions in terms of lack of eye contact. This was in contrast to her actual presentation however, in which no obvious differences in the amount of eye contact were noted. Her concerns seemed to reflect her underlying emotions and inner worries about how she appeared to others, rather than her actual outer appearance.

'I, c', can't even look at people ... and discuss it, you know, emm, like, you've probably noticed that my eyes are going everywhere. I can't, I can't even look at you' Katy

Two participants talked of having integrated MRKH into their overall sense of self and reported that it no longer affected them. In both instances, however, contradictions were noted throughout the interviews with these participants also describing active attempts to suppress thoughts and feelings about MRKH. This alludes to a sense of managing how they present themselves to others in order to preserve their previous sense of self and other's opinions of them.

The women were keen to discuss their experiences in the interviews, however the social nature of the interaction caused them to manage how they presented themselves in order to

preserve their self-esteem. Some participants appeared to make light of the impact of MRKH throughout the interviews which seemed to be a means of managing how they were perceived and to help protect against their emotional reactions. Susan in particular often used humour at emotionally salient points of the interview. In the example below, she talks about her experience of talking to older women who have MRKH. Humour is used to buffer her concern about not having a partner or children in the future, controlling the emotions she portrayed to the researcher.

'they'd obviously come out the other end and had children and husbands and stuff like that so (right, okay) that was helpful to know that I wasn't (yeah) going to die lonely with a herd of cats (Susan laughs).' Susan

Susan also described using humour in other situations as a way of managing people's reactions to her diagnosis. It seemed that she wished to indicate to others how they were to respond in order to protect herself from being faced with their emotional reactions, and from potential negative reactions having an effect on her sense of self.

'Emm ... I think ... some people, because I joked about it I think some of them were a bit shocked, and that irritated me a little bit because it's not really anything to do with them ... it's me, you know, it's not them that's going through it, so, if I want to make jokes about it then they should just accept that and shut up (Susan laughs).' Susan

Discussion

This paper has presented the lived experiences of five young women who had received a diagnosis of MRKH within the past five years. Interviews were interpreted in the context of the transition to adulthood, resulting in four emerging themes; hindering

independence, a sensitivity to difference, managing intimacy and managing threat to identity; dealing with a disrupted developmental process.

As found by previous research, the women reported that their diagnosis caused them to feel down, different from others and to fear partner rejection (Bean et al., 2009). Being unable to carry a child was considered to be the most distressing aspect of having MRKH, again supporting previous literature (Bean et al., 2009; Kimberley et al., 2011). Bean et al.'s (2009) and Holt and Slade's (2003) findings that MRKH poses a challenge to women's core female identity and self-esteem, seeing themselves as being incomplete as women because they are unable to fulfil what they consider to be their fundamental role of childbearing, were also corroborated in the current study. This may be particularly problematic in the transition to adulthood where identity formation and reaching reproductive potential are key developmental tasks, and where gender role inadequacy has been found to relate to poor adjustment in later life (Aubé & Koestner, 1992; Brindthaupt & Lipka, 2002; Erikson, 1967; Marcia, 1966; O'Heron & Orlofsky, 1990).

In addition to supporting key concepts from previous literature, the themes identified interlink to offer a unique insight into the subtleties of the complex experience of MRKH in the transition to adulthood. At a stage where peer acceptance is highly sought and one is increasingly affected by other's perceptions (Engels & van den Eijnden, 2007; Parker et al, 1995; Seiffge-Krenke, 1998), the young women's sensitivity to the social impact of their diagnosis was apparent in their concerns about others knowing or 'finding out' they had MRKH, and the effect that others' reactions may have on their sense of self. Despite the pervasive impact of MRKH on their personal identities and emotional well-being, the women went to huge efforts to keep their diagnosis private, to present themselves as 'typically' female, and to avoid situations which may single them out. This is in keeping with Goffman's

(1959) notion of a hidden stigma where one tries to manage information about one's self in order to avoid being devalued by society.

The women's attempts to manage the disclosure of their condition and to maintain a sense of privacy about this personal and sensitive area were, however, complicated by their mothers taking the lead in telling family members about their diagnosis and, in some instances, professionals directing information to their parents. Not only did this undermine their desire for privacy, but it also hindered the re-negotiation of the parent-child relationship towards independence that typically occurs at this life stage (Bee, 1975, Engels & van den Eijnden, 2007; Larson & Richards, 1991). This may be problematic in the longer term as independent decision making in mid to late adolescence has been found to relate to better adjustment in adulthood (Smetana, Campione-Barr & Daddis, 2004).

When the young women were in situations where they had to discuss their MRKH with others, they again described using strategies to manage the discourse. This was evident in the interviews themselves, with some participants using humour as a means of managing the conversation. Managing the disclosure of MRKH in intimate relationships was also described as a particular challenge for these young women at a life stage where romantic and sexual relationships typically begin to develop, and hence where individuals need to negotiate the particulars of this new type of interpersonal situation (Collins, Welsh & Furman, 2009). The intimate nature of these relationships meant they wanted to discuss MRKH with partners from the outset to avoid awkward situations, but at the same time they wanted to wait until they had developed a sense of trust in their partners to avoid potential rejection or becoming subject of gossip. For some, this negotiation was so difficult that they avoided developing romantic relationships. This is of concern as having such relationships is commonly associated with feelings of positive self-worth (Connolly & Konarski 1994, Harter 1999). Their coping strategies may therefore have led to greater distress. Avoidance of wider social

situations may have similarly been ineffective in the longer term, by reducing the opportunity to build relationships with their peer group, and hence may have heightened their feelings of difference.

The interviews therefore showed that, although the young women felt that being unable to carry a child would have a significant effect on them in the future, during the transition to adulthood it was the challenges that MRKH posed in social settings which cause the most distress. Although Heller-Boersma et al.'s (2007, 2009b) cognitive behavioural model of MRKH addresses some of the self-appraisals evident in these young women's reports, and to an extent their coping strategies in terms of appearing as a 'proper' woman to others, the social context which largely influenced these is not considered. The model's focus on failure to adequately process the diagnosis did not come across as a core feature of these young women's experiences, suggesting it does not adequately described the lived experience of MRKH in the transition to adulthood. Further, the model's assumption that idiosyncratic negative appraisals are partly due to cognitive immaturity at the time of diagnosis is not supported by a review of infertility literature which showed women of various ages to report similar self beliefs (Espie, this volume).

Strengths and weaknesses

Although within the sample size recommended for doctorate level research employing IPA, the study sample was lower than initially intended. Despite there being a national database proposed to hold information on all those diagnosed with a disorder of sex development, the number of individuals with MRKH was lower than would be expected given the population of Scotland and reported prevalence rates of the condition. This suggests that a large proportion of women with MRKH are not in contact with services. This study therefore represents a particular sub-group of young women who remain engaged with

services. The views and experiences of those who have chosen not to have contact with services may be different, hence they are an important group to include in future studies.

Secondly, only one interview was conducted with each participant. Although all were offered the opportunity to raise additional areas they wished to discuss at the end of the interview, participants had no opportunity to broach any further dimensions that came forward after reflecting on the interview. This paper therefore explores their experiences as described at a single moment in time. Finally, although the researcher reflected on her impact on the process throughout, her interpretations may have been influenced by how important having a child in the future is to her, and by how she imagines she would feel if this was not possible.

In terms of study strengths, considering the participants' experiences in their developmental context has allowed for a detailed understanding of MRKH in the transition to adulthood. This experience has not been captured by quantitative research or in studies looking at women from a broad age range and at different life stages. Those who took part were keen to discuss their experiences and the richness of data collected suggests that they felt comfortable to talk openly and honestly despite the personal nature of issues covered. It may be that the researcher herself being a young woman and having no clinical agenda facilitated this process. This being said, however, the impact of dealing with MRKH in social settings was evident during the interviews, with the participants containing their emotional reactions and managing how they presented themselves to the interviewer.

Exploring topics as they were brought to the interview by participants also enabled them to discuss their experiences without being restricted by the researcher's own agenda or beliefs. Reaching a point of data saturation further validated that the study has explored a full range of issues which help to explain young women's experiences of MRKH. Every attempt was also made to ensure a transparent and rigorous process through having two transcripts

analysed by a second researcher as a means of audit, discussing emerging themes in research meetings, and keeping a reflective diary.

Implications

This study suggests that psychologically informed care for women with MRKH needs to consider adjustment within a developmental context. Support groups specifically for young women with MRKH may be beneficial in creating a safe environment for them to talk to others of a similar age to increase a sense of peer group acceptance and to help reduce feelings of inadequacy and difference. Group based psychological interventions have indeed been found to be effective for reducing psychological distress in women with MRKH (Heller-Boersma et al., 2007; Weijenborg & terKuile, 2000), however the active components of these interventions have not been considered. It may be that social support alone contributes to the effectiveness of these groups.

Professionals should also consider how best to manage their communication with young women and their families, rather than assume a straight forward parent-child interaction in which the parent takes the lead. Given the growing need for autonomy in the transition to adulthood, removing the young women from the centre of their care is potentially damaging to their development. Efforts should therefore be made to keep the young women informed and to allow them to have an active role in decision making. Given the strong emotional reactions on the part of the women's mothers, and their need to discuss their daughters' diagnosis with others, a support network for parents may also be beneficial.

Further, psychologically informed support in the short term after diagnosis which focuses on identity, social competence and adaptive coping strategies is advised.

Future Directions

Further qualitative work exploring the specific experiences of MRKH at different life stages would be warranted to develop holistic longer term care needs. The development of psychologically informed interventions targeting the specific effects of MRKH during the transition to adulthood is required in order to provide timely care and avoid potential longer term negative consequences.

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CHAPTER 3: ADVANCED PRACTICE 1: REFLECTIVE CRITICAL ACCOUNT

Case complexity, a tick box exercise? The role of clinical psychologists in physical health settings

(Abstract only)

Abstract

Introduction: This reflection focuses on the thoughts and feelings I experienced during a meeting to discuss the role of clinical psychologists in physical health settings. The recent government HEAT access target for psychological therapies and The Matrix, A Guide to Delivering Evidence-Based Psychological Therapies in Scotland, are discussed in order to set the context of my reflection. I discuss the recommendations for clinical psychologists to work with the complex cases which require intensive intervention. Gibbs' (1988) model was used to guide my reflection, however not wanting to be restricted by this model, I also followed my natural train of thought, as promoted in psychoanalysis.

Reflection: I first describe the meeting that my reflection stemmed from, detailing the thoughts and feelings I experienced. I then draw on experiences of my clinical work in physical health settings to help me evaluate and make sense of these reactions. From my initial concern that as a health aligned trainee I had little opportunity to work with 'complex' cases, I come to recognise the true value of my training experiences and the developments in my clinical practice. I also recognise the importance of the wider role of a clinical psychologist in teaching and providing consultancy and supervision for other professionals, particularly when working in settings which largely adopt a medical model.

Reflective review: This reflection has allowed me to recognise the need for clinical psychologists to be involved in commissioning services and the value of reflection for personal learning.

CHAPTER 4: ADVANCED PRACTICE 2: REFLECTIVE CRITICAL ACCOUNT

Providing consultancy in medical settings: challenge or opportunity?

(Abstract only)

Abstract

Introduction: The current reflection stems from my thoughts and feelings in response to a conversation with a nursing colleague regarding her management of, and reaction to, a difficult situation. The extended roles of a clinical psychologist in terms of providing supervision, consultancy and training, as promoted in *The Matrix, A Guide to Developing Evidence-Based Psychological Therapies in Scotland* and *Applied Psychology and Psychologists in Scotland*, are discussed in order to provide the context for the reflection. As previously found helpful, Gibb's (1988) reflective model was used to guide the reflection, however I also followed my natural flow of thought in order to freely explore this experience.

Reflection: I describe the conversation which triggered my reflections and consider my thoughts and feelings in relation to three elements of this discussion, namely the practical, emotional and personal levels. I then consider why I found this situation challenging, highlighting the need to develop my skills in providing consultancy regarding staff reactions to client presentations, and recognising the importance of formalising supervision arrangements with future supervisees. Finally, I consider the subsequent development of my competence in offering advice and training on emotional responses to professional work. My reflections are concluded by a recognition of these extended roles being an opportunity to be grasped in order to promote psychologically informed care in medical settings.

Reflective review: The current reflection has also encouraged me to think more fully of my own emotional reactions to my clinical work and to be mindful of these as potential barriers to effective intervention.

APPENDICES

Appendix 1: Systematic Review

Appendix 1.1: Instructions for authors - Health Psychology Review

Instructions for authors

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Acknowledgements

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|-------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
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| Design | <ul style="list-style-type: none"> - Method/design apparent - Above consistent with research intent - Rationale given - Data collection strategy apparent - Data collection strategy appropriate |
| Sampling strategy | <ul style="list-style-type: none"> - Sample and sampling method explained - Above justified - Above appropriate |
| Analysis | <ul style="list-style-type: none"> - Analytic approach explained - Above appropriate - More than one researcher involved if appropriate - Participant involvement in analysis - Evidence of data saturation/discussion or rationale if did not |
| Interpretation | <ul style="list-style-type: none"> - Context described - Context taken account of in interpretation - Clear audit trail (sufficient so others can follow decision trail) - Data used to support interpretation |
| Reflexivity | <ul style="list-style-type: none"> - Researcher reflexivity demonstrated |
| Ethical dimensions | <ul style="list-style-type: none"> - Ethical approval granted - Documentation of how consent was managed - Documentation of how confidentiality and anonymity were managed |
| Relevance and transferability | <ul style="list-style-type: none"> - Relevance and transferability evidence - Links to theories and literature - Limitations/weaknesses outlines - Outlines further directions for research |

Appendix 2: Major Research Project

Appendix 2.1: Instructions for authors – Psychology and Health

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- Papers are accepted only in English. British spelling and punctuation is preferred. Please use single quotation marks, except where ‘a quotation is “within” a quotation’. A typical article will not exceed 30 pages (inclusive of tables/references/figure captions/footnotes/endnotes), with a font size of 12 in New Times Roman, and all margins should be at least 2.5cm. Papers that greatly exceed this will be critically reviewed with respect to length. Authors should include a word count with their manuscript. Manuscripts should be double-spaced throughout (including tables and references), and each page should be numbered consecutively.
- Manuscripts should be compiled in the following order: title page; abstract; keywords; main text; acknowledgments; appendixes (as appropriate); references; table(s) with caption(s) (on individual pages); figure caption(s) (as a list).

- Abstracts of no more than 200 words are required for all papers submitted.
- Each paper should have three to six keywords or phrases . These will be used for indexing and data retrieval, and so where appropriate we recommend using standard MeSH terms (the terms used for indexing articles for MEDLINE).
- Search engine optimization (SEO) is a means of making your article more visible to anyone who might be looking for it. Please consult our guidance [here](#) .
- All the authors of a paper should include their full names, affiliations, postal addresses, telephone numbers and email addresses on the cover page of the manuscript. One author should be identified as the corresponding author. The affiliations of all named co-authors should be the affiliation where the research was conducted. If any of the named co-authors moves affiliation during the peer review process, the new affiliation can be given as a footnote. Please note that no changes to affiliation can be made after the article is accepted.
- For all manuscripts non-discriminatory language is mandatory. Sexist or racist terms should not be used.
- Authors must adhere to SI units . Units are not italicised.
- When using a word which is or is asserted to be a proprietary term or trade mark, authors must use the symbol ® or TM.
- Reports of statistical tests should include an indication of effect size whenever possible. Reports of randomised controlled trials should state any registration details of the trial and should follow CONSORT guidelines where relevant (see Moher, D., Schulz, K.F. & Altman, D.G. for the CONSORT group, 2001. The CONSORT statement: Revised recommendations for improving the quality of reports of parallel-group randomized trials. *Annals of Internal Medicine*, 134, 657-662).

2. Style guidelines

- Description of the Journal's article style , quick guide
- Description of the Journal's reference style , quick guide
- Guide to using mathematical symbols and equations
- Word templates are available for this journal. If you are not able to use the template via the links or if you have any other template queries, please contact authortemplate@tandf.co.uk

3. Figures

- It is in the author's interest to provide the highest quality figure format possible. **Please be sure that all imported scanned material is scanned at the appropriate resolution: 1200 dpi for line art, 600 dpi for grayscale and 300 dpi for colour.**
- Figures must be saved separate to text. Please do not embed figures in the paper file.
- Files should be saved as one of the following formats: TIFF (tagged image file format), PostScript or EPS (encapsulated PostScript), and should contain all the necessary font information and the source file of the application (e.g. CorelDraw/Mac, CorelDraw/PC).
- All figures must be numbered in the order in which they appear in the paper (e.g. Figure 1, Figure 2). In multi-part figures, each part should be labelled (e.g. Figure 1(a), Figure 1(b)).
- Figure captions must be saved separately, as part of the file containing the complete text of the paper, and numbered correspondingly.

- The filename for a graphic should be descriptive of the graphic, e.g. Figure1, Figure2a.

4. Colour

The Journal has no free colour pages within its annual page allowance. Authors of accepted papers who propose publishing figures in colour in the print version should consult Taylor & Francis at proof stage to agree a financial contribution to colour reproduction costs. Figures that appear in black-and-white in the print edition of the Journal will appear in colour in the online edition, assuming colour originals are supplied.

5. Reproduction of copyright material

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For further information and FAQs, please see
<http://journalauthors.tandf.co.uk/preparation/permission.asp>

Copies of permission letters should be sent with the manuscript upon submission to the editors.

- [Copyright permission letter template](#)

6. Supplementary online material

Authors are welcome to submit animations, movie files, sound files or any additional information for online publication.

- [Information about supplementary online material](#)

Appendix 2.2: Information sheet

Dr R McGowan
Specialist Registrar Clinical Genetics
Dept of Clinical Genetics,
Ferguson-Smith Centre
Yorkhill, Glasgow, G3 8SJ
Tel: 0141 201 0808 Fax: 0141 201 0361
e-mail: ruth.mcgowan@ggc.scot.nhs.uk

PARTICIPANT INFORMATION SHEET

Title of Project: Study Investigating Women with Variations in the Development of the Reproductive System

Name of Principal Investigator: Dr Ruth McGowan

Dear

You are invited to take part in a research Study Investigating Women with Variations in the Development of the Reproductive System.

This project is based at the Yorkhill Hospital and is supported by The Scottish Genital Anomalies Network (SGAN).

Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully. Talk to others if you wish.

Part 1 of this information sheet tells you the purpose of this study and what will happen if you take part

Part 2 of this information sheet gives you more detailed information about the conduct of this study

Please feel free to ask us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part.

We thank you for taking the time to read this.
Sincerely

Dr Ruth McGowan
SpR Clinical Genetics, RHSC, Yorkhill

PART 1

What is the aim of the study?

The aim of this study is to find new mechanisms involved in the development of the female reproductive system in women. We hope to find new genes and proteins involved. This may allow us to work out how variations in known genes and proteins have their effect and offer women with variations in development of the reproductive tract more effective genetic counselling about risks of recurrence.

We also wish to document changes that have occurred in other body systems such as bone, kidneys, etc.

We would like to investigate the psychological impact of having a variation in the development of the reproductive system using standardised questionnaires.

We also wish to interview a smaller group of women about their experiences of having abnormal development of the female reproductive tract. We want to find out their views as experts on their own lives.

Why have I been chosen?

We are approaching individuals identified through the SGAN register or by their local gynaecologist who have abnormal development of the female reproductive tract.

Why is this study being done?

Within the past twenty years, we have started to understand some of the processes that control the development of the reproductive system.

These processes are controlled by **proteins**, such as growth factors in the testis or ovary, or hormones in the blood. These proteins are encoded by **genes**, which are regions of each person's DNA blueprint or genetic code found on the **chromosomes**. Like DNA, RNA is made up of a long chain of building blocks. As in DNA, the sequence of building blocks allows RNA to encode genetic information. RNA molecules play an active role in cells, controlling gene activity and carrying the genetic information that directs the synthesis of proteins.

By analysing the proteins/hormones, genes (DNA and RNA) and chromosomes of women with variations in reproductive development in more detail we hope to find new factors involved in reproductive development and help us to improve the diagnosis and management of individuals and families with reproductive system disorders.

What will happen to me if I am interested in taking part?

If you agree to participate in the study, we would obtain a small blood sample (one to two teaspoonfuls). This DNA (and RNA) will be stored and analysed for small changes in chromosome structure that may not be picked up on routine clinical tests. We hope this will uncover areas where there are specific genes related to the development of the female reproductive system. Some of these genes are currently known; others we hope to identify in the future. Wherever possible, we will take these blood samples at the same time that routine clinical tests are being done.

It is already known that women with variations in development of the reproductive tract can have variations in other areas (kidney, bone and hormone levels.) If basic investigations of these areas has not been done we ask for your consent for us to arrange a renal ultrasound, and X-Ray of the spine and to collect urine and blood samples for hormone tests. This blood sample will be collected at the same time as blood is taken for DNA extraction.

Usually only single blood samples are requested, although we may request an extra sample from you if we find something we would like to investigate in more detail, and we may ask at a later date for additional blood samples to look at the hormone or protein pattern. We also may request a small sample of tissue to look at the DNA, RNA and protein pattern within the tissue.

With your consent, we plan to keep the samples for other relevant studies in the future, if there is enough sample left.

You will be invited to complete questionnaires that ask about quality of life and other factors. The answers will be reviewed by a Clinical Psychologist to see if variation in the development of the reproductive tract has any influence over general wellbeing. Further follow-up by Clinical Psychology can be arranged as is necessary.

Your participation in this study will only require an additional clinic visit if you are not currently being seen at a clinic, although if you would like us to tell you any of our findings this may require slightly longer outpatients appointments, or a special appointment arranged with myself.

We would also like to speak to a smaller number of you about your experiences of getting a diagnosis. It will only be possible to interview a smaller number of women because this will be more in-depth and will last between 45 minutes to an hour. The aim is to find out your experiences as an expert in your own life. We will ask questions about getting your diagnosis, talking to others about your diagnosis, how you feel about yourself, how others have reacted to you and dealing with intimacy. However we are also interested in other issues you might have.

If you are happy to take part, and are satisfied with the explanations, you will be asked to sign a consent form. If you take part in the interview you will be asked to sign a form consenting to the interview being recorded. Interviews do not have to be recorded if you do not want them to be. You will be given a copy of the signed information sheet and consent/assent forms to keep for your records.

What are the alternatives for diagnosis or treatment?

There is now sufficient evidence that the tests proposed in this study would be considered in part of a standard of good clinical care. No routine tests or treatment would be withheld on the basis of whether you decide to participate in this study or not.

What are the potential risks and disadvantages of taking part?

No significant physical risk can be foreseen. We will try to take any blood samples at the same time that routine clinical tests are being performed. If a blood sample is being taken specifically for this study there will be some discomfort from the needle prick. If at any time you feel that the actual or perceived distress is too great, please don't hesitate to tell your research investigator.

As part of this study, you will be exposed to radiation from the bone X-rays. X-rays are used to form pictures of your body and provide more information to help your doctor. The level of dose from the X-rays that you will receive is 0.5mSv compared to the dose of a chest X-ray, one chest X-ray being the equivalent of 0.03mSv. The dose is equivalent to what you would normally be exposed to in the UK in 3 month's natural background radiation. The main risk of exposure to X-rays is that a cancer may occur many years after the exposure. The risk from the amount of radiation you receive from these diagnostic tests is considered to be **very low**.

Investigations may uncover some degree of abnormality and this could cause distress if participants are not prepared for this. Genetic studies may reveal an abnormality that could have implications for

the individual and their family members. It is possible that an abnormality identified may not be related to the study. The importance or implications of our genetic findings may not be known for some time. Further information related to genetic tests is provided in Part 2. Analysis of blood and urine samples may identify abnormal hormone levels and X-rays could reveal bony abnormalities.

Any results will first be communicated to the individual by their main clinician. Subsequently this clinician will seek the advice from the multidisciplinary team (which includes several specialists) in discussing the results in more detail. Specific opportunity will be provided for counselling about any genetic abnormalities identified with a clinical geneticist. Women involved in the study will be given contact details of two clinicians who they can contact at any point during the study if they have any queries.

Effort will be made to ensure that you feel as comfortable as possible as we understand that we are covering sensitive issue, however should you begin to feel distressed, you will be offered to opportunity to discuss this with a clinical psychologist.

What are the possible benefits of taking part?

There may be no immediate benefit to you taking part in this study, but our research will help us to understand the development of the reproductive tract in women better. This information may help other women with conditions affecting the reproductive system ensuring that they have appropriate investigations and when diagnosed. There is a small possibility, however, that we might find out more information related to your specific condition. You will have the option of how the results are given to you. You can choose whether we give you an aggregate result (for example, 3 out of 20 participants had interesting findings). You also have the **option of not being informed about results**.

We will have the results of the “diagnostic” tests i.e. X-ray, renal ultrasound and hormone tests fairly quickly but the genetic tests may take up to 1 year as results need to be validated.

Understanding more about people’s experiences should help services provide sensitive support and develop suitable interventions where required.

What happens when the research study stops?

We would like to keep any samples and relevant information related to this study for future research analysis beyond this current project. Undertaking further research work will be dependent on future funding.

What will happen if I don’t want to carry on with the study?

Your participation in this study is voluntary. If you decide now or at a later stage that you do not wish to participate in this research project you are free to withdraw at any time, without giving any reason. Your medical treatment or legal rights will not be affected. Stored samples or any data collected will be destroyed if you wish.

What if there is a problem?

Any complaint about the way you have been dealt with during the study or any possible harm you might suffer will be addressed. The detailed information on this is given in Part 2.

Will my taking part in the study be kept confidential?

Yes. All information about your participation will be kept confidential. The details are included in Part 2.

This completes Part 1 of the Information Sheet. If you are interested in participating, please continue to read the additional information in Part 2 before making any decision.

PART 2 – Detailed information

What if relevant new information becomes available?

Your participation in this research study will not interfere with you being offered any relevant new tests or management approaches that might become available in the future.

Will my taking part in this study be kept confidential?

All information that is collected about you during the course of the study will be kept strictly confidential and in compliance with the Data Protection Act 1998. Any paper records will be stored securely and any electronic records bearing “identifiers” (e.g., names) will be password protected.

Only the researchers and a representative of the Research Ethics Committee will have access to the data collected during the study. You will be asked to agree that appropriate sections of your medical notes may be looked at by responsible individuals from the researchers where it is relevant to this study.

Notification of your General Practitioner (GP) and specialists involved in your care

We will ask you for your permission to inform your GP and other specialists about your participation in this study. In some situations it will be important to discuss potentially significant research findings with other members of the clinical and research team, and other specialists involved in your care.

If you show any signs of distress your GP will be notified and a referral can be made to a clinical psychologist.

What will happen to any samples I give?

Any samples provided will be processed and stored in the Clinical Genetics Laboratory at Yorkhill (DNA extraction and chromosomes), or in the Clinical Biochemistry Laboratory (serum/plasma/urine).

Everyone taking part in this study will be given a study number (“coded”) and samples kept in the research laboratory will be handled on a day-to-day basis using this number. Only the research team involved in this study will have access to these samples, and only the principal investigator and designated individuals will be able to identify whose sample it is.

We like to view any samples provided as “gifts” for the purposes of research. It is hoped that the samples will be kept for analysis by the research team on an ongoing basis, but it is quite possible that the samples will be used up during the course of this study. We also ask your permission to distribute coded samples and anonymous information to collaborators involved in related research within the UK, within the European Economic Area, and to countries outside of Europe. No names will be provided and you are free to choose whether you agree to this or not.

We do not anticipate that this work will lead to any outcome of commercial significance and you would not benefit financially if this research does lead to the development of a new treatment or medical test.

Information gathered in interviews and completed questionnaires will be kept in a locked filing cabinet for 10 years. Transcribed interviews will also be held on an encrypted computer for 10 years.

Will any genetic tests be done?

The aim of this study is to identify new genes involved in development of the female reproductive system, or variations in known genes involved in these systems.

We will not be able to tell you the genes we plan to look at when you start the study but may be able to as the study progresses if a certain gene(s) is highlighted as being important in reproductive

development. We will also ask you whether we can analyse additional factors that may be relevant in reproductive biology as they are identified. You may allow us to do this without contacting you further, or you may request we contact you to get your agreement each time.

Although this is a research study, we are willing to share any potential positive findings with you. If you would prefer not to know the results of any of our investigations then that is your choice. We recommend checking any positive findings in a separate sample, and – if you wish – trying to confirm our findings with further tests. However, it is possible that the significance of our findings is unclear at present. Finally, it is not currently possible to analyse all genes and proteins in their entirety, so we might overlook subtle changes in our studies.

In a few instances, we are also trying to analyse many genes at once using a specially designed “gene chip” or similar technology. This new technology might be useful in the future for obtaining more rapid results for patients. If we are interested in including your DNA, RNA or protein in our study to test this new technology we will discuss it with you.

What will happen to the results of the research study?

Our aim is that the results of these studies will be presented to the scientific community and important advances will be published in peer-reviewed scientific and medical journals. This is the best way of having our work reviewed by experts in the field, and allowing other doctors and patients to benefit from our findings.

You **will not be identified** in any report or publication unless you have consented to the release of such information. Clinical data, biochemical data (e.g. hormone results) and radiological (e.g. X-rays, scans), histological or whole-organ images (e.g. pictures of the gonad tissue), external photographs may be used.

Direct quotations from interviews may be used in published reports, however it will not be possible to identify individuals from this as any identifiers will be removed and all information will be anonymous.

What if there is a problem?

If you are worried about any aspect of this study please discuss this in the first instance with the chief investigator (Dr Ruth McGowan, 0141 201 0808) who will do her best to answer your questions. There will also be a local specialist for you to contact and their details are at the end of this information sheet. If the problems are not resolved, or you wish to comment in any other way, please contact the Research and Development Office at the Tennent Institute, 38 Church Street, Glasgow (Kirsty Theron, 0141 211 6372).

This project has been approved by an independent Research Ethics Committee who believe that it is of minimal risk to you. However, research can carry unforeseen risks and we want you to be informed of your rights in the unlikely event that any harm should occur as a result of taking part in this study.

This research is covered by a no-fault compensation scheme, which may apply in the event of any significant harm resulting to you from involvement in the study. Under this scheme it would not be necessary for you to prove fault. You also have the right to claim damages in a court of law. This would require that you prove fault on the Hospital/Institute and/or any manufacturer involved.

Who is organizing and funding the research?

This study is being organized by Dr Ruth McGowan (Specialist Registrar in Clinical Genetics, RHSC) with other co-investigators including: Dr John Tolmie (Clinical Genetics, RHSC), Dr Wayne Lam (Clinical Geneticist, Western General Hospital, Edinburgh), Dr Faisal Ahmed (RHSC, Glasgow), Dr Miriam Deeney (Consultant Gynaecologist, Glasgow Royal Infirmary), Dr Edward Tobias (University of Glasgow), Dr Susie Logan (Consultant in Sexual and Reproductive Healthcare, NHS Grampian), Dr Rebecca Crawford (Clinical Psychologist, Glasgow Royal Infirmary), Ms Carolyn Espie (Trainee Clinical Psychologist, University of Glasgow), Professor Andrew Jahoda (University of Glasgow). Funding for the study shall be sought from organisations such as SGAN and other sources.

Who has reviewed the study?

This study has been reviewed by the West of Scotland 4 Research Ethics Committee.

If you are interested in taking part you will be given a copy of this information sheet and any signed consent form to keep.

Please feel free to contact us if you have any further questions, or if your care moves to a different hospital or clinic.

We thank you for considering taking part and for taking the time to read this information sheet.

Contact:

Dr Ruth McGowan

Specialist Registrar Clinical Genetics
Ferguson-Smith Centre, Yorkhill

Glasgow, G3 8SJ

(Tel: 0141 201 0808 Fax: 0141 0361)

e-mail: ruth.mcgowan@ggc.scot.nhs.uk

Dr Susie Logan MD (comm),MRCOG,MFSRH

Consultant in Sexual and Reproductive Medicine
Square 13, 13 Golden Square

Aberdeen, AB10 1RH (Tel 01224 642711)

Dr Wayne Lam

Consultant Clinical Geneticist
Western General Hospital

Edinburgh, EH14 1JF

(Tel 0131 651 1013)

Appendix 2.3: Consent form

Centre: RHSC, Yorkhill

Study Number:

Patient Identification Number for this study:

CONSENT FORM FOR PARTICIPANTS IN RESEARCH STUDIES

Title of Project: Study Investigating Women with Variations in the Development of the Reproductive System

Name of Principal Investigator: Dr Ruth McGowan

Please initial box
Yes No

- | | | |
|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------------|--------------------------|
| 1. I confirm that I have read and understand the information sheet entitled, <u>“Study Investigating Women with Variations in the Development of the Reproductive System.”</u> for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily by _____. | <input type="checkbox"/> | <input type="checkbox"/> |
| 2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected. | <input type="checkbox"/> | <input type="checkbox"/> |
| 3. I understand that relevant sections of any of my Medical Notes and data collected during the study may be looked at by employees from Regulatory Authorities or from Royal Hospital for Sick Children, Glasgow where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records. | <input type="checkbox"/> | <input type="checkbox"/> |
| 4. I agree to my GP to be informed of my participation in the study. | <input type="checkbox"/> | <input type="checkbox"/> |
| 5. I agree to take part in the above study. | <input type="checkbox"/> | <input type="checkbox"/> |

Name

Date of Birth

Hospital number

Date

Signature

Name of Person taking consent
(if different from Investigator)

Date

Signature

Investigator

Date

Signature

One copy for participant; one copy for R&D section in the Medical Notes; original to be kept in the PI's site file

Centre: RHSC, Yorkhill
Study Number:
Patient Identification Number for this study:

CONSENT FORM FOR PARTICIPANTS IN RESEARCH STUDIES

Title of Project: Study Investigating Women with Variations in the Development of the Reproductive System

Name of Principal Investigator: Dr Ruth McGowan

| | Please initial box | |
|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------------|--------------------------|
| | Yes | No |
| 6a. I agree to have a blood/saliva sample taken for DNA extraction. | <input type="checkbox"/> | <input type="checkbox"/> |
| 6b. I agree to my DNA sample being stored for future analysis. | <input type="checkbox"/> | <input type="checkbox"/> |
| 6c. I agree to my DNA sample being analysed on a research basis for other genes that may be related to reproductive biology: | | |
| I) without further notification | <input type="checkbox"/> | <input type="checkbox"/> |
| OR | | |
| II) I would like to be informed prior to additional analysis (if not in clinic, I would prefer telephone/letter/email [delete]) | <input type="checkbox"/> | <input type="checkbox"/> |
| 6d. I agree to my DNA being used to analyse multiple genes with the "Gene Chip" | <input type="checkbox"/> | <input type="checkbox"/> |
| 6e. I agree to DNA/chromosome analysis for small rearrangements, deletions or duplications | <input type="checkbox"/> | <input type="checkbox"/> |
| 7a. I would like to be informed of the results of any positive research findings. | <input type="checkbox"/> | <input type="checkbox"/> |
| 7b. If yes (7a), I would like to receive any results in an aggregate format (for example, 3 out of 20 people assessed were found to have interesting results) | <input type="checkbox"/> | <input type="checkbox"/> |
| 7c. If yes (7a), I would like to receive results specific to myself (please note, these are research findings and do <u>not</u> represent a clinically approved test) | <input type="checkbox"/> | <input type="checkbox"/> |

Name

Date of Birth

Hospital number

Date

Signature

Investigator

Date

Signature

One copy for participant; one copy for R&D section in the Medical Notes; original to be kept in the PI's site file

Centre: RHSC, Yorkhill
Study Number:
Patient Identification Number for this study:

CONSENT FORM FOR PARTICIPANTS IN RESEARCH STUDIES

Title of Project: Study Investigating Women with Variations in the Development of the Reproductive System

Name of Principal Investigator: Dr Ruth McGowan

Please initial box
Yes No

- | | | |
|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------------|--------------------------|
| 8a. I agree to have a blood sample(s) taken for serum/plasma extraction, stored and analysed for hormones and proteins that may be related to reproductive biology. | <input type="checkbox"/> | <input type="checkbox"/> |
| 9a. I agree to have a urine sample(s) taken, stored and analysed for hormones and proteins that may be related to reproductive biology. | <input type="checkbox"/> | <input type="checkbox"/> |
| 10a. I agree to my samples (for example, DNA, serum/plasma) being kept and used in future ethically approved research projects. | <input type="checkbox"/> | <input type="checkbox"/> |
| 11a. I agree to other radiological investigations being carried out to look for any abnormality in the renal system and with the spine, including: | | |
| I. X-rays of the spine | <input type="checkbox"/> | <input type="checkbox"/> |
| II. Renal ultrasound | <input type="checkbox"/> | <input type="checkbox"/> |
| 12a. I agree to my clinical information including the use of external photographs and research results being used in scientific presentations and journal publications. No names or specific identifiers will be used. | <input type="checkbox"/> | <input type="checkbox"/> |

Name

Date of Birth

Hospital number

Date

Signature

Investigator

Date

Signature

One copy for participant; one copy for R&D section in the Medical Notes; original to be kept in the PI's site file

Centre: RHSC, Yorkhill
Study Number:
Patient Identification Number for this study:

CONSENT FORM FOR PARTICIPANTS IN RESEARCH STUDIES

Title of Project: Study Investigating Women with Variations in the Development of the Reproductive System

Name of Principal Investigator: Dr Ruth McGowan

Please initial box
Yes No

13a. I agree to have a blood/saliva sample taken for RNA extraction.

☐ ☐

13b. I agree to my RNA sample being stored for future analysis.

☐ ☐

13c. I agree to my RNA sample being analysed on a research basis for other genes that may be related to reproductive biology:

I) without further notification

☐ ☐

OR

II) I would like to be informed prior to additional analysis
(if not in clinic, I would prefer telephone/letter/email [delete])

☐ ☐

14a. I agree to my tissue being taken to analyse genes/protein patterns

☐ ☐

14b. I agree to my tissue being stored for later analysis

☐ ☐

Name

Date of Birth

Hospital number

Date

Signature

Investigator

Date

Signature

One copy for participant; one copy for R&D section in the Medical Notes; original to be kept in the PI's site file

Centre: RHSC, Yorkhill
Study Number:
Patient Identification Number for this study:

CONSENT FORM FOR PARTICIPANTS IN RESEARCH STUDIES

Title of Project: Study Investigating Women with Variations in the Development
of the Reproductive System

Name of Principal Investigator: Dr Ruth McGowan

Please initial box

Yes No

14a. I agree to complete questionnaires about general psychological well-being
and other factors ☐ ☐

14b. I agree to the answers being reviewed by a Clinical Psychologist with expertise in working with women
with variations in the development of the reproductive tract ☐ ☐

14c. I agree to be contacted by telephone or letter by Clinical Psychology after completing the
questionnaires if required ☐ ☐

15. I agree to taking part in an interview concerning my experience of having a diagnosis ☐ ☐

(There will still be an opportunity to request an appointment with Clinical Psychology even if this is declined at
this stage)

Name

Date of Birth

Hospital number

Date

Signature

Investigator

Date

Signature

One copy for participant; one copy for R&D section in the Medical Notes; original to be kept in the PI's site file

Appendix 2.4 Invitation letter and reply slip for interview

Carolyn Espie
Trainee Clinical Psychologist
Department of Mental Health and Wellbeing
Gartnavel Royal Hospital
1055 Great Western Road, Glasgow, G12 0XH
e-mail: c.espie.1@research.gla.ac.uk

Dr R McGowan
Specialist Registrar Clinical Genetics
Department of Clinical Genetics
Ferguson-Smith Centre
Yorkhill, Glasgow, G3 8SJ
Tel: 0141 201 0808
email: ruth.mcgowan@ggc.scot.nhs.uk

Title of Project: Study Investigating Women with Variations in the Development of the Reproductive System

Name of Principal Investigator: Dr Ruth McGowan

Dear

Thank you for consenting to taking part in an interview regarding your experiences of your diagnosis as part of the above study.

I have enclosed the study information sheet to remind you of what is involved in this interview. Areas relevant to this interview have been underlined. If you are still interested in taking part, please return a completed contact details form in the envelope provided. You will then be contacted to arrange a suitable time to attend for interview.

Please feel free to ask us if there is anything that is not clear or if you would like more information.

We thank you for taking the time to read this.

Sincerely

Carolyn Espie
Trainee Clinical Psychologist
Co-investigator for research study

Contact Details Form

Please complete this form and return it in the stamped addressed envelope. You will then be contacted to arrange a suitable time to participate,

Surname:

First name(s):

Address:

Contact number:

Appendix 2.5: Topic Guide

Topic Guide

Introduction

- *I am extremely grateful for you coming along today. I know you will have seen various medics since your diagnosis and will have had medical treatment, however I am really interested in hearing about your experiences and what having MRKH has meant for you. This area has not been talked about as much and it would be really helpful for us to learn more about this. So today I am keen to learn from your experiences and about what has been important to you.*

Context

- *I thought it might be helpful to start off by finding out about you and your life in general and wondered whether you could tell me a bit about you, those who are close to you and what you enjoy doing.*

Diagnosis

- *It has been really helpful to hear about your life and what is important to you. I would be really interested to hear about what happened when you first noticed anything different?*
 - *What made you concerned that something may be wrong?*
 - *What did you do when you became concerned/noticed this?*
 - *Who was the first person you spoke to about it?* (link back to those people previously mentioned)
- *What was your experience of being given a diagnosis of MRKH?* (Information given, by whom, understanding, how felt)

Affect on life

- *It's interesting to hear about what happened when you first had concerns and about when you were initially diagnosed. I wonder how would feel that having MRKH has affected you or your life?*

(if multiple aspects are noted these will be addressed in detail one by one)

Talking to others

- *I've heard about when you first learned of your diagnosis and how it has affected you. I am also curious about your experiences of telling people about MRKH?*

If don't tell people – What do you think they would say or think about you if you told them?

(Relate to those people already mentioned and to general family and peers)

- *How, if at all, would you say MRKH affected things with your family and friends?*
- And how, if at all, would you say it affects you current relationships?

Identity

- *I wonder what your diagnosis, and all these experiences you have been telling me about, mean to you as a person?*
- How does having MRKH make you feel about yourself?
- How do you feel about other people's reactions to MRKH?

Intimacy

- *It's been really helpful to hear about your experiences of MRKH in relation to yourself and to others. I wonder if you could tell me about how your experience of intimate relationships may have been affected by MRKH?*
- How have you discussed your diagnosis with partners/ how do you feel about discussing your diagnosis with future partners?

Future

- *Everything we have discussed has been so helpful to help me understand you experiences of having MRKH. I am also interested to hear whether you anticipate that MRKH will affect you in the future?*

Other areas

- *I am keen to make sure that we have as full as understanding as possible about living with MRKH. Are there other areas which we have not yet discussed which are important in your experiences of having MRKH?*

Appendix 2.6 Consent form – recording and use of quotations

Centre: RHSC, Yorkhill

Study Number:

Patient Identification Number for this study:

CONSENT FORM FOR PARTICIPANTS IN RESEARCH STUDIES

**Title of Project: Study Investigating Women with Variations in the
Development of the Reproductive System**

Name of Principal Investigator: Dr Ruth McGowan

I would like to tape record this meeting with you. Anything that you say will be kept private and I will be the only one who will listen to the tape. If you do not want me to record the meeting that is okay, I will write down the things you say instead.

Please initial box

Yes No

1. I am happy for this interview to be tape-recorded.

☐ ☐

2. I am happy for anything I say to be used in the final report (it will not be possible to identify you from what you say).

☐ ☐

Name

Date of Birth

Participant Number

Date

Signature

Name of Person taking consent
(if different from Investigator)

Date

Signature

Investigator

Date

Signature

One copy for participant; one copy for R&D section in the Medical Notes; original to be kept in the PI's site file

Appendix 2.7: Sample analysed interview extract

Interviewer's notes

*Altered confidence
Interest other sex
Partner at time*

Louise: it, it stopped me, like I was a confident girl, I would go out I'd do the flirting and have a good laugh and, it's mostly guy friends I have and I was with a boy when I got told all about it

CE: mm hmm

*Ended relationship
Low self-worth
Worthless
Confidence in
relationships*

Louise: and that kind eh split us up, like I had split up wae him because I didn't feel as if he'd want to be with me anymore and, because I couldn't do anything, and I think it's really, it did, it took a lot off, eh, me, like relationship wise as well. Still to this day I find it hard to even get in a relationship because it, I don't think I'm ready for it yet

CE: okay

Difficult to talk about

Louise: tae tell the person my problem.

CE: mm hmm

*Concern about
partner reactions
will end relationship*

Louise: When I say, 'oh', to a guy, 'oh look I've got this problem', I've, I feel as if they are just going to be like that 'ooft no thanks, don't want to go - - through it with you', do you know what I mean

CE: okay

*Knocked confidence
- relationship
Mixed emotions
Hard to deal with*

Louise: so I was kind of like, relationship wise it did knock me back. Em, my confidence. But it's, it started playing about wae my mind so I was like down one day and I'd be happy the next and then just really depressed and, I don't know, it just really took a lot from me so it did. It, like, it was bad, it was really bad.

CE: Okay. And you were talking a wee bit there about telling other people

Louise: Mm hmm

CE: about your diagnosis and you were saying that you've, that you eh, kind of, worry about partners

Louise: mm hmm

CE: and telling them. What is it that you think you are worried might happen if you did tell a partner?

*Previous positive
partner reaction
Knocked confidence
Not good enough
Feel unfair on partner/
holding back*

Louise: well, as I said I was going wae a boy, .. when I got told about my diagnosis and he was just totally fine with it, he was like 'listen, I'm not with you just because of the sex part of it, I'm not with you because of that' he says 'I'll help you through it'. But I think it was because it did knock confidence off me I was just like 'no, I want you eh have someone better, I want you be able to have a sex life, I don't want you to be able to, I don't want to be, like, not go through the stages that every man goes through, I don't want eh take that away from you'. So when I do like, go eh, like I'm, I was seeing a boy a few month ago and when I was going to tell him it was just, I was like, I couldn't, and I was like 'no'. Don't think, I think it's to do

Need trust
Personal/ don't want others to know with trust and could I trust a boy not to run about telling everybody about it because it's really personal thing to me I don('t), I don't want *everybody* knowing about it

CE: sure

Those close to know **Louise:** it's mostly just family that know and my, one of my friends

CE: mm

Hard to talk about
Worry about reactions
- end relationship **Louise:** so, I, I just, I do find it hard tae .. opening up about it. But I think what would happen like, if I did tell somebody that, they'd be like 'oh no, I don't want to go wae you'

CE: okay

End as can't have sex
Worry re reactions
- difficult **Louise:** like 'I can't have a sex life with you' and then go. So I do find it hard.

CE: Mm hmm. But you were saying you did tell the, the person you were with

Louise: yeah

CE: yeah, m, yeah.

Effect of lowered confidence on relationship **Louise:** I ruined that myself (laughing) because I didn't feel confident enough and I was with a boy for a year and a half

CE: mm hmm

Previous positive reaction **Louise:** and he didn't bother, he was like 'I don't care, I'll help you through it. If we need to do what we need to do then that's what we need to do' kind of thing, and then if we were sitting talking about having kids and stuff and [name of boyfriend] really wanted a ch, child, so I've, I kind a took that away from him as well so I was kind of hurt by that and,. But then, when I told him about not being able to carry he was like 'we could get somebody else to carry the baby, we can adopt, we can' and he'd be saying so many things and I just think I didn't feel, I just felt as if 'you should be able to have a normal life and just not have to worry about me constantly'

CE: mm

Confidence affected relationship worthless **Louise:** So I think that's why I kind of finished it and stuff, coz I, my confidence, it just totally went, I just felt as if I was worthless.

CE: Okay, okay. And in what way, do you think, you fe(el), you feel or felt wor, worthless?

Unable to have sex **Louise:** ... Em, I don't know, I just felt as if, like, because I couldn't have a sex life, like most women *do*

CE: mm hmm

Can't have child,sex
Not complete woman **Louise:** So I felt as if because I couldn't do that and (be)cause I couldn't, like, have children, that, I just felt, I didn't feel part women, if you

CE: can you

Range of emotions
Worthless
Unfair on partner

Louise: known what I mean. I don't know, I, it was, it was weird like, the way I felt, like a totally different feeling running through me every day. So I think it was, just weird for me, like I just felt worthless in a way, like I couldn't give my boyfriend the best, or couldn't have a sex life with him

CE: mm hmm

Difficult in
relationships
Hard consequences
in relationships
Hurt

Louise: so I found that quite hard for him and me because, even though I did love him, I loved him to pieces, like, everybody says 'you don't know what love is when you're younger', like I did really love him, my mum noticed that. When we finished I think my mum knew it was because of my problem and my diagnosis and stuff. I think she realised, she's hurt

CE: yeah

Hold partner back
Not normal

Louise: like, she doesn't, she wants to let him have a normal life, so.

Appendix 2.8: Ethical approval letters

**West of Scotland REC 4
West of Scotland Research Ethics Service**

Ground floor, Tennent Institute
Western Infirmary
38 Church Street
GLASGOW
G11 6NT
e-mail: evelyn.macfadyen@ggc.scot.nhs.uk
Telephone: 0141-211-1722
Facsimile: 0141-211-1847



22 September 2009

Dr Ruth McGowan
SpR Clinical Genetics
Royal Hospital for Sick Children
Dalnair Street
Glasgow
G3 8SJ

Dear Dr McGowan

| | |
|------------------------------|----------------------------------------------------------------------------------------------------------------------|
| REC reference number: | 09/S0704/43 |
| Protocol number: | 2 |
| Study Title: | Spectrum of Clinical & Genetic Abnormalities In Women With Congenital Abnormalities Of The Female Reproductive Tract |

Thank you for your letter of 4 September 2009, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

Confirmation of Ethical Opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation, as revised, subject to the conditions specified below.

Ethical Review of Research Sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

The Committee has not yet been notified of the outcome of any site-specific assessment (SSA) for the non-NHS research site(s) taking part in this study. The favourable opinion does not therefore apply to any non-NHS site at present. I will write to you again as soon as one Research Ethics Committee has notified the outcome of a SSA. In the meantime no study procedures should be initiated at non-NHS sites.

Continued...../

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22 September 2009

Letter to Dr R McGowan, Royal Hospital for Sick Children...../

Conditions of the Favourable Opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

For NHS research sites only, management permission for research ("R&D approval") should be obtained from the relevant care organisation(s) in accordance with NHS research governance arrangements. Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at <http://www.rdforum.nhs.uk>. *Where the only involvement of the NHS organisation is as a Participant Identification Centre, management permission for research is not required but the R&D office should be notified of the study. Guidance should be sought from the R&D office where necessary.*

Sponsors are not required to notify the Committee of approvals from host organisations.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Approved Documents

The final list of documents reviewed and approved by the Committee is as follows:

| Document | Version | Date |
|---------------------------------------------|----------------|------------------|
| Covering Letter | - | 11 May 2009 |
| Investigator CV | - | - |
| Research Study Dose and Risk Assessment | - | 16 June 2009 |
| Letter from Sponsor | - | 17 June 2009 |
| REC application | - | - |
| Protocol | 2 | - |
| Participant Information Sheet | 2 | - |
| Participant Consent Form | 2 | - |
| Response to Request for Further Information | - | 4 September 2009 |

Statement of Compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

Continued...../

22 September 2009

Letter to Dr R McGowan, Royal Hospital for Sick Children...../

After Ethical Review

Now that you have completed the application process please visit the National Research Ethics Service website > After Review

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

The attached document "*After ethical review – guidance for researchers*" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Progress and safety reports
- Notifying the end of the study

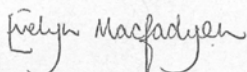
The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

We would also like to inform you that we consult regularly with stakeholders to improve our service. If you would like to join our Reference Group please email referencegroup@nres.npsa.nhs.uk.

09/S0704/43

Please quote this number on all correspondence

Yours sincerely



for **Dr Brian Neilly**
Chair

Enclosures: "After ethical review – guidance for researchers"

Copy to: Ms Kirsty Theron, R&D Office, Tennant Institute, Western Infirmary

WoSRES

West of Scotland Research Ethics Service



**Greater Glasgow
and Clyde**

West of Scotland REC 4

Ground Floor, Tennent Building
Western Infirmary
38 Church Street
Glasgow
G11 6NT
www.nhsggc.org.uk

Dr Ruth McGowan
SpR Clinical Genetics
Royal Hospital for Sick Children
Yorkhill
Dalnair Street
Glasgow
G3 8SJ

Date 19 September 2011
Direct line 0141-211-1722
Fax 0141-211-1847
e-mail evelyn.jackson@ggc.scot.nhs.uk

Dear Dr McGowan

| | |
|--------------------------|---------------------------------------------------------------------------------------------------------------------------------|
| Study title: | Spectrum of Clinical & Genetic Abnormalities In Women With Congenital Abnormalities Of The Female Reproductive Tract |
| REC reference: | 09/S0704/43 |
| Amendment number: | AM03 |
| Amendment date: | 29 August 2011 |

The above amendment was reviewed at a meeting held on 1 July 2011 when it was decided that the Committee had no objections to the following amendments to the above study:

Change 1 - The Committee have no objection to the proposed addition of four collaborators as follows – Dr E Tobias, Dr G Tydeman, Dr R Crawford and Carolyn Espie.

Change 2 - The Committee have no objection to the proposed change that women recruited to the study will be offered the opportunity to complete standardised and validated questionnaires, including the Hospital Anxiety and Depression Score (HADS) as a method of screening for distress associated with congenital Mullerian abnormalities.

Ethical opinion

The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

Approved documents

The documents reviewed and approved at the meeting were:

| Document | Version | Date |
|----------------------------------------------|---------|----------------|
| Notice of Substantial Amendment (non-CTIMPs) | AM03 | 29 August 2011 |

Membership of the Committee

The members of the Committee who took part in the review are listed on the attached sheet.

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www.nhsggc.org.uk

WoSRES

West of Scotland Research Ethics Service

Dr Ruth McGowan
SpR Clinical Genetics
Royal Hospital for Sick Children
Dalnair Street
Glasgow
G3 8SJ

West of Scotland REC 4

Ground Floor, Tennent Building
Western Infirmary
36 Church Street
Glasgow
G11 6NT
www.nhs.uk/ggc.org.uk

Date 27 October 2011
Direct line 0141-211-1722
Fax 0141-211-1847
e-mail evelyn.jackson@ggc.scot.nhs.uk

Dear Dr McGowan

| | |
|--------------------------|---------------------------------------------------------------------------------------------------------------------------------|
| Study title: | Spectrum of Clinical & Genetic Abnormalities In Women With Congenital Abnormalities Of The Female Reproductive Tract |
| REC reference: | 09/S0704/43 |
| Amendment number: | AM01/1 |
| Amendment date: | 29 August 2011 |

Thank you for submitting the above amendment, which was received on 13 September 2011. It is noted that this is a modification of an amendment previously rejected by the Committee (our letter of 18 July 2011 refers).

The modified amendment was reviewed by the Sub-Committee in correspondence. A list of the members who took part in the review is attached.

Ethical opinion

I am pleased to confirm that the Committee has given a favourable ethical opinion of the modified amendment on the basis described in the notice of amendment form and supporting documentation.

The approved amendments are as follows:

1. Increase number of study participants from 30 to 70.
2. Collection of additional amount of blood (3-5mls) for RNA studies.
3. Collection of a small tissue sample when women are having surgery, or skin biopsy using local anaesthetic if surgery is not planned.

Approved documents

The documents reviewed and approved are:

| Document | Version | Date |
|----------------------------------------------|---------|----------------|
| Participant Consent Form | 3 | 18 April 2011 |
| Participant Information Sheet | 3 | 09 May 2011 |
| Protocol | 2 | 28 August 2011 |
| Modified Amendment | - | 29 August 2011 |
| Participant Consent Form: For tape recording | 1 | 13 June 2011 |

R&D approval

All investigators and research collaborators in the NHS should notify the R&D office for the relevant NHS care organisation of this amendment and check whether it affects R&D approval of the research.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

| | |
|--------------|------------------------------------------------|
| 09/S0704/43: | Please quote this number on all correspondence |
|--------------|------------------------------------------------|

Yours sincerely



Dr Ken James
Vice-Chair

Enclosures: List of names and professions of members who took part in the review

Copy to: R&D Office, Tennent Building, Western Infirmary
Ms Pamela Shand, NHS Research Scotland Co-ordinating Centre

Research & Development
R&D Management Office
1st Floor, Tennent Institute
Western Infirmary
GLASGOW, G11 6NT



Our Ref: MB/LR
Enquiries to: Dr Michael Barber
Direct Line: 0141 211 8548
e-mail: Michael.Barber@ggc.scot.nhs.uk

15th March 2010

Dr Ruth McGowan
SpR in Clinical Genetics
Ferguson Smith Centre
Royal Hospital for Sick Children
Dalnair Street
Glasgow
G3 8SJ

R&D Management Approval

Dear Dr McGowan

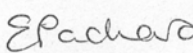

R&D Reference: GN09MG277
GG&C Site: Yorkhill Hospital
Chief Investigator: Dr Ruth McGowan
Project Title: Spectrum of Clinical & Genetic Abnormalities In Women With Congenital Abnormalities of the Female Reproductive Tract
Protocol Ref: Version 2 detailed in REC letter 22/09/09.

I am pleased to confirm that Greater Glasgow & Clyde Health Board is now able to grant **Management Approval** for the above study.
As a condition of this approval the following information is required during the lifespan of the project:

1. SAES/SUSARS – If the study is a **Clinical Trial** as defined by the Medicines for Human Use Clinical Trial Regulations, 2004 (CTIMP only)
2. Recruitment Numbers on a quarterly basis (not required for commercial trials)
3. Any change of Staff working on the project named on the ethics form
4. Change of CI
5. Amendments – Protocol/CRF etc
6. Notification of when the Trial / study has ended
7. Final Report
8. Copies of Publications & Abstracts

Please add this approval to your study file as this letter may be subject to audit and monitoring.

Yours sincerely


 Dr Michael Barber
Research Co-ordinator

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www.nhsggc.org.uk

Cc: NRSPCC, R&D Office, NHS Aberdeen

Queen's Medical Research Institute
47 Little France Crescent, Edinburgh, EH16 4TJ

DEN/JK/approval

25 February 2010

Dr Wayne Lam
Clinical Genetics Service
Molecular Medicine Building
Western General Hospital
Crewe Road
EH4 2XU



Research & Development
Room E1.12
Tel: 0131 242 3330
Fax: 0131 242 3343
Email:
R&DOffice@luht.scot.nhs.uk

Director:
Professor David E Newby

Dear Dr Lam,

Lothian R&D Project No: 2009/W/GEN/02

Title of Research: Spectrum of Clinical & Genetic Abnormalities in Women With Congenital Abnormalities of the Female Reproductive Tract

MREC No: 09/S0704/43

LREC No: N/A

CTA No: N/A

Eudract: N/A

PIS: Version 1 dated May 2009

Consent: Version 1 dated May 2009

Protocol No: no version number or date

I am pleased to inform you that this study has been approved for NHS Lothian and you may proceed with your research, subject to the conditions below. This letter provides Site Specific approval for NHS Lothian.

Please note that the NHS Lothian R&D Office must be informed if there are any changes to the study such as amendments to the protocol, recruitment, funding, personnel or resource input required of NHS Lothian.

Substantial amendments to the protocol will require approval from the ethics committee which approved your study.

Please inform this office when recruitment has closed and when the study has been completed.

I wish you every success with your study.

Yours sincerely

A handwritten signature in black ink, appearing to read 'David Newby', written over a horizontal line.

Professor David E Newby
R&D Director

enc Research Governance Certificate ☒ (to be signed and returned)
Tissue Policy (if applicable) ☒

cc Dr Ruth McGowan, Ferguson-Smith Centre, Yorkhill Hospital, Glasgow, G3 8SJ
Stewart Morgan, NHS Research Scotland
Labs Co-ordinator, Microbiology Office, RIE
Caroline Brydon, Radiology

Research and Development

Foresterhill House Annexe
Foresterhill
Aberdeen
AB25 2ZB



Dr Susie Logan
Consultant in Sexual and
Reproductive Health
Square 13
13 Golden Square
Aberdeen
AB10 1RH

Date 19/02/10
Our Ref 2009OG010

Enquiries to
Extension 51121
Direct Line 01224 551121

Dear Dr Logan

Management Approval for Non-Commercial Research

MREC Ref: 09/S0704/43
NRS Ref: NRS09/GY04
Project title: Spectrum of Clinical & Genetic Abnormalities In Women With
Congenital Abnormalities of the Female Reproductive Tract

Thank you very much for sending all relevant documentation. I am pleased to confirm that the above project is now registered with the NHS Grampian Research & Development Office. The project has R & D Management Approval to proceed locally. This is based on the documents received from yourself and the relevant Approvals being in place.

All research with an NHS element is subject to the Research Governance Framework for Health and Community Care (2006, 2nd edition), and as Chief or Principal Investigator you should be fully committed to your responsibilities associated with this.

It is particularly important that you inform us when the study terminates.

The R&D Office must be notified immediately and any relevant documents forwarded to us if any of the following occur:

- A change of Principal Investigator, Chief Investigator or any additional research personnel
- Premature project termination
- Any amendments – substantial or non-substantial (particularly a study extension)
- Any change to funding or any additional funding
- Any Serious Adverse Events

We hope the project goes well, and if you need any help or advice relating to your R&D Management Approval, please do not hesitate to contact the office.

Yours sincerely

A handwritten signature in black ink, appearing to be 'S. Ridge', with a stylized flourish at the end.

Susan Ridge
Business Development Officer

Cc: Dr Ruth McGowan, Chief Investigator
NHS Research Scotland Co-ordinating Centre (NRSCC)

Operational Division
Victoria Hospital

Recd

Hayfield Road
Kirkcaldy
Fife KY2 5AH
Telephone 01592 643355
www.show.scot.nhs.uk/fiht



Dr Ruth McGowan
SpR Clinical Genetics
Royal Hospital for Sick Children
Dalnair Street
GLASGOW
G3 8SJ

Date: 22 March 2011
Our ref: 11-017 09/S0704/43
NRS09/GY04
Enquiries to: Aileen Yell
Direct Dial: 01383 565110
Fax No:
E-mail: aileenyell@nhs.net

Dear Dr McGowan

Project Title: Study investigating women with variations in the development of the reproductive system (Mullerian Study)

Thank you for your application to carry out the above project. Your project documentation (detailed below) has been reviewed for resource and financial implications for NHS Fife Operational Division and I am happy to inform you that NHS permission for the above research has been granted on the basis described in the application form, protocol and supporting documentation. The documents reviewed were:

| Document | Version | Date |
|-----------------------------------------------------------------------|---------|-------------------|
| Protocol | | |
| SSI Form | | 26 May 2009 |
| Sponsor letter | | 17 June 2009 |
| CV of investigator | | 25 June 2009 |
| IRAS R&D Form | | 4 September 2009 |
| REC favourable opinion letter | | 22 September 2009 |
| Funding confirmation | | 21 December 2009 |
| REC letter confirming evidence of compliance with approval conditions | | 8 February 2010 |
| NRS-CC Certificate of Compliance | | 11 February 2010 |

The terms of the approval state that you are the investigator authorised to undertake this study within NHS Fife with assistance from Dr Graham Tydeman.

No separate Site Specific Review is required in this case.

The sponsors for this study are Greater Glasgow & Clyde Health Board.

Details of our participation in studies will be included in annual returns we are expected to complete as part of our agreement with the Chief Scientist Office. Regular reports of the study require to be submitted. Your first report should be submitted to Dr A Wood, R&D Manager, R&D Resource Centre, Lynebank Hospital, Halbeath Rd, Dunfermline, KY11 4UW (Amanda.wood3@nhs.net) in 12 months time and subsequently at yearly intervals until the work is completed. A Lay Summary will also be required upon completion of the project.

In addition, approval is granted subject to the following conditions, where applicable:-


Chief Executive Mr John Wilson
Victoria Hospital is part of The Operational Division
of NHS Fife

Vsn 1 - 01.11.09

- All research activity must comply with the standards detailed in the Research Governance Framework for Health & Community Care (<http://www.cso.scot.nhs.uk/publications/resgov/resgov.htm>)
- Health & safety regulations, data protection principles, other appropriate statutory legislation and in accordance with Good Clinical Practice (GCP).
- Any amendments which may subsequently be made to the study should also be notified to Aileen Yell, Research Governance Officer (aileenyell@nhs.net), as well as the appropriate regulatory authorities. Notification should also be given of any new research team members post approval and/or any changes to the status of the project.
- This organisation is required to monitor research to ensure compliance with the Research Governance Framework and other legal and regulatory requirements. This is achieved by random audit of research. You will be required to assist with and provide information in regard to monitoring and study outcomes (including provision of recruitment figures to the R&D office when required).
- As custodian of the information collated during this research project you are responsible for ensuring the security of all personal information collected in line with NHS Scotland IT Security Policies, until the destruction of this data.
- Permission is only granted for the activities for which a favourable opinion has been given by the REC (and which have been authorised by the MHRA where applicable).
- The research sponsor or the Chief Investigator or local Principal Investigator at a research site may take appropriate urgent safety measures in order to protect research participants against any immediate hazard to their health or safety. The R&D office (aileenyell@nhs.net) should be notified that such measures have been taken. The notification should also include the reasons why the measures were taken and the plan for further action. The R&D office should be notified within the same time frame of notifying the REC and any other regulatory bodies.

I would like to wish you every success with your study and look forward to receiving a summary of the findings for dissemination once the project is complete.

Yours sincerely



Dr Gordon G Birnie MD FRCP
Medical Director, Operational Division

*Cc : Aileen Yell, Research Governance Officer, NHS Fife, Lynebank Hospital, Dunfermline
Pamela Shand, NHS Research Scotland C, R&D Office, Foresterhill House Annex, Foresterhil, Aberdeen AB25 2ZB
Dr Graham Tydeman, Consultant Obstetrician, Forth Park Hospital, Kirkcaldy*

Appendix 2.9 Themes and dimensions

| Theme | Dimensions |
|-----------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Hindering independence | <ul style="list-style-type: none"> (i) Personal nature of MRKH (ii) Mothers involvement in diagnostic process (iii) Professionals disregard for growing autonomy (iv) Undermined wish for privacy |
| A sensitivity to difference | <ul style="list-style-type: none"> (i) Emotional reactions to difference (ii) Social situations as triggers for thinking about diagnosis (iii) A need to talk to others with MRKH |
| Managing intimacy | <ul style="list-style-type: none"> (i) Negotiating when to tell partners (ii) Concerns about negative reactions (iii) Avoidance of intimacy |
| Managing threat to identity; dealing with a disrupted developmental process | <ul style="list-style-type: none"> (i) Affected female identity (ii) Affected self-esteem (iii) Managing presentation of self |

Appendix 2.10 Major Research Project Proposal

Exploring the short term psychological impact of Mayer-Rokitansky-Kuster-Hauser (MRKH) syndrome; an interpretational phenomenological analysis approach

Abstract

Background Mayer-Rokitansky-Kuster-Hauser (MRKH) is a congenital condition diagnosed in adolescence when a female's vagina and uterus do not develop. Many studies have noted a strong psychological impact of MRKH however few have examined this explicitly. As gender identity and social acceptance are particularly salient during adolescence it is proposed MRKH may provide unique challenges at this life stage.

Aims This study aims to examine young people's experience of having MRKH, particularly the social and emotional challenges and impact on their identity.

Methods A semi-structured interview following a topic guide will be completed with 8-10 women diagnosed between 6 months and 5 years previously and will be analysed using Interpretational Phenomenological Analysis.

Applications Understanding the challenges a diagnosis of MRKH places on adolescents should inform management of women with the condition, and enhance the development of sensitive and timely interventions.

Introduction

Mayer-Rokitansky-Kuster-Hauser (MRKH) is a congenital condition where, although females have normally functioning ovaries, the vagina and uterus do not develop (Edmonds, 2000). The condition is relatively rare, occurring in around 1 in 5000 females (Edmonds 2003). Diagnosis tends to be in mid-adolescence when secondary sexual features have developed but menstruation has not begun (Aittomaki, Eroila & Kajanoja 2001; Mungadi, Ahmed, Yunuse et al. 2010).

Many studies have noted a strong psychological impact of having MRKH with Edmonds (2003) recommending that management should involve supporting women to cope with the impact of the condition. However, the majority of studies have employed a medical approach, looking at outcomes in terms of surgical and non-surgical treatments to create a neovagina (Bean et al. 2009). Hence, enhancing the understanding of the psychological impact of MRKH should assist services in developing more sensitive and effective care for this population.

The majority of research concerning diagnosis of long-term conditions has focused on those which have continuing, often serious, health implications, such as cancer. Leventhal's (1984) 'self-regulation model', is a commonly used model of adjustment to chronic illness and proposes that people's interpretation of the health threat has an effect on adjustment. According to this model, five areas determine an individual's cognitive representation of the health threat, namely the identity or label they give the condition, the perceived cause, believed consequences, timeline of the illness, and belief about controllability and cure. These cognitive representations have been found to predict illness outcome, including psychological well-being (Hagger & Orbell, 2003). This model however has not been applied to conditions such as MRKH which, although life changing, do not have serious health consequences or require long term pervasive treatment. It is therefore important to look at

the unique challenges of MRKH and whether adjustment to this may differ from the conditions Leventhal's model applies to.

Using Interpretational Phenomenological Analysis (IPA), Holt and Slade (2003) found four themes to emerge for women who had been diagnosed with MRKH between two and 22 years previously, namely dealing with loss, experience of medical services, sharing with others, and the role of time. In a recent review, Bean et al (2009) found only 11 articles focused on the psychological effects of MRKH with 'infertility', diagnosis, adjustment and coping strategies, family responses, self-concept and view of self as female being found to be the most challenging aspects. Kimberley, Hutson, Southwell & Grover (2011) found 79% of their sample, aged between 16 and 71 years old, reported that being unable to carry a pregnancy the most distressing feature of their diagnosis. These studies suggest that having a diagnosis of MRKH challenges women's identities. Indeed, in a review, Heller-Boersma (2006) concluded that adjusting to MRKH is difficult and traumatic, causing women to question their female identity and feel confused about their social and sexual roles. From these studies, it seems that adjustment to MRKH does not easily follow Leventhal's model. Although MRKH is a physical condition, the impact appears to be psychological and largely relate to social roles rather than health per say. Having MRKH is not visible to others, but it fits with Goffman's (1959) notion of a hidden stigma. Consequently, women with MRKH may try to keep their diagnosis secret in order to avoid anticipated negative consequences from others.

The studies reviewed by Bean et al. (2009) have a wide range of ages. However, it is likely that different challenges may be salient at different life stages, for example being unable to carry a child may be more distressing for older females than adolescents. Indeed, time came out as a theme in Holt and Slade's study and Heller-Boersma found women to be most distressed immediately after diagnosis; unsurprisingly accounts changed as time passed. It

is therefore important to explore the unique challenges faced at different phases following diagnosis, including the initial period of adjustment.

As previously mentioned, diagnosis usually occurs in adolescence where Erikson (1968) proposed that the key developmental task for young people is forming a consistent identity and avoiding role confusion; being sure of who they are and how they fit into society. Marcia (1966) further developed Erikson's concepts and proposed that adolescents move through identity stages, beginning with identity diffusion and reaching identity achievement once crisis has been resolved and the person is committed to their identity. Marcia found those who have progressed to identity achievement were more resistant to stress and less affected by receiving negative information about themselves (1966, 1967). Both Erikson and Marcia therefore propose that individuals experience an identity crisis before their identity is formed and that individuals are vulnerable during this period.

Adolescence has been consistently thought to be the life stage associated most with transition and rapid cognitive, social and physical changes, and where sense of self is more salient (Jessor, 1984; Descombe, 2001; Brown, 2004; Sebastian, Burnett & Blakemore, 2008). For example, physical changes in adolescence involve individuals becoming reproductively mature, beginning when secondary sexual characteristics develop and finishing when fully capable of reproduction (Brinthaup & Lipka, 2002). Social changes include becoming more independent, spending less time with their parents, and more time with their peers, and developing more romantic and sexual relationship (Larson & Richards, 1991, Bee, 1975, Engels & van den Eijnden, 2007).

These physical changes have been found to cause many concerns, for example physical changes relate to worry about gender adequacy and sense of self as male or female, with a sense of inadequacy being related to depression and anxiety as well as poorer adult

adjustment (Brinthaup & Lipka, 2002; O'Heron & Orlofsky, 1990; Aubé & Koestner, 1992). Sex role identity, determined by whether the individual fits the roles used within their culture to determine male or female behaviour, is also a salient issue (Paul & White, 1990). The increased focus on social and romantic relationships has been found to relate to being more susceptible to feeling inadequate, fearing peer rejection, and to increased expectations for closeness (Seiffge-Krenke, 1998, Engels, 2007). Adolescents may worry about other's reactions to their diagnosis of MRKH as it breaks social norms relating to gender identities. Consequently they may try to conceal their diagnosis from others to avoid being stigmatised. The social impact of MRKH for adolescents has, however, not been studied.

It therefore seems that irrespective of whether an identity crisis is experienced, adolescents face many challenges and this physical and social process can cause them to question their identity. As gender identity and social acceptance are salient, a diagnosis of MRKH which truly challenges an individual's identity may cause an additional trauma. There may be particular challenges for adolescents with MRKH which lead to greater distress around the time of diagnosis and prolonged identity confusion. Kroger (2004, 2010) proposes that identity development may not be achieved by early adolescence, because particular circumstances may mean many additional hurdles have to be overcome. This study will therefore specifically explore young people's experience of having a diagnosis of MRKH in order to understand their experiences at this critical stage of adolescent development

Qualitative approaches aim to explore individuals' lived experiences. The particular approach to be taken in this study is IPA (Smith, 1996), a method that often deals with significant and emotional life changing experiences (Smith & Osborne, 2003). IPA has been found to be effective in exploring health and illness, sexuality, psychological distress, life transitions and identity (Smith & Osborne, 2008). IPA should be employed with a discrete, homogeneous

group (Smith & Osborne, 2007), therefore this study will focus on those diagnosed with MRKH the past five years to capture the experiences of adolescents at this specific stage.

It is hoped that findings from this study should inform the management of adolescents with MRKH and aid the development of sensitive and timely interventions.

Aims

The study aims to explore young women's experiences of having MRKH to address the following broad research questions;

- What are young people's experiences of having a diagnosis of MRKH?
- What are the social and emotional challenges faced by young people with a diagnosis of MRKH (including family and peer relationships)?
- How do individuals manage a diagnosis of MRKH in relation to their developing identities?

Plan of Investigation

Design

Data will be analysed using Interpretational Phenomenological Analysis (IPA). This approach was chosen to allow descriptive and detailed reports of individual experiences (Smith & Osborne, 2007). IPA is phenomenological in that it concerns individual's personal reports of their experiences yet recognises the interpretative element of the researcher analysing data.

Participants

8-10 females with MRKH will participate. As this is a qualitative study a power calculation was not appropriate, and the sample size follows the recommendations for IPA studies. Smith, Jarman & Osborne (1999) recommend 10 participants to be at the larger end of appropriate sample size and Smith (2003) reports typical sample sizes of 6-12 participants. These small sample sizes allow for a detailed interpretative account of each participant's

experiences (Smith and Osborne, 2008). The number should also allow for data saturation, that is when no new themes are emerging.

Inclusion and Exclusion Criteria

Participants must be female, aged 16 and over, and have had a formal diagnosis of MRKH at least 6 months prior to being interviewed to allow for the initial shock to be resolved.

Participants must not have had a diagnosis for over 5 years. Those aged over 30 and those who do not speak English as their first language will be excluded from the study. As individuals usually receive a diagnosis of MRKH in mid-adolescence and we are looking to recruit only those diagnosed within the last 5 years, it is likely that in reality the age range of the sample will be closer to 16 – 25 years. The sample will have as narrow an age range as possible. These criteria are being used to ensure the group is homogenous and to determine particular experiences at this time.

Recruitment Procedures

This study will be linked to a multicentre genetic study for women with congenital abnormalities with sites in Glasgow, Edinburgh, Aberdeen and Leeds. Individuals on the Scottish Genetic Anomaly Network (SGAN) database are being invited to participate in this genetics study and it is expected that around 40 women will be recruited from the Scottish sites and a further 40 from Leeds. Within the ethical approval for the multicentre study, participants will be asked to consent to both genetic and psychological assessments. They will also be asked whether they agree to being contacted about taking part in an open-ended interview to describe their experiences.

For the current study, those who have consented to being contacted and who received their diagnosis between 6 months and 5 years previously will be sent an information pack about this study with a telephone number and a stamped addressed envelope to contact the

researcher, should they be interested in participating. The information sheet will inform potential participants that, if possible, we would like to record interviews. As participants have already agreed to being contacted about this study, they will be telephoned to ask if they would like to participate if there has been no response after two weeks.

Those who consent to taking part will be asked to meet the researcher at the study site which is closest to them. Participants will be given at least 24 hours to consider the information sheet and consent form before being asked to give consent to taking part and to the interview being recorded. It is anticipated that the interview will last approximately 45 minutes.

Semi-Structured Interview

In line with IPA, a non-directive, semi-structured interview will be used. As this study concerns a very sensitive topic with young women, pilot work will be carried out to find ways to develop a dialogue that allows them to express their views openly. Tasks such as using cards with different statements and asking the participants to sort them into piles depending upon whether it is information they would or would not share with their friends or family might be included as a way of stimulating discussion. This sorting type task would be used as a prompt for discussion rather than to generate data, therefore this data will not be analysed. The first two interviews will be considered as pilots, but if successful it may be possible to include the data in the final analysis. Care will be taken to make it clear to the young women that the researcher is interested in their views as experts on their own lives.

A paper topic guide, based on guidance from experienced clinicians working in the field and from past research, will be used to help structure the dialogue. However the interview will remain alive to different topics raised by the participants themselves and will be discussed fully. It is planned that the draft topic guide will be as follows:

- (i) Social context (family composition, family life, friendships)
- (ii) Diagnosis (initial understanding, information given,)
- (iii) Talking to family (family reactions and communication)
- (iv) Talking to friends (puberty and diagnosis)
- (v) Identity (personal meaning, how the diagnosis makes them feel about themselves and the reactions of others to them, both actual and anticipated)
- (vi) Dealing with intimacy

The social context will be covered at the start of the interview to gain insight into individual contexts and to help participants relax before discussing more sensitive topic. Beyond this, topics will be covered as they are raised by the participant. Participants will also be given the opportunity to discuss any other issues not yet raised.

Measures

As the proposed study is linked to the wider genetics study, there will be permission to access participant information held on the SGAN database such as ages, post codes (to determine socio-economic status using the Scottish Index of Multiple Deprivation and the English Index of Deprivation), and time since diagnosis. The Hospital Anxiety and Depression Scale (Zigmond and Snaith, 1983), a widely used self-report measure, will be completed as part of the genetics study and therefore scores for anxiety and depression will be available. In line with previous literature it is not expected that large numbers of participants will have clinical levels of difficulty, however a characteristic of the sample is likely to be elevated levels of distress compared to the general population (Laggari et al, 2009, Heller-Boersma, Schmidt Edmonds, 2009). This background information will be used to situate the sample.

Research Procedures

Interviews will be held in a clinic room at one of the sites where the genetics study is being run. The interview will last approximately 45 minutes.

Settings and Equipment

Interviews will take place in clinic rooms at the various study centres. Where consent is given, interviews will be recorded and transcribed verbatim.

Data Analysis

Transcripts will be analysed using IPA, as described by Smith & Osborne (2008). Transcripts will be closely read and points of interest and significance will be noted line by line.

Transcripts will then be re-read and themes will be noted. Emerging themes for each interview will be compared and integrated. The key concepts that emerge will be used as the basis for writing the final report, with appropriate quotations being selected to serve as illustration.

To ensure the process is rigorous and transparent a reflective journal will be kept to record decisions made when selecting themes and key concepts. Themes and concepts will also be compared against summaries of each individual interview, including reflective written notes, to ensure individual experiences are described. Identified themes will be discussed in research meetings as a means of audit (Elliott, Fischer & Rennie, 1999; Reid, Flowers & Larkin, 2005). Two transcripts will also be marked by the academic supervisor as a second rater.

Health and Safety Issues

No health and safety issues are anticipated. In order to maintain a safe and private environment where the participant can feel comfortable, interviews will be held in a NHS clinic room within working hours when other staff will be in the building.

Ethical Issues (including where submissions will be made)

An application will be made to the West of Scotland Research Ethics Committee 4 for ethical approval. This application will be linked to the study 'Spectrum of Clinical and Genetic Abnormalities in Women with Congenital Abnormalities of the Female Reproductive Tract'.

Informed written consent will be sought from all participants entering the genetic study and, if asked to attend a semi-structured interview, participants will be asked to sign a consent form for the interview to be recorded. All participants will be given a copy of their signed form. Participants will be informed that they can withdraw consent at any point.

Information will be treated in a confidential manner, with all participants being assigned a participant number. Direct quotations from interviews will be included in the final report, however no identifiable information will be given. Data will be stored on a University of Glasgow laptop, encrypted to NHS standards. Paper copies of anonymous data will be stored in a locked filing cabinet at the Department of Psychological Medicine, University of Glasgow.

The researcher understands that this is a very sensitive topic area and participants may feel embarrassed or find it hard to discuss these issues. The researcher will remain aware of this and will approach topics in a sensitive and incremental fashion. Pilot work will address this issue and tasks may be used within the interview to help prevent participants from feeling too self-conscious. At the end of the interview time will be taken to ask if there is anything else the participant would like to talk about and to check how the participant is feeling. All participants will be provided with a summary of the findings from the study and will be given the opportunity to contact the researcher to discuss this. If any participants show signs of distress, an onward referral will be made to the clinical psychologist linked to the study at the

relevant centre and their GP will be notified with the participant's consent. This procedure will be discussed with participants prior to consenting to taking part in the study.

Financial Issues

Rooms will be available at the centres involved in the study with no cost. A voice recorder and foot pedal will be required to record and transcribe interviews. There will also be costs of posting information and letters to participants. No monies are being requested for travel costs to Leeds.

Timetable

| | |
|----------------------------------------|-------------------------------|
| Submission for ethical approval | February/March 2011 |
| Pilot study to develop interview guide | September 2011 – October 2011 |
| Data Collection | October 2011 – March 2012 |
| Analysis and write up | March 2012 – June 2012 |
| Submission of project | June 2012 |

Practical Applications

Knowing more about the social and emotional challenges a diagnosis of MRKH places on adolescents should inform service development. Further, this greater understanding should enhance the development of therapeutic approaches for this population.

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