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McKillop, Kirsten Ann (2013) *An exploration of the psychosocial consequences of delayed puberty in children who attend the Royal Hospital for Sick Children Endocrine Clinic: A qualitative study and clinical research portfolio*. D Clin Psy thesis.

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VOLUME 1

An exploration of the psychosocial consequences of delayed puberty in children who attend the Royal Hospital for Sick Children Endocrine Clinic

A Qualitative Study and Clinical Research Portfolio

(Volume II bound separately)

Kirsten McKillop

MA (Hons)

Submitted in partial fulfilment of the requirements for the degree of
Doctorate in Clinical Psychology (DClinPsy)

Mental Health and Wellbeing

University of Glasgow

March 2013

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Acknowledgements

I can't believe I am finally completing this section, I thought this day would never come! First and foremost, thank you to all the young people who gave up their time to participate in this research. It was a privilege to hear your stories; thank you for being so open and honest. Thank you also to the Endocrine Team at the RHSC in Yorkhill for all of your support throughout this project.

I would like to give a sincere thanks to my supervisors, Sarah Wilson and Kathleen McHugh, for their unfaltering support, wise words and encouragement throughout this bumpy journey. Thank you especially to Sarah for your never ending patience with my many dramas and poor use of the English language! I really appreciate everything you have both done for me. I am also very grateful to Dougie Fraser and Alison Robertson for giving me the time I needed to finally cross the finish line. Thank you especially to Alison for your understanding, encouragement and humour. All 3 were greatly needed!

Thank you to my classmates for making the last few years so enjoyable! It has been a pleasure getting to know you all. Thank you especially to my study group: Karra, Susan and Aisha. Our 'study dinners' were my lifeline! Thank you also to Lesley for all you have done throughout the past few years!

Without my friends and family I would not have made it this far. A special thanks to Lisa, Gillian, Vicky, Amy, Nikki, Ali..... to name but a few! Thank you for encouraging me and consoling me! Thank you especially to Phil who has been a tremendous support, from chauffeur & chef to editor & motivator, throughout all my years of study.

Also, what a team of support I had at the last hurdle!! Thank you to Jenny, Mum, Dad and Irene for all your help pulling things together. Thank you especially to my Aunt Irene and Uncle John for the incredibly kind and special incentive you gave me to reach the end.

Finally, and most importantly, I would like to say an extra special thank you to my parents for all you have done. Thank you for always believing in me and encouraging me in everything I do. Your unfailing love and support have got me where I am today. Without both of you this would not have been possible. I am very lucky to have such amazing parents!! Thank you!!

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CHAPTER ONE: SYSTEMATIC LITERATURE REVIEW

Psychosocial Aspects of Congenital Disorders of Sex Development:

A Qualitative Systematic Review

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Keywords: Qualitative Systematic Review, Disorders of Sex Development, Psychosocial Impact, Psychological Impact, Patient Experience

Submitted in partial fulfilment of the requirements for the degree of

Doctorate in Clinical Psychology (DClinPsy)

Written in accordance with the requirements for submission to British Journal of Health Psychology (Appendix 1)

Abstract

Aim: To explore the psychosocial aspects of people born with a Disorder of Sex Development (DSD) by conducting a systematic review of relevant qualitative studies using meta-ethnography which will incorporate assessment of the methodological quality of these studies.

Method: The following electronic databases were searched: PsychINFO (EBSCO), Scopus, Embase (Ovid), CINAHL (EBSCO), Web of Science (Web of Knowledge) and Medline (Ovid). The reference lists of the included papers were searched and, in addition, two journals were hand searched for relevant articles. Meta-ethnography was utilised to synthesise the qualitative literature.

Results: The systematic search of the literature identified five articles that were deemed appropriate to be included in the review. All the studies were appraised and considered to be of an acceptable standard or higher. The following themes were identified from the synthesis: Management of Diagnosis; Dissatisfaction with Medical Interventions; Impact on Sense of Self; Emotional, Psychological and Social Impact; Long-term Adjustment.

Conclusions: The synthesis highlighted the importance of open, honest communication between healthcare professionals, the individuals and their families, throughout the process of diagnosis and treatment. The review also has implications for health professionals who can play a key role in giving psychological support to families at various stages of their journey

Introduction

The diagnosis and management of Disorders of Sex Development (DSD) has seen a number of significant changes in the last few years. Advances in medical knowledge and an increased awareness of patients' experiences of medical management, and voiced concerns regarding their treatment, prompted a review of the diagnosis and management of DSD (Frader et al., 2004). This led the European Society for Paediatric Endocrinology (ESPE) and the Lawson Wilkins Paediatric Endocrinology Society (LWPES) to endorse a meeting in Chicago in 2005 which was attended by fifty international experts in the field. They considered the long term care of DSD and issued a consensus statement making a number of recommendations regarding the management at birth and the long term care of individuals born with a DSD (Hughes, Houck, Ahmed & Lee, 2006). The statement recommended the use of a new definition: Disorders of Sex Development (Hughes et al., 2006). Historical terms that were used regularly, including intersex and hermaphrodite, were considered controversial and thought to be derogatory by patients (Conn, Gillam & Conway, 2005).

DSD conditions are not always identified immediately at birth. Some are not diagnosed until adolescence when they enter puberty and some conditions are diagnosed even later (Preves, 2003). Hughes et al. (2006) report that genital anomalies occur in approximately 1 in 4500 live births. Ahmed, Chalmers, Picton & Youngson (2006) however reported that, in Scotland, genital anomalies occur in 5.8 per 1000 live births. Usual practice has been to carry out genital correction surgery as soon as possible and normally before the child is two years of age. The goal of surgery is to normalise appearance and subsequently reduce any feelings of difference the child may have (Gough, Weyman, Alderson, Butler & Stoner, 2008). Hughes et al. (2006) note, however, that there is great uncertainty regarding the youngest age at

which a sound appraisal of gender identity can be achieved, therefore they advise that the “generalisation that the age eighteen months is the upper limit of imposed gender reassignment should be treated with caution.” (p4).

The Psychologist, John Money, in the 1950's and 1960's, proposed that nurture had more influence on the development of gender identity than nature (Money, Hampson & Hampson, 1957). He added that assigning a gender and raising a child as that gender will guide their development because “genetic, hormonal, or other prenatal determinants do not exercise a gross, inevitable predisposing effect” (Money, 1975, p609). Money's influential theory has subsequently guided the treatment and intervention services provided to children born with a DSD. It was thought that any uncertainty surrounding the child's gender would negatively influence the development of their gender identity. As such parents were encouraged to consistently raise their child as either male or female and the importance of their genitalia appearing ‘normal’ and in line with the gender they were assigned was stressed (Carmichael & Alderson, 2004). This promoted a culture of secrecy around DSD conditions as well as a sense of urgency to establish a gender and perform corrective surgery. Perves (2003) argues that this in fact reinforces feelings of stigma surrounding their ambiguous genitalia. He suggests that carrying out surgery that is seen to be corrective and not life-saving promotes a culture of secrecy and shame. He goes even further to suggest that this is unethical. Furthermore, the culture of secrecy is thought to have made it difficult for individuals to explore their diagnosis and seek the appropriate support (Carmichael & Alderson, 2004).

The Chicago Consensus statement stresses the importance of on-going open communication with families. It states that individuals should receive a sex assignment only after expert evaluation and that management should be carried out by a multi-disciplinary team that

includes mental health clinicians (Paterski et al., 2010). Lee (2004) noted that a multidisciplinary team approach would provide the optimal care. This would involve many professionals from various disciplines including medical staff from endocrinology, surgery, gynaecology, genetics and neonatology as well as mental health staff. Furthermore, he notes that representation from social work, nursing and medical ethics would also be helpful, if available, to inform and support the decisions and management of care regarding individuals with a DSD.

In Meyer-Bahlburg's (1999) review of the literature exploring quality of life in individuals with a DSD, he concluded that DSD "differ markedly in their impact on psychological functioning and the combinations of problems encountered" (p 114). Since then research in the area has continued to conclude varying results. In Kleinemeir, Jurgensen, Lux, Widenka & Thyen's (2010) review of the literature they report that studies with adult participants appear to predominantly focus on sexual orientation and psychological well being. When the participants are children, studies appear to focus predominantly on gender development. Furthermore, they report that negative outcomes appear to be found more in studies with adult samples and note a number of studies have concluded that adults suffer from functional, sexual and psychological difficulties (Nordenskjold et al., 2008; Gastaud et al., 2007; Johannsen, Ripa, Mortensen & Main, 2006). Further studies however conclude that there are no significant differences (Warne et al., 2005; Stikkelbroeck et al., 2003; Migeon et al., 2002). The inconsistencies within the DSD literature may be a result of a lack of control groups, small sample sizes or varying assessment approaches (Kleinemeir et al., 2010).

Gough et al. (2008) concluded that, in order to design services that best meet the needs of individuals with a DSD and their families, a greater understanding of sex and gender is required. Hughes (2008) notes that the Chicago Consensus statement “raised more questions than it answered” (p 130). He concluded that further research is required to improve not only the diagnosis in DSD cases but the subsequent medical, surgical and psychological management of these conditions as well. Reiner (2004) also highlights the importance of studying the experiences of those with a DSD to aid ethical interventions. Synthesising the qualitative research that has explored the psychosocial experiences of individuals born with a disorder of sexual development will add to the existing knowledge base and aid service development and provision.

Aim

The aim of this systematic review is to explore the experiences of people born with a DSD, to add to the knowledge base of DSD conditions, to methodologically critique the identified studies and to discuss the implications of the results by conducting a systematic review of relevant published studies.

Review Question

What are the emotional, cognitive, behavioural and social experiences for those born with a DSD, from the point of view of the individuals in question?

Method

Search Strategy

The following electronic databases were searched: PsychINFO (EBSCO), Scopus, Embase (Ovid), CINAHL (EBSCO), Web of Science (Web of Knowledge) and Medline (Ovid). The search terms were also entered into Google Scholar to identify any further articles which may be relevant that were not identified in the databases. There were no limits placed on the time span and index terms were used where available when searching EMBASE and MEDLINE as well as the key terms. Boolean operators (OR and AND) were then used to combine search strings. The reference lists of the included papers were searched and, in addition, two journals that published two of the included papers were selected and hand searched for relevant articles: the British Journal of Health Psychology and the Journal of Clinical Nursing.

Search Terms

When searching EMBASE and MEDLINE the following search terms were used: disorders of sex development mapped to index term OR disorder* of sex* development or intersex or hermaphrodite or ambiguous genitalia AND psychosocial or psychological or experience* or consequence* or impact AND qualitative mapped to index term OR qualitative AND research or method* or analys*.

When searching PsychINFO, Scopus, CINAHL and Web of Science the following search terms were used: disorder* of sex* development or intersex or hermaphrodite or ambiguous genitalia AND psychosocial or psychological or experience* or consequence* or impact AND qualitative AND research or method* or analys*.

Inclusion Criteria

This review included studies if they were published in English, published in peer reviewed journals, described original data and used qualitative methods and analysis to focus on the impact of being born with a DSD, from the point of view of the individuals in question.

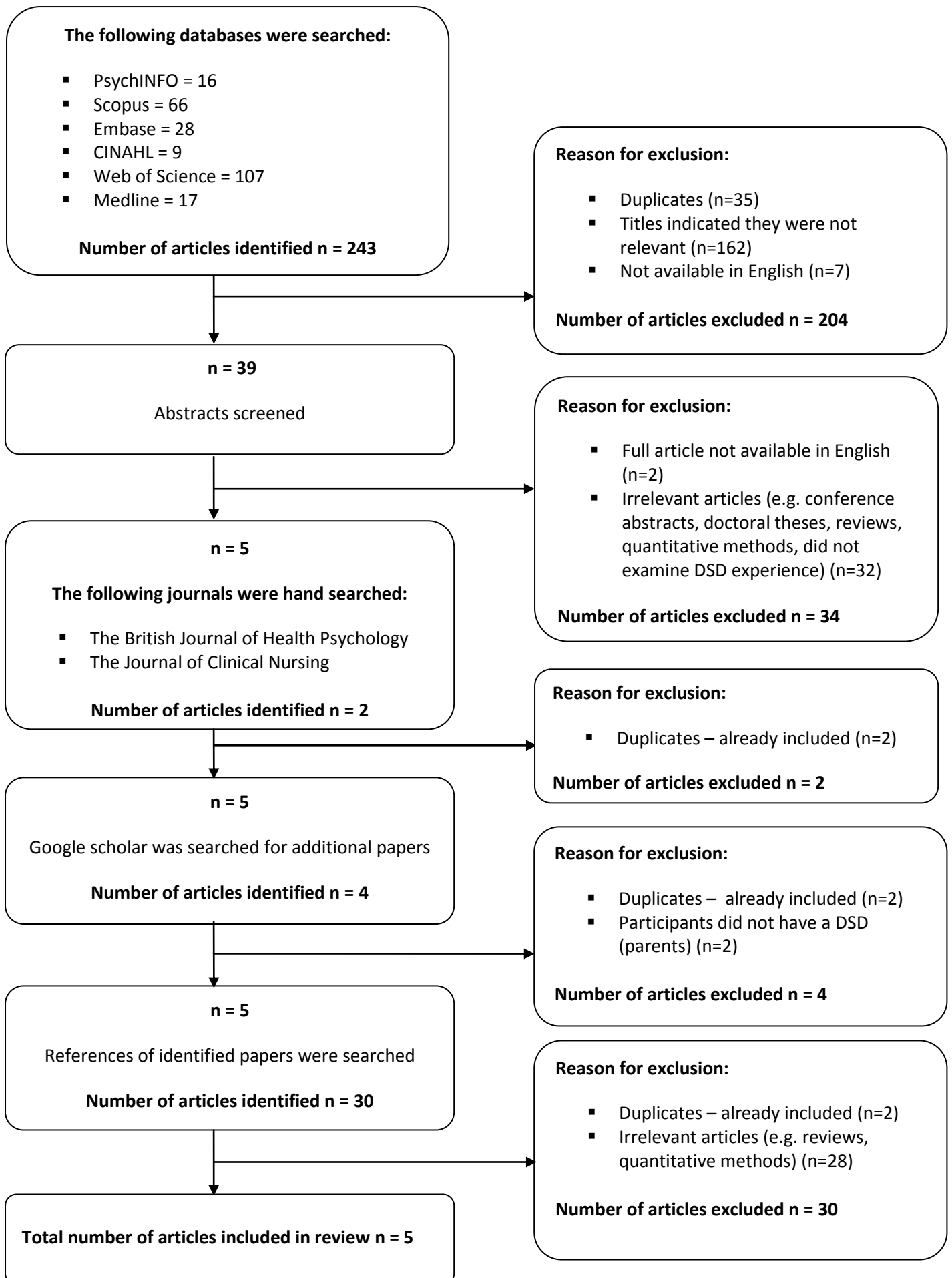
Exclusion Criteria

This review excluded studies that were published in a language other than English, were not published in a peer reviewed journal or if they used quantitative methods or participants who do not have a DSD diagnosis.

Results of Search Strategy

The search yielded 243 articles. 238 articles were discarded because their title/abstract indicated they were not relevant or they did not meet inclusion criteria or they were duplicates. No further articles were identified from Google Scholar, searching the reference lists of the included papers or hand searching two journals. A flow diagram of this process can be seen in Figure 1 which was prepared in accordance with PRISMA guidelines (Moher, Liberati, Tetzlaff & Altman, 2009).

Figure 1: Flow diagram of search strategy process



In total five articles were considered appropriate to be included in this systematic review: Malouf, Inman, Carr, Franco & Brooks, 2010; Mackenzie, Huntington & Gilmour, 2009; Guth, Witchel, Witchel & Lee, 2006; Boyle, Smith & Liao, 2005; Alderson, Madill & Balen, 2004. An overview of the methodological details of the five included articles is given in Table 1.

Quality Appraisal

The quality of each study was assessed on 30 items based on Walsh & Downe's (2006) quality criteria for qualitative studies. Studies were awarded 1 point if the criterion was met and 0 if the criterion was not met. Papers were given a score out of 30 and given a rating of good (>75%), acceptable (>50%) or poor (<50%). The papers were then rated using the same scale by another researcher. Disagreements were resolved through discussion. The quality criteria can be found in Appendix 2.

Synthesis

There are no standard tools or methods to synthesise qualitative research; however a variety of approaches have been identified (Denzin, 1989; Yin, 1989; Miles & Huberman, 1994). The most developed method is Noblit & Hare's (1988) meta-ethnography (Britten, Campbell, Pope, Donovan, Morgan & Pill, 2002). This approach has been applied in various settings (Jensen & Allen, 1994; Paterson, Thorne & Dewis, 1998). Meta-ethnography allows meanings from a variety of studies using qualitative methods to be synthesised and reinterpreted (Atkins, Lewin, Smith, Engel, Fretheim & Volmink, 2008).

Meta-ethnography involves 7 steps: Getting started; deciding what is relevant for initial interest; reading the studies and extracting data; determining how the studies are related; translating the studies; synthesising translations and expressing the synthesis (Noblit & Hare, 1988).

Noblit and Hare (1988) advise recording the themes reported in each of the reviewed studies as a way to begin exploring how the themes relate. The studies' individual interpretations were then compared and the similarities and differences between them were examined in order to explore what could be said about the psychosocial experiences of having a DSD as a whole based on the individual interpretations. It became apparent that the themes identified by the individual authors were not clearly understood by the label alone attached to them. Also, although the authors identified different labels to describe their themes, the content appeared to be synonymous. These findings were integrated to develop a new level of interpretation (Pope, Mays & Popay, 2007). More specifically, overarching concepts (super-ordinate themes) that applied across all the studies were identified that were felt to be most representative of the different labels reported by the individual authors. Table 2 outlines the list of original themes in each of the papers and illustrates the overarching themes each label is best represented by.

Results

Table 1: Methodological overview of included articles

Authors	Year	Country	Data Collection	Recruitment	Setting	Participants	Study Aim	Quality Ratings
Malouf et al.	2010	USA	Consensual Qualitative Research	Advertisements at support group, on support forums and other research related websites	Setting not specified	8 Age range: 23 – 51 All female All have a diagnosis of Congenital Adrenal Hyperplasia (CAH)	To investigate health related quality of life, mental health concerns of women with CAH and their recommendations for therapists	25/30 83% Good
Mackenzie et al.	2009	New Zealand	Thematic Analysis	Recruitment was recorded as being difficult. Participants were recruited through one contact	Setting not specified	3 Age range: 30s/ 40s 2 identified as intersex, 1 identified as female	To explore the experiences of people with intersex conditions and contribute to the knowledge of the condition	24/30 80% Good

Guth et al.	2006	USA	Content Analysis	Identified by a physician	Setting not specified	5 Age range: 23- 34 All female All had diagnosis of Congenital Adrenal Hyperplasia (CAH)	To explore self-perceptions of adult women who have CAH and the impact of this condition on their relationships, sexuality, gender identity and self-concept as well as their perceptions of medical treatment	21/30 70% Acceptable
Boyle et al.	2005	UK	IPA and Foucauldian Discourse Analysis	Recruited from a support group for women with DSD following presentation from researcher and information on support group website	Women were given the choice of being interviewed at home or in a hospital psychology department	6 Age range: 27 – 52 All female Variety of DSD conditions	To explore the experiences of women with intersex conditions who chose genital surgery as adults	24/30 80% Good

Alderson et al.	2004	UK	Grounded Theory	Recruited through AIS UK wide support group media and network for people with AIS	7 women were interviewed in their home and 1 was held in a conference centre	8 Age range: 20 – 54 All female All had diagnosis of Androgen Insensitivity Syndrome (AIS)	To provide an understanding of the psychological sequel of AIS in phenotypic females in order to inform psychosocial health care services	24/30 80% Good
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Table 2: List of original themes and synthesised overarching themes

Original Themes	Synthesised Overarching Themes				
Malouf et al., 2010	Management of Diagnosis	Dissatisfaction with Medical Intervention	Impact on Sense of self	Emotional, Psychological and Social Impact	Long-term Adjustment
Physical Functioning		X	X	X	
Psychological Functioning	X	X	X	X	X

Interpersonal Processes and Relationships			X	X	X
Healthcare Experiences of Women with CAH	X	X			X
Recommendations for Health Care Professionals		X			X
Systemic Considerations	X	X		X	
Mackenzie et al., 2009	Management of Diagnosis	Dissatisfaction with Medical Intervention	Impact on Sense of self	Emotional, Psychological and Social Impact	Long-term Adjustment
Managing Silence	X	X		X	X
Coping with Difference			X	X	
Development of Acceptance		X	X	X	X
Guth et al., 2006	Management of Diagnosis	Dissatisfaction with Medical Intervention	Impact on Sense of self	Emotional, Psychological and Social Impact	Long-term Adjustment
Perceptions of Medical Treatment	X	X		X	
Perceptions of Visits to Physicians		X		X	X
Relationships and Sexuality		X	X	X	

Gender identity and Gender Roles			X	X	
Personal Characteristics			X	X	X
Body Image		X	X		X
Boyle et al., 2005	Management of Diagnosis	Dissatisfaction with Medical Intervention	Impact on Sense of self	Emotional, Psychological and Social Impact	Long-term Adjustment
Conferring Normality	X	X	X	X	
Conferring Sexual and Relationship Entitlement		X	X	X	
Doctor Knows Best		X			
Just Another Fanny		X			
The Imperfect Results		X		X	
Surgery is Not Experienced as Conferring Normality or Satisfactory Intimate Relationships		X	X	X	
Two Fixed Sexes			X	X	
The Conflation of Sex with Intercourse		X	X	X	
The Absence of Pleasure Talk		X	X	X	

Alderson., et al 2004	Management of Diagnosis	Dissatisfaction with Medical Intervention	Impact on Sense of self	Emotional, Psychological and Social Impact	Long-term Adjustment
AIS Distress			X	X	
Compromised Womanhood			X		
Fear of Devaluation			X	X	
Talking about AIS				X	X
Medical Management	X	X		X	X
AIS Support Group					X
Adaption	X				X

This synthesis highlighted the following key themes:

- ❖ Management of Diagnosis
- ❖ Dissatisfaction with Medical Interventions
- ❖ Impact on Sense of Self
- ❖ Emotional, Psychological and Social Impact
- ❖ Long-term Adjustment

This review will now go on to explore the psychosocial impact under the heading of each theme that has been identified in the synthesis.

Management of Diagnosis

The first theme to emerge from the synthesis was the management surrounding receiving a DSD diagnosis (Malouf et al., 2010; Mackenzie et al., 2009; Guth et al., 2006; Boyle et al., 2005; Alderson et al., 2004). Some participants reported positive interactions with their medical clinicians (Malouf et al., 2010; Guth et al., 2006; Alderson et al., 2004). The majority of participants, however, spoke of information surrounding their diagnosis being withheld, incomplete or inappropriately communicated.

“They would explain it to me in these very complicated, physiological terms from a scientist’s perspective.....” (Unknown, Guth et al., 2006, p65)

“What I did feel angry about was the secrecy.” (Barbara, Alderson et al., 2004, p90)

Some participants reported that they were aware something was not as it should be; however, they were unsure what it was because they were not given a full explanation of their diagnosis. This subsequently contributed to feeling uncomfortable and concerned.

“I wasn’t the same as other women and it absolutely terrified me.” (Douglas, Mackenzie et al., 2009, p.1778)

Some participants stressed that the way people are told of their diagnosis will influence their adjustment and how they cope (Alderson et al, 2004). Some participants also reported that they would prefer to have all of the information at once where as others said it would be helpful to receive bits in stages (Alderson et al., 2004).

It appears that the onus was primarily on some of the individuals to gather information for themselves (Mackenzie et al., 2009; Guth et al., 2006; Alderson et al., 2004). They tried to seek answers to their questions from a number of sources; other clinicians, parents, reading, television and the internet.

The role of parents in the management of diagnosis was also acknowledged. Some of the participants noted that their parents struggled to answer questions or avoided the topic altogether. Parents were reported to have expressed feelings of guilt regarding their child's diagnosis (Malouf et al., 2010; Alderson et al., 2004). Some participants wondered if the secrecy surrounding their diagnosis was instigated by the medical clinicians or if it was because their parents were also struggling with the news (Mackenzie et al., 2009).

“The nurse bent down and picked you up and said ‘oh god it’s a hermaphrodite’. Now as Mum said that she started screaming and crying and ran out of the room.... so she comes back into the room about 10 minutes later and she’s clearly been crying” (Douglas, Mackenzie et al., 2009, 1778)

Some participants noted the positive changes over the years with regards to the amount of information given to children and their families and how medical staff interact with them (Malouf et al., 2010).

Dissatisfaction with Medical Intervention

The dissatisfaction of ongoing treatment and medical intervention was reported by a number of participants (Malouf et al., 2010; Mackenzie et al., 2009; Guth et al., 2006; Boyle et al., 2005; Alderson et al., 2004). They spoke of regular hospital appointments and procedures

contributing to their feelings of difference (Guth et al., 2006; Boyle et al., 2005). Some participants also noted that they were not always told what procedures involved or why they were taking place (Mackenzie et al., 2009; Boyle et al., 2005; Alderson et al., 2004). Some said they felt ‘on show’ during medical appointments, especially if there were a number of clinicians present (Guth et al., 2006). Examinations were also reported to have been traumatic at times.

“I felt violated at the hospital after being vaginally dilated.....” (Unknown, Malouf et al., 2010, p6).

“I felt like I am being used..... I felt like I was more embarrassed than anything. I don’t mind one or two coming in but not six, seven doctors looking at you.” (Unknown, Guth et al., 2004, p65)

A number reported misconceptions regarding genital surgery. Some participants hoped it would improve their feelings of difference and positively impact their intimate relationships (Boyle et al., 2005). Mackenzie et al. (2009) reported that some think surgery removed what was unique about them and they questioned why surgery was required at all. Participants noted that they think the emphasis placed on surgery comes from the lack of acceptance in society for anything that differs from the norm. A general dissatisfaction regarding surgery was reported and it did not appear to resolve feelings of difference or difficulties within relationships (Boyle et al., 2005).

“One thing I would say is unless there is an absolute need for it, keep the scalpels away”.
(Kate, Mackenzie et al., 2009, p1780)

Some participants noted they thought they were functioning normally apart from having to take medication every day (Malouf et al., 2010). A number of side effects from medication were reported. Most notable was weight gain (Malouf et al., 2010; Guth et al., 2006). Some side effects were also reported to contribute to ambivalence or non-adherence (Malouf et al., 2010; Alderson et al., 2004). A few of the participants from the United States noted they were concerned about the financial implications of ongoing treatment (Malouf et al., 2010).

Impact on Sense of Self

The impact on their sense of self was another theme to emerge from the synthesis. Some participants spoke of a tension between the gender they were assigned and how they felt internally. Some identified with the gender they were assigned, whereas others had some underlying uncertainties and a few identified as neither male nor female (Mackenzie et al., 2009; Guth et al., 2006).

“I feel like I am a neutral party... neither male nor female. But what if you are born in the middle? Who are you? How are you supposed to identify? Am I a motherly figure or aren't I? Is that my role in life or isn't it?” (Unknown, Guth et al., 2006, p67)

A number of participants noted concerns surrounding their fertility and whether or not they would be able to have children (Malouf et al., 2010; Mackenzie et al., 2009; Guth et al., 2006; Alderson et al., 2004).

Some women also spoke about their appearance being mismatched to their assigned gender as well. They reported a number of characteristics that they perceived to be predominantly

masculine: facial hair, excessive sweat, lower voice, strength (Malouf et al., 2010; Guth et al., 2006; Alderson et al., 2004).

“I was quite muscular and I felt I looked quite masculine, and you know, I just felt I looked awful.” (Barbara, Alderson et al., 2004, p 87)

Some participants spoke of tension between their assigned gender and how they behave. Parents were reported to sometimes prompt them to engage in activities that were seen as more specific to their assigned gender (Malouf et al., 2010; Mackenzie et al., 2009; Guth et al., 2006).

“Mum tried to push me into girl things. I had formal lessons on how to arrange flowers. In the end Mum gave up, but it clearly distressed her.” (Kate, Mackenzie et al., 2009, p1778)

The perceived mismatch in their physical appearance and the awareness of their diagnosis appears to contribute to the ongoing reminder that they are different. Some participants appear to feel uncomfortable with who they are. A number of participants used negative words like ‘freak’ to describe themselves (Boyle et al., 2005; Alderson et al., 2004).

“Every time I see a reflection of myself it’s almost as if it’s a reminder of the AIS.” (Fiona, Alderson et al., 2004, p87)

Emotional, Psychological and Social Impact

Participants reported a range of emotions throughout the process of receiving the diagnosis, treatment and ongoing adjustment. Some participants reported anxiety and fear when they did

not know what was going on (Mackenzie et al., 2009). This led to anger for some patients when they felt situations were badly managed (Alderson et al., 2004). A number reported feelings of shame as a result of the secrecy or awareness of being different (Guth et al., 2006).

“Having CAH is very shameful and humiliating. I feel it has greatly impacted my psychological abilities....” (Unknown, Guth et al., 2006, p66)

Many reported difficulties within relationships (Malouf et al., 2010; Mackenzie et al, 2009; Guth et al., 2006; Boyle et al., 2005; Alderson et al., 2004). Some reported positive sexual experiences (Mackenzie et al., 2009; Guth et al., 2006) while others noted difficulties (Malouf et al., 2010; Boyle et al., 2005). Several participants reported ongoing fears that they’ll be ‘found out’. They noted that they were worried about other peoples’ reactions. As such some participants are very selective in what they share and with whom in order to protect themselves (Malouf et al., 2010; Guth et al., 2006; Boyle, et al., 2005; Alderson et al., 2004). Some participants appear to withdraw and be more isolated as a way to protect themselves because of their fear of discovery (Mackenzie, et al., 2009).

“I didn’t allow myself to get close to them and em because it was this massive thing and I couldn’t bear anyone to find out so I, so I deliberately I suppose really built and kept that difference between me and them.” (Fiona, Alderson et al., 2004, p89)

Mackenzie et al. (2009) proposed that the difficulties participants experienced in relationships were a result of avoidance and withdrawal from social experiences as a child.

Boyle et al. (2005) and Alderson et al. (2004) both proposed that it was the individual's own anxieties and feelings regarding their diagnosis that caused relationship difficulties.

“I felt.... like.... I hadn't learned all the social sort of skills that were needed to... you know to... to establish a relationship and that maybe that was the main problem, and having a vagina wouldn't really help... there's more going on than just vaginal length.” (P5, Boyle et al., 2005, p579)

Some participants reported their diagnosis, or the way it was managed, had an impact on their concentration and learning at school (Malouf et al., 2010). DSD does not appear to have had a global impact on the participants' education status. Many of the participants were noted to have achieved varying levels of qualifications. Some participants however, did report being bullied at school (Mackenzie et al., 2009). This likely exacerbated fears of how others would react. Although some reported positive responses, participants continue to be wary of others and have a fear of discovery (Alderson et al., 2004).

Long-term Adjustment

Another theme to emerge was the long-term adjustment to their diagnosis. Individuals adopted a number of coping strategies to help manage their difficulties and long-term adjustment. Some of these were maladaptive; many avoided situations and relationships and limited what information they shared about themselves (Mackenzie et al., 2009; Boyle et al., 2005; Alderson et al., 2004). Other strategies, however, were helpful; positive thinking, focussing on the benefits and sharing their feelings (Malouf et al., 2010; Guth et al., 2006; Alderson et al., 2004).

“It’s given me a different perspective on life... I am a very deep, analytical person with incredible convictions in the sense of justice” (Unknown, Guth et al., 2006, p69)

Mackenzie et al. (2009) proposed the importance of understanding their condition to aid long-term adjustment. Alderson et al. (2004) concluded that while knowledge of their diagnosis is important, support from others is also required.

Some participants reported that they received positive support from their friends, families, co-workers and professionals. Others reported a lack of support from family and co-workers (Moulf et al., 2010; Mackenzie et al., 2009; Alderson et al., 2004).

“They had a really good understanding of it, and were really fantastic about it.” (Barbara, Alderson et al., 2004, p89)

“I want people to understand where I am coming from, but I feel frustrated because people tell me I do not look like I have a problem.” (Unknown, Malouf et al., 2010, p7)

The majority of participants had contact with support groups or online forums, although the extent of their involvement varied (Malouf et al., 2010; Mackenzie et al., 2009; Guth et al., 2006; Alderson et al., 2004). Some found it helpful to know there were others out there and they found the experience empowering. Others found it overwhelming and upsetting. Alderson et al. (2004) reported that overall, participants found it beneficial.

“I think the best thing I found was this website where we all share and now that makes me feel less alone.” (Unknown, Malouf et al., 2010, p6)

The role of Mental Health services was acknowledged and the participants who had received their input reported that it was helpful; those who had not received any input noted that therapeutic support would have been beneficial at some point during their journey (Malouf et al., 2010; Mackenzie et al., 2009; Guth et al., 2006; Alderson et al., 2004). Some reported that they felt support for their mental health was forced upon them after a suicide attempt (Malouf et al., 2010). Other participants reported that they felt their medical clinicians underestimated the psychological impact of their diagnosis (Guth et al., 2006; Alderson et al., 2004).

“I mean, it has been 34 years and nobody has ever been interested in the psychological aspects.” (Unknown, Guth et al., 2006, p66)

Therapeutic support for the family was also identified as being potentially helpful.

“You have two healthy parents and then they suddenly have got a sick baby that’s dependant on drug therapy and mean doctors and stuff, but the kid just has it; the parents have to deal with it”. (Unknown, Malouf et al., 2010, p8)

Discussion

The aim of this systematic review was to explore the psychosocial aspects of DSD. The management of the diagnosis was the first theme to emerge from the synthesis. Most participants reported that they felt their diagnosis was not managed as well as it could have been. Sutton, Young, McInerney-Leo, Bondy, Gollust & Biesecker (2006) explored the importance of diagnostic disclosure and concluded that women were more likely to interpret the way their diagnosis was shared as negative if they felt that information had been

withheld. This highlights the importance of transparency and honesty during the diagnostic process. A few participants identified that they were aware of a change in recent years with regard to the way information is shared when someone is diagnosed. This is echoed in Liao, Green, Creighton, Crouch & Conway's (2009) study exploring service users' experiences of obtaining information about DSD. They concluded that older participants were less likely to report that they had been adequately informed of their diagnosis compared to younger participants.

A number of participants reported that their parents appeared to find it difficult to answer their questions about their diagnosis or that they prompted them to behave more in line with their chosen gender. How well parents cope and adjust to their child's diagnosis will likely impact their ability to support their child and ultimately the child's coping and adjustment. A number of qualitative studies have been conducted exploring the parental experience of having a child with a DSD. In Sanders, Carter & Goodacre's (2011) study parents reported that they thought the medical clinicians involved in their child's care were vague and reluctant to discuss their child's diagnosis. They noted that this exacerbated their concerns. Parents highlighted the importance of early and ongoing support from healthcare professionals in supporting them to manage their child's DSD. Crissman, Warner, Gardner, Carr, Schast, Quittner, Kogan & Sandberg (2011) concluded that parents found the uncertainty surrounding their child's diagnosis and treatment particularly stressful. Furthermore, parents spoke of a tension between wanting to seek support while also being concerned that if they shared their child's diagnosis they were risking their child being stigmatised. Many participants also recognised that individual therapeutic support would have been helpful at different points during their journey. This stresses the importance of support being offered to the whole family and not just the individual, starting with the

diagnosis and at various stages of their journey. The DSD Consensus recommends early planning with the parents with regular, gradual medical information and counselling (Hughes et al., 2006).

Many participants spoke of dissatisfaction with medical interventions and a number of them noted that they found certain procedures traumatising. Schutzmann, Brinkmann, Schacht & Appelt (2009) explored psychological distress in people with DSD. They concluded that adults with DSD report suicidal tendencies and self-harming behaviours on a level comparable to women with a history of physical or sexual abuse. They stress though that recruitment procedures prevent their results being generalised. Johannsen et al. (2006) investigated the quality of life experienced by 70 women with DSD. They found an overall impaired quality of life and suggested traumatic/distressing medical procedures may play a role.

It is of interest that a number of participants reported a general dissatisfaction with genital surgery. Fagerholm et al., (2011) explored attitudes regarding genital surgery among 24 females who had surgery in childhood. They concluded that the majority of women thought it was performed at the right age, only 3 thought it was carried out too late and no one expressed the opinion that they had undergone the procedure too young. Only 2 women regretted the operation. Furthermore, sexual functioning was generally similar to that of the controls.

Many participants noted a tension between the gender they were assigned and the atypical behaviours that they engaged in or how they felt internally. Some participants also reported

that they were confused about their gender or they identified as intersex. This stresses the importance of not only a thorough assessment process to assign gender but also the need for ongoing involvement and support of health professionals. It also raises the question of how society as a whole views 'gender'. Hird (2003) noted that "the patient's gender is always read as either male or female. There is no discussion of the possibility of both, neither, or a third gender, despite the sustained discussion of these possibilities in the intersex, transgender, feminist, psychoanalytic and social psychology literatures" (p188).

The medical, surgical and ethical management of individuals with a DSD appears to be an ongoing discussion. Although the DSD consensus does offer some guidance surrounding the medical/surgical management, individuals and parents can still have decisions to make regarding treatment (e.g. timing of surgery and/or the use of vaginal dilators). This can be a difficult choice for them to make because for some individuals it is not always clear what would be the best course of action (Cohen-Kettenis, 2010).

Limitations and Future Directions

Synthesising qualitative literature can provide a greater, more in-depth understanding of the area being explored (Atkins et al, 2008). There are however a number of limitations. Given that this is a synthesis of qualitative research the participants within the studies are self-selecting and homogenous. It is possible that individuals who are finding life more difficult are unlikely to volunteer to discuss an area of life that is distressing. This is especially true for this topic area given that there is a historic attitude of secrecy surrounding a DSD diagnosis. As such, the key themes cannot be generalised to all adults with DSD and instead they only offer an insight into the experiences that those with DSD may have.

Pope et al. (2007) highlight that a weakness of meta-ethnography is that new interpretations are formed based on themes identified by the authors of the articles rather than the original data. As with all qualitative research the interpretation of the data in every study will have been influenced by the researchers own beliefs and values. It is important to note that this will also be true for the author's interpretations within this synthesis.

The limited number of papers identified by the systematic search should be acknowledged. It is recognised that there are a number of difficulties encountered when searching for qualitative literature (Atkins et al., 2008). Qualitative literature is not thought to be adequately indexed within medical databases. It is also often published in books rather than peer reviewed journals (Walsh & Downe, 2005). Evans (2002) notes that the titles of qualitative research are often descriptive, subsequently making it difficult to identify if the research is qualitative. Furthermore, he reports that the abstracts of qualitative studies do not always indicate that qualitative methods were used. Pope et al. (2007) stress however that when conducting a systematic review of qualitative research, "searching and selection is purposive, and it may not need to be exhaustive or comprehensive since the goal is theoretical not statistical generalisation" (p81).

It is however possible that the limited number of papers identified is indicative of the limited qualitative research that is available. Given the historic secrecy that has surrounded the management of this diagnosis it is understandable that individuals with a DSD may be reluctant to participate in research. The ongoing debate regarding ethical, medical and surgical management stresses the importance of understanding the experience of DSD from the individuals perspective. Subsequently this review, synthesising what qualitative literature is available, will make a significant contribution to the existing body of research and

highlight the importance of continuing to explore this field. Further qualitative research into DSD would increase the awareness of these conditions and reduce some of the stigma and secrecy that surrounds them.

Implications for Clinical Practice

Psychosocial care should be an integral part of the on-going management of individuals with a DSD. According to Hughes et al. (2006), in their review of the Consensus and its suggested management of intersex disorders, involvement from mental health clinicians can inform decisions regarding gender assignment, hormone treatment and the appropriate time for surgery. They can also offer support to the individual and their family regarding adjustment and coping with the diagnosis and treatment.

Conclusion

This systematic review adds to the existing body of research and ongoing discussion regarding the medical, surgical and ethical management of individuals born with a DSD. The identified key themes give an insight into the impact of DSD from individuals who have experienced it. The review highlights the experiences of individuals with a DSD from diagnosis to long-term adjustment. The results suggest that uncertainty surrounding diagnosis, along with the dissatisfaction of medical interventions, can impact their sense of self and result in emotional, psychological and social consequences. This synthesis highlights the importance of open, honest communication between healthcare professionals, the individual and their families throughout the process of diagnosis and treatment. The review also has implications for health professionals who can play a key role in giving psychological support to families at various stages of their journey to aid their management of the emotional, psychological and social consequences.

References

Ahmed, S. F., Chalmers, J., Picton, S. & Youngson, G. (2006). *Scottish Audit of Genital Anomalies*. NHS Quality Improvement Scotland

Alderson, J., Madill, A. & Balen, A. (2004). Fear of devaluation: Understanding the experience of intersexed women with androgen insensitivity syndrome. *British Journal of Health Psychology*. **9**, 81-100.

Atkins, S., Lewin, S., Smith, H., Engel, M. Fretheim, A. & Volmink, J. (2008) Conducting a meta-ethnography of qualitative literature: Lessons learnt. *BMC Medical Research Methodology*, **8** (21).

Boyle, M., Smith, S. & Liao, L. (2005). Adult Genital Surgery for Intersex: A Solution to What Problem? *Journal of Health Psychology*. **10**, 573-584

Britten, N., Campbell, R., Pope, C., Donovan, J., Morgan, M. & Pill, R. (2002). Using meta ethnography to synthesise qualitative research: a worked example. *Journal of Health Service Research Policy*. **7** (4), 209- 215.

Carmichael, P. A. & Alderson, J. (2004). *Psychological care of children and adolescents with disorders of sexual differentiation and determination*. In A. Balen (Ed.) *Paediatric and adolescent gynaecology: a multidisciplinary approach*. Cambridge: Cambridge University Press.

Cohen-Kettenis, P. T. (2010). Psychosocial and psychosexual aspects of disorders of sex development. *Best Practice & Research Clinical Endocrinology & Metabolism*. **24**, 325-334.

Conn, J., Gillam, L. & Conway, G. (2005). Revealing the diagnosis of androgen insensitivity syndrome in adulthood. *British Medical Journal*. **331**, 628-630.

Crissman, H., Warner, L., Gardner, M., Carr, M., Schast, A., Quittner, A., Kogan, B. & Sanberg, D. (2011). Children with disorders of sex development: A qualitative study of early parental experience. *International Journal of Paediatric Endocrinology*. **10**, 1-11

Denzin, N. K. (1989). Interpretive *interactionism*. *Applied social research methods series, vol 16*. Newbury Park: Sage.

Evans, D. (2002). Database searches for qualitative research. *Journal of the medical library association*. **90** (3), 290-293.

Fagerholm, R., Santtila, P., Miettinen, P. J., Mattila, A., Rintala, R. & Taskinen, S. (2011). Sexual function and attitudes toward surgery after feminizing genitoplasty. *Journal of Urology*. **185** (5), 1900-1904

Frader, J., Alderson, P., Asch, A., Aspinall, C., Davis, D., Dreger, A..... Parens, E. (2004). Health Care Professionals and Intersex Conditions. *Archives of Paediatric & Adolescent Medicine*. **158** (5), 426-428

Gastaud, F., Bouvattier, C., Duranteau, L., Brauner, R., Thibaud, E., Kuttenn, F. & Bougnères, P. (2007). Impaired sexual and reproductive outcomes in women with classical forms of congenital adrenal hyperplasia. *Journal of Clinical Endocrinology & Metabolism*. **92**, 1391-1396.

Gough, B., Weyman, N., Alderson, J., Butler, G. & Stonder. (2008). “They did not have a word”: The parental quest to locate a ‘true sex’ for their intersex children. *Psychology and Health*. **23** (4), 493-507

Guth, L., Witcher, R., Witcher, S. & Lee, P. (2006). Relationships, Sexuality, Gender Identity, Gender Roles, and Self-Concepts of Individuals who have Congenital Adrenal Hyperplasia: A Qualitative Investigation. *Journal of Gay & Lesbian Psychotherapy*. **10** (2), 57-75.

Hird, M. J. (2003). A typical gender identity conference? Some disturbing reports from the therapeutic front lines. *Feminism & Psychology*. **13**, 181-199.

Hughes, I. A. (2008). Disorders of sex development: a new definition and classification. *Best Practice & Research Clinical Endocrinology & Metabolism*. **22** (1), 119-143.

Hughes, I. A., Houk, C., Ahmed, S. F. & Lee, P.A. (2006). Consensus statement on management of intersex disorders. *Archive of Disease in Childhood*. **91**, 554-563.

Jensen, L. A. & Allen, M. N. (1994). A synthesis of qualitative research on wellness-illness. *Qualitative Health Research*. **4**, 349-369.

Johannsen, T., Ripa, C., Mortensen, E. & Main, K. (2006). Quality of life in women with disorders of sex development. *European Journal of Endocrinology*. **155**, 877-885.

Kleinemeier, E., Jurgensen, M., Lux, A., Widenka, P. & Thyen, U. (2010). Psychological Adjustment and Sexual Development of Adolescents with Disorders of Sex Development. *Journal of Adolescent Health*. **47**, 463-471.

Lee, P. A. (2004). A perspective on the approach to the intersex child born with genital ambiguity. *Journal of Paediatric Endocrinology & Metabolism*. **17**, 133-140.

Liao, L. M., Green, H., Creighton, S. M., Crough, N. S. & Conway, G. S. (2009). Service users' experience of obtaining and giving information about disorders of sex development. *International Journal of Obstetrics and Gynaecology*. **117**, 193-199.

Mackenzie, D., Huntington, A. & Gilmour, J. (2009). The experiences of people with an intersex condition: a journey from silence to voice. *Journal of Clinical Nursing*. **18**, 1775-1783.

Malouf, M., Inman, A., Carr, A., Franco, J. & Brook, L. (2010). Health-Related Quality of Life, Mental Health and Psychotherapeutic Considerations for Women Diagnosed with a Disorder of Sexual Development. *International Journal of Paediatric Endocrinology*. Article ID: 253465, 11 pages.

Meyer-Bahlburg, H. F. L. (1999). Health-related quality of life in intersexuality. *Acta Paediatrica Supplement*. **88**, 114–115.

Migeon, C. J., Wisniewski, A. B., Gearhart, J. P., Meyer-Bahlburn, H. F., Rock, J. A., Brown, T. R., Casella, S. J., Maret, A., Ngai, K. M., Money, J. & Berkovitz. (2002). Ambiguous genitalia with perineoscrotal hypospadias in 46, XY individuals: Long-term medical, surgical and psychosexual outcomes. *Pediatrics*. **110** (3).

Miles, M. B. & Huberman, A. M. (1994). *Qualitative data analysis: an expanded source book*. Thousand Oaks, CA: Sage.

Money, J., Hampson, G. & Hampson, J. L. (1957). Imprinting and the establishment of gender role. *Archives of Neurology and Psychiatry*. **77**, 333-336.

Money, J. (1975). Psychological Counselling: Hermaphroditism. In L. Gardner (Ed). *Endocrine and genetic diseases of childhood and adolescence*. 2nd ed. London: Saunders. p609-618.

Mother, D., Liberati, A., Tetzlaff, J. & Altman, D.G. (2009). Preferred reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. *PLoS Med* **6**(6)

Noblit, G. W. & Hare R. D. (1988). *Meta-ethnography: Synthesizing Qualitative Studies*. Vol 11. Newbury Park, CA: Sage.

Nordenskjold, A., Holmdahl, G., Frisen, L., Falhammar, H., Filipsoon, H., Thoren, M., Janson, P. O. & Hagenfeldt. (2008). Type of mutation and surgical procedure affect long-term quality of life for women with congenital adrenal hyperplasia. *Journal of Clinical Endocrinology & Metabolism*. **93**, 380-386.

Pasterski, V., Prentice, P. & Hughes, I. A. (2010). Consequences of the Chicago consensus on disorders of sex development (DSD): current practices in Europe. *Archive of Disease in Childhood*. **95** (8), 618-23.

Paterson, B. L., Thorne, S. & Dewis, M. (1998). Adapting to and managing diabetes. Image: *Journal of Nursing Scholarship*. **30**, 57-62.

Pope, C., Mays, N. & Popay, J. (2007) *Synthesizing qualitative and quantitative health evidence. A guide to methods*. England: Open University Press; 1 Edition.

Preves, S. E. (2003). *Intersex and identity: the contested self*. New Brunswick, New Jersey: Rutgers University Press.

Reiner, W. (2004). Mixed-method research for child outcomes in intersex conditions. *BJU International*. **93** (3), 51-53.

Sanders, C., Carter, B. & Goodacre, L. (2012). Parents need to protect: influences, risks and tensions for parents of prepubertal children born with ambiguous genitalia. *Journal of Clinical Nursing*. **21**, 3315-3323.

Schutzmnn, K., Brinkmann, L., Schacht, M. & Richter-Appelt, H. (2009). Psychological Distress, Self-harming Behaviour, and Suicidal Tendencies in Adults with Disorders of Sex Development. *Archives of Sexual Behaviours*. **38**, 16-33.

Stikkelbroeck, N. M., Beerendonk, C.C., Willemsen, W. N., Schreuders-Bais, C.A., Feitz, W. F., Rieu, P. N., Hermus, A. R. & Otten, B. J. (2003). The long term outcome of feminizing surgery for congenital adrenal hyperplasia: Anatomical, functional and cosmetic outcomes, psychosocial development and satisfaction in adult female patients. *Journal of Paediatric and Adolescent Gynaecology*. **16**, 289-296.

Sutton, E. J., Young, J., McInerney-Leo, A., Bondy, C. A., Gollust, S.E. & Biesecker, B. B. (2006). Truth- telling and Turner Syndrome: the importance of diagnostic disclosure. *Journal of Paediatrics*. **148** (1), 102-107.

Walsh, D. & Downe, S. (2005). Meta-synthesis method for qualitative research: a literature review. *Journal of Advanced Nursing*. **50** (2), 204-211.

Walsh, D. & Downe, S. (2006). Appraising the quality of qualitative research. *Midwifery*. **22**, 108-19.

Warne, G., Grover, S., & Hutson, J., Sinclair, A., Metcalfe, S., Northam, E. & Freeman, J. (2005). A long term outcome study of intersex conditions. *Journal of Paediatric Endocrinology & Metabolism*. **18**, 555-567.

Yin, R.K. (1989). Case Study Research: design & methods. Newbury Park: Sage

CHAPTER TWO: MAJOR RESEARCH PROJECT

An exploration of the psychosocial consequences of delayed puberty in children who attend the Royal Hospital for Sick Children Endocrine Clinic: A Qualitative Study

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KEYWORDS: Delayed Puberty, Psychosocial, Psychological, Patient Experience, IPA, Qualitative

Submitted in partial fulfilment of the requirements for the degree of

Doctorate in Clinical Psychology (DClinPsy)

Written in accordance with the requirements for submission to British Journal of Health
Psychology (Appendix 1)

Lay Summary

When adolescents enter puberty they have a growth spurt and develop secondary sexual characteristics. If this doesn't happen by the time girls are 13 or boys are 14, then their puberty is thought to be delayed. Research so far has focused on the impact of delayed growth and so it is unclear what impact delayed puberty has. The aim of this study was to explore how delayed puberty affects the lives of those who have it. Five young people diagnosed with delayed puberty and attending the Endocrine Clinic at Royal Hospital for Sick Children were interviewed about their experiences. The interviews were recorded, typed out and analysed. Five themes were identified: Confusion Surrounding Diagnosis, Adolescents' Internal Experience, Adolescents' External Experience, Coping and Future Prognosis. Delayed puberty appears to impact the adolescents emotionally, psychologically and socially. The adolescents used a number of coping strategies to help them manage the consequences of their diagnosis. They reported feelings of confusion surrounding their diagnosis and treatment which stresses the importance of adolescents receiving clear information about delayed puberty and how it is treated as soon as possible when they first attend the Endocrine Clinic. Overall they reported that delayed puberty only affects certain areas of their life and that they are generally happy with who they are.

Abstract

Background: Puberty is considered to be delayed if sexual maturation occurs beyond 13 years in girls and 14 years in boys. Physical consequences of delayed puberty include short stature and immature appearance, relative to their chronological age. Psychosocial consequences include social withdrawal and isolation, teasing and bullying, parental over protection, poor body image, low self-esteem and declining academic performance. Research findings in this area can be conflicting with most of the focus being upon delayed growth. Consequently, the psychosocial impact of delayed puberty remains unclear. This study aimed to explore adolescents' experiences of delayed puberty from a psychosocial perspective.

Method: Five adolescents with delayed puberty attending the Royal Hospital for Sick Children were recruited to the study. Semi-structured interviews were conducted, recorded and transcribed. The data were analysed using Interpretative Phenomenological Analysis.

Results: Analysis identified five super-ordinate themes: Confusion Surrounding Diagnosis, Adolescents' Internal Experience, Adolescents' External Experience, Coping and Future Prognosis. They reported a range of emotional, behavioural, social and psychological affects. Adolescents utilised various maladaptive and adaptive coping strategies to help them manage the consequences they experience. They reported feelings of confusion and uncertainty surrounding their diagnosis and treatment which may be contributing to the emotional and psychological symptoms.

Conclusion: Adolescents reported that delayed puberty only affects certain areas of their life and that they are generally happy with who they are. This study emphasises the importance of adolescents receiving clear information about delayed puberty and its treatment as soon as possible when they first attend the endocrine clinic to help manage the psychological and emotional consequences reported.

Introduction

Adolescence is a time of significant psychological, social and physical changes (Christie & Viner, 2005). The “growth spurt and appearance of secondary sexual characteristics are the hallmarks of adolescent physical development” (Apter, Galatzer, Beth-Halachmi & Laron, 1981, p501). Puberty is considered to be delayed if sexual maturation occurs beyond 13 years in girls and 14 years in boys (Sedlmeyer & Palmert, 2002). It has been suggested that the cut off ages should be lowered because of “a downward trend in pubertal timing in the United States and other countries” (Palmert & Dunkel, 2012, p443). Approximately 2.5% of adolescents will have delayed pubertal onset. No cause for delay can be identified for the majority of these adolescents, who will successfully progress through puberty albeit later than their peers (Rosen & Foster, 2001).

More boys than girls present with delayed puberty. This is thought to be because delayed growth and an immature appearance have a greater psychosocial impact on boys than on girls (Rosen & Foster, 2001). Boys who are treated as their height age rather than their chronological age experience low self-esteem and difficulty separating from their parents (Suris, Michaud & Viner, 2004). They report feelings of unpopularity due to the reduction in age appropriate social opportunities (Rosen & Foster, 2001). In their review of the literature Rosenfeld, Gregory, Northcraft & Hintz (1982) report that short stature in males can lead to feelings of vulnerability and incompetence as well as anxiety and depression, especially if they suffer from delayed puberty as well.

Girls will present with fewer psychosocial difficulties and some are reported to value their immature physique (Rosen & Foster, 2001). Delayed puberty in girls is not accompanied by delayed growth as often as it is in boys (Apter et al., 1981). Pubertal induction is offered to boys over 14 years old and girls over 13 years old who show no signs of endogenous puberty. The rationale for offering timely progression through puberty is to normalize physical appearance and psychosocial development (Palmert & Dunkel, 2012).

The specific impact of delayed growth out with delayed puberty is widely reported; however, findings are conflicting with regard to the impact of short stature on psychosocial development. Approximately 60-70% of children with short stature have reported experiencing teasing and juvenilization, which has been shown to be related to behavioural difficulties (Sandberg & Michael, 1998). Some studies, however, have shown that the self-esteem of children with short stature is not adversely affected and they display normal patterns of behaviour (Voss, Bailey, Mulligan, Wilkin & Betts, 1991; Downie, Mulligan, Stratford, Betts & Voss, 1997). Furthermore, Sandberg (2000) reported that psychosocial difficulties that have been attributed to short stature appear to be less problematic than originally thought; also it is not clear if treatment improves the adolescents' psychological experience.

Given the possible increased risk of ridicule from peers, parents can become overprotective of their adolescent (Holmes, Hayford & Thomson, 1982). This can lead to further social immaturity and difficulties expressing independence or assertiveness (Meyer-Bahlburg, 1990). Zimet, Cutler, Litvene, Dahms, Owens & Cuttler (1995) however assessed the psychosocial functioning of adults who were medically referred as children for short stature

but were not treated with human growth hormone. It was reported that despite the negative experiences endured by adolescents with short stature and immature appearance, they are not thought to suffer long term from clinically significant problems of psychosocial adaptation.

Parents have also reported that children of short stature exhibit more limited academic abilities compared to their peers who do not have delayed growth (Visser-van Balen, Sinnema & Green, 2006). Sandberg, Colsman & Voss (2004) in their review, however, reported that some studies were unable to identify academic deficits among children with short stature. Furthermore, Holmes, Karlsson & Thomson (1985) found that adolescents with delayed growth *and* delayed puberty were academically performing as expected for their chronological age and concluded that any difficulties experienced were likely to be short-term.

Adolescents who appear younger than they are may also lack social experiences appropriate to their age. This can lead to isolation and withdrawal from their peers (Sandberg & Voss, 2002). Apter et al. (1981) investigated the impact of delayed puberty and delayed growth on self-image in adolescents. Fifty-six adolescents with varying combinations of delayed puberty and delayed growth were given a Self-Image Questionnaire. It was found that, although isolated delayed puberty did not have a significant impact on self-image, delayed growth did. It was concluded that delayed growth is potentially more problematic than delayed puberty.

The specific focus on short stature within the literature, along with the methodological diversity in studies, makes identifying the psychosocial impact of delayed puberty problematic. It is not surprising that research findings conflict. Previous research in this area has been quantitative, using questionnaires written by health professionals without taking into account the experiences of affected individuals. An exploration is therefore needed of the problems concerning delayed puberty from the perspective of those who are experiencing the delay.

Aim

This study aims to explore adolescents' experiences of delayed puberty from a psychosocial perspective.

Method

Ethical Considerations

Before the study commenced, approval was sought from the University of Glasgow, the West of Scotland Research Ethics Committee and Greater Glasgow and Clyde's Research and Development Department (Appendix 3).

Design

This study utilised a qualitative design with in-depth, semi-structured interviews. Interpretative Phenomenological Analysis (IPA) was employed to explore the experiences of the individuals without forming hypotheses or attributing meaning about the area being

explored (Smith, Flowers & Larkin, 2009). The aim of IPA is to explore how the individual has understood and interpreted a significant event in their life (Smith et al., 2009). This is done through collating the accounts that participants have given of their experience. IPA attempts to offer an understanding of the area being explored through identifying themes and forging links within the individuals' accounts (Smith, 1996). These links provide evidence and a greater understanding of the phenomenon being explored (Willig, 2001).

Sample/Participants

Purposive sampling was used to recruit adolescents attending the Endocrinology outpatient clinic at The Royal Hospital for Sick Children (RHSC) who presented with delayed puberty. Recruitment took place from July 2012 to February 2013. Consultant Endocrinologists identified adolescents according to the following inclusion and exclusion criteria:

Inclusion Criteria

Pubertal induction is offered to girls older than 13 and boys older than 14. There are many markers for delayed puberty. Therefore girls older than 12 years and boys older than 13 years, that were identified by a Consultant Endocrinologist as clinically pre-pubertal and had not yet started treatment or had been receiving treatment for less than 4 months, were included.

Exclusion Criteria

Adolescents who had been receiving treatment for more than 4 months were excluded. Those with a psychiatric diagnosis or impaired cognitive functioning, which would have prevented them from being able to give informed consent, were not included. Adolescents were also excluded if they were not in mainstream education or fluent in English or aged 18 years or older.

Whilst over 10 adolescents were identified only 8 agreed to take part and only 5 attended for interview. The main reason given for adolescents declining to participate was that having delayed puberty is something that makes them feel uncomfortable/upset and they did not wish to discuss it in-depth. Participant characteristics are detailed in Table 1. All adolescents were allocated a number and will be referred to by their number throughout in order to ensure anonymity.

Table 1: Participant Characteristics

Participant	Age at time of interview (year and month)	Gender	Clinics attended at RHSC
1	13.3	Male	<ul style="list-style-type: none">• Endocrinology for Delayed Puberty
2	13.11	Male	<ul style="list-style-type: none">• Endocrinology for Delayed Puberty• Clinical Psychology for Anger Management• Infectious Diseases for Allergies• Asthma Clinic
3	13.3	Female	<ul style="list-style-type: none">• Endocrinology for Delayed Puberty and another endocrine condition• Gastroenterology
4	14.11	Female	<ul style="list-style-type: none">• Endocrinology for Delayed Puberty• Gastroenterology• Dietetics
5	14.6	Male	<ul style="list-style-type: none">• Endocrinology for Delayed Puberty

Recruitment Procedures

Adolescents who were identified as matching the inclusion criteria and not the exclusion criteria were given, either in clinic or mailed, a study information pack. All packs included an invitation letter and information about the study for the adolescent (Appendix 4) and for their parent/guardian (Appendix 5). An opt-in form (Appendix 6) was also included for the

adolescent to register their interest in participating. Members of the clinical team, whose name appeared on the invitation letters, contacted the adolescents and their parent/guardian via telephone to ensure they had received the study pack. Adolescents who agreed to participate gave permission for their contact details to be passed on to the primary researcher. Interviews were then arranged at a mutually-convenient time and, where possible, they were scheduled for the same day as the adolescent's next returning hospital appointment. Four interviews took place in the RHSC and one was arranged in a nearby GP surgery for the convenience of the participant.

Research Procedures

Prior to the interview all participants and their parents were given the information sheet for a second time. There was also the opportunity to discuss the study with the researcher and ask any questions. Written informed consent was then gained from both the adolescent and their parent (Appendix 7) before the interview took place. Interviews were individual and semi-structured. They were recorded and lasted no more than 45 minutes.

A topic guide was devised which covered the areas to be explored (Appendix 8). The Self-Regulatory model of Illness Behaviour (Leventhal et al., 1992) provided a framework for the topic guide. It included an exploration of the participants' understanding of delayed puberty as a health problem and their perceptions of how the condition impacts on the most salient aspects of their lives.

Adolescents were asked how they viewed the condition and the symptoms they perceived to be a part of it. Their ideas regarding the cause of delayed puberty and how long it would last were explored before moving on to explore their perceptions of expected outcomes and how responsive they believe the symptoms will be to treatment. Having elicited their perceptions of delayed puberty as a health problem, the interview then set out to explore the positive and negative consequences of delayed puberty at home, in school, with friends, with romantic attachments and how they view themselves, in order to establish the effect it is having on their life. Lastly their perception of their controllability regarding the reported consequences was explored.

Questions were open-ended and non-directive and acted as triggers to encourage participants to talk about their experiences. Prompts in the form of specific and/or focused questions were used to encourage the adolescents to expand and elaborate. The topic guide was reviewed after the first interview by both the primary researcher and supervising researchers. After piloting, the topic guide was deemed effective and revisions were not required. It must be noted that the topic guide was not prescriptive and it did not explicitly control the structure of the interview. Instead it guided the interviewer and provided prompts. Interviews were then transcribed and anonymised for all references to people and places.

Data Analysis

To analyse the data, the 6 steps in IPA described by Smith et al. (2009, p82-101) were followed. This begins by reading and re-reading the transcripts to gain a holistic understanding of the participants' experience. Then semantic content and points of interest are initially coded in each transcript. By doing this, emergent themes are developed and

connections are explored between the themes. This process is completed for all the transcripts before patterns and overarching themes across all the participants' narratives are identified. A sample of one of the transcripts and initial coding is available in Appendix 9.

Reflexivity

This involves the researcher reflecting on their own beliefs, values and life experiences and the impact these will have on how the research is interpreted. It is important the primary researcher acknowledges and reflects upon this throughout the study, given that the interactions with those participating had the potential to impact the analysis and interpretations of the transcribed interview (Willig, 2001). Reflexivity was addressed through regular discussions in research supervision and a proportion of the transcripts (80% in total) were also analysed by other researchers who are experienced in IPA. The researchers analysed the transcripts independently of each other. Themes identified by all analysts were compared for consistency to ensure reliability of the analysis and there was a high level of agreement across all the themes identified by all the researchers.

Results

Five super-ordinate themes emerged from the narratives with regard to the psychosocial impact of delayed puberty on adolescents. The first super-ordinate theme to emerge from the data was confusion surrounding their diagnosis of delayed puberty. The second super-ordinate theme was the adolescents' internal experience followed by the adolescents' external experience, which was identified as the third super-ordinate theme. The fourth super-ordinate theme to surface from the data was the adolescents' coping styles. The final super-ordinate

theme to emerge from the narratives was their thoughts regarding their future prognosis. Sub-themes were identified within each of the five super-ordinate themes. The themes are listed in Table 2.

Table 2: Table of super-ordinate and sub-themes

Super-ordinate Themes	Sub-themes
1. Confusion Surrounding Diagnosis	<ul style="list-style-type: none"> • Reliance on Information and Advice from Medical Professionals • Personal Models of Cause
2. Adolescents' Internal Experience	<ul style="list-style-type: none"> • Comparison and Feeling Different • Dissatisfaction with Physical Appearance • Emotional Impact
3. Adolescents' External Experience	<ul style="list-style-type: none"> • Impact on Behaviour • Effect on Family Life • Consequences at School • Impact on Social Life • Effect on Romantic Relationships

<p>4. Coping</p>	<ul style="list-style-type: none"> • Maladaptive • Adaptive • Role of Others
<p>5. Future Prognosis</p>	<ul style="list-style-type: none"> • Lack of Clarity Regarding Treatment and Timing • Reliance on Medication • Sense of Self

Super-ordinate Theme 1: Confusion Surrounding Diagnosis

Reliance on Information and Advice from Medical Professional

The adolescents all reported that they noticed they were different to their friends and they were not growing like they thought they should be. They reported that they were unsure about what was going on or why they were different. Four participants noted that attending the endocrine clinic and speaking to health professionals gave them some understanding of the differences they had identified.

“Being told by [Name] that I have delayed puberty wasn’t really a shock, but it was kind of like, it was more a kind of relief because then I know what’s going on. So like I know it’s not something that won’t be able to be fixed, it’s something that can be fixed. It’s when I didn’t know I just thought.... like they said like that I am growing but they didn’t..... I just didn’t know what was happening.”

(Participant 3, P16: L506)

Personal Models of Cause

Despite gaining some understanding of their diagnosis from attending the clinic, the adolescents still remain uncertain about the cause of their delayed puberty. Only one adolescent was confident in the underlying cause for her delay. The remaining four adolescents expressed uncertainty regarding why they had not entered puberty and all five reported different explanations. These explanations included: genetics, premature birth, a low BMI and a side effect from taking medication for other endocrine difficulties or an unknown condition that had not yet been diagnosed.

“I thought it was because I was born a month early... like a whole month... because my body wasn’t fully ready when I came out. I thought that’s why.”

(Participant 1, P20: L575)

“My parents are both quite small so obviously I’m not going to grow as such..... I understand that I’m not going to be the tallest man in the world, but it’s still.....”.

(Participant 5, P1: L28)

Super-ordinate Theme 2: Adolescents' Internal Experience

Comparison and Feeling Different

Comparison played a significant role in the adolescents' growing awareness of their condition. All adolescents still compare themselves to their peers and report feeling different: not only with regard to their physical appearance and appearing younger but also in terms of things they feel they can't do but their friends can. The boys predominantly reported not being able to perform as well in sports and the girls focused mainly on not being able to wear the same clothes.

"I was a wee bit self-conscious about certain things, I didn't really..... like when my friends were able to wear like certain clothes and things and where we were going to a lot of parties, I wasn't able to like wear like the same things. When I was in pictures with them I just thought I looked a bit out of place, like I looked younger than them all."

(Participant 3, P3: 96)

All adolescents noted that they are aware of other people who are also small. Although no one knew anyone who has a definite diagnosis of delayed puberty, they noted that it can be helpful to think there are others who are similar to them.

"It makes you feel like you're not alone and there's other people as well like that. So it's not just you."

(Participant 1, P17: L484)

Dissatisfaction with Physical Appearance

All five adolescents reported dissatisfaction with their physical appearance. All five wish they were taller and the girls reported that they would like to develop breasts while two of the boys reported that they would like to be stronger.

“Well sometimes I don’t feel like a girl, because I don’t have pretty underwear and I don’t have the boobs”.

(Participant 4, P12: L 395)

“I’ve not really bulked out at all, which is quite disappointing”.

(Participant 5, P3: L74)

Emotional Impact

Delayed puberty was reported to have an impact on all of the adolescents’ emotional state. They reported variations of the following: anxiety, frustration, low mood, anger, helplessness and embarrassment.

“I just sometimes get a bit frustrated. And like sometimes I get like a wee bit upset because like I’m a bit smaller”.

(Participant 3, P14 L: 448)

Super-ordinate Theme 3: Adolescent's External Experience

Impact on Behaviour

All adolescents reported that delayed puberty has an impact on their behaviour, mainly with regard to avoidance. Two of the boys noted that they avoid attending theme parks because they are unable to go on any of the rides. One adolescent spoke about having to think twice before contributing to conversations or engaging in certain activities because he thinks people are more likely to reject his contribution because of his height. Another adolescent reported that he avoided hanging out with people who are taller than him because he thinks they are more likely to make fun of him. Both girls spoke of feeling isolated when their friends speak about bras and periods and they will subsequently disengage from conversations and move away to avoid feeling embarrassed or uncomfortable. The girls also reported avoiding wearing certain clothes or trying on any clothes when out shopping with their friends because they feel uncomfortable with how they look.

“They’ll say ‘oh well, come back when your voice has broken’ and that, they can take the rip sometimes and I have to be careful what I say”.

(Participant 5, P14: L465)

“Like, I don’t like to get changed in the changing room where people can see because I don’t think I look like I should at my age.”

(Participant 4, P1: L11)

Effect on Family Life

Four of the adolescents have siblings. They all appear to compare themselves to their siblings and three of them noted that although their sibling is younger than them, they are the same height or even taller. Three then reported that their siblings have made unhelpful comments about their height. The adolescent who did not have any siblings reported that they thought it was helpful they were an only child because they would find it difficult to have a sibling go through puberty before them.

“My sister’s always saying ‘Ha ha, I’m taller than you, I’m taller than you’. So that gets quite annoying.”

(Participant 2, P2: L56)

All five adolescents reported that delayed puberty does not have a significant impact on their relationship with their parents.

“I don’t think it has really made any difference. I think they still love me... or I hope they do anyway.... (laughing).... love me for who I am and not really if anything is different about my body.”

(Participant 5, P6: L179)

Four participants, however, noted some specific difficulties in family life. One boy reported not being able to help his father in the garden because of his lack of strength and one boy noted they felt a lack of support from his mother when his sibling made comments about his height. With regard to the girls, one girl noted she felt uncomfortable speaking with her dad about her delayed puberty and one noted she needs additional support from her parents to reach certain items around the house.

“My mum’s there when she says it, but she doesn’t really say anything”.

(Participant 2, P16: L450)

“It’s just wee silly things like when I can’t reach things... I have to climb on top of a chair, or I have to wait until someone, like my mum and dad, comes in and helps me reach stuff”.

(Participant 3, P6: L184)

Consequences at School

Delayed puberty was not reported to have a global effect on school or school work. Three adolescents, however, spoke of negative comments or questions from peers and one reported being pushed around by peers when she is moving between classes.

“If they’re not friends with me they sometimes say that I’m ‘flat chested’, ‘oh you’re not tall’, ‘you’re really tiny’, ‘you’re a good arm rest’ and things like that.”

(Participant 4, P9 L: 298)

“They can say stuff to me because they know I won’t punch them back for a joke because I’m not strong enough or they know I’m not a threat to them”.

(Participant 5, P1: L8)

Impact on Social Life

All five adolescents reported that delayed puberty does not have a general impact on activities outside of school, however, they all acknowledged specific difficulties with regard to some of the hobbies they engage in. The girls predominantly spoke about the impact delayed puberty has on shopping because they feel unable to try on clothes with their friends. The boys focused on the impact delayed puberty had on playing sport. Two participants noted they can have difficulties attending the cinema because they appear younger than they are and one adolescent also noted difficulties when learning to play the guitar because their hands are smaller.

“A friend had a go-karting party and I wasn’t tall enough to go..... uh I didn’t go”

(Participant 1, P3: L85)

“I’ve been let go by a couple of teams because of my height and my size”.

(Participant 5, P1: L10)

Effect on Romantic Relationships

Currently, delayed puberty does not appear to have an effect on romantic relationships. All five adolescents reported that this was not a concern at present but they were all unsure if it would cause difficulties in the future. Two of the boys reported that they think men should be taller than women and therefore it may cause problems for them at some point. One of the girls noted that boys may not be attracted to her because she does not look like other girls her age.

“Well men are like supposed to be like taller than like the girls, so it’s like, quite eh..it can be quite awkward... because if you’re small, then eh, they might think that ‘oh no, he’s small, I don’t want to go out with him.”

(Participant 2, P11: L300)

“Well some of the boys might like me, but they wouldn’t tell me, because they might get judged by the more popular boys in our year....um...because they don’t think that I’m what they should go out with because I’m not the same.”

(Participant 4, P13: L437)

Super-ordinate Theme 4: Coping

All adolescents reported utilising both maladaptive and adaptive coping strategies to help them manage some of the consequences of delayed puberty.

Maladaptive

Some of the maladaptive strategies reported were avoiding thinking about it or talking about it, avoiding certain places, punching the wall to vent anger and wearing high heels to mask a height difference or wearing certain clothes to hide their body.

“Sometimes I may take my anger out on some stuff.... like sometimes I punch the walls, sometimes I punch.... eh... my bed”.

(Participant 2, P3: L65)

“Even on holiday I’ll wear a cardigan to cover up and I’ll always walk with my arms crossed so no one... cause I feel out of place sometimes”.

(Participant 4, P6: L194)

Adaptive

Adaptive strategies included setting achievable goals, engaging in hobbies, speaking to people (friends, teachers, parents and health professionals), distraction techniques, focusing on the positive, using humour and identifying the benefits of delayed puberty.

“I think... kind of thinking positively like just helps a lot”.

(Participant 1, P23: L678)

“Use your disadvantage and try to turn it into an advantage....cause if you’re eh, like in basketball if you’re small then you can eh... eh... like ease your way through... eh... taller people.”

(Participant 2, P12: L352)

Role of Others

All adolescents noted the role of others in helping them manage the impact of delayed puberty. This included: parents offering reassurance, encouragement and sharing their personal experience: teachers addressing bullying comments at school: friends and family understanding and being supportive: health professionals answering questions and giving the adolescents an understanding of their delayed puberty.

“So if you have your class behind you, or you have a group of friends behind you, it helps.....knowing if there’s people out there to help you.”

(Participant 4, P16: L517)

Super-ordinate Theme 5: Future Prognosis

Lack of Clarity Regarding Treatment and Timing

All adolescents reported a desire to see improvements as soon as possible but they all recognised that it would take time for things to improve. All five adolescents reported that they are uncertain of the specifics regarding possible treatments and when these would be considered and how long it would be before they see a change.

“I don’t really know... because they haven’t really told me what these things [blood tests and tablets] do, but I think when I go back and they tell me what they do, I think I will understand it more.... and how it’s going to help me”.

(Participant 3, P5: L164)

Three participants noted being concerned that things may never improve.

“You think you’re small, and then you think like, eh... that you might not hit puberty, and that you’re just gonna stay small for the rest of your life.”

(Participant 2, P9: L259)

Reliance on Medication

Although the adolescents reported being uncertain of the exact details regarding treatment, they all reported that taking medication is one of the most important things required to improve their situation.

“I’m just hoping... to find a possible solution like, something that would help me grow like, a medication or something because it... I’m really... it’s really getting desperate”.

(Participant 5, P4: L125)

Sense of Self

Despite wishing to change physical attributes the adolescents reported that delayed puberty only impacts certain aspects of their life and they are otherwise generally happy with themselves and who they are.

“I would still like to be the same person, I would still like to be a nice boy, but it would help if I were stronger and things and bigger built”.

(Participant5, P14: L457)

Discussion

The illness perception components of the Self-Regulatory model of Illness Behaviour (Leventhal et al., 1992) informed the development of the topic guide used during the interviews with the young people. It is important to acknowledge the potential influence using the Self-Regulatory model as a framework may have had on the narratives. The illness

perception components were used in order to illicit information regarding the young people's understanding of their condition. Although Leventhal's terminology wasn't used explicitly during the interviews, and the topic guide acted as a prompt for the interviewer and did not control the structure of the interview, the Self-Regulatory model did inform the content of the questions the interviewer asked. Asking questions based on the Self-Regulatory model may have potentially influenced how the young people thought about their health and subsequently shaped the answers they gave. This would, however, have also been true had the topic guide been developed using another model or questions suggested by staff from the Endocrinology Clinic. In order to counter the effects of this, the young people were encouraged to expand and elaborate on their answers in order to fully explore the impact of their diagnosis from their point of view. An audio recording of the first interview was also reviewed by an independent researcher to ensure, as much as possible, that the way the questions were asked did not explicitly shape the answers given. In accordance with Smith et al. (2009), with the aim of gaining an understanding of the consequences of delayed puberty for the perspectives of the young people concerned, the narrative were analysed bottom-up.

This study set out to explore the psychosocial impact of delayed puberty. The five super-ordinate themes were identified in all five of the narratives. The results of the interviews indicated that delayed puberty does not globally impact their lives, however they all reported specific psychological and social difficulties. The gender balance within this study was 40% female and 60% male. This appears to be in line with the overall gender balance of young people who present with delayed puberty. As noted earlier, more boys than girls present with delayed puberty (Rosen & Foster, 2001).

All five adolescents spoke of being aware that something was not right but they were unsure what was wrong until they attended the clinic. Bramswig & Dubbers (2009) stress the importance of the medical team providing the adolescent with information and supporting/reassuring them throughout the process of puberty to contain the anxiety they will feel regarding developing at a different pace to their peers.

The most common cause of delayed puberty is constitutional delayed puberty. This is thought to be an extreme variation of normal pubertal timing (Palmert & Dunkel, 2012). What specifically triggers puberty remains unknown. There are thought to be a number of things that could contribute to a delayed pubertal onset. These include: nutrition, chronic illness, psychosocial stress and eating disorders (Christie & Viner, 2005). The adolescents all gave different explanations for the cause of their delayed puberty. It is unclear if the explanations given by the adolescents for their delayed onsets were correct and given to them by the medical team. Only one adolescent was confident in her explanation. The remaining four were uncertain which suggests that, even if the medical team had previously given them an explanation, they could not remember it and they remained uncertain.

The adolescents' internal experience predominantly involved feelings of difference. They compared themselves to their peers and subsequently reported feeling unhappy with their physical appearance. All 5 noted they wished they were taller. Wit, Reiter, Ross, Saenger, Savage, Rogol & Cohen (2007), in their recent review into the management of short stature, reported that there is an inconsistency within the literature with regard to adolescents' dissatisfaction with their physical appearance. They note that some studies concluded that their satisfaction was lower than controls while other studies concluded that there wasn't a

difference. They stress that the adolescents' perceived height, rather than their actual height was a more significant predictor for psychosocial distress.

It is of interest that both of the female participants reported being dissatisfied with their physical appearance and sexual immaturity. This is in contrast to Rosen & Foster's (2001) report that some girls are thought to value their immature physique. Rosenblum & Lewis (1999) recognise however that girls are particularly vulnerable with regard to noticing if they are developing at a different pace to others, which can then subsequently impact their body image.

Four of the participants reported unhelpful things their peers say or do. These appeared to significantly impact their emotional state and contribute to how they feel about their delayed puberty, and ultimately themselves. Issues of bullying have been identified in other studies. Voss & Mulligan (2000) explored bullying in a UK school and concluded that boys with short stature were more than twice as likely to be bullied compared to controls. Adolescents utilised both helpful and unhelpful strategies to cope with impact of delayed puberty. Wit et al., (2007) report that to date, there are not any published studies that explore the coping strategies short statured children adopt.

Sandberg & Colsman (2005) in their review of the literature note that, while adolescents with short stature can have negative social experiences which can cause psychosocial stress, their global emotional and behavioural functioning is not significantly impaired. This was reflected in the accounts of the participant group, despite having the additional diagnosis of

delayed puberty. Although all five participants expressed a desire to change their physical attributions, they appeared to be relatively content with the life they lived and who they were.

Ferguson (2005) questions whether the use of growth hormones is necessary or if it is a “designer therapy” (p24). Current guidelines regarding the prescribing of growth hormone for children with short stature note that the decision to commence treatment should be informed by the improvement it would have on the adolescents’ quality of life (Furlanetto & Drug, 1995). Rosen & Foster (2001) reported that adolescents with delayed puberty are at risk of psychosocial difficulties and they should therefore be “sought actively” at assessment (p314). This raises the question of how psychosocial stress and the adolescents’ quality of life is currently being assessed to ensure that those who are at risk are being identified in a timely manner and offered the appropriate support.

Limitations

Although this study provides a unique insight into the problems and issues surrounding delayed puberty as seen by the young people concerned it is not without limitations. Adolescents were recruited from the endocrine clinic at the Royal Hospital for Sick Children therefore they are already engaged with services and have received varying degrees of explanation with regard to their symptoms. Despite reporting ongoing uncertainties regarding their diagnosis and treatment, this may have positively influenced their narratives regarding the emotional consequences of delayed puberty. Also, as noted earlier, the main reason given for adolescents declining to participate was that having delayed puberty is something that makes them feel uncomfortable/upset and they did not wish to discuss it in-depth. The

inclusion of these adolescents may have provided a different, more negative perspective on the impact of delayed puberty.

The sample size recruited was smaller than originally anticipated; however, five participants were sufficient to conduct a detailed IPA. The sample was also homogeneous and therefore the results cannot be generalised to the wider population (Smith et al, 2009). It would therefore be important to recruit a wider demographic of adolescents in order to gain a more in-depth understanding of how delayed puberty impacts the lives of young people.

Implications for Clinical Practice

The individuals' experiences can be used to inform both research and clinical practice with other patients as well as the participant group. The feelings of confusion reported surrounding diagnosis and treatment stresses the importance of adolescents receiving information about delayed puberty as soon as possible when they first attend the endocrine clinic. Information regarding what delayed puberty is, the cause and prevalence of the condition, as well as specific details of treatment and the timescales involved, play a significant role in managing the psychological and emotional consequences reported.

All adolescents reported an emotional, behavioural, social and psychological impact of delayed puberty. This, along with their dissatisfaction with their physical appearance and the use of maladaptive coping strategies, highlights the importance of psychological input for this patient group. This could support the adolescents in managing the difficulties identified in the 5 super-ordinate themes. Psychological input could usefully be applied with regard to the

following issues raised by the participants: confusion around the diagnosis, body image, self-esteem and managing difficulties at school such as bullying.

Conclusion

This study has provided an insight into the experiences of adolescents with delayed puberty. Delayed puberty does not appear to globally impact the adolescents' lives, however, a range of emotional, behavioural, social and psychological consequences were reported. This study emphasises the importance of adolescents receiving clear information about delayed puberty and its treatment as soon as possible when they first attend the Endocrine Clinic to help manage the feelings of confusion they report surrounding their diagnosis. This uncertainty may be contributing to the emotional symptoms they experience. Further research is needed into the diagnostic process from the service users' perspective in order to explore their expectations and the limitations of present practice: such a focus would play a major role in informing and developing the clinical process.

References

Apter, A., Galatzer, A., Beth-Halachmi, N. & Lavron, Z. (1981). Self-Image in adolescents with delayed puberty and growth retardation. *Journal of Youth and Adolescence*. **10** (6). 501-505.

Bramswig, J. & Dubbers, A. (2009). Disorders of Pubertal Development. *Deutsches Arzteblatt International*. **106** (17), 295-304.

Christie, V. & Viner, R. (2005). ABC of Adolescent Development. *British Medical Journal*. **330**, 301-304.

Downie, B., Mulligan, J., Stratford, R., Betts, P. & Voss, L. (1997). Are short normal children at a disadvantage? The Wessex Growth Study. *British Medical Journal*. **314**, 97-100.

Ferguson, L. (2011). Growth Hormone use in children: Necessary or designer therapy? *Journal of Paediatric Health Care*. **25**, 24-30.

Furlanetto, R. & Drug, A. (1995). Guidelines for the use of growth hormone in children with short stature. A report by the Drug and Therapeutics Committee of the Lawson Wilkins Paediatric Endocrine Society. *Journal of Paediatrics*. **127**, 857-867.

Holmes, C., Hayford, J. & Thomson, R. (1982). Parents and teachers differing views of short children's behaviour. *Child: Care, Health and Development*. **8**, 327-336.

Holmes, C., Karlsson, J. & Thomson, R. (1985). School and school competencies in children with short stature. Longitudinal patterns. *Journal of Developmental and Behavioural Paediatrics*. **6**, 263-267.

Leventhal, H., Diefenbach, M. & Leventhal, E. A. (1992). Illness cognition: using common sense to understand treatment adherence and affect cognition interactions. *Cognitive Therapy and Research*. **16**, 143-163.

Meyer-Bahlburg, H. (1990). *Short stature: psychological issues*. In F. Lifshitz (Eds). *Paediatric Endocrinology. A Clinical Guide 2nd edn*. pp 173-196. New York. Marcel Dekker.

Palmert, M. & Dunkel, L. (2012). Delayed Puberty. *New England Journal of Medicine*. **366**, 443-53.

Rosen, D. & Foster, C. (2001). Delayed Puberty. *Paediatrics in Review*. **22** (9), 309-315.

Rosenblum, G. & Lewis, M. (1999). The relations among body image, physical attractiveness, and body mass in adolescence. *Child Development*. **70** (1), 50-64.

Rosenfeld, R. G., Gregory, B., Northcraft, B. A. & Hintz, R. L. (1982). A Prospective, Randomized Study of Constitutional Delay of Growth and Development in Male Adolescents. *Paediatrics*. **69** (6), 681-687.

Sandberg, D. (2000). Should short children who are not deficient in growth hormone be treated? *Western Journal of Medicine*. **172**, 186-189.

Sandberg, D. & Colsman, M. (2005). Growth hormone treatment of short stature: status of the quality of life rationale. *Hormone Research*. **63**, 275-283.

Sandberg, D., Colsman, M. & Voss, L. (2004) *Short Stature and Quality of Life: A Review of Assumptions and Evidence*. In O. Pescovitz, E. Eugster (Eds). *Paediatric Endocrinology: Mechanisms, Manifestations, and Management*. Philadelphia: Lippincourt. Williams & Wilkins. pp 191–202.

Sandberg, D. & Michael, P. (1998). *Psychosocial stresses related to short stature: Does their presence imply psychiatric dysfunction?* In D. Drotar (Ed): *Assessing Paediatric Health-Related Quality of Life and Functional Status: Implications for Research*. Mahwah, Lawrence Erlbaum Assoc. p 287–312.

Sander, D. & Voss, A. (2002). The psychosocial consequences of short stature: a review of the evidence. *Best Practice and Research Clinical Endocrinology and Metabolism*. **16** (3). 449-463.

Sedlmeyer, I. & Palmert, M. (2002). Delayed Puberty: Analysis of a Large Case Series from an Academic Centre. *The Journal of Clinical Endocrinology & Metabolism*. **87** (4), 1613-1620.

Smith, J. A. (1996). Beyond the divide between cognition and discourse: using interpretative phenomenological analysis in health psychology. *Psychology and Health*. **11**, 261-271.

Smith, J. A., Flowers, P. & Larkin, M. (2009). *Interpretative Phenomenological Analysis: Theory, Method and Research*. SAGE Publication Ltd. London

Smith, J. A. & Osborn, M. (2003). *Interpretative Phenomenological Analysis*. In J. A.

Smith (Ed.). *Qualitative Psychology: A Practical Guide to Research Methods*. London: Sage. p.51-80.

Suris, J. C., Michaud, P. A. & Viner. (2004). The adolescent with a chronic condition. Part 1: developmental issues. *Archives of Disease in Childhood*. **89**, 938-942.

Visser-van Balen, H., Sinnema, G. & Geenen, R. (2006). Growing up with idiopathic short stature: psychosocial development and hormone treatment: a critical review. *Archives of Disease in Childhood*. **91**, 433–439.

Voss, L., Bailey, B., Mulligan, J., Wilkin, T. & Betts, P. (1991). Short stature and school performance: The Wessex Growth Study. *Acta Paediatr Scand. Suppl.* **370**, 29-31.

Voss, L. & Mulligan, J. (2000) Bullying in school: are short pupils at risk? Questionnaire study in a cohort. *British Medical Journal*. **320**, 612–613.

Willig, C. (2001). *Introducing Qualitative Research in Psychology: Adventures in Theory and Method*. Open University Press: Buckingham.

Wit, J., Reiter, E., Ross, J., Saenger, P., Savage, M., Rogol, A. & Cohen, P. (2008). Idiopathic Short Stature: Management and Growth Hormone Treatment. *Growth Hormone & IGF Research*. **18**, 111-135

Zimet, G., Cutler, M., Litvene, M., Dahms, W., Owens, R. & Cuttler, L. (1995). Psychological adjustment of children evaluated for short stature: a preliminary report. *Journal of Developmental & Behavioural Paediatrics*. **16**, 264–270.

CHAPTER THREE: ADVANCED PRACTICE I: REFLECTIVE ACCOUNT

(Abstract only)

Common Practice is Not Necessarily Best Practice

Abstract: Clinical Psychologists are professionals who support individuals to make sense of, and manage, their distressing thoughts and feelings in order to promote psychological wellbeing. They enable individuals to communicate difficult experiences and, through using evidenced-based theories and models, help them to process their thoughts and feelings to reduce the level of distress and the negative impact they are having on their lives.

The British Psychological Society's National Occupational Standards (BPS, 2006) are helpful when considering what the key roles of a Clinical Psychologist are. Regularly reflecting on practice is important for all Clinical Psychologists, especially when training, to not only be aware of their own feelings/actions and the impact they have on the therapeutic process, but also to consider how their skills are developing in line with the key roles of their profession.

Writing this account has highlighted that reflective practice has become an integral part of my clinical work. I am more mindful of my thoughts and feelings as events are unfolding and I am able to make sense of them there and then to inform my next action. Supervision is then key in enabling me to, not only reflect further on my thoughts/feelings and on the ongoing development of myself as a professional, but also to consider the implications for the wider system.

In this account I consider being mindful of standard common procedures within departments and reflecting on whether these are best practice. I consider that doing so can impact a patient's experience of a service and ultimately their engagement and intervention. As I do in this account, striving to reflect on routine procedures and exploring whether they are the most appropriate will enable me to best contain the distress patients experience and therefore focus on supporting them to move forward.

CHAPTER FOUR: ADVANCED PRACTICE II: REFLECTIVE ACCOUNT

(Abstract only)

Clinical Psychologists: Jack of all Trades, Master of None?

Abstract: When I embarked upon my doctorate training I thought I would be predominantly working with patients in direct therapy, however, as I approach the end of my training I recognise now that there are many other roles I will also be engaging in. These include training, research and management. These roles, as defined by the British Psychological Society's National Occupation Standards (BPS, 2006), have implications within the wider professional and political context. Rolfe, Freshwater & Jasper's (2001) framework, as well as supervision, has been helpful in enabling me to consider these roles, not only their implication for me as a Clinical Psychologist, but also Clinical Psychology as a profession. I have learned that the skills and qualities required to deliver effective therapeutic interventions are transferable to the other roles that I will take on. I recognise that the profession needs to engage in Continual Professional Development and be regulated by the Health Professionals' Council in order to continue developing and consolidating knowledge of psychological theories and models as well as the skills to apply these within each of their roles.

Although it could be thought that because psychologists take on so many roles they are a 'Jack of all trades and a master of none', I am now of the mindset that Clinical Psychologists are 'masters' of disseminating psychological theories and models to promote psychological well being. This can be done in many different ways and through many different roles. Therefore they are no longer always the best person to engage in direct clinical work with patients.

CHAPTER FIVE: APPENDICES

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Appendix 1: Author Publication Guidelines

British Journal of Health Psychology

Author Guidelines

The aim of the British Journal of Health Psychology is to provide a forum for high quality research relating to health and illness. The scope of the journal includes all areas of health psychology across the life span, ranging from experimental and clinical research on aetiology and the management of acute and chronic illness, responses to ill-health, screening and medical procedures, to research on health behaviour and psychological aspects of prevention. Research carried out at the individual, group and community levels is welcome, and submissions concerning clinical applications and interventions are particularly encouraged.

The types of paper invited are:

- papers reporting original empirical investigations;
- theoretical papers which may be analyses or commentaries on established theories in health psychology, or presentations of theoretical innovations;
- review papers, which should aim to provide systematic overviews, evaluations and interpretations of research in a given field of health psychology; and
- methodological papers dealing with methodological issues of particular relevance to health psychology.

1. Circulation

The circulation of the Journal is worldwide. Papers are invited and encouraged from authors throughout the world.

2. Length

Papers should normally be no more than 5000 words (excluding the abstract, reference list, tables and figures), although the Editor retains discretion to publish papers beyond this length in cases where the clear and concise expression of the scientific content requires greater length.

3. Editorial policy

The Journal receives a large volume of papers to review each year, and in order to make the process as efficient as possible for authors and editors alike, all papers are initially examined by the Editors to ascertain whether the article is suitable for full peer review. In order to qualify for full review, papers must meet the following criteria:

- the content of the paper falls within the scope of the Journal
- the methods and/or sample size are appropriate for the questions being addressed
- research with student populations is appropriately justified
- the word count is within the stated limit for the Journal (i.e. 5000 words)

4. Submission and reviewing

All manuscripts must be submitted via [Editorial Manager](#). You may like to use the [Submission Checklist](#) to help you prepare your manuscript. The Journal operates a policy of anonymous peer review. Authors must suggest three reviewers when submitting their manuscript, who may or may not be approached by the Associate Editor dealing with the paper. Before submitting, please read the [terms and conditions of submission](#) and the [declaration of competing interests](#).

5. Manuscript requirements

- Contributions must be typed in double spacing with wide margins. All sheets must be numbered.
- Manuscripts should be preceded by a title page which includes a full list of authors and their affiliations, as well as the corresponding author's contact details. A template can be downloaded from [here](#).
- **Statement of Contribution:** All authors are required to provide a clear summary of 'what is already known on this subject?' and 'what does this study add?'. The 2-3 (maximum) sentences for each point should identify existing research knowledge relating to the specific research question/topic and a summary of the new knowledge added by your study. Under each of these headings, please provide 2-3 clear outcome statements (not process statements of what the paper does); the statements for 'what does this study add?' should be presented as bullet points of no more than 100 characters each. The Statement of Contribution should be a separate file.
- Tables should be typed in double spacing, each on a separate page with a self-explanatory title. Tables should be comprehensible without reference to the text. They should be placed at the end of the manuscript with their approximate locations indicated in the text.
- Figures can be included at the end of the document or attached as separate files, carefully labelled in initial capital/lower case lettering with symbols in a form consistent with text use. Unnecessary background patterns, lines and shading should be avoided. Captions should be listed on a separate sheet. The resolution of digital images must be at least 300 dpi.
- For articles containing original scientific research, a structured abstract of up to 250 words should be included with the headings: Objectives, Design, Methods, Results, Conclusions. Review articles should use these headings: Purpose, Methods, Results, Conclusions.
- For reference citations, please use APA style. Particular care should be taken to ensure that references are accurate and complete. Give all journal titles in full and provide doi numbers where possible for journal articles.
- SI units must be used for all measurements, rounded off to practical values if appropriate, with the imperial equivalent in parentheses.
- In normal circumstances, effect size should be incorporated.
- Authors are requested to avoid the use of sexist language.
- Authors are responsible for acquiring written permission to publish lengthy quotations, illustrations, etc. for which they do not own copyright. For guidelines on editorial style, please consult the [APA Publication Manual](#) published by the American Psychological Association.
- Manuscripts describing clinical trials are encouraged to submit in accordance with the CONSORT statement on reporting randomised controlled trials (<http://www.consort-statement.org>).

6. Supporting Information

Supporting Information can be a useful way for an author to include important but ancillary information with the online version of an article. Examples of Supporting Information include appendices, additional tables, data sets, figures, movie files, audio clips, and other related nonessential multimedia files. Supporting Information should be cited within the article text, and a descriptive legend should be included. Please indicate clearly on submission which material is for online only publication. It is published as supplied by the author, and a proof is not made available prior to publication; for these reasons, authors should provide any Supporting Information in the desired final format.

For further information on recommended file types and requirements for submission, please visit: <http://authorservices.wiley.com/bauthor/supinfo.asp>

7. Copyright

Authors will be required to assign copyright to The British Psychological Society. Copyright assignment is a condition of publication and papers will not be passed to the publisher for production unless copyright has been assigned. To assist authors an appropriate copyright assignment form will be supplied by the editorial office and is also available on the journal's website at http://www.blackwellpublishing.com/pdf/CTA_BPS.pdf. Government employees in both the US and the UK need to complete the Author Warranty sections, although copyright in such cases does not need to be assigned.

8. Colour illustrations

Colour illustrations can be accepted for publication online. These would be reproduced in greyscale in the print version. If authors would like these figures to be reproduced in colour in print at their expense they should request this by completing a Colour Work Agreement form upon acceptance of the paper. A copy of the Colour Work Agreement form can be downloaded [here](#).

9. Pre-submission English-language editing

Authors for whom English is a second language may choose to have their manuscript professionally edited before submission to improve the English. A list of independent suppliers of editing services can be found at http://authorservices.wiley.com/bauthor/english_language.asp. All services are paid for and arranged by the author, and use of one of these services does not guarantee acceptance or preference for publication.

10. OnlineOpen

OnlineOpen is available to authors of primary research articles who wish to make their article available to non-subscribers on publication, or whose funding agency requires grantees to archive the final version of their article. With OnlineOpen, the author, the author's funding agency, or the author's institution pays a fee to ensure that the article is made available to non-subscribers upon publication via Wiley Online Library, as well as deposited in the funding agency's preferred archive. For the full list of terms and conditions, see http://wileyonlinelibrary.com/onlineopen#OnlineOpen_Terms. Any authors wishing to send their paper OnlineOpen will be required to complete the payment form available from our website at: https://authorservices.wiley.com/bauthor/onlineopen_order.asp. Prior to acceptance there is no requirement to inform an Editorial Office that you intend to publish your paper OnlineOpen if you do not wish to. All OnlineOpen articles are treated in the same way as any other article. They go through the journal's standard peer-review process and will be accepted or rejected based on their own merit.

11. Author Services

Author Services enables authors to track their article – once it has been accepted – through the production process to publication online and in print. Authors can check the status of their articles online and choose to receive automated e-mails at key stages of production. The author will receive an e-mail with a unique link that enables them to register and have their article automatically added to the system. Please ensure that a complete e-mail address is provided when submitting the manuscript. Visit <http://authorservices.wiley.com/bauthor/> for more details on online production tracking and for a wealth of resources including FAQs and tips on article preparation, submission and more.

12. The Later Stages

The corresponding author will receive an email alert containing a link to a web site. A working e-mail address must therefore be provided for the corresponding author. The proof can be downloaded as a PDF (portable document format) file from this site. Acrobat Reader will be required in order to read this file. This software can be downloaded (free of charge) from the following web site: <http://www.adobe.com/products/acrobat/readstep2.html>. This will enable the file to be opened, read on screen and annotated direct in the PDF. Corrections can also be supplied by hard copy if preferred. Further instructions will be sent with the proof. Hard copy proofs will be posted if no e-mail address is available. Excessive changes made by the author in the proofs, excluding typesetting errors, will be charged separately.

13. Early View

British Journal of Health Psychology is covered by the Early View service on Wiley Online Library. Early View articles are complete full-text articles published online in advance of their publication in a printed issue. Articles are therefore available as soon as they are ready, rather than having to wait for the next scheduled print issue. Early View articles are complete and final. They have been fully reviewed, revised and edited for publication, and the authors' final corrections have been incorporated. Because they are in final form, no changes can be made after online publication. The nature of Early View articles means that they do not yet have volume, issue or page numbers, so they cannot be cited in the traditional way. They are cited using their Digital Object Identifier (DOI) with no volume and issue or pagination information. Eg Jones, A.B. (2010). Human rights Issues. *Journal of Human Rights*. Advance online publication. doi:10.1111/j.1467-9299.2010.00300.x

Further information about the process of peer review and production can be found in this document. [What happens to my paper?](#)

Appendix 2: Walsh & Downe's (2006) Quality Criteria for Qualitative Studies.

Studies were awarded 1 point if the criterion was met and 0 if the criterion was not met.

Papers were given a score out of 30 and given a rating of good (>75%), acceptable (>50%) or poor (<50%).

Stage	Essential criteria	Specific prompts	Scoring 1 if present, 0 is
Scope and purpose	<p>Clear statement of focus, and rationale for research question/aims/purposes</p> <p>Study thoroughly contextualised by existing literature</p>	<p>Clarity of focus demonstrated?</p> <p>Rational of purpose?</p> <p>Link between research and existing knowledge based?</p>	
Design	<p>Method/design apparent, and consistent with research intent</p> <p>Data collection strategy apparent and appropriate</p>	<p>Method/design apparent?</p> <p>Rational explored for specific method?</p> <p>Appropriate - consistent with research intent?</p> <p>Data collection strategy apparent?</p> <p>Appropriate - likely to capture the complexity/diversity of experience?</p>	
Sampling strategy	<p>Sample and sampling method appropriate</p>	<p>Description of how sampling was undertaken?</p> <p>Justification for sampling strategy given?</p> <p>Appropriate - thickness of description likely to be achieved?</p>	
Analysis	<p>Analytic apparent and appropriate</p>	<p>Approach made explicit?</p> <p>Appropriate for the qualitative method chosen?</p> <p>Evidence of more than one researcher involved in stages?</p>	

		<p>Did research participants have any involvement in analysis?</p> <p>Evidence provided that data reached saturation or discussion/rationale if it did not?</p>	
Interpretation	<p>Context described and taken account of in interpretation</p> <p>Clear audit trail given</p> <p>Data used to support interpretation</p>	<p>Description of social/physical and interpersonal contexts of data collection?</p> <p>Sufficient discussion of research processes such that others can follow 'decision trail'?</p> <p>Extensive use of field notes entries/verbatim interview quotes in discussion of findings?</p> <p>Clear exposition of how interpretation led to conclusions?</p>	
Reflexivity	Researcher reflectivity demonstrated	<p>Demonstration of researcher's influence on stages of research process?</p> <p>Evidence of self-awareness/insight?</p>	
Ethical dimensions	Demonstration of sensitivity to ethical concerns	<p>Ethical committee approval granted?</p> <p>Documentation of consent recorded?</p> <p>Description of how autonomy and confidentiality were managed?</p>	
Relevance and transferability	Relevance and transferability evident	<p>Analysis interwoven with existing theories and other relevant explanatory literature drawn from similar settings and studies</p> <p>Interpretation plausible and 'makes sense'?</p> <p>Provides new insights and increases understanding?</p>	

		Limitations/weaknesses of study clearly outlined? Outlines further directions for investigation?	
Total (out of 30)			

Appendix 3: Letters of Approval from WoSREC and Research and Development

WoSRES

West of Scotland Research Ethics Service



Dr Sarah Wilson
Senior Lecturer in Health Psychology
Mental Health & Well Being
Gartnavel Royal Hospital
1055 Great Western Road
Glasgow
G12 0XH

West of Scotland REC 5

Ground Floor - Tennent Building
Western Infirmary
38 Church Street
Glasgow
G11 6NT

Date 24 May 2012

Direct line 0141 211 2102
Fax 0141 211 1847
E-mail sharon.macgregor@ggc.scot.nhs.uk

Dear Dr Wilson

Study Title: An exploration of the psychosocial consequences of delayed puberty
REC reference: 12/WS/0128

The Research Ethics Committee reviewed the above application at the meeting held on 16 May 2012. Thank you to Ms McKillop for attending to discuss the study.

Documents reviewed

The documents reviewed at the meeting were:

Document	Version	Date
Covering Letter	-	01 May 2012
Interview Schedules/Topic Guides	1	24 April 2012
Investigator CV	-	01 May 2012
Letter of invitation to participant	1	24 February 2012
Other: Dr Kathleen McHugh's CV	-	18 April 2012
Other: Ms Kirsten Ann McKillop's CV	-	01 May 2012
Other: Letter to Parent/Guardian	1	24 February 2012
Other: Opt-in form	1	24 February 2012
Other: Parent Assent Form	1	24 February 2012
Other: Letter re study from Professor Tom McMillan	-	27 February 2012
Other: Flow Chart	1	24 February 2012
Participant Consent Form	1	24 February 2012
Participant Information Sheet	1	24 February 2012
Participant Information Sheet: Parent/Guardian	1	24 February 2012
Protocol	1	24 February 2012
REC application	-	13 April 2012

Provisional opinion

To summarise the main points of Ms McKillop's discussion with the Committee:

- Ms McKillop was asked how representative is this sample group of the condition and she advised that, since this is not known, this is what the researchers hope to find out during this study.
- Since data shows that only 1 to 2% of patients attending the Endocrine Clinic at Yorkhill have only delayed puberty, it is likely that most patients will have other conditions in which delayed puberty is a symptom. Therefore, anyone from the Endocrine clinic with delayed puberty will be invited to attend.
- With regards to the questions to be asked at the interview, Ms McKillop advised that they will be asked in much the same way that they are written in the topic guide but will be asked in a warm, non-judgemental and non-direct way. The children will be asked only what their view is of the issues.
- The questions have been checked by the clinic consultants and psychologists and have a readability score of 9 years old. The children will be familiar with the term "delayed puberty".

The Committee would be content to give a favourable ethical opinion of the research, subject to receiving a complete response to the request for further information set out below.

The Committee delegated authority to confirm its final opinion on the application to the Chair and lead reviewer.

Further information or clarification required

1. It is suggested that the title of the study should be amended to be more representative of the study population and include "in children who attend the Yorkhill Endocrine Clinic".
2. Although it is noted that the researchers will try to schedule the interviews for the same day as the returning clinic appointment, the Committee felt that it is very important to ensure this as it will reduce time out of school, parental time of work and travelling time. Since the hospital has a large catchment area, it can involve long journeys for some families.

Invitation letters:

3. The first paragraph implies that the interviews are routine and therefore it should instead clearly state that this for a research study being undertaken for Ms McKillop's PhD.
4. The participant letter should also state that they are being asked to *consider* arranging an interview, as it is stated in the Parent/Guardian letter.
5. The sentence in both letters that states "This will help us make services better.." is rather overstated, as this is not known. It should be changed to "*may* help us..".

Participant Information Sheet:

6. With regards to some of the words such as "psychologically" and "romantic attachments", 13-18 year olds may not use or understand these terms and it was suggested that these are changed.

Parent/Guardian Information Sheet:

7. On page 2 of the document, the word "help" is missing from the first heading.
8. Both sheets:
9. There is no mention of a possible second interview, as stated in A21 of the application.
10. There is no mention of what will happen to the audio tapes after the interview.
11. Since all studies are voluntary, the answer to "Will I be paid for taking part?" should simply state that no payment will be made.
12. In the "Who has reviewed the study?", this should state "by the West of Scotland Research Ethics Committee". There are no multicentre and local RECs now.
13. The document should state that the researcher will try to arrange the interview for the same day as a normal clinic appointment.

Participant Consent form:

14. It is not necessary for the parent/guardian to sign the document and therefore this section should be removed.
15. In the second last statement, the word "of" is missing from the end of the first line.

Parent Assent Form:

16. A sub-header of "for under 16 year olds" should be inserted below the main heading of "Parent Assent Form".

Interview Topic Guide:

17. It is suggested that a prompt for the researcher is added to advise the child that they don't have to answer a question.
18. It is also suggested that question 5 is asked before question 3.

If you would find it helpful to discuss any of the matters raised above or seek further clarification from a member of the Committee, you are welcome to contact Mrs Sharon Macgregor, Committee Coordinator on any of the contact details on this letter.

When submitting your response to the Committee, please send revised documentation where appropriate underlining or otherwise highlighting the changes you have made and giving revised version numbers and dates.

If the committee has asked for clarification or changes to any answers given in the application form, please do not submit a revised copy of the application form; these can be addressed in a covering letter to the REC.

The Committee will confirm the final ethical opinion within a maximum of 60 days from the date of initial receipt of the application, excluding the time taken by you to respond fully to the above points. A response should be submitted by no later than 21 September 2012.

Membership of the Committee

The members of the Committee who were present at the meeting are listed on the attached sheet.

Statement of compliance

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

12/WS/0128

Please quote this number on all correspondence

Yours sincerely

A handwritten signature in black ink, appearing to read 'S Macgregor'.

Dr Gregory Ofili
Chair

Enclosures: List of names and professions of members who were present at the meeting and those who submitted written comments.

Copy to: Ms Joanne McGarry, Research & Development Management Office

Dr Sarah Wilson
Senior Lecturer in Health Psychology
Mental Health & Well Being
Gartnavel Royal Hospital
1055 Great Western Road
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G12 0XH

West of Scotland REC 5

Ground Floor - Tennent Building
Western Infirmary
38 Church Street
Glasgow
G11 6NT

Date 03 July 2012

Direct line 0141 211 2102

Fax 0141 211 1847

E-mail sharon.macgregor@ggc.scot.nhs.uk

Dear Dr Wilson

Study title: An exploration of the psychosocial consequences of delayed puberty
REC reference: 12/WS/0128

I refer to Ms McKillop's letter of 18 June 2012, 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 and Lead Reviewer.

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

NHS 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).

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.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study 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 a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

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:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Covering Letter	-	01 May 2012
Interview Schedules/Topic Guides	2	13 June 2012
Investigator CV	-	01 May 2012
Letter of invitation to participant	2	13 June 2012
Other: Dr Kathleen McHugh's CV	-	18 April 2012
Other: Ms Kirsten Ann McKillop's CV	-	01 May 2012
Other: Letter re study from Professor Tom McMillan	-	27 February 2012
Other: Parent/Guardian Invitation letter	2	13 June 2012
Other: Participant opt in form	2	13 June 2012
Other: Parent/Guardian Assent Form	2	13 June 2012
Participant Consent Form	2	13 June 2012
Participant Information Sheet	2	13 June 2012
Participant Information Sheet: Parent/Guardian Information sheet	2	13 June 2012
Protocol (front cover)	2	13 June 2012
REC application	-	13 April 2012
Response to Request for Further Information		18 June 2012
Summary/Synopsis	2	13 June 2012

Statement of compliance

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

After ethical review

Reporting requirements

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
- Notification of serious breaches of the protocol
- 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.

Feedback

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.

Further information is available at National Research Ethics Service website > After Review

12/WS/0128

Please quote this number on all correspondence

With the Committee's best wishes for the success of this project

Yours sincerely



for
Dr Gregory Ofili
Chair

Enclosures: "After ethical review – guidance for researchers"

Copy to: Ms K McKillop, NHS GGC
Ms Joanne McGarry, Research & Development Management Office

R&D Management Office
Western Infirmary
Tennent Institute
1st Floor, 38 Church Street
Glasgow, G11 6NT



Coordinator/Administrator: JMcG/LR
Direct Line: 0141 211 8548
E-mail: Joanne.McGarry@ggc.scot.nhs.uk
Website: www.nhsggc.org.uk/r&d

5th July 2012

Dr Kathleen McHugh
Principal Clinical Psychologist
Dept of Clinical Psychology
RHSC
Dalnair Street
Glasgow
G3 8SJ

NHS GG&C Board Approval

Dear Dr McHugh

Study Title: An exploration of the psychosocial consequences of delayed puberty
Chief Investigator: Dr Sarah Wilson
GG&C HB site: RHSC
Sponsor: NHS GG&C Health Board
R&D Reference: GN12KH106
REC Ref: 12/WS/0128
Protocol no: V2 dated 13/06/12

I am pleased to confirm that Greater Glasgow & Clyde Health Board is now able to grant **Approval** for the above study.

Conditions of Approval

1. **For Clinical Trials** as defined by the Medicines for Human Use Clinical Trial Regulations, 2004
 - a. During the life span of the study GGHB requires the following information related solely to this site
 - i. Notification of any potential serious breaches.
 - ii. Notification of any regulatory inspections.

It is your responsibility to ensure that all staff involved in the study at this site have the appropriate GCP training according to the GGHB GCP policy (www.nhsggc.org.uk/content/default.asp?page=s1411), evidence of such training to be filed in the site file.

2. **For all studies** the following information is required during their lifespan.
 - a. Recruitment Numbers on a monthly basis
 - b. Any change of staff named on the original SSI form
 - c. Any amendments – Substantial or Non Substantial

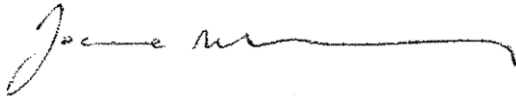
- d. Notification of Trial/study end including final recruitment figures
- e. Final Report & Copies of Publications/Abstracts

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

Your personal information will be held on a secure national web-based NHS database.

I wish you every success with this research study

Yours sincerely

A handwritten signature in black ink, appearing to read 'Joanne McGarry', with a long horizontal flourish extending to the right.

Joanne McGarry
Research Co-ordinator

Cc: Dr Sarah Wilson, Chief Investigator, Glasgow
Ms Kirsten McKillop, Student, Glasgow

Ms Kirsten McKillop
Trainee Clinical Psychologist
Specialist Children's Services
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West of Scotland REC 5

Ground Floor - Tennent Building
Western Infirmary
38 Church Street
Glasgow
G11 6NT

Date 11 January 2013

Direct line 0141 211 2102
E-mail sharon.macgregor@ggc.scot.nhs.uk

Dear Ms McKillop

Study title: An exploration of the psychosocial consequences of delayed puberty
REC reference: 12/WS/0128
Amendment number: Minor
Amendment date: 10 January 2013
IRAS project ID: 101819

Summary of Amendment: Extension of study to end of March 2013

Thank you for your email of 10 January 2013, notifying the Committee of the above amendment.

The Committee does not consider this to be a "substantial amendment" as defined in the Standard Operating Procedures for Research Ethics Committees. The amendment does not therefore require an ethical opinion from the Committee and may be implemented immediately, provided that it does not affect the approval for the research given by the R&D office for the relevant NHS care organisation.

Documents received

The documents received were as follows:

Document	Version	Date
Notification of a Minor Amendment		10 January 2013

Statement of compliance

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

12/WS/0128:

Please quote this number on all correspondence

Yours sincerely

A handwritten signature in black ink, appearing to read 'S Macgregor'.

**Mrs Sharon Macgregor
Committee Co-ordinator**

Copy to: Ms Joanne McGarry, Research & Development Management Office
Dr Sarah Wilson, NHS GGC

Dr Sarah Wilson
Senior Lecturer in Health Psychology
University of Glasgow
Mental Health & Well Being
Gartnavel Royal Hospital
1055 Great Western Road
Glasgow
G12 0XH

West of Scotland REC 5

Ground Floor - Tennent Building
Western Infirmary
38 Church Street
Glasgow
G11 6NT

Date 17 January 2013

Direct line 0141 211 2102
E-mail sharon.macgregor@ggc.scot.nhs.uk

Dear Dr Wilson

Study title: An exploration of the psychosocial consequences of delayed puberty
REC reference: 12/WS/0128
Amendment number: 1
Amendment date: 10 December 2012
IRAS project ID: 101819

The above amendment was reviewed by the Sub-Committee in correspondence.

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
Protocol	3	10 December 2012
Notice of Substantial Amendment (non-CTIMPs)	1	10 December 2012
Covering Letter		01 May 2012

Membership of the Committee

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

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 and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at <http://www.hra.nhs.uk/hra-training/>

12/WS/0128:

Please quote this number on all correspondence

Yours sincerely



for
Dr Gregory Ofili
Chair

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

Copy to: Ms Joanne McGarry, Research & Development Management Office

West of Scotland REC 5

Sub-Committee Members who took part in the review

Written comments received from:

<i>Name</i>	<i>Position</i>
Professor Pauline Banks	Reader (Older Persons' Health)
Mr Sandy Morton	Retired (Teacher)
Dr Gregory Ofili (CHAIR)	Consultant Gynaecologist

R&D Ref: GN12KH106 - AM01

Reid, Lorraine [Lorraine.Reid2@ggc.scot.nhs.uk]

Sent: 21 January 2013 11:40

To: McHugh, Kathleen [Kathleen.McHugh@ggc.scot.nhs.uk]

Cc: Sarah Wilson; Kirsten McKillop

Dear Dr McHugh

R&D Ref: GN12KH106

Ethics Ref: 12/WS/0128

Chief Investigator: Dr Sarah Wilson

Project Title: An exploration of the psychosocial consequences of delayed puberty

Protocol Number: V3 dated 10/12/12

Amendment: AM01 dated 10/12/12

Sponsor: NHS GG&C Health Board

I am pleased to inform you that R&D have reviewed the above study Amendment and can confirm that Management Approval is still valid for this study.

Reviewed Documents:	Version	Dated
Notification of Amendment form	V3.4	20/12/12
Protocol	V3	10/12/12
REC favourable opinion letter	Not versioned	17/01/13

I wish you every success with this research project.

Yours sincerely

Research & Development

R&D Management Office

1st Floor, Tennent Institute


Western Infirmary

Glasgow

G11 6NT

Tel: 0141 211 6372

Please visit our website for further information

 www.nhsggc.org.uk/r&d

 Please consider the environment before printing this e-mail or its attachment(s)

CC: Dr Sarah Wilson, Chief Investigator, Glasgow
Ms Kirsten McKillop, Student, Glasgow

NHSGG&C Disclaimer

The information contained within this e-mail and in any attachment is confidential and may be privileged. If you are not the intended recipient, please destroy this message, delete any copies held on your systems and notify the sender immediately; you should not retain, copy or use this e-mail for any purpose, nor disclose all or any part of its content to any other person.

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R&D Ref: GN12KH106 - Minor Amendment dated 30/01/13

Reid, Lorraine [Lorraine.Reid2@ggc.scot.nhs.uk]

Sent: 14 February 2013 12:08

To: McHugh, Kathleen [Kathleen.McHugh@ggc.scot.nhs.uk]

Cc: sarah.wilson@glasgow.ac.uk ['sarah.wilson@glasgow.ac.uk']; kirsten.mckillop@ggc.scot.nhs; Kirsten McKillop

Dear Dr McHugh

R&D Ref: GN12KH106

Ethics Ref: 12/WS/0128

Chief Investigator: Dr Sarah Wilson

Project Title: An exploration of the psychosocial consequences of delayed puberty

Protocol Number: V3 dated 10/12/12

Amendment: Minor Amendment dated 30/01/13

Sponsor: NHS GG&C Health Board

I am pleased to inform you that R&D have reviewed the above study Amendment and can confirm that Management Approval is still valid for this study.

Reviewed Documents:	Version	Dated
Email summarising Minor Amendment	Not versioned	30/01/13

I wish you every success with this research project.

Yours sincerely

Research & Development

R&D Management Office

1st Floor, Tennent Institute


Western Infirmary

Glasgow

G11 6NT

Tel: 0141 211 6372

Please visit our website for further information

 www.nhsggc.org.uk/r&d

 Please consider the environment before printing this e-mail or its attachment(s)

CC: Dr Sarah Wilson, Chief Investigator, Glasgow
Ms Kirsten McKillop, Student, Glasgow

NHS GG&C Disclaimer

The information contained within this e-mail and in any attachment is confidential and may be privileged. If you are not the intended recipient, please destroy this message, delete any copies held on your systems and notify the sender immediately; you should not retain, copy or use this e-mail for any purpose, nor disclose all or any part of its content to any other person.

All messages passing through this gateway are checked for viruses, but we strongly recommend that you check for viruses using your own virus scanner as NHS Greater Glasgow & Clyde will not take responsibility for any damage caused as a result of virus infection.

Appendix 4: Participant Invitation and Information Sheet



Specialist Children's Services

Confidential

Insert address of recipient

Insert date

Clinical Psychology Department

2nd Floor, Above A&E

Royal Hospital for Sick Children

Dalnair Street

Glasgow, G3 8SJ

Tel: 0141-201-0644

Dear (insert young person's name),

We are trying to find out how it makes young people feel when they have delayed puberty. We are asking everyone who comes to the Endocrine Clinic at Yorkhill to consider taking part in a research study being carried out by Kirsten McKillop as part of her Doctorate in Clinical Psychology. This study involves arranging an interview with Kirsten to answer some questions about what it is like to have delayed puberty. This may help us make services better as we can offer more support.

Before you make up your mind, it is important for you to understand what we are asking you to do and why. Please find enclosed an information sheet to tell you all about the study. Please read it and ask your parent/carer to read theirs as well. If you have any questions you can call Ethel McNeill. She is a nurse who works in the clinic but she is not involved in our research. She will be able to answer any questions you have.

Asking you to take part in this study does not mean we think you are not coping with your condition. We are asking everyone who comes to clinics and you can say "no" if you don't want to take part.

If you decide you do want to take part after reading the information sheet, you can either return the form in the enclosed envelop to the Clinical Psychology Department in Yorkhill, call Ethel on 0141 201 0245 or email Kirsten McKillop at Kirsten.McKillop@ggc.scot.nhs.uk.

When you call or email, please tell us whether you would prefer us to call you or write to you to arrange an appointment to meet at Yorkhill.

When you attend this appointment, you can find out more about the study and ask more questions before taking part. If you decide you don't want to take part, then you can say no and you won't be asked anything else. If you do decide to take part, you will then be asked some questions about the effects that having delayed puberty is having on your life. This appointment will last roughly one hour.

If you want to ask anything else about the study you can call Ethel or email Kirsten.

Thank you for reading this.

Yours sincerely,

Professor Syed Faisal Ahmed

Consultant Endocrinologist

Dr Donaldson

Consultant Endocrinologist

Dr Shaik

Consultant Endocrinologist

Dr Kathleen McHugh

Clinical Psychologist

Ms. Kirsten McKillop

Trainee Clinical Psychologist

**An exploration of the psychosocial
consequences of delayed puberty in children
who attend the Yorkhill Endocrine Clinic:**

A qualitative study
Researcher: Kirsten McKillop

Participant Information Sheet

This information sheet describes a study exploring the consequences of delayed puberty in young people that you are being invited to take part in. This study is being carried out by Kirsten McKillop as part of her Doctorate in Clinical Psychology at the University of Glasgow.

Before you decide, it is important for you to understand what we are asking you to do and why. If anything is not clear or you want more information please ask us. Thank you for reading this.

Why is this study being done?

It is known that delayed puberty can affect a young person both physically and emotionally, however, the exact impact of delayed puberty on everyday life and relationships is unknown.

What is the aim of this research project?

This study aims to explore young people's experiences of delayed puberty. This study will ask questions about your understanding of delayed puberty. We will also ask you questions about the impact it has on your family life, school life and education, social life, having a boyfriend/girlfriend and how you view yourself. The answers we get will help us plan services to offer help and support to people when they need it most.

Who are we looking for to participate in this study?

We are looking for young people attending the Endocrinology outpatient clinics at The Royal Hospital for Sick Children (RHSC), Yorkhill, who are diagnosed with delayed puberty.

Why am I being asked?

You are being asked because you have delayed puberty and come to an endocrine clinic at Yorkhill. In asking you to take part, we are not saying that you have any problems with how you feel or how you act or how you cope. We are asking everyone who comes to clinic.

Do I have to take part?

No, you do not have to take part in the study and it will not affect your medical care and treatment in any way if you say “no”. It is up to you to decide whether or not you take part. If you decide to take part in the study, you will be asked to sign a consent form, as will any grown-ups in your family if you agree to take part. You can change your mind at any time and pull out without giving a reason.

Will the study help me?

It may help the doctors, nurses and other people working in RHSC to understand some of the things that can worry or upset young people with delayed puberty. The study might help us learn about why some people find things hard and when. This could help us make services better as we can offer more support.

What will happen to you if you decide to take part in the project?

If you decide to take part you can contact the main researcher, Kirsten McKillop by phone, email or by returning the slip on the enclosed letter in the stamped addressed envelope included. You will be asked to say how you would like to be contacted by Kirsten to organise a time to meet at Yorkhill. Kirsten will try to arrange the interview for the day you will next be at Yorkhill Hospital for an appointment.

The appointment will take roughly one hour of your time and it will involve answering questions about your experience of having delayed puberty. If after the interview there is still more to talk about, you may be asked to arrange one more interview to talk about your experiences further.

We will ask your permission to record your interview to help us remember what you have told us. Your answers will then be typed up, but your name will be removed so no one will know what you said. Your recording and paper copy of your interview will be kept in a locked filing cabinet on University Grounds.

Will I be paid for taking part?

No

Who has reviewed this study?

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

Want more information?

More information about the study is available from the main researcher, Kirsten McKillop, Trainee Clinical Psychologist: Kirsten.McKillop@ggc.scot.nhs.uk

You can also contact Ethel McNeill. She is an Endocrine Nurse Specialist who works in the hospital. She is not involved in our research so she is someone you can speak to about any questions or concerns you have. You can contact her on 0141 201 0245.

Appendix 5: Parent/Guardian Invitation and Information Sheet



Specialist Children's Services

Confidential

Clinical Psychology Department

2nd Floor, Above A&E

Insert address of recipient

Royal Hospital for Sick Children

Insert date

Dalnair Street

Glasgow, G3 8SJ

Tel: 0141-201-0644

Dear Parent/Guardian,

We are trying to find out how it makes young people feel when they have delayed puberty. We are asking everyone who comes to the Endocrine Clinic at Yorkhill to consider taking part in a research study being carried out by Kirsten McKillop as part of her Doctorate in Clinical Psychology. This study involves arranging an interview with Kirsten to answer some questions about what it is like to have delayed puberty. This may help us to improve services.

Before you and your child make up your mind, it is important for you both to understand what we are asking them to do and why. Please find enclosed an information sheet to tell you all about the study. Please read it and ask your child to read it as well. If you have any questions you can call Ethel

McNeill. She is a nurse who works in the Endocrine Clinic although she is not directly involved in this research. However, she will be able to answer any questions you have.

Asking your child to take part in this study does not mean we think they are not coping with their condition. We are asking everyone who comes to clinics and they can say "no" if you don't want to take part.

If they decide they want to take part after reading the information sheet, you can either return the form in the enclosed envelop to the Clinical Psychology Department in Yorkhill, call Ethel McNeill on 0141 201 0245 or email Kirsten McKillop at Kirsten.McKillop@ggc.scot.nhs.uk.

When you call or email, please tell us whether you would prefer us to call you or write to you to arrange an appointment to meet at Yorkhill.

When your child attends this appointment with Kirsten, they will have time to find out more about the study and ask more questions before taking part. If your child decides they don't want to take part, then they can say no and they won't be asked anything else. If they do decide to take part, they will be asked some questions about the effects that having delayed puberty is having on their life. This appointment will last roughly one hour.

If you want to ask anything else about the study you can call Ethel or email Kirsten.

Thank you for reading this.

Yours sincerely,

Professor Syed Faisal Ahmed

Consultant Endocrinologist

Dr Donaldson

Consultant Endocrinologist

Dr Shaik

Consultant Endocrinologist

Dr Kathleen McHugh

Clinical Psychologist

Ms. Kirsten McKillop

Trainee Clinical Psychologist

**An exploration of the psychosocial
consequences of delayed puberty in children
who attend the Yorkhill Endocrine Clinic:
A qualitative study
Researcher: Kirsten McKillop**

Parent/Guardian Information Sheet

This information sheet describes a study exploring the consequences of delayed puberty in young people that your child has been invited to take part in. This study is being carried out by Kirsten McKillop as part of her Doctorate in Clinical Psychology at the University of Glasgow.

Before your child decides to take part, it is important for you to understand what we are asking your child to do and why. If anything is not clear or you want more information please ask us. Your child has also been provided with an information sheet. Thank you for reading this.

Why is this study being done?

It is known that delayed puberty can affect a young person both physically and psychologically. However, the exact impact of delayed puberty on everyday life and relationships is unknown.

What is the aim of this research project?

This study aims to explore young people's experiences of delayed puberty. This study will ask questions about your child's understanding of delayed puberty. We will also ask questions about the impact it has on their family life, school life and education, social life, romantic attachments and how they view themselves. The answers we get will help us plan services to offer help and support to people when they need it most.

Who are we looking for to participate in this study?

We are looking for young people attending the Endocrinology outpatient clinics at The Royal Hospital for Sick Children (RHSC), Yorkhill, who are diagnosed with delayed puberty.

Why is my child being asked?

Your child is being asked because they have delayed puberty and come to an endocrine clinic at Yorkhill. In asking them to take part, we are not saying that they have any problems with how they feel or how they act or cope. We are asking everyone who comes to clinic.

Do they have to take part?

No, your child's participation is voluntary and it will not affect their medical care and treatment in any way if they say "no". If they decide to take part in the study, they will be asked to sign a consent form. Your signature will also be required on this form. You will also be asked to sign your own form indicating that you are happy for your child to participate. Your child can change their mind at any time and withdraw from the study at any stage without giving a reason.

Will the study help my child?

It may help the doctors, nurses and other people working in RHSC to understand some of the things that can worry or upset young people with delayed puberty. The study might help us learn about why some people find things hard and when. This could help us improve services offered to young people.

What will happen next if my child decides to take part in the project?

If your child decides to take part they can contact the main researcher, Kirsten McKillop by phone, email or by returning the slip on the enclosed letter in the stamped addressed envelope included to tell her. They will be asked to say how they would like her to contact by Kirsten them to organised a time to meet at Yorkhill. Kirsten will try to arrange the interview for the same day as their next appointment at Yorkhill Hospital.

The appointment will take roughly one hour and it will involve answering questions about their experience of having delayed puberty. If after the interview there is still more to talk about, they may be asked to attend for one more appointment to explore their experiences further.

We will ask their permission to record the interview to help us remember what they have told us. Their answers will then be typed up, but all personal identifiers will be removed so their responses will be anonymous. Both the recording and transcript of their interview will be kept in a locked filing cabinet on University Grounds. Only the members of the research team will have access to these.

Will my child be paid for taking part?

No

Who has reviewed this study?

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

Want more information?

More information about the study is available from the main researcher, Kirsten McKillop, Trainee Clinical Psychologist: Kirsten.McKillop@ggc.scot.nhs.uk

You can also contact Ethel McNeill. She is an Endocrine Nurse Specialist who works in the hospital. She is not involved in our research, so she is someone you can speak to about any questions or concerns you have. You can contact her on 0141 201 0245.

Appendix 6: Participant Opt in



An exploration of the psychosocial consequences of delayed puberty in children who attend the Yorkhill Endocrine Clinic: A qualitative study

I have read the information sheet and I would like to participate in this study.

Name: _____

Signed: _____

Please contact me to arrange an appointment by:

Email: my email address is: _____

Phone: my telephone number is: _____

Post: my address is: _____

Appendix 7: Participant Consent Form and Parent /Guardian Assent Form

**An exploration of the psychosocial
consequences of delayed puberty in children
who attend the Yorkhill Endocrine Clinic:**

A qualitative study

Name of researcher: Kirsten McKillop



Consent Form

Thank you for agreeing to take part in our study into the consequences of delayed puberty. The purpose of this form is to make sure that you are happy to take part in the research and that you know exactly what this involves.

Please initial

I have had the chance to ask questions about the study

I am happy with the answers given to all of my questions

I have been told enough about the study

I agree for my interview to be digitally recorded. I also understand
it will be typed out word for word, on a computer, and made
anonymous, so that all names of people and places will be removed
so that no one can tell it was me.

I understand that this information will be kept securely so that only the research team can see it.

☐

I agree for my interview to be read by the research team and for some of what I said to be used in the report, as long as it is anonymous.

☐

I understand that I am free to say no to taking part in this study:

- at any time without having to give a reason**
- and without affecting future medical care**

☐

Name of participant

Date

Signature

Researcher

Date

Signature

1 copy to be retained by participant and 1 copy to be retained by researcher



**An exploration of the psychosocial
consequences of delayed puberty in
children who attend the Yorkhill**

Endocrine Clinic:

A qualitative study

Name of researcher: Kirsten McKillop



Parent Assent Form

For under 16 years olds

The purpose of this form is to make sure that you are happy for your child to take part in the research and that you know exactly what this involves.

I have had the chance to ask questions about the study that my child will participate in and I am happy with the answers given to all of my questions.

☐

I agree for my child's interview to be digitally recorded, anonymised and transcribed. I understand that this information will be stored in a locked filing cabinet in secure university grounds.

☐

I agree to my child's interview transcript being read and analysed by the research team and quotations being used in any reports or publications (once identifying information has been removed).

☐

I understand that my child's participation is voluntary and they are free to say no to taking part in this study:

☐

- at any time without having to give a reason**
- and without affecting future medical care**

Name of child

Name of parent or carer

Date

Signature

Researcher

Date

Signature

1 copy to be retained by parent/carers and 1 copy to be retained by researcher

Appendix 8: Interview Topic Guide



An exploration of the psychosocial consequences of delayed puberty in children who attend the Yorkhill Endocrine Clinic **A qualitative study** Researcher: Kirsten McKillop



Interview Topic Guide

These questions are intended to be used as a guide to initiate discussion.

Prompt – remember to inform the participant at the start of the interview that they do not need to answer every question. They can ask to move on to the next one if they do not want to answer.

1) Cause

- Why do you visit this clinic?
- If you had to explain to someone who didn't know what delayed puberty meant, how would you described it?
- What do you think is the cause of your delayed puberty?
- Do you know anyone else who has delayed puberty?
- What do you think caused their delayed puberty?

Prompts: can you tell me more?

2) Length

- When did you first think that puberty was delayed?
- What made you think it was delayed?
- How long do you think it will stay like this?

Prompts: can you tell me more?

3) Curability

- Will it ever get better?
- What will make it better?

Prompt: Can you tell me more?

4) Consequences

- What effect does having delayed puberty have on your family life?

Prompts: How does it effect your relationship with your parents, with your brothers/sisters/ other family members? What are the benefits (if any)? What are the bad effects (if any)?

- What effect does having delayed puberty have on your school life and education?

Prompts: How does it affect your relationships with your friends/teachers? Does having delayed puberty make things harder or easier at school in any way? What are the benefits (if any)? What are the bad effects (if any)?

- What effect does delayed puberty have on you doing things outside of school?

Prompts: Does it stop you from doing anything? Can you tell me more? Does it affect you doing anything with your friends?

- Does having delayed puberty affect what you think of yourself?

Prompts: Would you think different things about yourself if you didn't have delayed puberty? What would you change about yourself? What are the benefits of having delayed puberty (if any)? What are the bad effects of having delayed puberty (if any)?

- What effect does delayed puberty have on having a boyfriend/girlfriend?

Prompts: Do you think it would have an effect when you have a boyfriend/girlfriend?

Can you tell me more? Would having delayed puberty stop you from speaking to someone that you like? Can you tell me more?

5) Controllability

- Can you do anything to improve the consequences you experience of delayed puberty?

Prompts: What can you do to improve it? What hasn't helped? What has helped?

Appendix 9: Example of Transcription and Initial Encoding

Participant 5, P2-3, L65- 97:

5 – Participant, I - Interviewer

	Transcript	Initial Notes/Codes
I	When did you first think that your puberty might be delayed? When did you first start to think that maybe it was?	
5	Well, I think 2 nd .. S2 wasn't that bad, but now, a lot of the boys in my year started to grow and taken growth spurts and things and when they started to get in to S3, it started to.... then it started to hit me that something's not right. Because S2 and S1 okay, I hadn't hit puberty, and I mean that's ok. But I think now I'm getting into 3 rd year, I'm getting.. everyone's turning 15 and I'm still quite small. I'm one of the small boys. So think something wasn't up..right when...	Aware something was wrong Uncertain what Feelings of difference Comparison to peers Dissatisfaction with height
I	But you didn't think anything of it in first or second year?	
5	Not really, I just think.. I thought I was.. I was still to happen and it would just take it's time. I mean I knew I wasn't really going to grow that much anyway. Obviously I said that my mum and dad weren't quite tall, but like, I've not really bulked out at all, which is quite disappointing	Model of cause – genetic comparison to parents Dissatisfaction with appearance/strength Emotional impact - disappointed
I	So it's only just when you went into S3 you'd noticed?	
5	Yeah.. that kind of... yeah.....	

I	And you said there that your friends had had a growth spurt but you hadn't and you've not bulked out	
5	Yeah	
I	Was there anything else that made you think that maybe it was delayed?	
5	Well yeah.. it's a bit embarrassing... but a lot of the boys in PE and things are starting to get like arm pit hair and stuff like that, and starting to get bulk and be really strong and then.. I'm not really strong at all, I don't really have any armpit hair. I'm quite weak, I've got really small arms. So like.. yeah..	<p>Comparison to peers</p> <p>Dissatisfaction with strength, arm pit hair, small arms</p> <p>Emotional impact – embarrassed, helpless?- weak</p>
I	Was that again something else you noticed in 3 rd year and not before?	
5	yeah.. yeah I think it was all in 3 rd year that I've really started ... it hit me the end of S2 so I can remember maybe I started to think, but I think now that I'm in 3 rd , that it's really to approach me... that things... it's worrying that I've not really hit puberty	<p>Recognition of difference – growing awareness</p> <p>Emotional impact – worry</p>
I	And when was your appointment at the.. at yorkhill?	
5	I think it was October, early November I think	
I	Ok. So and that was after you'd started, so you've been in 3 rd year, for a wee bit?	
5	Yes., for about 3 months.	

I	Ok. And how long do you think it will be like this for?	
5	What.. me not..	
I	Yeah.. all the things you've described... how long do you think it will stay like that for?	
5	I hope it's not too long. I hope... it's a bit embarrassing... I have got some hairs, but hopefully things will start to change.. I hope.. I really hope so	<p>Desire to change</p> <p>Emotional impact – embarrassed</p> <p>Dissatisfaction with appearance</p> <p>Future prognosis - uncertain</p>

Appendix 10: Major Research Project Proposal

An Exploration of the Psychosocial Consequences of Delayed Puberty in Children Who Attend the Yorkhill Endocrine Clinic

Abstract

Background: The growth spurt and appearance of secondary sexual characteristics are the hallmarks of adolescent physical development. Puberty is considered to be delayed if sexual maturation occurs more than 2 to 3 standard deviations later than the normal population. Clinically, this equates to pubertal onset beyond 13 years in girls and 14 years in boys. Physical consequences of delayed puberty include short stature and immature appearance, relative to their chronological age. Psychosocial consequences include social withdrawal and isolation, teasing and bullying, parental over protection, poor body image, low self esteem and declining academic performance. Research findings in this area can be conflicting with a focus on delayed growth. Consequently, the psychosocial impact of delayed puberty remains unclear.

Aim: This study aims to explore adolescents' experiences of delayed puberty from a psychosocial perspective.

Methods: All children with delayed puberty who present at The Royal Hospital for Sick Children in Glasgow (RHSC) or attend for continuing care will be identified by a medical clinician. Those who meet inclusion criteria will be invited to attend for interview. The study will aim recruit between 6-12 participants. Semi-structured interviews and subsequent interpretative phenomenological analysis (IPA) will be used to provide evidence and a greater understanding of the psychosocial consequences of delayed puberty.

Application: The findings of this preliminary study will encourage and inform future research in this area. Clinicians will be informed of the psychosocial consequences of delayed puberty from the point of view of the adolescent. Data gathered can also be used to design appropriate and effective interventions for the psychological impact of delayed puberty.

Introduction

The “growth spurt and appearance of secondary sexual characteristics are the hallmarks of adolescent physical development” (Apter et al, 1981, p501). Puberty is considered to be delayed if sexual maturation occurs more than 2 to 3 standard deviations later than the mean of the normal population. Clinically this equates to pubertal onset beyond 13 years in girls and 14 years in boys (Rosen & Foster, 2001). Approximately 2.5% of adolescents will have delayed pubertal onset. No cause for delay can be identified for the majority of these adolescents, who will successfully progress through puberty, albeit later than their peers (Rosen & Foster, 2001).

More boys than girls present with delayed puberty. This is thought to be because delayed growth and an immature appearance have a greater psychosocial impact on boys than on girls (Rosen & Foster, 2001). Boys who are treated as their height age rather than their chronological age experience low self-esteem and difficulties separating from parents. They report feelings of unpopularity due to the reduction in age appropriate social opportunities (Rosen & Foster, 2001).

Girls will present with fewer psychosocial concerns and some are reported to value their immature physique (Rosen & Foster, 2001). Delayed puberty in girls is not accompanied by delayed growth as often as it is in boys (Apter et al, 1981).

Pubertal induction is offered to boys over 14 years old and girls over 13 years old showing no signs of endogenous puberty, achieving dual effects of growth acceleration and physical maturity without sacrificing adult height potential (Rosen & Foster, 2001). The rationale for offering timely progression through puberty is to normalize physical appearance and psychosocial development (Rosen & Foster, 2001).

The specific impact of delayed growth out with delayed puberty is widely reported, however, findings are conflicting with regards to the impact of short stature on psychosocial development. Approximately 60-70% of children with short stature have reported they have experienced teasing and juvenilization, which has been shown to be related to behaviour problems (Sandberg & Michael, 1998). Some studies though have shown that the self-esteem of children with short stature is unimpaired and they display normal patterns of behaviour (Voss et al, 1991, Downie et al, 1997).

Given the possible increased risk of ridicule from peers, parents can become overprotective of their adolescent (Holmes et al 1982). This can lead to further social immaturity, lack of independence and unassertiveness (Meyer-Bahlburg et al 1990). Zimet et al (1995), however, assessed the psychosocial functioning of adults who were referred as children for short stature but were not treated with human growth hormone. It was reported that despite the negative experiences endured by adolescents with short stature and immature appearance, they are not thought to suffer from long term clinically significant problems of psychosocial adaptation.

Parents have also reported that children of short stature exhibit more limited academic abilities compared to their peers who do not have delayed growth (Visser-van Balen et al, 2006). Sandberg et al (2002), however, in their review of the evidence reported that some studies were unable to identify academic deficits among children with short stature. Furthermore, Holmes et al (1985) found that adolescents with delayed growth *and* delayed puberty were academically performing as expected for their chronological age. He concluded that any difficulties experienced were likely to be short-term.

Adolescents who appear younger than they are may also lack social experiences appropriate to their age. This can lead to isolation and withdrawal from their peers (Sandberg & Voss, 2002). Apter et al (1981) investigated the impact of delayed puberty and delayed growth on self-image in adolescents. Fifty-six adolescents with varying combinations of delayed puberty and delayed growth were given a Self-Image Questionnaire. It was found that isolated delayed puberty did not have a significant impact on self esteem. Delayed growth, however, did have a significant impact. It was concluded that delayed growth is potentially more problematic than delayed puberty.

The specific focus on delayed growth within the literature, along with the methodological diversity in studies, results in difficulties identifying the psychosocial impact of delayed puberty. It is not surprising that research findings in this area can be conflicting. Any previous research in this area has been qualitative and used questionnaires written by health professionals who have not taken in to account the problems and issues as seen by the individual. An exploration is needed of the problems around delayed puberty from the perspective of those who are experiencing the delay.

Aim

This study aims to explore adolescents' experiences of the psychosocial aspects of delayed puberty from a psychosocial perspective.

Objectives.

- ☐ To explore adolescents' understanding of delayed puberty in terms of their beliefs related to cause, how long it will last, and controllability/curability (Leventhal et al, 1992).
- ☐ To explore the perceived consequences of delayed puberty for their family life, school life and education, social life, romantic attachments and how they view themselves.

Design

Qualitative, using semi-structured interviews and subsequent interpretative phenomenological analysis (IPA).

Plan for Investigation

Participants

Adolescents attending the Endocrinology outpatient clinic at The Royal Hospital for Sick Children (RHSC), Yorkhill, who present with delayed puberty.

Inclusion and Exclusion Criteria

Pubertal induction is offered to Girls older than 13 and Boys older than 14. There are many markers for delayed puberty therefore Girls older than 12 years and boys older than 13 years that are identified by a Consultant Endocrinologist/Specialist Nurse as clinically pre-pubertal and have not yet started treatment or have been receiving treatment for less than 4 months will be included.

Adolescents who have been receiving treatment for more than 4 months will be excluded. Those with a psychiatric diagnosis or impaired cognitive functioning which would prevent them from being able to give informed consent will not be included. Adolescents will also be excluded if they are not in mainstream education or fluent in English or aged 18 years or older.

Recruitment Procedures

All children with delayed puberty who present at The Royal Hospital for Sick Children in Glasgow (RHSC) or attend for continuing care who meet the inclusion criteria will be identified by a Consultant Endocrinologist or a Specialist Nurse who assess and treat children referred to the service.

If a potential participant is identified by the medical clinician at their first appointment with the service, information regarding the study will be handed to both the adolescent as well as their parents/carers at their appointment. Information regarding the study will also be posted to all parents/carers of potential participants who attend for continuing care that are identified as meeting the inclusion criteria. Information for the adolescent will be included in the envelope as well.

As well as information about the study, adolescents will also be given an invitation to take part. Should they wish to participate, they will be asked to contact the main researcher either by returning the opt-in slip included with their invitation or by emailing to indicate their willingness to participate as well as identifying how they would prefer to be contacted to arrange a convenient time for the adolescent to attend for interview. They will also be given the option of telephoning the Endocrine Clinic within RHSC to indicate their willingness and leave their details for the main researcher to contact them.

If possible, interviews will be scheduled for the same day as the adolescent's next returning appointment to the clinic.

Measures

The interview topic will include exploration of the participants' understanding of delayed puberty as a health problem and also their perceptions of how the condition impacts on the most salient aspects of their lives. The self-regulatory model of illness behaviour (Leventhal et al, 1992) will provide a framework for the topic guide. Adolescents will be asked how they view the condition and the symptoms they perceive to be a part of it. Their ideas regarding the cause of delayed puberty and how long it will last will be explored before moving on to explore their perceptions of the positive and negative consequences and expected outcomes. Lastly their perceptions of controllability will be explored and how responsive they believe the symptoms will be to treatment. Having elicited their perceptions of delayed puberty as a health problem, the interview will then set out to explore the impact of delayed puberty at home, in school, with friends, with romantic attachments and how they view themselves, in order to establish the effect it is having on their life.

Questions will be open-ended and non-directive and will act as triggers to encourage participants to talk about their experiences. Prompts in the form of specific and/or focused questions will be used to encourage the children to expand and elaborate.

Design

The design is qualitative using Interpretative Phenomenological Analysis (IPA) to explore the experiences of the individuals without forming hypotheses or attributing meaning about the area being explored (Smith, Flowers & Larkin, 2009).

Research Procedure

Semi-structured interviews will be carried out based on the aims of the study. Each interview will last no more than an hour and one further interview may be undertaken, if needed, to gain a full understanding of a participant's perceptions.

Conducting Interviews

When the participant arrives, they will be given the information sheet for a second time. They will be given the opportunity to discuss the study with researcher and ask any questions immediately before

the interview. If they are not willing to continue, they will be thanked for their time and the interview will not take place. There will be no further contact.

If they are willing to continue, written informed consent will be gained from both the adolescent and their parent/carer and the recorded interview will begin. Parents/carers will be given the option of remaining in the waiting area for the duration of the interview.

Interview recordings will be transcribed and anonymised for references to place and person. The analysis will commence after a transcript has been checked both for accuracy and for complete anonymisation of references to persons and places. Following transcription and reading through of the first two interviews, the topic guide will be reviewed and amended, if necessary, to improve data elicitation.

Sample Size

The study will aim recruit 10 participants. This number of participants is noted as being at the higher end of the recommended sample size for Interpretative Phenomenological Analysis by Smith, Jarman & Osborn (1999).

Setting and Equipment

The adolescents and their parents/carers will be met at the RHSC. To save time, expense and the adolescent missing school, the scheduled interviews will be co-ordinated with other hospital visits. A room will be booked on hospital grounds to conduct the interview.

A Sony digital voice recorder (ICD-V60 512mb) will be required for each interview as well as a Transcription foot pedal.

Data Analysis

The aim of IAP is to explore how the individual has understood and interpreted a significant event in their life (Smith, Flowers & Larkin, 2009). IPA attempts to offer an understanding of the area being explored through identifying themes and forging links within the individuals' accounts (Smith, 1996). Through these links an understanding of the psychosocial impact of delayed puberty in adolescents can be gained. Interviews will be transcribed as soon as they have been completed and analysis will begin soon thereafter (Reid, Flowers & Larkin, 2005).

The aim of data gathering in IPA is to collate the accounts that participants have given of their experience. These experiences provide evidence and a greater understanding of the phenomenon being explored (Willig, 2001).

Personal Reflexivity

This involves the researcher reflecting on their own beliefs, values and life experiences and the impact these will have on how the research is interpreted. It also involves the researcher reflecting throughout the research process on how the research has impacted them and influenced their beliefs and values (Willig, 2001). It is important the primary researcher acknowledges and reflect upon this throughout the study, given the interactions with those participating had the potential to impact the analysis and interpretations of the transcribed interview.

A proportion, 30%, of the transcripts will be analysed by another researcher who is experienced in IPA and themes identified by both analysts will be compared for consistency to ensure reliability of the analysis.

Health and Safety Issues

Researcher safety issues: the interviews will be held in a clinical setting with numerous health professionals on site. Home visits will not be conducted and interviews will only be carried out during working hours. Given the principal researcher's level of experience (Trainee Clinical Psychologist) and interviews are only taking place in NHS Greater Glasgow & Clyde clinical settings, local health and safety protocols will also apply. Regular supervision will support the monitoring of these throughout the study.

Participant safety issues: all participants and their parents/carers will be given adequate information about the study to allow for informed consent. All interviews will be recorded and the transcripts will be reviewed by an experienced health psychologist.

Participants can decline to talk about particular topics during the interview. Every participant will be informed that they can ask the researcher to move on to the next question should they be asked something that makes them feel uncomfortable. All of the questions within the topic guide will be non-intrusive and they will be asked in an open, non-judgemental manner. Through out the interview, both non-verbal and verbal communication will be observed to monitor any possible signs of anxiety or fatigue.

Although it is very unlikely that the interview will have any adverse effect, participants levels of distress will be monitored during the interview. Should signs of distress be identified, the following procedure will be followed:

1. The interview will be suspended.
2. The interviewer will remain with the participant until their distress decreases.
3. Should they remain distressed, the interviewer will seek assistance from a member of the clinical team.
4. The adolescent will be offered the opportunity to talk to someone they feel comfortable with. This may be the researcher, a member of the clinical team such as a named specialist, their parent/carer or another individual.
5. The interviewer will inform their lead clinician through whose clinic the participant was recruited.
6. If appropriate, a referral to the Clinical Psychology department would be discussed and filled out.

Should the need to use the procedure arise, the adolescent will be made aware of every action in the procedure as it is taken. Parents/carers will also be given contact details for a named nurse specialist should the child become distressed after the interview.

Ethical Issues

The study will seek approval from the West of Scotland Research Ethics committee and the Greater Glasgow and Clyde NHS Research and Development department.

During recruitment, the primary researcher will ensure that each participant is fully informed of the aims, procedures and limits of confidentiality. Written consent for participation, recording and transcribing of interviews will be gained from all participants and their parent/carer, as well as consent to use and publish anonymised quotations.

Adolescents will only be included in the study if consent is given from both the adolescent and their parent/carer.

It will be made clear that participants are able to withdraw their consent at any stage. Every participant will be informed that all data will be anonymised, transcripts will be anonymised and identifiable only by means of a numeric code. The information linking participant and transcript will be held securely, adhering to NHS and Glasgow University Data Protection guidelines.

Financial Issues

The recording and transcribing equipment will be provided by the Department of Psychological Medicine, University of Glasgow. In addition, there will be the postage costs of opt-in slips, appointment slots as well as the cost of photocopying information sheets and consent forms. Travelling expenses for the principle researcher will be met by NHS Education for Scotland (NES).

Timetable

Submission of proposal for ethical approval – April/May 2012

Data collection – June 2012

Data analysis and write up – July 2012

Final draft to supervisor – August 2012

Practical Application

The individuals' experiences can be used to develop and communicate an understanding with other patients. Data from this study can also be used to design appropriate and effective interventions for the psychological impact of delayed puberty. The findings of this preliminary study will encourage and inform future research in this area. They may also inform the development of suitable measures that can be used in future research.

References

Apter, A., Galatzer, A., Beth-Halachmi, N & Lavron, Z. (1981). Self-Image in Adolescents with Delayed Puberty and Growth Retardation. *Journal of Youth and Adolescence*. 10. (6). 501-505.

Downie, B., Mulligan, J., Stratford, R., Betts, P., Voss, L. 1997. Are short normal children at a disadvantage? The Wessex Growth Study. *British Medical Journal*. 314:97-100.

Galatzer, A., Rosenblith, E., Laron, Z. 1978. Psychological and rehabilitation aspects of short stature and delayed puberty, in Gedda L, Parisi P, (eds): *Auxology: Human Growth in Health and Disease*. London. Academic Press.

Holmes, C., Hayford, J. & Thomson, R. 1982. Parents and teachers differing views of short children's behaviour. *Child: Care, Health and Development*. 8: 327-336.

Holmes, C., Karlsson, J. & Thomson, R. 1985. School and school competencies in children with short stature. Longitudinal patterns. *Journal of Developmental and Behavioural Paediatrics*. 6: 263-267.

Leventhal, H., Diefenbach, M., Leventhal, EA. (1992). Illness cognition: using common sense to understand treatment adherence and affect cognition interactions. *Cognitive Therapy and Research*. 16: 143–163.

Moss, L. & Goldstein, H. (1979). The recall method in social surveys, 1979 In: Blane, D.B. *Collecting retrospective data: development of a reliable method and a pilot study of it's use*. *Soc Sci Med* (2006); 42: 751-757.

Meyer-Bahlburg, H. 1990. Short stature: psychological issues. In Lifshitz F (ed). *Paediatric Endocrinology. A Clinical Guide*, 2nd edn, pp 173-196. New York. Marcel Dekker.

Norris, F.H. & Kaniasty, K. (1992). Reliability of delayed self-reports in disaster research. *J Trauma Stress*. 5: 575-588.

Reid, K., Flowers, P., & Larkin, M. (2005). Explored lived experience. *The Psychologist*, 18, (1), 20-23.

Rosen, D., & Foster, C. (2001). Delayed Puberty. *Pediatrics in Review*. 22, (9), 309-315

Rosenfeld, R.G., Gregory, B., Northcraft, B.A & Hintz, R. L. (1982). A Prospective, Randomized Study of Constitutional Delay of Growth and Development in Male Adolescents. *Pediatrics*. 69. (6). 681-687.

Sandberg, D. Colman, M & Voss, L. (2004) *Short Stature and Quality of Life: A Review of Assumptions and Evidence*, *Pediatric Endocrinology: Mechanisms, Manifestations, and Management*. Philadelphia. Lippincourt. Williams & Wilkins. pp 191–202.

Sandberg, D & Michael, P. (1998). Psychosocial stresses related to short stature: Does their presence imply psychiatric dysfunction? Drotar D (ed): *Assessing Pediatric Health-Related Quality of Life and Functional Status: Implications for Research*. Mahwah, Lawrence Erlbaum Assoc. p 287–312.

Sander, D & Voss, A. (2002). The psychosocial consequences of short stature: a review of the evidence. *Best Practice and Research Clinical Endocrinology and Metabolism*. 16. (3). 449-463.

Smith, J.A. (1996). Beyond the divide between cognition and discourse: using interpretative phenomenological analysis in health psychology. *Psychology and Health*, 11, 261-271.

Smith, J.A., Flowers, P. & Larkin, M. (2009). *Interpretative Phenomenological Analysis: Theory, Method and Research*. SAGE Publication Ltd. London

Smith, J.A., Jarman, M., & Osborn, M. (1999). Doing Interpretative Phenomenological Analysis. In M. Murray, & K. Chamberlain (Eds.). *Qualitative Health Psychology. Theories and methods* (pp.218-240). London. Sage Publications Ltd.

Smith, J.A., & Osborn, M. (2003). 'Interpretative phenomenological analysis, in J.A. Smith (ed.), *Qualitative Psychology: A Practical Guide to Research Methods* (pp.51-80). London: Sage.

Suris, J.C., Michaud, P.A. & Viner. (2004). The adolescent with a chronic condition. Part 1: developmental issues. *Archives of Disease in Childhood*. 89: 938-942

Visser-van Balen, H. Sinnema, G & Geenen, R. (2006). Growing up with idiopathic short stature: psychosocial development and hormone treatment: a critical review. *Arch Dis Child*. 91: 433–439.

Voss, L., Bailey, B., Mulligan, J., Wilkin, T., Betts, P. 1991. Short stature and school performance: the Wessex Growth Study. *Acta Paediatr Scand*. Suppl 370:29-31.

Willig, C. (2001). *Introducing Qualitative Research in Psychology: Adventures in Theory and Method*. Open University Press: Buckingham.

Zimet, G. Cutler, M. Litvene, M. Dahms, W. Owens, R. Cuttler, L. (1995). Psychological adjustment of children evaluated for short stature: a preliminary report. *J Dev Behav Pediatr*. 16: 264–270.