PARENTS OF CHILDREN WITH AMBIGUOUS GENITALIA: STORIES OF EXPERIENCES OF RECONSTRUCTIVE GENITAL SURGERIES AND FINDING HARMONY

by

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ABSTRACT

Title: Parents of children with ambiguous genitalia: Stories of experiences of reconstructive genital surgeries and finding harmony.

Aim: To explore and understand parents' experiences of their child's genital ambiguity and the reconstructive surgeries for ambiguous genitalia that occurred in infancy and middle childhood

Background: The determination of sex and gender for a child born with ambiguous genitalia is an extremely complex medical and social process. Academic debate, professional practice, the law and increased political and ethical debate have all more recently challenged the evidence base for practice. Currently the 'optimal gender policy' and the 'informed consent policy' drive treatment options. Little research has been conducted to understand the significance gender ambiguity has in parents' lives and how the child's genital surgery affects parents.

Methods: An exploratory design of narrative inquiry was chosen and data were collected through eighteen in-depth narrative interviews with a purposive non-random sample of fifteen parents of 11 children (aged 0-11 years).

Findings: Narrative analysis resulted in three keystone stories which contained in total eight aggregate stories and twenty foundational stories. The three keystone story themes were firstly, parents' stories about their child. Secondly, stories about being a parent of a child with AG and finally stories about healthcare professionals. Interpretation and synthesis of the three keystone stories revealed three core elements fundamental to parents stories; shock, protection and anxiety. Parents had to develop new skills in order to deal with the challenges of living with a child with AG. Parents endeavoured to find a sense of harmony from their experiences of shock, anxiety and the need to protect their child. Harmony is a concept that brought consistency and agreement together resulting in parents embracing their experiences holistically and giving their experiences meaning.

Conclusion: Parents overcame the tensions inherent in their experience of their child's AG and found a sense of harmony which has not previously been described in the literature.

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GLOSSARY OF TERMS / ABBREVIATIONS

The following glossary is provided to facilitate understanding of the sometimes complex terminology that is used to describe ambiguous genitalia (Parents Handbook, Consortium DSD/ISNA 2006)

Androgens are hormones (molecules or chemical messengers) made mostly by the testes, but also made to a lesser extent in the adrenal glands located above the kidneys, and in the ovaries. They stimulate male reproductive organ (sex organ) development and secondary sex characteristics such as facial hair and lower pitch of voice. The two major types of androgens involved in sex development are testosterone and dihydrotestosterone.

Androgen Insensitivity Syndrome (AIS) is an X linked autosomal recessive genetic condition and can be either complete or partial. Previously this condition was referred to as testicular feminisation syndrome. The XY foetus develops testes but due to androgen insensitivity there is little or no virilisation. The testes produce anti-mullerian hormone which inhibits the development of fallopian tubes and the uterus. The vagina can be short or be present as a small dimple on the perineum. Female body shape develops at puberty there is no underarm hair or pubic hair and the child needs long-term hormone replacement therapy. The internal testes are often removed at or before adulthood to eliminate the risk of cancerous changes. In the foetus with partial androgen insensitivity there is some response to androgens this results in ambiguous genitalia.

Ambiguous Genitalia (AG) - Ambiguous genitalia in the newborn suggests the baby has a phallus too small to be a penis but too large to be a clitoris (Lee & Donahoe, 1994). The ambiguity of the external genitalia leads to the initial uncertainty about the child's sex.

Persistent Cloaca is a urogenital malformation in girls occurring during embryonic development when the cloaca, situated at the far end of the hindgut, fails to develop. Therefore the development of the urinary tract and

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reproductive tract are incomplete resulting in the girl having a single urinary, vaginal and anal opening. These girls experience life saving surgery and reconstructive genital surgeries have historically been undertaken in infancy.

Congenital Adrenal Hyperplasia (CAH) are autosomal recessive conditions which result from a mutation of the genes that control the enzymes that produce cortisol from cholesterol by the adrenal glands. Most of these conditions involve the excessive production of sex steroids and can alter development of primary or secondary sex characteristics in some affected female infants, resulting in ambiguous genitalia.

Disorder of sex development (DSD) when the more common path of sex development (male or female) does not take place, the condition is often called a disorder of sex development or DSD.

Oestrogen is a hormone (molecules, or chemical messengers) mainly produced in the ovaries, but also produced to a lesser extent in the adrenal glands and testes. It plays a part in the development of secondary sex characteristics, such as breast development. Oestrogen is also important in female reproduction, such as regulating the menstrual cycle.

Gender usually refers to mental, social, and cultural characteristics, regardless of anatomy, related to being a boy, girl, man, or woman in any society.

Gender identity Disorder (GID) is a formal diagnosis usually given by a psychologist to describe someone who experience significant gender dysphoria. The person is discontent with the biological sex they were born with.

Gonads are a general term for the sex glands. The term gonad can refer to an ovary, a testis (testicle), an ovotestis, or a streak gonad. Mature ovaries usually release eggs until menopause, while mature testes usually produce sperm. In addition, the gonads release hormones that affect the development of the reproductive organs at puberty and affect other physical traits that, after

puberty, usually make men and women look different, such as pitch of the voice and body shape and size.

Healthcare professionals (HCP) include medical doctors such as a paediatrician or an endocrinologist. A surgeon is a paediatric general surgeon, urologist or gynaecologist. Often these groups meet as a multidisciplinary team and provide care to the child and their family. This team also includes specialist nurses, geneticists and psychologists.

Intersex is a term sometimes used to refer to the condition of having a sex anatomy that is not typically male or a female. Like disorders of sex development, it is an umbrella term that covers many different conditions.

A **karyotype** is a picture of the chromosomes in a cell. A karyotype is used to see what kinds of chromosomes a person has. It is determined by taking a blood or tissue sample from a person, and then staining the chromosomes with dye and photographing them through a microscope. The photograph is then cut up and rearranged so that the chromosomes are aligned into corresponding pairs and the XX or XY or variance can be determined.

Mullerian ducts are present in all embryos in early development, in most females the Mullerian ducts develop into the uterus, fallopian tubes, and the upper part of the vagina. In males this development is inhibited by AMH.

Mullerian inhibiting substance is also called anti-Mullerian hormone (AMH). A hormone normally produced by the testes in the early stages of male foetal development that prevents the Mullerian ducts from developing into the fallopian tubes, uterus and upper part of the vagina.

Ovotestes are gonads (sex glands) containing both ovarian and testicular tissue. These are sometimes present in place of one or both ovaries or testes in people with DSDs.

Sex usually refers to a person's external visual and internal physical anatomy. Females have a clitoris, vagina, uterus, fallopian tubes and ovaries while males have a penis, scrotum, testis and ejaculatory mechanisms.

CHAPTER 1: PROLOGUE

As an introduction to my thesis this prologue outlines my rationale for exploring parents' experiences of their child's reconstructive genitalia surgeries in addition to outlining the structure of my thesis. As a clinical practitioner faced with the day-to-day challenges of wanting to provide an evidenced based quality service to the children and parents I support, I began to examine the literature surrounding the management of children born with ambiguous genitalia. Having witnessed parents within hospital settings prior to and following their child's reconstructive surgeries for ambiguous genitalia I recognised that I understood little about their experiences. This limitation on my part and paucity in the literature I believed influenced my ability to be able to develop and deliver the evidenced based quality care I considered these parents and their children deserved.

My thesis uses narrative inquiry as the means to elicit, examine, interpret and synthesise parents' stories about their experiences of their child's reconstructive genital surgeries for ambiguous genitalia. I wanted my thesis to have as narrative a structure as possible. To this end I have a prologue that introduces the thesis and an epilogue providing an ending. As my study adopted a narrative approach stories are at the core of my work. I made a considered decision to embrace a narrative approach throughout my thesis in order to facilitate purposeful engagement with my findings and discussion. The overall structure of my thesis follows a broadly 'traditional' style and within this chapter I set out the structure of it to provide the reader with a map of the story of my study thereby affording the reader some direction. I have provided a glossary of terms (p 15-18) to share definitions of the often complex terminologies and conditions which present with ambiguous genitalia.

In my second chapter I start to explore and address the core issues that help to inform an understanding of the complex issues that need to be addressed to understand this study. In considering parents of children with ambiguous genitalia I discuss biological differentiation and determination, aetiology and diagnosis. However, as my thesis is not a biological or scientific exploration of

sexual determination of differentiation this subject area is only briefly covered to provide understanding and share the complexity of biological development. The historical, professional and academic debate surrounding ambiguous genitalia coupled with the terminology and language used to explain ambiguous genitalia are explored to provide context and widen understanding of the intricacies of ambiguous genitalia. The two current key approaches to managing ambiguous genitalia are discussed. The core issues of sex, gender, intersex and disability are explored within the context of ambiguous genitalia. I also address psychosexuality and the related issues of gender identity disorder, transgender and deviance. However, my thesis is not an exploration of sex, gender and intersexuality within the context of feminist or queer-theory and I therefore signpost the reader towards more in-depth reading. I show how the two areas of secrets and surgical decision making as described by adults with DSD could be bound to the literature of parents of a child with ambiguous genitalia. Other areas of the literature that are addressed include long-term adult outcomes from childhood reconstructive genital surgeries. The limited case studies which explore parents' experiences of having a child with ambiguous genitalia along with their levels of stress and coping are discussed. In exploring these I examine the broader issues of ambiguous genitalia in accordance with the Law, politics and ethics.

Having critically explored these areas I demonstrate the complexity surrounding ambiguous genitalia and the uncertainty of sex or gender. I demonstrate how the lack of literature and knowledge based on parents' experiences demonstrates the clear direction for the framing of my study.

Chapter three addresses the design of my study and tackles my clinical role and research and the importance of reflexivity and how my philosophy and beliefs were integral to all of the decisions I made. I present the methodology of my study, the research aims and the rationale for undertaking this in two phases. I examine my own clinical practice and experiences as matched against the literature. Reflexivity as a result of my engagement with my study and my early beliefs are also discussed. I explore the theoretical perspective and epistemological base which guided the research paradigm most appropriate to answer my research question and explain why I adopted narrative inquiry.

Within this chapter I also explain my study design and method, including how I identified the parent target population and developed a recruitment matrix. I explain narrative in-depth interviews and how I used these as a method of generating data. I explore the ethical complexities that were both considered before commencement of my study but also those that arose throughout my study. In conclusion, I address the trustworthiness in qualitative data. In the following chapter I explore how I understood and analysed these data.

Within chapter four I aim to make clear both how and why I managed parents' stories as data. The approaches I used within both phases one and two are considered. I provide insight into the development of how I explored these data. How I made sense of parents' stories is explained and made clear to ensure that a 'trail' of thinking is visible to the reader within this section of analysis and critical thinking. I also explore the impact these data had on my own thoughts and issues of reflexivity. Continuing the narrative theme within my thesis I explain the reasoning behind how I present stories as data and the emergence of foundational, aggregate and keystone stories. In conclusion, chapter four leads the reader towards the vignettes and findings chapters.

Vignettes of the parents who participated in the study are presented in chapter five. These vignettes are included to provide context and insight into the lives of parents. These vignettes have been built from my fieldnotes and are written in a 'story style'. The aim is to present the parents as people who are living their lives as well as parenting a child born with ambiguous genitalia who has experienced reconstructive genital surgeries.

In the following four chapters I present the findings from my study and the stories that are fundamental to my thesis. The first findings chapter (Chapter 6) is the introductory findings chapter which addresses the participant results ratios and the findings from the interview process. In the subsequent three chapters, seven, eight and nine I present a different keystone story and the contributing aggregate and foundation stories that underpin each keystone story. In chapter seven I concentrate on the keystone story which focuses on parents' stories about their children. These stories broadly related to stories

about the beginning of their experiences of having a child with ambiguous genitalia and how these impact on them. These stories include the uncertainty it created, how they came to make decisions about sharing the news of their child's birth and sex as well as addressing their experiences of danger, worry, risk and the need to protect their child's future. Within chapter eight I move from a focus on parents' stories about their children to the stories that parents told about being a parent of a child with ambiguous genitalia. The stories I present and explore in this chapter express the parents' search for normality, the ways in which they create emotional bonds with their child and the place surgical intervention has within their lives. In chapter nine my focus moves to the stories parents tell about healthcare professionals and how parents develop trust and how this contributes to their decision making. The stories also address parents need for good communication and the importance of information.

In the previous three keystone story chapters I have demonstrated how the three core elements of shock, protection and anxiety bring about tensions for parents and are evident to a lesser or greater degree in all the parents stories. In chapter ten I summarise these tensions and present my discussion of the central concept of my thesis, which is the parents' search for harmony. In presenting the parents tensions within the core elements and the central concept of harmony I clearly demonstrate how this constitutes a contribution to new knowledge both conceptually and in its potential for guiding practice. In this chapter I link through to the existing relevant literature to place my discussion and synthesis in context.

In the epilogue I address the limitations and the implications for practice as a result of my study.

CHAPTER 2: LITERATURE REVIEW

2.1 Introduction

In this chapter I explore and address ten issues pertaining to the literature[1] which helped inform my understanding of the complexity of ambiguous genitalia (AG)[2]. I present these issues under separate sections to promote clarity. However, the complexity of AG is such that these core issues are interrelated. Where necessary these sections are split into subsections. The issues are:

- Biological differentiation, determination and brain sex. Incidence of intersex/disorders of sexual differentiation (DSD) and AG. Aetiology of AG, congenital adrenal hyperplasia (CAH), androgen insensitivity (AIS) and persistent cloaca;

- Historical, professional and academic debate surrounding AG;

- The current language and terminologies used to describe AG;

- The 'optimal gender policy' and 'informed consent policy';

- Social construction of sex, gender and intersex and disability in relation to AG;

- Psychosexuality including gender identity disorder (GID), transgender and deviance;

- The approach and consequences of parents and professionals keeping secrets from the child or adult with AG;

- Evidence based surgical decision making, liminality and outcomes from early reconstructive genital surgeries;

- Parents experiences of having a child with AG and similarities to other conditions; and

- The law, politics and ethics in relation to AG.

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[1] Search parameters included key words; intersex, DSD, AG, parents experiences, children and genital surgery, gender. Data search history was recorded on data star. I narrowed articles / books to key publications which encompassed key words. Six key research papers explored concepts of parents and their child's DSD. I also explored sociological, narrative, philosophical, qualitative methods, parenting, coping and adjustment literature. I used many search engines including Pubmed, Science Direct, CINHAL. I also contacted (via email) key authors e.g. Creighton, Diamond, Dreger, Feder, Holmes, Jürgensen, Slijper, Weyman, Warne, Zucker; in order to obtain unpublished or referenced materials. English translations of articles were collected from authors directly e.g. Hester. I met with leaders in the field at International meetings and study events e.g. Reissman, Frank, Carmichael, Thomas.

[2] See glossary page 4-7 as a reference guide for terminologies and abbreviations used throughout my thesis and within the context of the wider literature.

2.2 Biological differentiation, determination and brain sex. Incidence of intersex/disorders of sexual differentiation (DSD) and AG and the specific conditions included in my study.

The following section is split into three subsections to firstly, establish a biological understanding of the scientific nature of sex development. Secondly, to establish an awareness of the incidence of AG and the challenges in determining the 'true' numbers of affected individuals. Finally, to establish an understanding of the specific conditions included in my study.

2.2(a) Biological differentiation, determination and brain sex.

The sex chromosome present within the embryo is influenced by various molecular events that determine the development and migration of the germ cells in the "bi-potential gonads'"(Lam, 2008; Ogilvy-Stuart & Brain, 2004). In the presence of a Y (male) chromosome (46, XY) and several genes including the SRY gene, these cells form testes. An ovary develops in the absence of a Y chromosome and is perhaps influenced by the HOX gene and as yet unknown genes[3] (Lam, 2008). This process is called sex determination with sex differentiation being the second stage following determination. MacLaughlin & Donahoe (2004) suggest that sex differentiation is "the specific response of tissues to hormones produced by the gonads" (p 367). Both these processes are complex and are still the focus of research. Scientific and genetic researchers continue to investigate sex determination and differentiation (Migeon, 2004; Hines, 2004). Sex differentiation (boy or girl) occurs during the fourth to fifth week of embryonic life (Pajkrt & Chitty, 2004). The mesoderm gives rise to the nephrogenic cord, which forms the urogenital ridge and the mesonephric ducts (Wolffian - male structures). Developing lateral to the mesonephric ducts are the paramesonephric ducts (Mullerian - female structures). The presence of a complete Y chromosome and gene set influences these primitive gonads to secrete human chorionic gonadotropin (hCG), which consequently triggers the primitive gonadal tissue to secrete testosterone. Dihydrotestosterone stimulates the development of male internal and external anatomy. Anti-Mullerian hormone produced by the testes inhibits

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[3] Females are commonly 46XX however, Turners is a female 46X DSD condition that does not present with AG.

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and subsequently degenerates the Mullerian ducts thereby halting the development of female structures in the male embryo.

Biological sex has traditionally been determined by the presence or absence of genitalia and gonads (phenotype) and the sex chromosomes (karyotype) (Gurney, 2007). More recently, with the advancement of in-depth screening techniques, the differences in brain structure and development between men and women (brain sex) is gaining "wider recognition" (Gurney, 2007). Gurney (2007) suggests that the "brain's sex" is a "biological factor" which can influence the psychological and psychosexual development of the individual. Gurney (2007), a human rights activist, with a background in both science and law explores the context of brain sex and sex determination. She examines the scientific and biological evidence that have been used within the law courts in order to provide treatments or withhold treatment for individuals with AG. [The complexities of scientific and biological uncertainty surrounding genital ambiguity are discussed later in this chapter]. Fausto-Sterling (2000) as a biologist, feminist and historian of science explores the debate of sexing the brain within a chapter of her book 'sexing the body'. She discusses the scientific and historical literature in relation to the difference of the corpus callosum between men and women and suggests that "not everyone believes in this difference in brain anatomy" (p 115). Scientists describe a difference in the size and the innervation of the corpus callosum between men and women. These differences could possibly contribute to the distinction between male and female behaviours and abilities. In summary Fausto-Sterling (2000) acknowledges the limitations of scientific inquiry and the need for ongoing research programmes. However, she believes that for her:

> "the real excitement of studies on the corpus callosum lies in what we can learn about the vastness of human variation and the ways in which the brain develops as part of a social system" (p145).

Although there is an acceptance that science has provided some insight into the differences between boys and girls the biological nature of sex is not the sole determinate of gender.

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2.2(b) Incidence of intersex/disorders of sexual differentiation (DSD) and AG

Haas (2004) suggests that between 1.7% and 4% of the world population have primary and secondary sexual characteristics which are ambiguous and are neither clearly boy nor girl. In addition, Gough et al (2007) explain that:

> "not all intersex conditions are identified at birth, and some may not be identified until puberty or later. This complicates the task of calculating numbers affected, and it may not be unreasonable to cite an incidence figure as high as 4%" (p2).

Gurney (2007) also suggests that issues of "sterility" or "sexual identity" may highlight incidences of intersex that are currently not investigated. She concludes that: "more than half of patients with incomplete sex differentiation are not properly diagnosed" (p627).

Fausto-Sterling (2000) is in agreement with the higher incidence of intersex while Sax (2002) disagrees suggesting that the term intersex has been used too broadly. The debate surrounding terminologies is discussed later in this chapter. The incidence of DSD in the general population as determined by biological factors remains unknown and methods that determine occurrence are not discussed further within the context of my study.

2.2(c) Aetiology of AG, congenital adrenal hyperplasia, androgen insensitivity and cloaca

There are a variety of different conditions that can give rise to individuals possessing characteristics of both male and female sex. I describe those which were included within my study.

Ambiguous Genitalia (AG)

Ambiguous genitalia in the newborn suggests the baby has a phallus too small to be a penis but too large to be a clitoris (Lee & Donahoe, 1994). Micropenis is a label given to a male child who is born with a penis too small for normal sexual function (Looy & Bouma 2005). Measurements of the external genitalia (Hughes et al, 2006), Prader classification or staging (Forest et al, 2004; Ogilvy-Stuart & Brain, 2004) and Tanner stage ratings (Migeon et al, 2002) are tools which aid in the classification of ambiguous genitalia and later pubertal

development.   Invasive and radiological examinations undertaken on the newborn to determine internal anatomy are described in the literature (Al-Agha et al, 2001; Mouriquand, 2004).

Congenital Adrenal Hyperplasia (CAH)

The two most common types of CAH are 21-hydroxylase (salt losing) and 11 beta hydroxylase (non-salt losing). In both types, excess androgens are secreted in utero (Forest et al, 2004). Therefore (XX) girls with CAH have been exposed to high levels of prenatal androgens (dihydrotestosterone) resulting in masculinisation of their external genitalia. Internal female organs are formed; ovaries, fallopian tubes and uterus. However there can be a common channel for the urethra and vagina (urogenital sinus). High level pre-natal androgens in an XX female potentially have the capacity to impact upon the girls psychosocial and psychosexual development (Kuhnle & Bullinger, 1997; Hines, 2004).

Androgen Insensitivity (AIS)

Androgen insensitivity syndrome (AIS) can either be partial or complete. AIS accounts for the largest group of XY women: people with a female phenotype (who look externally female) and XY karyotype (chromosomes). AIS is an X linked autosomal recessive gene with a risk ratio of 1:4 of having an affected child (Slijper et al 2000). The differences between the partial or complete AIS relate to the degree of masculinisation of the child, in CAIS the child is completely phenotypically female[4] but has no internal female sex structures. Children with PAIS have a degree of masculinisation and genitalia ambiguity; some are raised as male while others are raised as female depending on the degree of masculinisation.

Persistent Cloaca

Cloaca is a urogenital malformation in females occurring during embryonic development when the cloaca, situated at the far end of the hindgut, fails to

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[4] The visual appearance of the infant is female, normal sized clitoris and labia and no ambiguity is noted. This infant or child could present with bilateral or unilateral hernia, which on assessment are found to be testes. Alternatively the testes could be intra-abdominal and the young woman presents at adolescence with a history of primary amenorrhea.

develop. The embryonic failure of differentiation results in a persistent cloaca in which the rectum, vagina and urinary tract are fused together in a single common channel. A single perineal orifice exists and the genitalia are reported as smaller than normal (Levitt & Peña, 2005). The infant will experience genital surgeries due to the 'incomplete nature' of their genito-urinary tract.

Within the context of my thesis the term ambiguous genitalia (AG) will be used. Rather than referring to the underlying sexual determination or differentiation models I have adopted DSD (rather than intersex) as a generic term which includes the children with CAH, PAIS, and incomplete XY. Also DSD is the terminology currently adopted within my clinical practice.

2.3 Historical, professional and academic debate surrounding AG

Prior to medical advances the inability to diagnose and treat life limiting DSD conditions such as CAH resulted in infant mortality. Children who did not have a life limiting DSD condition prior to modern surgical techniques were "generally left as they were born" although they did often "perplex society" (Gurney, 2007, p631). Looy & Bouma (2005) explored the history of AG and suggest that:

> "we do know that, for thousands of years, human cultures have reacted with fear and horror at the birth of a sexually-ambiguous child" (p171).

They continue to describe excerpts from Babylonian historical documents and conclude that in societies where AG was feared the child was left to die. However, not all societies view AG with fear as they explain:

> "gender ambiguity and intersexuality were sometimes attributes of powerful, divine beings, as in the case of the ancient pagan goddess Cybele, the 'Bearded Aphrodite' (whose child with the god Hermes, Hermaphrodites, gives us the word used to describe a form of intersexuality), the Hindu Shiva and Shakti, and possibly ancient Neolithic divinities. It is therefore difficult to know for certain the various ways in which intersexed and transgendered persons were treated historically" (Looy & Bouma, p171).

Gurney (2007) describes succinctly the development of both medical and surgical management of AG over the last century:

"[D] during the second half of the nineteenth century the developing science of medicine began assigning sex in terms of gonads and their histology rather than genitalia, and from the mid-twentieth century on, often also in terms of the sex chromosomes" (p631).

However, within a global context not all individuals with DSD are treated the same. In some cultures such as Native American they have unique social roles consistent with a 'third gender' as Looy & Bouma (2007) explain and may be considered ""two-spirited" and may be highly regarded, especially in religious roles" (p172). This open cultural acknowledgement of AG coupled with little or no attempt to alter or treat it is different to Western approaches and management. The medicalisation of AG, was until recently, the basis for surgical management (Looy & Bouma, 2005). Reis (2007) describes this concisely:

"the ways in which intersex bodies have been scrutinized and pathologized have been negative, harmful, and based, not on medical necessity but on social anxieties about marriage, heterosexuality, and the insistence on normative bodies" (p539).

The clinical and medical approach adopted over the last quarter of a century was guided by Money's theorising. A firm belief that gender identity is malleable during the first two years of life was proposed by Money, Hampson & Hampson, in 1955. This resulted in doctors believing they could surgically "normalise the genitalia" allowing parents to rear their child "in the corresponding gender" confident that their child would be "both physically and psychologically comfortable" with their sex and gender (Looy & Bouma, 2005). Academic debate is driven from a sociological, feminist and bioethics perspective. Holmes (2008) suggests that for individuals with intersex the problem is "not a lack of who will provide treatment" but the enthusiasm of some surgeons to focus on "invasive treatments" as the management of choice. Holmes (2008) continues to explore the basis for the attitudes that guide surgeons and parents towards invasive treatments. He believes that the challenge lies in trying to change "the parents, the wider family, clinicians, social workers" attitudes towards intersex. By moving towards a new attitude of acceptance of intersex Holmes (2008) believes the intersexed "adult's sense of authenticity as persons, and as gendered subjects" (p175) is preserved.

The historical debate that surrounds intersex illustrates that AG is not a new phenomenon. The 'true' incidence of intersex within the context of a global population is unclear. Understanding incidence is made more difficult because terminologies remain confusing. A shift in professional medical thinking brought about by theorising and advances in surgical technologies is evident in the literature. The arguments against the medicalisation of intersex is driven by those exploring and trying to understand the complexity of sex and gender within social systems. Furthermore, Holmes (2008) suggests that the lesson learnt from some intersexed adults should inform practice since these individuals believe they have lost their "authenticity" as people as a consequence of interventions.

2.4 The current language and terminologies used to describe AG

Many different classifications of AG exist and these are described concisely in a key paper by Hughes et al (2006). This is a key paper which presents the complete consensus statement of the management of intersex made by professional societies representing paediatric endocrinology (LWPES1/ESPE2 Consensus Group, 2006). The consensus group consisted of a group of over 50 international experts involved in research and clinical care of individuals with DSD (including members of the Lawson Wilkins Pediatric Endocrine Society and the European Society for Paediatric Endocrinology). They met in Chicago in 2005 to work on an interdisciplinary consensus paper on the optimal management of intersex disorders based on an evidence-based literature review (Brinkmann et al, 2007). In a later paper Hughes (2008) expands further the negative impact of previous language used in relation to AG:

> "the nomenclature 'intersex', 'hermaphrodite' and 'pseudohermaphrodite' is anachronistic, unhelpful, and perceived to be pejorative by some affected families. In its place, a consensus statement recommends the term 'disorder of sex development' (DSD), a generic definition encompassing any problem noted at birth where the genitalia are atypical in relation to the chromosomes or gonads" (p119).

Therefore words such as 'hermaphrodite' and 'pseudohermaphrodite' are no longer used by many professional groups. As Reis (2007) rightly describes hermaphrodite as a historical term that depicts intersex individuals as "mythical

creatures". If language is used that conjures ideas of intersex individuals as "monsters and freaks" Ries (2007) is not surprised "that people would want to avoid" such individuals (p536). The need to develop and use language and terminology that is inclusive for affected individuals as well as professionals is important but as suggested by Ries (2007) this remains complicated as it raises:

> "political as well as medical questions. The choice of nomenclature influences not only how doctors interpret medical situations but, equally as important, how parents view their affected children, how intersex people understand themselves, and how others not directly involved in medical settings—such as gender and legal scholars, historians, and media commentators—conceive of and theorize about gender, sex, and the body" (p536).

Although DSD has been suggested as the umbrella term some professionals remain reluctant to change their language and this is based on their own beliefs and experiences (Holmes, 2008). Therefore I will expand the term intersex, its origins and the challenges that have been discussed in relation to it use in practice. Since the 1990s some activists advocated intersex as the term to replace hermaphroditism. Reis (2007) clarifies that intersex was first introduced by biologist Richard Goldschmidt in 1917. However, she continues to describe the difficulties that the use of the term intersex has had for some groups:

> "[S] some parents, though, were uncomfortable with the intersex label for their affected children. To them, *intersex* meant a third gender, something in-between male and female. They wanted to see their newborn babies as girls or boys, not as intersex" (Reis, 2007 p537).

Many professional groups have accepted the term, disorders of sex development (DSD), following the consensus meeting in 2005 and use this in clinical practice and professional dialogue. Reis (2007) suggests that supporters of this term:

> "believe it deemphasizes the identity politics and sexual connotations associated with *intersex* and the degradation associated with *hermaphrodite* and instead draws attention to the underlying genetic or endocrine factors which cause prenatal sex development" (p537).

Dreger (2005) also supports the recent changes to previous "medicalised" terminology suggesting that individuals with DSD considered previous terms controversial, insensitive and potentially uncomplimentary.

Yet debate surrounding an agreed terminology continues. Reis (2007) discusses terminologies at length and argues that "divergence of sex development" is more appropriate as this does not medicalised intersex as a disorder. Similarly Diamond & Beh (2008) suggest that:

> "[T] this [DSD] terminology is no less stigmatizing than many other terms. The largest intersex support group in the world—Organisation Intersex International—considers such terms negative, supports the use of non stigmatizing expressions" (p5).

Clearly no global agreement exists with respect to terms and I am confident that debate will continue and the terminology will evolve. Within the context of my thesis I use the same terminology I use in clinical practice. I have accepted DSD as the generic (albeit contested) terminology for all intersex conditions, which result in a baby with visible ambiguous genitalia (AG). Ambiguous genitalia will continue to be used as a key descriptive term since many parents used this within their stories.

2.5 The 'optimal gender policy' and 'informed consent policy'

Two approaches to the management of infants with ambiguous genitalia are discussed in the literature and operationalized in real life; the "optimal gender policy" and the "informed consent policy" (Thyen et al, 2005).

The optimal gender policy (OGP) focuses centrally on the theorising by John Money in the 1950s (Money et al, 1955). Money and his colleagues proposed that early sexual reassignment of "atypically sexed children" was possible (Roen, 2004). However, this needed to be carried out before a critical age and the child needed to be raised unambiguously within a gender role consistent with their new or clear sex. The cornerstone of the OGP is the nurture not nature theory (Gurney 2007). Gurney (2007) discusses this theory and summarises clearly the salient issues:

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"the theory became the sole justification in many parts of the world for assignment of sex and gender in genital intersex cases. Early interventional surgery for maximising the sex-appropriate appearance of the external genitalia was recommended to facilitate gender appropriate rearing and the chosen sex, at least in part, based on sex role stereotypes" (p633).

Essentially the optimal gender policy assumes a reductionistic relationship between genital appearance and identity (Kessler, 1990). Holmes (2008) suggests that:

"[I] intersexed infants and children face a prevailing perception that they are so seriously damaged it is impossible even to conceive of admitting them to the category of personhood without performing extensive and immediate medical and surgical intervention on them" (p170).

Therefore, ambiguous genitalia are reduced to biology; the distinction between male and female creates the impetus for early genital surgeries to maximise gender identity and minimise stigma (Money et al, 1955). Furthermore once a sex is established the assigned gender pathway is followed throughout childhood to adult life. Within the OGP it is suggested that a "non-disclosure of ambiguity" to the child is essential (Money, Hampson & Hampson 1955). Adults treated in this manner have described their experiences as "well kept secrets" (Liao, 2003). Studies on adults have also revealed the negative impact of unanticipated disclosure of their genital ambiguity and their previous surgery has had on them (Preves, 2003).

The full consent policy (FCP) proposes the postponement of surgery until the child can give their consent (Dreger, 1998; Wilson & Reiner, 1998; ISNA, 2005). The focus of concern is not on medically necessary treatment but treatments carried out for cosmetic effect (Roen, 2004). Roen (2004) continues to describe the core problems and debates in relation to early surgery:

"the treatment offered may not only be inadequate but may also cause harm. There is also concern that 'normalising' treatment renders intersexuality invisible and maintains it as shameful: something that needs to be hidden or erased. This is counterproductive from the point of view of those seeking to build positive identities and communities around notions of sexual diversity" (p127).

Currently no evidence base exists for non-treatment and the proposal of waiting for the child to decide seems "a very poor philosophy" according to those supporting the OGP such as Ahuja (2004). Opponents of the FCP suggest the possibility of later psychological problems believing that untreated children can be left questioning their gender (Nihoul-Fekete, 2005). Melton (2001) describes how advocates of the OGP believe that early surgery is necessary to prevent longer term consequences and she suggests that:

> "children left to grow up in gender limbo will encounter many obstacles in our highly sexually dimorphic society. For example, what toilet should a girl with a small phallus use at school? They will be teased and bullied and may grow into adolescence feeling frustrated and sexually inadequate" (p 2110).

Morland (2001) responds to this by suggesting that there is "no published documentation of the extent and nature of such teasing" (p 2085). Furthermore, supporters of the FCP believe early cosmetic reconstructive surgeries have the capacity to alter the child's self-perception resulting in the child believing:

> "their intersexed bodies were unacceptable, perhaps unlovable, and certainly unrecognizable as persons" (Holmes, 2008 p170).

The FCP clearly focuses on the social construction and interpretation of AG as well as the child's inclusion in the decisions making process. For this to happen in practice the child needs to know the truth about their AG. Influential advocates of the FCP, Alice Dreger and Bruce Wilson, express strong beliefs and debate the ethical issues in their publications in relation to the "child deserving to know the truth" (Koyma, 2004).

The two approaches to managing AG are oppositional. A shift in medical thinking as a consequence of evidenced based medicine (EBM) coupled with a rise in bioethical debate has resulted in the OGP being questioned in practice. Although the long term research evidence for the FCP is absent from the literature the surgical practice of cosmetic irreversible reconstructive genital surgeries in infancy is approached more cautiously (Melton, 2001) and she concludes that:

"what is best for an intersexed baby remains a contentious issue in need of good long-term follow-up data" (p2110).

2.6 The social construction of sex, gender, intersex and disability in relation to AG

The role, function and image of the body in society is a very complex concept discussed in great detail in the fields of gender studies and humanities (Looy & Bouma, 2005). Risman (2004) sums up the four key social scientific traditions developed to explain gender:

> "The first tradition focuses on how individual sex differences originate, whether biological (Udry, 2000) or social in origin (Bem, 1993). The second tradition, perhaps portrayed best in Epstein's (1988) *Deceptive Distinctions*, emerged as a reaction to the first and focuses on how the social structure (as opposed to biology or individual learning) creates gendered behavior. The third tradition, also a reaction to the individualist thinking of the first, emphasizes social interaction and accountability to others' expectations, with a focus on how "doing gender" creates and reproduces inequality (West and Zimmerman, 1987). While we do gender in every social interaction, it seems naive to ignore the gendered selves and cognitive schemas that children develop as they become cultural natives in a patriarchal world (Bem, 1993). The [fourth and] more recent integrative approaches (Connell 2002; Lorber, 1994; Ferree, Lorber, and Hess 1999; Risman, 1998) treat gender as a socially constructed stratification system" (p430).

Many authors such as Kessler & McKenna (1978), Fausto-Sterling (2000), and Kitzinger (2004) have spent their academic careers exploring the concept of gender and the body. In this subsection I aim to establish a clear meaning of sex and gender within the context of my study, rather than debate at length and theorise the impact that genital ambiguity has on society's understanding of sex and gender.

The physical appearance of the body is important since the visual body and the social values associated with it are attached to both appearance and ultimately self-understanding (Howson, 2004). Looy & Bouma (2005) suggest that within Western society:

> "most of us take for granted a harmony between our biological sex and our psychological experience of being female or male, our gender identity. We just *are* women and men, and the relative effortlessness of this identity can lead us to reify a simple, dichotomous view of gender. People are

meant to be either female or male, both physically and psychologically" (p166).

Therefore sex and gender are concepts that are dependent upon what a body 'looks like' and how it 'functions' as either male or female based on the belief of "binary opposites" (Roen, 2004). Howson (2004) suggests that "we discern sex from genital appearance" and that additional physical characteristics, for example, physical frame or presentation of gender such as dressing femininely "tell a story about sex". She continues that:

"[T] the contemporary dimorphic model of sex is invariant, in that sex is assigned according to a fixed set of scientifically derived criteria, which we assume will be constant across the life course. Western societies do not on the whole accommodate individuals whose sex is either ambiguous or altered" (p41).

Zucker (1999) supports the above and suggests that society's expectations are challenged when individuals fall outside "the certainty" that is behind the binary model of sex or their sexuality is in opposition to either their sex or gender. Looy & Bouma (2005) discuss the effects of a gender-dichotomous culture and suggest that:

"[O] one question that is regularly raised by intersexed and transgendered persons, and by some feminist scholars, is whether it is our strongly gender dichotomized culture that *creates* 'disorder' of gender identity. In a context where everything from pronouns to dress to expected interests and roles is defined as *either* female or male, people who do not feel comfortable in either of the two available categories indeed have a problem. But is the problem the context or the person? Is it possible that gender is, both biologically and psychologically, a much more varied
- phenomenon than human cultures generally allow?" (p171).

Feminist sociology has previously sought to establish a conceptual distinction between sex and gender, although Kitzinger (2004) suggests this model is now being challenged. However the prevailing discourse within feminist research considers that 'sex' is defined by biology while 'gender' refers to the established psychological, social and representational differences between men and women therefore "gender represents a set of learned social attributes" (Oakley, 1985).

Therefore when no clear delineation between biological sexes exists (AG) both the 'sex' and 'gender' of the child become increasingly complicated since biology no longer informs the "social parenting" and in turn the "identity" of the child as either a boy or girl (Stein, 2004). Therefore a possible difficulty exists for parents regarding how to support the development of their child's self-identity when they are uncertain about their child's sex and gender. Preves (2003) explored the significance of self-identity within her study of 37 intersex adults all of whom were recruited from support groups. Interestingly, within the qualitative text analysis Preves (2003) paid attention to the issue of the concepts of "self and stigma" as a consequence of AG. Preves used Mead's (1934) theory in her work with adults born with AG. She proposed that since individual 'self' is a product of society shaped by social experience, individuals with AG were likely to be considered abnormal.

Mead (1934) believed that children start to develop as social beings (that is, recognising who they are and what their relationship is to others), through a process of imitation. This involves, initially, the simple copying of behaviour and, as the child develops, experimentation and innovation. My reflection on her theory is that the child has to learn how to fit into the existing society he or she is born into. The child's learning is influenced by the parents' ideology of how to raise their child in line with the various moral and normative expectations of the social groups and society to which they belong. In this respect, gender preconceptions about how males and females should be raised and how they should respond to adult stimulation (such as toys, books and language) are in place from the moment a child is born. Already parents of children with AG are at a disadvantage since the immediate socialising of their child into a particular male or female gender, for example giving them a male or female name, is limited by the sex uncertainty.

Once established the child's belief of their gendered 'self' needs to be maintained by the parents and ultimately the child. Goffman (1968) considered that knowledge of and adherence to social rules and rituals helps to sustain the social interaction of self. Therefore as long as the child with AG performs, presents and appears male or female they can anticipate a degree of

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predictability in social encounters. These interactions have the capacity to further reinforce the child's belief in relation to their gendered 'self'. A possible risk exists when there is an incongruity between sex and gender in either how the child appears or the gender behaviours they adopt. Generally, physical attributes of a male or female body allow the individual to behave in socially expected patterns, such as boys stand to void, girls sit to void. Ambiguous genitalia can result in compromises to expected bodily behaviour, such as boys sitting to void. In such circumstances perhaps the body "betrays" the AG and the certainty of how a male or female body should behave is jeopardised (Howson, 2004). Goffman first proposed in 1968 that a "bodily betrayal" has the capacity to result in the individual being stigmatised. Cull (2005) from her own experiences of AG suggests that for people with AG, stigma was tied to risk with intimacy. She also discusses that individuals were stigmatised if they sought psychological support. Recent studies by Jürgensen et al (2006), Duguid et al (2007), Gough et al (2007) and Rebelo et al (2008) all make reference to parents' concerns surrounding their child being ridiculed or stigmatised.

The early work by Money et al (1955) found that children with AG could be stigmatised by unwarranted "peer curiosity and ridicule" and "neighbourhood gossip". To overcome such stigma Money et al (1955) encouraged parents to raise their child in line with generally held normative expectations associated with gender appropriate behaviour. Parents were actively encouraged to dissuade their child from activities or gender typical behaviours that did not match their assigned sex. As Money proposed "sexual neutrality at birth", the child with AG could be "nurtured into adopting a positive gender self-identity" that did not necessarily match biological sex (Sandberg & Mazur, 2004). As described earlier for this to be successful the uncertainty of 'sex' needed to be surgically resolved in infancy and brought in line with the child's gender, in order to limit stigmatisation. Money claimed that his theory was successful and his findings were focused on what was to become the high profile "John/Joan" case as Dreger (2002) reports:

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"[A] after a pediatrician accidentally destroyed the penis of an identical twin boy (who was not intersexed) during circumcision at eight months, Money recommended to the parents that the child be made into a girl. They decided to take his advice and for years Money claimed the sex reassignment had worked. We now know that that child, who grew up to take the name David Reimer, was never happy as a girl. John Colapinto tells his story--including his attempts to rebuild what he could of the male anatomy that was taken from him in "reassignment" surgeries in the book "As nature made him"" (Dreger, 2002 p1).

If stigma relates to the sense of 'self' the "John/Joan" case demonstrated the challenges experienced by this individual in unifying his feelings with his 'social presentation' of gender. It was Diamond and Sigmundson (1997) who revisited Money's "John/Joan" case and discovered the problems that David had faced during his childhood and adult life. The story of David Reimer is particularly sad since he took his own life as his twin brother had done two years earlier (Usborne, 2004). Traditional claims that in order to develop a stable gender the corresponding same set of visual sex genitals are necessary are beginning to be questioned and rejected by many professionals (Diamond & Beh, 2008). However as Feder (2002) describes, the legacy from Money's work continues:

"despite the fact that Money's reputation has been irrevocably damaged by the revelation of the deception he engineered in the now famous case of John/Joan the protocols of intersex management that everywhere bear his mark retain a powerful and enduring authority" (p295).

A challenge that the public face in understanding DSD is that successful adjustment stories maybe less interesting than the failures and hence are less reported. Also single case studies need to be considered with caution especially when comparisons are being made (Zucker, 2002). If only failures and single case studies are reported any negative outcomes associated with these have the capacity to influence the management of DSD as a group. If positive outcomes along with diverse experiences are not explored they are excluded from informing practice and research. The paradigm of AG is in a process of change. Yet despite this shift in opinion there is no evidence with regard to the child's or parents' experiences and long term outcomes from delaying childhood genital surgery for AG (Eugster, 2004; Warne, 2008).

Looy & Bouma (2005) suggest that within "our sexually dimorphic society" AG presents "awkward issues for the affected individuals, parents and professionals" (p168). These "awkward" issues include the inability to quickly and confidently assign a sex and gender and in turn, a name to the child. The ramification from this is the difficulty parents face when they struggle to share news about their child's sex with friends and family. Holmes (2008) describes the lack of "instantiate recognition" of sex at birth as challenging as this impedes the "performative pronouncement of a sex upon which to hang a subject/identity" (p172). Since at the point of 'birth' the baby does not yet have any personality traits or display any gendered behaviour perhaps the focus can only be visual and biological. Genital appearance, still for the majority of people, initially determines everything that follows in relation to gender.

Disability studies suggest Holmes (2008) rarely take into account intersex and it is through the "lens of feminist and queer theory orientated ethical discussions" that concepts of the value of intersex within society have been briefly discussed. Holmes (2008) continues and suggests that "intersex characteristics" are believed to be "inherently disabling". This informs clinical practice and drives approaches that aim to surgically reduce disability.

Holmes (2008) discusses intersex as disabling since it presents a shift away from the expected norms of a binary sex. Intersex bodies are considered "disabled" by their lack of congruity with clearly defined male or female. He compares the disability paradigm to intersex activists' search for the acceptance of "difference". He also explores the influence that labelling "intersex as disabling" has on prenatal selection. Interestingly, Holmes (2008) also briefly discusses intersex and illness and the impact this has on how medicine regards the intersexed body as diseased. He describes this very clearly:

> "with intersex, there is an extra layer of struggle as most of us who were treated as children did not experience ourselves as ill, and there is little agreement in the intersex population with the medical assessment of our bodies as diseased, rather than merely different. However, in common with those who have experienced illness, intersexed persons inhabit what Couser describes as "colonized bodies" (Couser, 1997) and granted little opportunity to speak authoritatively and granted little credence by medical audiences" (p171).

Holmes discusses an excerpt from Thomas (2004) in which he believes that intersexed individuals "narratives are discredited as overly personal, as a form of public self-pity" (p171). He believes that authors such as Thomas (2004) can discredit individual experiences which results in devaluing. This in turn impacts on the political drivers that influence the medical management of intersex.

Like Holmes (2008), Hester (2004b) suggests that it is the confusion that results from AG that assigns the ambiguity as an "illness" in the form of "social stigmatisation". Hester (2004b) continues to describe how illness becomes linked to "disease" which in turn places medicine in the position of needing to "cure and heal". Holmes' (2008) suggestion that the medical profession unites illness and disability with intersex resonates with my clinical practice. In practice I support and encourage parents and aim to minimise hospital visits in order to allow them to move away from the belief their child is ill since it attends hospital. Similarly I have found that most parents do not believe that their child is disabled as a consequence of their AG. Holmes last suggestion of discredited narratives ties into the value of 'evidence' and this is discussed further in subsection 2.9(a).

Sex and gender impact on other concepts such as gender identity and role and sexuality and sexual orientation. All are complex and are discussed in areas other than DSD including gender dysphoria (Zucker, 2004), childhood development (Carmichael & Alderson, 2004) and religion (Looy & Bouma, 2005). Furthermore, interest in the body and gender in the fields of "power" and "feminism" has seen an "enormous upsurge in interest" (Van Lenning, 2004). Van Lenning (2004) discusses the body within the context of power and contemporary feminist scholarship and suggests that:

> "there is no unequivocal interpretation of the body. At one end of the continuum is anti-essentialism, founded in constructionism....At the other end, we find an emphasis on somatic experiences and on the necessity of a revaluation of the body and of femininity" (p25).

The challenge to feminist researchers also includes what Hird (2000) describes as the "erosion of the binary model" and she believes that sex should be

considered as a "construction". Van Lenning (2004) argues against the model of sex as a construction and discusses the work from the two feminist philosophers mentioned in her earlier quote. She concludes that:

> "masculinity and femininity are unavoidable" as these are "caused by the force of construction. Given the enormous force and impact of these constructions, it is more effective to stretch the categories of femininity and masculinity than to try to subvert them" (p44).

The debate continues in relation to power, sex and the gendered body and the variation in-between the bipolar of either male or female. To offer clarity I have chosen to frame 'sex' as a visual representation within the context of my study. Gender is regarded as the maleness or femaleness "related to an imposed or adopted social and psychological conditioning" in accordance with Diamond (2002). Parents express the gender of their child through their actions, such as giving a child a name that is "stereotypically masculine or feminine" (Zucker, 1999). Gender is also reinforced through parents' social interactions and modelling behaviours towards their child.

Having discussed the concepts of sex, gender and demonstrated my chosen stance on sex and gender within the context of my thesis, in the following subsection I will review the literature surrounding psychosexuality.

2.7 Psychosexuality including gender identity disorder (GID), transgender and deviance;

Hughes et al (2006) in their consensus paper explore definitions of psychosexuality that are linked with sexuality. They suggest psychosexual development is:

> "traditionally conceptualised as three components. Gender identity refers to a person's self representation as male or female (with the caveat that some individuals may not identify exclusively with either). Gender role (sex-typical behaviours) describes the psychological characteristics that are sexually dimorphic within the general population, such as toy preferences and physical aggression. Sexual orientation refers to the direction(s) of erotic interest (heterosexual, bisexual, homosexual) and includes behaviour, fantasies, and attractions" (p554).

They conclude that psychosexual development is influenced by many factors including; exposure to androgens, sex chromosome and genes, and the sex of the brain. All these factors are influenced by the child's social development and family dynamics. For some AG individuals gender identity is also an awareness of their ambiguity coupled with the gender role they have chosen to adopt once they have determined 'who they are'. Looy & Bouma (2005) discuss that even with knowledge of the individual's prenatal hormone history predicting a concordant gender identity is not reliable. The timing and influencing factors that facilitate the development of a gender identity have been described as "fluid" by Hester (2004a). Kuhnle & Krohl (2002) suggest that:

> "[E] even though we know a lot about the biology of normal and defective sexual development, we know little about how and when normal psychosexual identity develops, and less about what are the essential factors in such development. In short, we do not know at this point how to determine in early life *psychologic* sex" (p95).

Biology is able to offer some understanding of the physiological changes that bring about psychosexual development although how this happens within intersexed individuals is not as clear (Looy & Bouma, 2005). However, when individuals who have no DSD condition believe they are a different gender than their biological sex, gender certainty based on unequivocal biology is brought into question. Looy & Bouma (2005) suggest that:

> "[T] transgendered persons who find this apparent contradiction between sex and gender identity a source of intense discomfort, or experience disgust with their bodies, or have difficulty socially as a result of their gender dysphoria, maybe diagnosed with Gender Identity Disorder (GID)"(p168).

Whilst GID is not generally believed to be a DSD condition it can emerge in early childhood or during adolescence. People who are transgendered can be heterosexual, homosexual, bisexual or asexual, as can any individual. GID and DSD are separate areas of study. However, the social complexities of sex and gender are perhaps areas in which they may share similarities. Transgender persons have "diverse experiences" which limit any conclusions being drawn in relation to biology and gender identity (Looy & Bouma, 2005). They continue to suggest that:

"the fact that many people diagnosed with childhood GID expressed, from as young as age two, a conviction that they were the other gender preferred to play with members of the other gender and to take on those gender roles, suggests that gender identity can form extremely early in development. Whether that identity is responsive to experience is difficult to determine" (p 170).

Psychosexual development is complex and is influenced by biological, psychological and social contexts. The body, how it is perceived by individuals and its role and function in society have been explored in many areas as described by Frost (2005). Women athletes' bodies and gender have been examined closely when suspicion about their sex has arisen (Carlson, 2005). Within the competitive arena of Olympic sport, close up visual examination of the external genitalia was introduced in 1967 and later "blanket chromosome screening" (Carlson, 2005).

Deviance has been applied to bodies that appear or function or behave outside expected societal norms. The consequence within sports, for example, is an end to the woman's athletic career. Liao (2003) discusses deviance in relation to sexual activity and intersex women. She suggests not only do some women believe their body is different but they assume a position of deviance based on painless penetrative intercourse being unlikely or impossible.

Having outlined psychosexual development and the challenges faced by some individuals with DSD I will review the literature surrounding how sharing information about DSD has taken place historically and the shift evident in current practice. I will also explore how sharing information is important within the context of surgical decision making.

2.8 The approach and consequences of parents and professionals keeping secrets from the child or adult with AG

Discussions between parents and their child and medical staff about genitalia are often guarded and obtaining information from individuals about their feelings towards their genitalia can be difficult (Kitzinger, 2004). The sensitivity of the subject of AG and the way in which AG is kept secret complicates the task of open discussion with parents and children. Within the literature secrets in

relation to AG fell into three broad categories. Firstly doctors not informing parents or children of the AG diagnosis. The second subsection focuses on the shift in practice and how doctors now advise parents to tell children the truth about their AG. Finally, the challenges parents face sharing the news of their child's AG to friends and family.

2.8(a) Doctors not informing parents or children of the AG diagnosis

As described previously, early reconstructive genital surgeries for AG in infancy and childhood were the accepted method of management under the OGP (Holmes, 2002). In order to promote successful psychological adjustment to the established sex and corresponding gender, the OGP demanded "secrecy regarding aspects of medical management" (Carmichael & Alderson, 2004). The importance of keeping the child's condition secret from them was conveyed to parents as necessary to reinforce the child's gender and promote "healthy psychosexual development" (Thyen et al, 2005). Hester (2004a) reported that the secrecy experienced by people with AG, created by doctors and parents, resulted in adults with DSD recalling feelings during childhood of isolation and upset. Carmichael and Alderson (2004) explain:

> "the imposition of secrecy has been employed without critical scrutiny of its value in each case. Its use appears to have reinforced feelings of stigma and shame for those affected by DSDD[5]. It can be argued that the perceived need for secrecy has protected healthcare practitioners from addressing some difficult aspects of care. This effectively precluded the opportunity for individuals to explore any concerns they may have had about their condition" (p159).

A secret is defined in the Oxford English Dictionary (1992) as "not open or public; kept private or not revealed" or "not openly made known" or "hidden from view" (p 474). Secrecy within the management of DSD became a principal approach rather than a considered way of promoting psychological well being of a child (Carmichael & Alderson, 2004). Long-term studies of the effect of such a "routine non-disclosure approach" are only just being proposed (Hester 2004b). Hester (2004b) explored adult DSD stories, available on the Internet, in which adults with DSD told of how they had heard their diagnosis and how

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[5] DSDD has been expanded in their text to mean disorders of sexual differentiation and determination

medical staff and parents had colluded in the "concerted practice of secrecy". Many of these stories reflected the paternalistic nature of medicine based on the belief that knowing the truth would be destructive for individuals with DSD. Liao and Boyle (2004) discuss the recurrent theme of secrecy which they encountered in both their clinical practice and also within their studies with DSD women. Non-discussion, inadequate information and deception left some women experiencing shame about their genitals and their sexuality (Alderson et al, 2004; Liao, 2003). Holmes (2002) reports receiving "regular communication" from families and patients in which they describe feelings of "isolation" and "embarrassment" in relation to the "non-discussion". Evidence from studies with adults has increased the realisation amongst professionals that secrecy and failure to reveal information at early stages might be associated with long-term psychological distress (Brinkmann et al, 2007).

Creighton and Liao (2004) highlight a secondary consequence of secrecy in relation to research opportunities and consent. They suggest that uninformed people with DSD will not be able to consent to research due to the nature of being unaware of the AG status. Limited access to potential study participants can result in an over use or even dramatisation of "key case studies" such as the John/Joan story (Kitzinger 2004). Fausto-Sterling (2000) further supports the suggestion that research based on limited high profile case studies using problematic methods remains questionable. Slijper (2003) also considers that the history of secrecy and "inferred shame" has the capacity to limit participants' "motivation" to participate in research "because of a fear of stigmatisation" or associated physical examinations.

2.8(b) Doctors advising parents to tell their children the truth about their AG condition the shift in medical practice

Changes to medical practice over the last decade now means that it is recommend that both parents and children be given full and complete information with "utmost honesty" (Diamond & Beh, 2008). Such a shift in thinking away from the OGP was considered long over due and Diamond & Beh (2008) believe that:

"utmost honesty challenged the belief that candor would damage the parent-child bond, and eventually cause psychological damage to the maturing patient. We now know, however, that the shame and stigma that secrecy and deception breed could be more psychologically scarring than the truth" (p5).

Furthermore the move towards complete sharing of information is driven by both more enlightened medical ethics and recognition that withholding information is perhaps not "legally defensible" (Beh & Pietsch, 2004). Frank (2004a), Dreger (2006) and Arana, (2005) believe that children with DSD conditions deserve to know the truth. However, the shift in healthcare professionals' practice towards 'no secrets' can only come to fruition if parents are open to sharing information with their child (Tyrrell, 2003). The challenge of sharing information focuses on both from 'whom' and 'how' the child should learn about their DSD (Goodall, 1991; Tyrrell, 2003). An additional complication relates to the parents' wishes about 'what' and how 'quickly' information is to be shared with their child. The ambition of full disclosure can sometimes cause direct conflict between parents and professionals (Eugster, 2004).

Parents and professionals now recognise that the concealment or non-disclosure of the nature of the child's AG could potentially isolate the child and bring about feelings of shame (Diamond, 2004). Retrospective studies with adult participants reveal stories from childhood reflecting emotions of sadness, falsehood and shame (Hester, 2004b). A study by Wisniewski et al (2000) with 14 CAIS women (mean age 45yrs) identified that 57% of their participants "exhibited no understanding of CAIS". Participants lack of knowledge about their condition perhaps reflects the "secret" nature of the management of their DSD. Brinkmann et al (2007) study of intersexuality (n=37 adults) reported the need for "general and truthful handling" of information. Poor delivery of information coupled with individuals' limited knowledge and a history of secrets surrounding DSD could impact on patients understanding (Brinkmann et al, 2007). Clearly individuals with DSD can experience a wide range of information ranging from non-disclosure and poor information; this is also true for their parents (Systma, 2002).

2.8(c) Parents not sharing the information outside of the family

Following the birth of the baby with AG, telling friends and family can raise parents' anxiety (Carmichael & Alderson, 2004; Warne, 2005) and parents need help "in deciding what to tell family and friends" (Ogilvy-Stuart & Brain, 2004). Carmichael & Alderson (2004) suggest that parents need time to think through the "implications of sharing information about their child's condition", although they do not make explicit these implications. Recent guidelines developed by the Intersex Society North America (ISNA) for parents and professionals are available on the internet and offer guidance on sharing information (ISNA, 2006).

In Hester's (2004b) study of adults with DSD he reports that they were deeply hurt as a consequence of their parents' keeping secrets about their DSD from them during their childhood, adolescence and young personhood. Adults with DSD also believed that these secrets had impacted on their ability to validate their identity. Furthermore, Hester (2004b) describes how boys with AG as a consequence of severe hypospadias reported memories of surgery which were traumatic. The support the boys received from their parents played a very important role in the boy's self-esteem and sense of "healing" after hypospadias surgeries. Being told the truth is apparently a very important element of adult DSD stories. Evidence of "sadness stories" surrounding the secrecy of childhood genital surgery for DSD exists in the literature as Frank (2004a) illustrates:

> "[B] both academic research and the website of the Intersex Society of
> North America (ISNA) present stories of people who feel mutilated by
> surgeries that sought to correct differences in genitalia" (p24).

Frank also suggests that "we lack the stories of the decision makers" in relation to AG surgery and perhaps the reasons for the stories they tell their children. Le Maréchal's (2001) AIS study[6] is both explored and cited heavily by Carmichael and Alderson (2004). Le Maréchal's (2001) study is one of a small number of studies undertaken with parents of children with either CAIS or PAIS.

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[6] However, Le Maréchal's study exists as an unpublished thesis and despite contact with the British Library and supervisors on the study, one of which was Carmichael, I have been unable to obtain a copy of the thesis from UCL.

Carmichael and Alderson (2004) focused on the study to provide them with data to write in a chapter relating to gynaecology and DSD in which they suggest parents overarching desire was to protect their child from harm. In an earlier publication, Carmichael & Ransley (2002) discuss these findings and offer some insight into the methodology behind the study. Semi-structured interviews were conducted with twenty parents, seven of whom were couples. These parents had a total of 17 children between them of which three had PAIS and the remaining 11 CAIS. The age range of the children was 3-21 years. Data were analysed using a grounded theory technique. Parents were "unanimous in their view" that their children had the right to know the full details of their clinical diagnosis and medical treatment, although they did not know how to deliver this information. Parents considered that their child was 'normal' and by giving information they would confer a view that their child was 'abnormal'. Parents also feared that giving information would mean that their child could or would blame them for their condition or be angry with the decisions that had been taken on their behalf (Carmichael & Alderson, 2004). Parents worried about how their child might react to their diagnosis. They believed that telling their child about their diagnosis too early could result in their child experiencing intense psychological distress. The emotional consequence and shame in telling other people is also seen in Slijper et al's, (2000) study of AIS. Reactions of shame have the capacity to impact on how the child views themselves in the future as they in turn then "keep secrets about their AG" from others (Preves, 2003).

Current practice guidelines (Hughes et al, 2006) suggest keeping secrets from either the parents of a child with DSD or the child is no longer an aim of treatment (Frimberger & Gearhart, 2005; Hughes, 2007). How and when to share information about AG and long term outcomes with children continues to be unclear yet recent studies report that parents are more inclined to share information with their child (Carmichael & Alderson, 2004; Berenbaum,et al 2004). If medical staff withhold information, fail to ascertain parents' understanding or are not truly honest with information there is the potential to impact on parents' capacity to be fully informed in the decision making process. Therefore keeping secrets is no longer believed to be a positive approach to the

management of DSD as keeping secrets from children and adults with DSD has a long-term emotional impact and can obstruct informed decision making.

Having discussed the complex nature of 'keeping secrets' in the following subsection I will review the literature surrounding surgical decision making and outcomes from early reconstructive genital surgeries

2.9 Evidence based surgical decision making, liminality and outcomes from early reconstructive genital surgeries

This section is divided into five subsections, firstly to explore the evidence base for DSD that informs practice and the inherent difficulties in deciding on the value of best practice where evidence does not exist. The second subsection explores parents' liminality and how this can be dependent on understanding and adapting to information, which has been informed from evidence based practice. Decision making is discussed in relation to theory in the third sub section. The fourth subsection explores studies that have discussed or examined parents' inclusion in surgical decision making. Finally the outcomes from early reconstructive genital surgeries are discussed.

2.9(a) Evidence based practice and AG

Often parents of children with AG seek reassurance that a male or female sex will "definitely be assigned" to their child based on bio-medical investigations (Ogilvy-Stuart & Brain, 2004). These investigations may take several weeks, during which time parents need to learn to live with "uncertainty" (Ogilvy-Stuart & Brain, 2004). Dinc & Terzioglu (2006) identified that waiting for results following genetic testing had a negative psychological impact on parents. They conclude by suggesting that providing information to parents about genetic testing and counselling are important aspects of care. Therefore, during the time spent waiting for test results to come back professionals should not assume that limiting information protects parents (Systma, 2002; Duguid et al, 2007). Systma proposes that:

> "[N] no attempt should be made to disguise the intersex condition, as has been done, by suggesting that the child's 'real sex' can be discovered.

Intersex is a reality. Furthermore, parents need to be informed about the current controversy within the medical profession" (p6).

Eugster (2004) like Myers & Lee (2004) supports the suggestion that all available information should be shared, "including highly publicized and arguably biased" information. Studies of adults with DSD report that participants have limited knowledge about their birth condition (Migeon et al, 2002; Morgan et al, 2005). In the UK a working group meeting of service users and professional people was convened as an 'intersex' forum and this group highlighted the importance of information sharing (Creighton et al, 2004). The significance of sharing information with parents in order to facilitate their decision making is crucial (Duguid et al, 2007; Gough et al, 2007). However tensions may exist between evidence based decision making and patient centred practice; this is briefly explored below.

Decision making within the structure of healthcare is currently focused on evidence based medicine (EBM). However, a few challenges exist with regard to the application of EBM for children with AG and their parents. Roen (2004) discusses EBM within a political context and suggests that there is an increasing demand for clinicians to position clinical practice in a "specific epistemological frame". She focuses on DSD specifically and suggests:

> "[T] there have been repeated calls for more evidence that the current practice does (or does not) work well enough to justify its continuation but the validity of any new given piece of evidence is contested (Diamond, 1999). The way the debate is framed is affected by the relative status of different kinds of 'evidence': scientifically validated 'evidence' about which surgical techniques work from a clinical perspective; qualitative research 'evidence' drawing from the experiences of those living with surgically altered bodies; and the 'evidence' offered by intersex people who choose to speak out publicly about their concerns" (p128).

Therefore challenges exist with reference to building an evidence base for the management of AG. Ideally, collaborative and cohesive partnerships across disciplines including medicine and science, social science and bioethics could support a philosophical and epistemological shift in both clinical practice and the lives of individuals with DSD (Roen, 2004). However, the current evidence base is limited and, as yet, still confusing with regard to the best treatment and

outcome for AG. Furthermore the application of evidence, no matter how small, is often constrained by doctors' limited time to access and understand the existing evidence (Scott et al, 2000). Evidenced based decision making should be a continuous interactive process involving open dialogue between doctors and parents using the best available evidence. Types of evidence available within healthcare include the empirical evidence from research, clinical practice and patients and carers (Rycroft-Malone at al, 2004). The value of focusing on a parent's individual experiences in relation to managing their child with AG is important especially against the backdrop of the limited nature of EBM in the treatment of AG. Evidence that informs philosophical ethics, such as parents' beliefs, is based on the evidence of reason, which as an epistemology is rationalism. Thus the beliefs and values of the parents and the child could be constructed as examples of rational evidence. The dilemma faced in building an evidence base for DSD management relates to the conflicts and tensions between the weight accorded to each form of 'evidence'. Roen (2004) recognises the complex debates in relation to 'evidence' and suggests "that there is too little genuine cross-pollination of ideas" between professions. She believes that each professional group within the team that cares for individuals with DSD "writes for their own audience" and there is limited listening between the team.

In order to develop a robust evidence base for DSD, partnerships need to be built and the political and social contexts of how we construct sex and gender need to be explored. Roen (2004) proposes that more attention should be given to dialogue and to the process of changing understandings and practices. Furthermore, parents need time to adjust to the event of having a child with AG and gain new knowledge and information.

2.9(b) Liminality
Wherever possible decision making is a process that should not be hurried. In the context of cosmetic surgery for AG there is no legitimate need to rush for surgery (Ahmed et al, 2004). The uncertainty surrounding a child born with AG often generates a medically driven approach that aims to reduce or overcome the parents unanticipated liminal state (Hester, 2004b). The liminal state is

characterised by the uncertainty of the child's sex which in turn raises questions about the parents' own identities. During this liminal period parents become disorientated and initially dependent upon healthcare professionals to provide information. Two options exist, firstly; the healthcare professional considers that the AG is a variant of 'normal' whereby the child is not recognised as having different genitalia from the majority. In such circumstances the outcome is that the child is non-medicalised; this results in their being 'accepted' for 'who they are' which inevitably minimises the parents' experience of liminality. Secondly, liminality is overcome when the AG is recognised and surgical intervention is sought to 'eradicate difference' (Hester, 2004b). Liminality becomes the period of transition that exists following the birth of the child with AG to the point at which parents have some perception and understanding of events and the meaning these have for them. Liminality allows for a period of time in which parents' 'normal' or 'expected' thoughts are safely challenged allowing them the opportunity to generate 'new perspectives' of AG. During periods of liminality new knowledge maybe sought and synthesised by parents and a shift in their belief systems may emerge. Therefore professionals are encouraged to recognise that they must give parents adequate information and time to reflect on their child's AG and diagnosis (Warne, 1998; Rangecroft, 2003). Carmichael & Alderson (2004) and Lee (2004) suggest parents will be experiencing overwhelming and difficult emotions at this time. Parents need to be encouraged to participate and ultimately lead discussions and decisions about their child. Some parents may need time to readjust back into their environment and make changes to their life style, such as shifting work patterns to accommodate a child that requires regular hospital visits. For some parents the liminal period allows them to address any feelings and emotions they may have in relation to the loss of a healthy child and future grandchildren (Carmichael & Alderson, 2004).

In the following section an explanatory framework of decision making is presented followed by the key papers and research studies that consider parents' decision making surrounding their child's AG.

2.9(c) Explanatory framework for decision making

Healthcare services base standards and the need for change on outcomes which in turn are based on evidence. This approach to the significance of measurable outcomes and effective management form the basis of professional decision making. Yet as Jürgensen et al (2006) highlights:

> "neither the "optimal gender policy" nor the "full consent policy" relies on evidence-based clinical outcome studies. Decision makers (health professionals as well as parents) have to act in a situation of extensive uncertainty about expected outcomes" (p360).

Decision theory is complex and not the primary focus of my thesis. Many authors have explored decisions and theory in relation to mathematics, utility and rational choice decisions (Savage, 1954), uncertainty and the disjunction effect (Tversky & Sharif, 1992) and both logic and social decision making (Messick, 1999). Classical decision theory focuses on how people can best achieve their desires in light of their own beliefs. Decisions take place under three conditions:

- certainty (outcomes of actions are certain);
- risk (outcomes are not certain but their probabilities are known); and
- uncertainty (probabilities of outcomes are unknown).

Carmichael & Alderson (2004) reviewed Le Maréchal's (2001) study which suggested parents of girls with AIS believed "they made little contribution to decisions regarding treatment" and they did not have the "relevant knowledge or status" to take an active role in decision making. Whereas, Schober (1998) suggests that parents chose early surgery in order to meet their own needs as genital surgery in infancy only affects "the infant's appearance to others". Whilst parents who consent for early surgical correction are criticised by supporters of the full consent policy the rationale behind their "collective" decisions are not reported in the literature (Hester, 2004b). Authors in favour of the full consent policy support waiting until the child can participate in the surgical discussion (Dreger, 1998; Daaboul & Frader, 2001). However, the argument in favour of "waiting for surgery" is predominantly based on adult

outcomes from early genital surgery (Hester, 2004b). Despite this a study by Meyer-Bahlburg et al, (2004) found that most adults in their study (n=72) "did not support major changes in the prevailing policy". The impact of untreated AG on child development or the parent-child relationship is rarely discussed (Carmichael & Alderson, 2004).

The surgical management of children born with ambiguous genitalia has been reported as difficult, evolving and controversial (Rangecroft, 2003). Guidelines that aim to standardise treatment have been devised both within the UK and the USA. Both highlight the need for early diagnosis and specialist treatment centres (British Association for Paediatric Surgery (BAPS), 2001; American Academy Pediatrics (AAP), 2000). The document published in the UK by BAPS focused on specific conditions with reference to relevant literature while highlighting the paucity of long term outcome studies. BAPS (2001) suggested that there were so many specific issues relating to DSD that a policy of no cosmetic surgery would be too prescriptive. BAPS continued to suggest that valid informed consent requires parents to be aware of both the known and perceived "operative" and "non-operative" outcomes. If parents were to maintain a strong bias in favour of surgery once fully informed, Eugster (2004) argues that there could be potential risks in denying surgery. Since no comparative studies of early childhood surgery vs. no surgery exist, Eugster suggests only two "safe" considerations. First, "the ultimate decision regarding early surgery rests with the parents" and should be made in the "context of their own cultural beliefs" but only after a detailed and comprehensive review of all the options, opinions, and available outcome data. Eugster (2004) further explains that in affording parents "surgical authority" no ethical dilemma exists, since in our society all major decisions regarding minor children are traditionally made by parents. Gurney (2007) discusses that until the age of majority in Australia parents have "decision making obligations" and that these include "making various medical decisions on their [child's] behalf" (p636). Kipins (2004) agrees with Eugster (2004) and suggests that if parents believe that surgery is necessary they are required to exercise their authority in ways that secure and promote their child's basic well-being.

Descriptive interpretations of classical decision theory offer some information about how people actually behave and are used in the study by Jürgensen et al (2006). They reported the narratives from two parents who had two children with DSD. Although part of a larger quantitative study these parents were also interviewed and their stories were explored. They were considered to be unique to the study population since they made different surgical decisions about their children's AG. Jürgensen et al (2006) explored how these two parents managed each child's DSD and the different decisions they came to in relation to treatments. Jürgensen et al (2006) found that:

> "[T] the parents decided to assign a gender to their children and to have the relevant corrective surgery done before a definitive diagnosis had been made. In their first child, they chose male sex of rearing; in the second, female. The decisions were made without advanced knowledge of their consequences and can therefore be seen as "risky choices" (p368).

The "risk" described by Jürgensen et al (2006) perhaps focuses more directly on normative interpretations of classical decision theory i.e. the way people are supposed to behave (Tversky & Sharif, 1992). In Jürgensen et al's (2006) study the lack of a "definitive diagnosis" to inform sex was the factor that should have caused parents to wait so they could make a decision based on evidence. However, Jürgensen et al (2006) describe the reasons why these parents decided to opt for genital surgeries:

> "[T] the reason why the parents were ready to take risky choices was their feeling of a great decision making pressure, based on the parental need to be able to see, address, and treat the child as a boy or a girl as soon as possible; their feeling of chronic stress in their everyday lives through uncertainty and indecision; the insecurity they felt in dealing with their friends and relations and their worry about their lack of understanding; their fear of stigmatization/of social isolation of the child/the whole family; and their feeling of being left alone with their responsibility" (p368).

Parents of a child born with AG are in a tenuous position in relation to making the "right" choice. They are limited by the initial medical hesitancy surrounding AG, uncertainty of the long term outcome and the life long consequences of their decisions.

Studies exploring parents' inclusion in decisions relating to their child's genital surgery are limited and comprise of three studies; Dayner et al (2004), Jürgensen et al (2006) and Rebelo et al (2008). These studies will be explored in greater detail in the following sections.

2.9(d) Studies exploring parents' inclusion in surgical decision making

Dayner et al (2004) retrospectively investigated parents' perspectives and participation surrounding their expectations and satisfaction with the medical management of their child's AG due to CAH. The study used a "detailed satisfaction questionnaire" to understand and appreciate parents' motivations, decisions and expectations of medical management. The questionnaire designed by the authors covered recollections of CAH education, initial medical management, discussions surrounding treatment option, details of their child's genital surgery and parents' rationale for surgery including their roles in decision making. Their sample consisted of 21 parents of 17 girls with CAH (8 months – 13 years); both parents responded for 4 children. Dayner et al (2004) reported that 52% of the parents were completely satisfied with the information provided in the neonatal period. All parents recalled being "advised to consent to genital surgery" and remembered a "thorough discussion behind early genital surgery" and being involved in the decision-making process. Two parents chose not to allow their child to have early genital surgery as they did not consider "cosmetic reasons" as a justifiable cause for surgery. Of the remaining parents (n=19) their rationale for surgery included a more "natural" appearance (89%), "better sexual function" (79%) and avoiding "ostracism from others". Parents' satisfaction with their child's genital surgery varied with 14 parents reporting "complete satisfaction" and four "partial satisfaction"; one parent's satisfaction was not reported. Twenty parents disagreed with postponing genital surgery until the child was old enough to decide independently, although two parents did not consent to surgery which suggests a discrepancy in their data analysis section. All parents agreed that the medical history and AG should be discussed with their child at a mean age of 10 years (3-13yrs) and 32% (7 parents) reported they "had begun this dialogue".

Jürgensen et al (2006) reported a case study of a mother's and father's decision making in respect of their two children born with DSD and AG. Both the parents and their children were part of a larger study of gender-role behaviour in children with XY karyotype. Jürgensen et al (2006) further explored this case study since both children were born with the same DSD condition and AG yet their parents decided to raise one as a boy and the other as a girl. Narrative interviews with both parents allowed the exploration of their children's "stories". Analysis focused on "text reduction" into "categories" and was descriptive in nature. The decision making of both parents was explored using descriptive decision-making theories. The mother's prenatal feeling about the sex and gender of her first child (boy) and the appearance of his genitalia influenced both parents decision about genital surgery. Chromosome results gave them a sense of "relief" about their decision, although these results came after their first child had had genital surgery. Not being able to share news about their child's sex was described as "stressful" and "upsetting". The parents believed genital surgery was "medically indicated" and, if postponed, they worried that their son would be rejected or teased by other people. Following the birth of their second child with AG the mother's prenatal feeling that she was carrying a girl influenced their surgical decisions. Lengthy discussions followed regarding genital surgery and a "wait for puberty" approach was adopted. However at six weeks old, their daughter required surgery for a hernia. Her parents wanted to reduce her future operations and chose to have her gonads removed at the time of hernia surgery. At twenty-two months she had reconstructive genital surgery. Although the DSD diagnoses for both children were the same, parents had made different decisions about their sex. These decisions were based on prenatal "feelings" and the appearance of their children's genitalia. For both children, parents adopted a "policy of discretion" regarding sharing information with family and friends in order to "prevent their children and themselves from being stigmatized" (p365). Retrospectively the parents highlighted feelings of being "alone" and that the "lack of information" and "social support" had been difficult.

Rebelo et al (2008) presented a detailed case report to illustrate the psychosocial challenges that existed for a South African family with a child born

with AG. The approach to managing AG in the townships generally followed the informed consent policy. However, the family and social dynamics were considered for this family as they were struggling to adapt to their daughters AG and were therefore offered genital surgery. Rebelo et al (2008), like Wiersma (2004, 2005) observed from practice that children in South Africa are "guided into strict gender categories" and "those that fall in-between run the risk of stigmatization" (p52). This case report highlighted the challenges a mother and father experienced in "struggling to protect" their daughter with AG from the "curiosity and focus of others". The parents measured their daughter's future inclusion in social and school life against the difficulties they were experiencing in their community. Both parents believed that they could only "rear their child properly" if she had reconstructive genital surgery. Rebelo et al (2008) discuss the cultural issues surrounding AG in South Africa's townships. Parents were "fully informed" and their decisions were documented in their child's medical notes.

Parents clearly face challenges in relation to decisions surrounding their child with AG (Howe, 1998). The importance of honest information sharing and discussion cannot be underestimated. In Dayner et al's (2004) study parents were satisfied, for the most part with the initial information they received about their daughters CAH condition. This information helped them in the surgical decision making process. The parents in Jürgensen et al's (2006) study had conflicting information from doctors and even without chromosome results they felt confident in making the "right" decision surgically for their children. Rebelo et al's (2008) parents were fully informed by the medical team caring for their daughter and made an informed decision for surgery based on their experiences and the impact AG had on their daughter's social integration and their relationship.

Where there is no scientific or EBM basis for parents to make surgical decisions, the focus of choice becomes grounded in parent centred practice. Parents use their feelings to help guide their decisions in relation to surgery.

Retrospective studies of surgical and functional outcomes of children and young people with DSD / AG are beginning to be reported more regularly in the literature (Al-Agha et al, 2001; Göllü et al, 2007). Varying techniques for reconstructive genital surgery are described in the literature (Alizai et al 1999; Crouch, 2004; Thomas & Brock, 2007). Nabhan & Lee (2007) suggest that surgery is discouraged for mild AG, although surgery during infancy should continue to be recommended for those with major AG. Surgery for girls with cloaca is complex with significant impact on girls urinary and reproductive systems if not managed effectively (Levitt & Peña, 2005). In line with BAPS and AAP, Eugster (2004) recommends that if early genital surgery is performed, it should be undertaken by surgeons in centres of excellence with particular expertise. Follow-up studies from early genital surgical intervention are limited to adult studies of quality of life (Vates et al, 1999), genital function (Krege et al, 2000; Creighton et al, 2001), reproductive health (Meyer-Bahlburg, 2001), sexual satisfaction, sensitivity, orientation (Dittmann et al, 1992; Minto et al, 2003; Woodhouse 2004) and long term outcomes (Warne et al, 2005; Creighton, 2004b). Longitudinal data are limited and this needs to be addressed so as to better inform practice (Rangecroft, 2003; Jürgensen et al, 2006).

2.10 Parents experiences of having a child with AG and similarities to other conditions

The following section is split into four subsections which include; parents' experiences of having a child with AG or being given the diagnosis of their child's AG. Secondly, how parents negotiate and cope with the uncertainty of their child's sex. Thirdly, parents' interpretations of their child's reconstructive genital surgeries are explored. Finally how parents' stories of their child's AG are similar or dissimilar to other parents' experiences of having a child with an anomaly, such as cleft lip and palate.

2.10(a) Parents' experiences of their child's AG

Slijper et al's (2000) study in the Netherlands explored the emotional reaction of parents to the clinical diagnosis of AIS. Recruitment from a primary population

of 19 families (21 children) generated 18 participating families (18 mothers and 17 fathers) with 20 children[7]. Three children had genital surgery which included clitoral reduction and reconstruction of labia minora and vaginoplasty (mean 3.8 years) for 2 girls and hypospadias surgery for 1 boy. Slijper claimed objectivity and used standardised questionnaires and two extensive semi-structured interviews in his research design. The first interview took place after diagnosis with AIS but before DNA testing, the second 3 months after DNA testing. It was unclear if the interviews were face to face or by telephone. Interviews were reported as "rated" by three independent clinical psychologists although the method of analysis is absent from the paper. Parents' responses to the semi-structured interviews appear to form the basis of analysis however no quotations are included in the paper. Descriptive statistics were employed due to the limited nature of the sample size and also a lack of control or normative data for the psychometric tests used.

Slijper et al (2000) assumed that following the birth of a child with AG parents would experience measurable post traumatic stress. They described that parents reacted to their child's diagnosis with shock, grief, anger, shame and mothers with guilt but they suggested that parents' emotional reactions were not influenced by length of time from diagnosis for either parent. Parents also disclosed variable levels of social support or "partner" support. There was no information regarding the sharing of information about AIS either inside or outside the family environment. Twelve parents reported a positive impact on their relationship as a consequence of AIS. Parents' knowledge in relation to AIS was reported as high with information delivered by a "range of professionals". This study is highly regarded as a significant study in exploring parents' emotions in relation to AG for AIS. However, only three of the children in the study experienced genital surgery and I wanted to explore the nature of parents' experiences in relation to these phenomena more closely. Other studies report parents' emotional responses to AG. Jürgensen et al (2006) report parents experiencing shock whilst Howe (1998) had previously reported parents' sense of shame. Similarly, Le Maréchal (2001) reported parents' grief

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[7] However, table 1 in the paper contradicts the number of participants.

and powerlessness. Clearly AG has an emotional impact on parents and many authors have highlighted the importance of early and ongoing psychological support for parents and children following discovery of AG or a diagnosis of DSD (Slijper et al, 1998, Carmichael & Alderson, 2004; Liao & Boyle, 2004). Rangecroft (2003) highlights that the BAPS guidelines recommend that families should have access to ongoing psychological support.

2.10(b) How parents' negotiate and cope with the uncertainty of their child's sex
Gough et al (2007) report the findings from Weyman's (2005) doctoral thesis. Her qualitative study used semi-structured interviews with 10 parents (three fathers and seven mothers) of children under 6 years of age born with AG. Data were explored using Interpretive Phenomenological Analysis (IPA) which is described as "grounded in text" yet having the capacity to be "more interpretive". IPA coding allowed for the discovery of "unique themes" yet also allowed for interpretation of commonalities across cases. Two key areas were discussed in relation to parents; their belief that intersex was an "unfathomable otherness" and their struggle to "recover a true sex". These core themes were consistent across all the interviews and resulted in two key concepts that informed the discussion which focused on parents' bewilderment of AG and their quest for an unambiguous sex for their child. These themes are explored in the following paragraph.

Firstly, parent and staff bewilderment "upon the detection of a problem" and the ensuing and ongoing "uncertainty" of the child's sex. Difficulty in understanding "intersex" was associated with confusion, disbelief and a "profound absence of knowledge". Uncertainty of their child's sex resulted in parents feeling their child had no status as a person. Parents described their distress and the challenge of having "no language" to describe their child's AG which resulted in a "limbo" phase. Uncertainty also resulted in parents experiencing difficulties "regarding disclosure to friends and family". Parents were disappointed with healthcare professionals as they had anticipated that they would "understand the nature" of AG. Parents had been hopeful that healthcare professionals would be able to offer information and communicate clearly with them. The second theme of "a quest" explored parents "desperate" and at times ongoing,

quest to discover the "true sex" of their child. Parents focused on "biological criteria within a medical context" and actively sought information from doctors. Gough et al (2007) suggest that some parents described surgery as a "means of increasing clarity". For one father surgery "offered" a "sense of relief" by reducing the possibility of his daughter being teased whilst playing in the garden. Parents explored the different physical features of their child and their feelings towards their baby in an attempt to assign 'sex' certainty. Gough et al (2007) also stressed how parents actively adopted gender stereotypes in order to reduce their child's ambiguity.

Gough et al's (2007) paper contributes to the very small number of studies that explore parents' experiences of AG and what this can mean for them. The study did not specifically focus on genital surgery although some of the children had had surgery. Gough et al (2007) challenged doctors to adopt a "more fluid understanding of sex and gender" which would "help parents cope with the initial impact of intersex". This study highlighted some parents' beliefs that doctors were poor at communicating the news of AG to them. Healthcare professionals with a greater appreciation of parents' experiences would undoubtedly communicate better with parents. Gough et al (2007) did not explore parents' concerns when the child had life-threatening conditions and as they tentatively suggest that this might provoke different worries for parents. They concluded by suggesting that:

> "more research is required into the perspectives of parents when their child is born with ambiguous genitalia. It is interesting that intersex is receiving more critical attention and we suggest that both staff and parents would benefit from education and training to promote more fluid and dynamic understandings of sex and gender" (Gough et al, 2007 p14).

Duguid et al (2007) explored 26 parents' experiences of coping following the discovery of their children's genital anomalies (age range 5 days -10.8 years). The significant majority of children in this study were 46XY and it is unclear if this genital anomaly equates to AG. The parent stress index (PSI/SF) (Abidin, 1995) and the coping health inventory for parents (CHIP) (Tak & McCubbin, 2002) were used; additionally 19 parents took part in a semi-structured interview. Statistical analysis did not demonstrate significant challenges to

parental stress or coping. Qualitative data were collected through interviews with 19 parents and separated into "meaning units", then descriptive categories which were assigned "descriptive labels". These labels were refined with subsequent transcripts and seven "principal themes" reported. These themes were:

> "general experience, handling the subject of genital anomalies, concomitant stressors, sources of social support, coping strategies used by the parents, the impact of the condition on the child and the family and suggestions on improving the clinical service" (p349).

Mothers reported "emotional vulnerability" in the post natal period and 60% of parents expressed difficulties in discussing the child's "condition" with relatives or friends. A fifth of parents reported the condition was difficult to discuss between them. For some parents (n=13/19) the genital anomaly resulted in worry about their child being ridiculed or stigmatised. The most common concern for parents related to their child's surgery and the anaesthetic risk. In the majority of cases, parents did not display abnormal levels of stress or coping on psychometric assessment.

Duguid et al's (2007) paper adds to the slowly growing body of knowledge about genital anomalies in relation to parents coping and stress as a consequence of genital anomalies. Sex uncertainty was only present in four of the children in the study; 21 boys had a 46XY karyotype, three children had a 45XO/46XY karyotype and 46XX in one girl. This study explores parents' stress within a week of a child being born with a genital anomaly and it would have been helpful to have compared these data to parents who had had a greater length of time to adapt. This study demonstrated that the parents needed specific and general information about their child and the potential outcomes from surgery. Additionally parents wanted information that was delivered gradually and steadily; written information and illustrated explanations were cited as being helpful. Just over a third of parents wanted local support from other parents with similar experiences.

2.10(c) Parents' interpretations of their child's reconstructive genital surgeries

Discussion of parents' visual impression of their child's genitalia is limited within the literature. The transcript recently discussed by Lee & Money (2004) is more than 25 years old. In this interview a father of a three week baby born with AG discussed relationships with doctors and how blood tests on his baby and waiting for results were distressing, particularly for his wife. In the story this father talks about looking at his child's genitalia following the diagnosis of AG. As he had "never seen too many male babies" he accepted the medical suggestion of abnormality. Lee & Money consider that 25 years later parents basic needs remain unchanged. The realization of "what is initially told to the parents has a major impact" upon them with the wording becoming "etched upon their memories" (Lee & Money, 2004). Kessler (1998) is in agreement that it is doctors that who "teach parents to see their child's anomalies" (p93).

Göllü et al (2007) summarizes medical reasons for early surgery as including maternal oestrogen offering flexibility to tissues, better compliance from the child and by parents with dilatation and the belief that early interventions are not remembered by the child. Göllü et al's (2007) paper also highlights the irreversibility of childhood genital surgery and the potential impact of no or late surgery on the child's "social, psychological and sexual function".

2.10(d) How parents' stories of their child's AG are similar or dissimilar to other parents experiences of having a child with an anomaly, such as cleft lip

Both Ogilvy-Stuart & Brain (2004) and Göllü et al (2007) suggest that AG noted at birth is "incomprehensible" and "distressing" for most parents. Having read literature that focused on reconstructive genital surgeries and the papers edited by Parens (2006) that focus on "surgically shaping children" I questioned if parents of a child with cleft lip and palate (CLP) reported similar experiences to parents of a child with AG. Although CLP is not tied to sex or gender CLP it is a visual deformity that is surgically shaped or altered early in the child's life, similar to AG since some surgeries are cosmetic. For both CLP and AG consent for cosmetic surgeries during childhood is given by their parents. Both CLP and AG are initially shocking and often unexpected events for parents in an otherwise healthy child. Johansson & Ringsberg (2004) suggest that giving

birth to a child with cleft lip and palate can be "emotionally traumatic for parents". They suggest that the deformity "awakens feelings and reactions in family and other people" (p165). Johansson & Ringsberg (2004) describe their approach to data collection and analysis as being qualitative phenomenographic. In-depth discussion of how they analysed the structured interviews is not provided. Two categories emerged from their findings; the first with three subcategories and the second with two subcategories. In the first category Johansson & Ringsberg (2004) describe the "unexpected event of having a child with CLP". This consisted of three subcategories; parents' experience when they saw their baby, support from professionals and treatment from the child health centre. Although some parents reported shock others suggested surprise, happiness or guilt. These concepts are not explored in any further detail within this paper. Parents' knowledge of CLP was low and some worried about future speech difficulties and surgery. Appearance following corrective surgery was more important if the child was a girl. The second subcategory, "support from professionals" highlighted that some parents believed staff had "difficulty in handling the situation" and "knowing how to treat parents" (p168). Although many parents believed that the staff were "very kind and helpful" they had little knowledge of how to support them and at times gave conflicting advice. The third subcategory reflected parents' satisfaction with the CLP team but a desire to see a psychologist for support. The second category of "reactions" included parents' experiences of other peoples' reactions towards their baby with CLP. Generally friends and family were very positive and supportive. Parents had some anxiety about going out with their baby as they felt that sometimes people would avoid them or stare and children would point. The second subcategory recalled parents' experiences once their child had had surgery. Some parents had "great expectations" in relation to the cosmetic outcome from surgery while others were more "realistic". However parents were generally satisfied with the surgical outcome.

This study has some resonance with those of parents of children with AG. Shock is described by parents of a child with CLP and parents of a child with AG. However shock is not explored any further in the CLP study by Johansson & Ringsberg (2004) or the AG studies by Slijper et al (2000) and Gough et al, (2007). Other than a recommendation for referral to a

psychologist, as suggested in all three papers, it remains unclear how parents managed their experiences of shock. Parents in the CLP study did not regard their children as handicapped but as having a defect or "flaw". To some extent this is similar to the study by Gough et al (2007) although parents' described their child's AG as a problem rather than a flaw. Although parents in the study by Johansson & Ringsberg (2004) did not discuss protectiveness towards their child a study by Coy et al (2002) did find that mothers demonstrated "extraordinary protectiveness" towards their child. Parents in the study by Johansson & Ringsberg (2004) described anxiety in relation to surgery and anaesthetic risk and later speech problems and eating difficulties; how they managed anxiety was not explored. It is clear that there is some resonance in terms of trauma and the needs to make surgical decisions between CLP and AG.

Other areas which can guide research include developing an understanding of DSD within the context of the law and children's rights and the political forces that drive issues related to DSD. Finally, the ethical complexities of DSD research in relation to parents and children needs to be considered. The following section will explore these issues within the context of DSD. However, as the main aim of the thesis is not the construction of an ethical paradigm in relation to the management of DSD the discussion will focus on the areas already noted in the literature.

2.11 The law, politics and ethics in relation to AG

The controversy surrounding the irreversible surgical treatment of children with DSD has seen an increase in the media (Usborne, 2004; Boggan, 2005). This has made more public the debate in some countries the legal implications surrounding early treatment (Arana, 2005). Gruney (2007) explores in depth specific case examples of sex determination and assignment in the family court in Australia. She describes how these cases have been dealt with "the up most sensitivity and care" and the outcomes were carefully considered in the context of "what constitutes their best interests in all circumstances" (p650). However, in the final section of her paper she deconstructs the judicial reasoning and the medical evidence and highlights inconsistencies. She concludes:

"the inherent danger of mistakes when infant assignments are performed soley to rehabilitate or improve the aesthetics of ambiguous genitalia can not be justified in the absence of an immediate threat to life or other physiological crisis which is a serious impediment to health" (p661).

Similarly Kipnis, (2004) suggests the law has a reasonable interest in the child but the law can also influence matters of professional ethics. Furthermore, some healthcare professionals may have "competing interest" when treating a child with DSD, they have to also consider the child within the context of their family (Beh & Pietch, 2004). Although parents are required to exercise their authority in ways that secure and promote the child's basic well being they need to try and work collaboratively with healthcare professionals. Meanwhile healthcare professionals must balance any "competing interest" while complying with professional standards, the law and their own ethical and moral convictions (Beh & Pietsch 2004).

Roen (2004) suggests that the surgical or hormonal treatments for DSD are political. She continues:

"[C] clinical process and clinicians themselves are, like everyone else, implicated in the greater 'political' process of making 'gender' work as it does in society. The decision to try and forge bodies and identities within heteronormative and gender normative parameters is not apolitical nor is its success clearly indicated by research" (p128).

Lebacqz, (1997) explores the political issues of DSD in relation to a shift in practice and the need for a louder and more determined voice of change from support groups. Daaboul & Frader (2001) acknowledge that traditionalist practices no longer conform to modern legal or ethical standards of care. However, they believe that some intersex activists.ignore the potential for psychosocial harm to intersex children which is brought about by our society's current understanding of a binary sex and gender. They argue for a middle way, which involves shared decision making with parents of children with intersex and honouring of parental preferences for or against surgery.

The ethical dilemmas involved in the management of children with DSD are complicated and entwined with the law, the social construction of sex and

gender and the issue of consent. The question remains whether surgical action (or inaction) is ethically sound. Although my thesis is not an in-depth study of the bioethical or moral debate surrounding DSD I do address the ethics of and around my study in the methodology section. The ethics involved in everyday practice are important and these include informed consent and the parents' role in the decision-making process. Roen (2004) suggests parents' decisions hinge on their "understanding of what treatment may entail in the long term" (p128). She continues to explore how informed parents are that following surgery:

> "their child's body may still have a substantially atypical appearance after treatment and treatments may lead to complications whose impact continues for many years"

She concludes by questioning if:

> "parents find it intolerable to raise an atypically sexed child, is it ethical practice for clinicians to cosmetically alter that child's genitalia in order to facilitate a parent-child relationship?" (p128).

How parents should act and respond in respect of their child's AG is complicated and what are considered 'reasonable behaviours' in relation to ethical decisions remain complicated. In the following section I will summarise the literature review and demonstrate how the lack of literature and knowledge base about parents' experiences provides the clear direction for the framing of my study

2.12 Conclusion

The biological development of sex is extremely complicated and continues to be driven by science and genetics. The role of biology in gender identity remains unclear. Scientists continue to explore the influence of sex hormones on the developing foetus and suggest there is a difference between the male and female brain. The reported incidence of DSD varies in the literature and the nature of late presentation of some DSD conditions makes it impossible to know the total number of people with DSD.

Historically, DSD has been reported for hundreds of years across different cultures. Some cultures have accepted DSD or even believed it to be a gift

while others have feared or been horrified by the genital ambiguity. The professional management approach towards DSD adopted over the last quarter of a century was guided by Money's (1955) theorising. He believed he had demonstrated, through his case studies, that gender identity was flexible during the first two years of life. Building from this concept, doctors believed they could surgically "normalise the genitalia" allowing parents to rear their child "in the corresponding gender" confident that their child would be "both physically and psychologically comfortable" with their sex and gender (Looy & Bouma, 2005). Academic debate, driven from a sociological, feminist and bioethics perspective, has explored the issues of the social construction of sex and gender. A shift in professional medical thinking brought about by theorising, advances in surgical technologies and the emergence of bioethics is evident in the literature. These areas have raised arguments against the medicalisation of intersex. The emergence of joint working groups consisting of professionals and support groups has helped drive both the dialogue and the debate surrounding DSD management (Hughes et al, 2006; Creighton, 2004). This has included a review of the terminology used within the context of DSD (Reis, 2007). Although many professionals agree with the new nomenclature some professionals and individuals have made a decision to continue to use the term intersex (Holmes, 2008).

Professional practice until the 1990s was guided by the optimal gender policy (OGP). This approach endorsed early reconstructive genital surgeries as necessary if the child was to attain concordant sexual function and gender identity. The cornerstone of the OGP is the 'nurture not nature' theory (Gurney 2007). However, the rise in self-help and support groups for individuals with DSD over the last twenty years has given impetus for a new approach to practice. The full consent policy (FCP) proposes the postponement of surgery until the child can give their consent (Dreger, 1998). A central component of the FCP is informed decision making, preferably in relation to the child but also for parents. The FCP recognises the power in the social construction of sex and gender and how this impacts on the social interpretation of AG. For the FCP to be realised in practice both the child and their parents need to know the truth about the child's AG. The OGP's and the FCP's approaches to managing AG are oppositional and traditionally medicine has followed the OGP. However, a

shift in medical thinking as a consequence of evidenced based medicine (EBM) coupled with a rise in bioethical debate within the law courts has resulted in the OGP being questioned in practice. Although long term research evidence for the FCP is absent from the literature, the surgical practice of cosmetic, irreversible, reconstructive genital surgeries in infancy is being approached more cautiously (Melton, 2001). A clear defecit in the literature is an understanding of where parents 'sit' in relation to either the OGP or the FCP and, if in practice, parents are provided with adequate information and resource to make informed choices for their child.

The role, function and image of the body in society are very complex and discussed in greater detail in the fields of gender studies and humanities. Sex and gender are concepts that are dependent upon what a body 'looks like' and how it 'functions' as either male or female based on the belief of "binary opposites" (Roen, 2004). Howson (2004) suggests that "we discern sex from genital appearance" and those additional physical characteristics, for example, physical frame or presentation of gender such as dressing femininely. Therefore when no clear delineation between biological sexes exists, as in AG, both the sex and gender of the child become increasingly complicated. Biology no longer informs social parenting and in turn the 'identity' of the child as either a boy or girl can be confusing. Holmes (2008) discusses that intersex persons become 'disabled' by medical confusion over their lack of congruity. Sex and gender impact on other concepts such as gender identity, sexuality and sexual orientation. Biology is able to offer some understanding of the physiological changes that bring about psychosexual development although how this happens within intersexed individuals is not as clear (Looy & Bouma, 2005). Absent from the literature are the bases on which parents gender their child and the significance that a concordant sex and gender have for them. Gough et al (2007) suggest that parents start a "quest" in order to find the "true sex" of their child but how they manage the competing emotions they experiences is unclear. Parents' experiences of caring for a child with AG throughout childhood and adolescence are unknown. Furthermore, how parents are influenced by their communities' social beliefs surrounding sex and gender are

uncertain. What factors influence parents' coping along with the aim of coping or adjustment to their child's AG are also absent from the literature.

Psychosexual development and sexual identity are perhaps a combination of biology and socialisation. Whether AG causes anxiety in parents in relation to their child's future psychosexual orientation is unknown. It also is unknown if professionals have a dialogue with parents about the worries they may have about their child's future psychosexual orientation. From a wider perspective the dialogue between professionals, parents and the child with AG has changed direction since a decline in the blanket adoption of the OGP. Whilst professionals no longer keep secrets from parents' about their child's AG, the degree to which parents understand information and how they use it is unknown. Parents are encouraged to share information about their child's AG with their child and relatives and/or friends but how this happens or does not happen in 'real life' is poorly understood.

Professionals are encouraged to recognise that they must give parents' adequate information and time to reflect on their child's AG and diagnosis (Warne, 1998; Rangecroft, 2003). While parents who consent to early surgery for their child are sometimes criticised the reasons behind their decisions remain unclear (Hester, 2004b). Currently studies exploring parents' inclusions in decisions relating to their child's genital surgery are limited (Dayner et al 2004, Jurgensen et al 2006 and Rebelo et al 2008). Longitudinal studies that explore parents' decision making and its effects over time and throughout their child's lives are not reported in the research literature. The perspectives and experiences of adults who experienced early surgery are discussed, for example by Hester (2004b), who suggests that many adults do not blame their parents for their earlier decisions. The significance of parents' surgical decisions and how these are informed needs to be further explored with a group of children with AG that have all had surgery. Furthermore, little is known about parents' decisions following initial 'life saving' surgery and if their experiences are similar to those who have sought cosmetic reconstructive surgeries for their children.

Once parents have made a decision for surgery they will have expectations in relation to the surgical experience and surgical outcome.

Parents' satisfaction with surgical outcome is discussed by Dayner et al (2004) however their experiences and the impact of surgical events on them are unknown. The four key exploratory studies of parents' experiences when confronted with their child being 'diagnosed' as having ambiguous genitalia are those by Slijper et al (2000), Le Maréchal (2001), Gough et al (2007) and Duguid et al (2007). Some of the emotional reactions reported by these authors reflect the findings from Johansson & Ringsberg's (2004) cleft lip and palate study. However, the CLP study clearly identified that parents experienced social support from friends and family. The significant difference between AG and CLP is the visual nature of the anomaly, cleft is highly visible whereas AG can be hidden. Gough et al (2007) suggested that some parents do not share information about their child's AG with friends and therefore their support networks are minimised. All the reasons which influence parents' decisions to keep information to themselves are unclear. Also how parents cope with the isolation resulting from not sharing information about their child's AG or surgeries is unknown.

Having explored the literature and discussed the limited evidence I began to generate ideas of 'what' needed to be explored. As a nurse working clinically with families, I acknowledged my own experiences and the limitations I had encountered in relation to being able to understand and support parents. I wanted to explore parents' experiences of AG and their child's reconstructive genital surgeries and what these experiences meant. The literature highlighted some of the challenges parents faced and reports some emotional responses but did not sufficiently explore parents' motivations and attitudes towards their child's reconstructive genital surgeries. Furthermore I hoped to explore how parents conceptualised their child's AG and how they managed to move forwards and 'cope' with their child's AG and surgeries. I recognised from practice that some parents were able, more quickly than others, to 'cope' but I was unclear how they then interpreted this in their lives. The following chapter explores my methodology and clearly explains how I moved forward from the stage of literature review to formulating a research question and the most appropriate paradigm in which to explore parents' experiences of their child's reconstructive genital surgeries for ambiguous genitalia.

CHAPTER 3: METHODOLOGY

3.1 Introduction

In this chapter, the research aims and questions are defined and my clinical background is both introduced and explored reflexively. The theoretical, philosophical, methodological and ethical issues pertaining to my study are discussed in relation to specific areas of my study. I present my methodology chapter under nine separate sections to afford the reader clarity. Where necessary these sections are split into subsections. The methodology chapter consists of the following sections:

- Aims and the research and the overview of the two phases of study;
- Relationships and paradigms within my study;
- Reflexivity in my research;
- The philosophy and beliefs guiding the research paradigm most appropriate to answer my research questions;
- Narrative inquiry as a methodology;
- The study design and method;
- Ethics;
- Data collection; and
- Trustworthiness in qualitative research.

3.2 Aims of the research and the overview of the two phases of study

The overall aim of the study was to explore and understand parents' experiences of their child's genital ambiguity and the reconstructive surgeries for AG that occurred in infancy and middle childhood. Following a traditional approach to doctoral research studies I was required to register for an MPhil and transfer to a PhD, therefore allowing my study to naturally fall into two phases.

The initial set of aims which guided phase 1 were developed from having read the literature, drawing on my own practitioner experience and from discussion with critical clinical colleagues and academic supervisors. These aims were to:

- Specifically examine the stories parents told of their experiences of their children's genital surgeries focusing on those surgeries occurring in infancy and middle childhood; and
- To focus on exploring aspects of the stories parents told about their children's genital surgeries that illuminated understanding of their motives, attitudes, beliefs and perceptions.

The first phase of my study examined stories from ten parents, focusing primarily on their experiences of their child's ambiguous genitalia (AG). The aim of the analysis from the first phase of data collection was to investigate the parents' stories and descriptions of 'what' their child's ambiguous genitalia meant to them and their motives, attitudes and beliefs surrounding their child's genital surgeries. Having completed the analysis the aims were refined for the second phase of the study to focus on how parents constructed meaning from their experiences. Furthermore I wanted to explore how parents' reactions to their child's AG helped them understand their experiences and in turn how they managed to move forwards and 'cope' with their child's AG and surgeries. The aims in the phase 2 were to:

- Investigate what parents understood about 'genital ambiguity' and 'genital surgery' including surgical decision making between parents and doctors;
- Explore how parents constructed meaning from their experiences;
- Search for how parents' beliefs brought about cohesiveness between genital ambiguity and parenting their child both within the context of family life and the wider social world; and
- Explore parents' relationships with healthcare professionals, including the level of engagement and how this was maximised or marginalised by either the parent or professional.

The second phase of my study focused on expanding seven of the existing parent stories (from phase 1) with second interviews and also collecting stories from an additional five parents. There were four joint interviews from across

both phases. The additional stories were drawn on to ascertain whether my primary interpretations were credible and also to explore parents' understanding and their role and relationships with their child and healthcare professionals (HCP)[8]. The time between data collection in phase 1 and 2 was twelve months. Interpretation and analysis of all parents' stories broadened my understanding of their perceptions, motives and beliefs in relation to their child's ambiguous genitalia and genital surgeries.

3.3 Relationships and paradigms within my study

This section is split into two subsections. The first explores my clinical practice and the limited evidence base that I discovered which allowed me to focus on the research opportunity. The second subsection examines nursing and the place my research has within nursing inquiry.

3.3(a) My clinical practice, evidence base and research opportunity

I believe that my own life experiences have impacted on and shaped my philosophy of being a nurse and 'nursing'. In this subsection I shall briefly make explicit my journey in becoming a nurse and how this led me to explore this area of research. My journey was the motivating factor that encouraged me to explore my practice by using research. A desire to continually improve healthcare for the children and families led me to explore the literature and discover the paucity of evidence available. I also believed that learning about peoples' experiences could help me advance my practice; all of these drivers encouraged me to pursue this research.

Life experiences and a change in circumstance led me to pursue a career in nursing immediately after I had left higher education. I qualified as a RGN/RSCN following a four-year combined nursing course. My interest in caring for children with disorders of sexual differentiation (DSD) and their families stems from my experiences as a urology nurse. I have practised in urology for more than fifteen years and over this time have witnessed an

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[8] Healthcare professionals include all staff involved in the care of the child and family and within my thesis comprise of doctors (medical and surgeons), nurses (ward staff and specialist practitioners), midwives, health visitors.

increase in the professional dialogue, support group networks and research surrounding individuals born with DSD. My clinical focus meant that I worked both with children and young people born with DSD and their parents. Engaging with parents, children and young people provided the opportunity to listen to a range of the experiences they chose to share. I became increasingly aware that I was actively listening specifically to parents' experiences surrounding their child's AG and genital surgery.

At this point I began to explore the literature surrounding parents' experiences and the evidence base for care and service delivery. As already described in Chapter 2 research of parents' experiences of having a child with AG were limited. Agreed medical management guidelines and consensus statements for managing children born with ambiguous genitalia existed (Rangecroft 2003; Lee et al, 2006) yet there was a lack of robust research literature especially focusing on parental experiences within the United Kingdom. This limited evidence, my clinical experience and my growing interest in research served as a motivating factor for me to explore, understand and ultimately share parents' experiences of their child's AG. I believed a research doctorate would nurture and foster my research abilities while also providing me with a concrete support structure to meaningfully investigate parents' experiences of their child's ambiguous genitalia and genital surgery.

3.3(b) Nursing and the place my research has within nursing inquiry

Aspects of nursing research seek to understand the complex nature of the research participants' experiences. Nursing research like other areas of research has the potential for the discovery of unanticipated or extraordinary outcomes. Holloway & Freshwater (2007) discuss the idea that nursing should not focus solely on science as such a positivist approach would neglect the biographical model of developing understanding. Even though my own existing experience was of using quantitative methods I agreed with Holloway and Freshwater (2007). Nursing research should aspire to facilitate meaningful nursing inquiry with a purpose of learning more about the experiences and the 'world view' of those we care for. I believe that listening to parents and formulating interpretations about their experiences within a research paradigm has the capacity to generate knowledge and

enhance practice. For these reasons I was drawn towards seeking out and understanding the changing experiences and outlooks of parents' in their everyday lives, wanting to explore what they saw as important. Parents management of their child's AG could have been assessed psychometrically providing what is considered as 'truly empirical evidence'. However, such approaches evaluate parents' skills, such as coping, at a particular point in time and do not provide insight into their experiences over time. My clinical experience was that parents themselves often told stories of their experiences within the context of their everyday lives. These stories would be added to each time I saw them in clinical encounters and sharing experiences appeared important to them. The questions that became important included; 'what' were parents' experiences, 'how' parents created meaning about their child's AG and 'how' meaning was assimilated into their lives. Therefore, I began to explore methods which would allow me to listen to parents' experiences and was drawn toward qualitative methods and these included narrative inquiry.

3.4 Reflexivity in my research

Within the qualitative paradigm I am viewed as an active component in the research process and a truly objective position is impossible to achieve. Therefore, information about my existing beliefs enables the reader to better understand my subjectivities (Etherington, 2004). In the following subsection I aim to reflexively explicate my values and influences which may relate to my thinking.

Reflexivity is used to indicate an awareness of the 'researcher's self' within the process of research (Elliott, 2005). I was committed to producing research that had the capacity to make a difference in my clinical practice and to parents. I recognised that my thinking would change during data collection and analysis therefore I used a reflexive approach throughout all phases of my study. Elliott (2005) suggests that:

> "while acknowledging that all research accounts will be partial and will be
> shaped by the intellectual biography of the author, there is a desire to
> make those accounts as informative as possible and to provide insights

into the means and circumstances of their production. An approach to conducting and writing up research which makes clear the perspective of the author and describes the practicalities of how the research has been conducted is therefore advocated" (p154-155).

Critical reflections taken from my notebook[9] (started prior to the start of my data collection) allowed me to explore my expectations and early beliefs about parents of a child with AG. Also influenced by the reading I had undertaken I speculated that parents would explore their sentiments about having a child with AG in some depth. I wondered if parents' would be influenced by 'scientific logic' and seek certainty from 'investigations' which would provide an explanation for their child's ambiguous genitalia. I anticipated that parents' would focus on medical staff to interpret and link information into a definite diagnosis of 'cause and effect'. I supposed that 'empirically verified knowledge' (Crotty, 2007) would help parents conceptualise their thoughts in an orderly manner similar to those described by Fisher & Goodley (2007) in relation to disability and illness. Once the initial phase of discovery of their child's genital ambiguity had passed I anticipated that parents would focus their actions on achieving future goals for their child in a similar way to Siegel (1988) where action in the present is assessed "in terms of the efficiency of means in achieving ends". Therefore I anticipated that parents would consider medical interventions as both certain and critical for their child, as a 'means to an end'. Furthermore, I wondered whether the social construction of a binary sex and the medicalization of AG drove parents to seek surgical solutions for their child with ambiguous genitalia, possibly placing surgery in the context of a curative role.

As previously discussed in the literature review a review of guidelines and consensus statements (Rangecroft, 2003; Hughes et al, 2006) highlighted the dilemma that there are no absolutes or guarantees for parents of children born with AG. I was interested in whether parents themselves functioned within a positivist paradigm expecting to receive care that was 'proven' to be the right approach in managing AG. I wondered if parents would seek evidence from scientific medical research and evidence based medicine (EBM).

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[9] Notebook – This applies to the place in which I wrote my thoughts prior to starting any data collection. Once I had commenced data collection I kept fieldnotes, which are referred to and drawn upon throughout the thesis.

If parents were relying on EBM how did healthcare professionals share the information about the uncertainty of treatment outcomes and how did they decide between the 'optimal' or 'informed' management approaches. This I felt was an enormous responsibility for healthcare professionals when they themselves operate in a world of constraints and uncertainty (Blakemore, 1998).

At this point in my thinking I began to very tentatively explore the methodology and the study design that would enable me to gather meaningful data from parents. I drew upon my clinical practice reflecting on how I already engaged with parents and how this could be used within my research. Koch (1998) recognizes the value that every day practice has to generate research inquiry. She states that:

> "[T] the phenomena of interest to nursing are those identified by nurses and patients in the course of everyday encounters" (p1184).

Combining my clinical experience, skills and research I aimed to develop a research inquiry that had the capacity to explore parents' experiences of their child's genital surgery and develop further my reflexive awareness. I then had to broaden my thinking again and consider what I had specifically hoped to learn. I wanted to learn about parents' experiences and their understanding not solely based on 'causal determinism or relationships' but by appreciating how parents related to their experiences (Snape & Spencer, 2004). I hoped to achieve this by using a method that allowed parents to say what was important to them. Furthermore, I wanted to explore the social influences that influenced and shaped parents' thinking. I was interested to explore if parents of children born with AG shared a common social reality or if multiple context-specific reality existed. Additionally I wanted to explore if the parents' social behaviours in relation to their child and gender were governed by 'social laws' that could be seen as immutable. Finally I sought insight and clarity from the parents about the healthcare professional (HCP) role at the moment of discovery that their child had ambiguous genitalia and also what support they required from HCP during their child's childhood.

I began to challenge my initial attitude to knowledge that had previously been informed by the modernist traditions of positivism. I shifted my thinking and recognised that empirical research methods were not the epistemological

stance from which to explore parents' experiences. Therefore in order to pursue my research study I started to consider other more relevant philosophical and epistemological frameworks. My aim was to expand my thinking and consider how best I could learn from parents' experiences and what philosophical base would best suit my study.

3.5 Philosophy and beliefs guiding the research paradigm most appropriate to answer my research questions

This section is divided into two subsections; firstly I outline my philosophical view of the value in human experience. Secondly, I frame my thinking within a theoretical and epistemological stance.

3.5(a) Philosophical view

Four areas within philosophy that are often identified by authors are logic, ethics, epistemology, and ontology (Flamming, 2004). Many philosophers have existed and will in the future exist; some will share beliefs, values and concepts while others will endeavour to possess a degree of singularity. For the majority of individuals 'human experience' includes physical and cognitive growth, exposure to social and emotional encounters and interactions that expand knowledge and create understanding. I believe human experiences are multifarious and complex. I also consider that elemental or core reasoning exists for individuals as a consequence of their beliefs, experiences and constructs.

To question the issues that surround the research idea I needed to explore and also justify my chosen methodology and method. The need to explore openly and honestly sent me in search of my assumptions about reality and the understanding I have of "what human knowledge" is and how do I believe I will attain this in my research (Crotty, 2007).

3.5(b) Theoretical perspective and epistemology

Philosophies shape inquiry through different ontological beliefs (the nature of reality), because what is understood to constitute reality drives the epistemology (how do we know what we know about the world) and the methodology (how we

gain knowledge about the world) (Denzin and Lincoln, 1998; Koch, 1998). By briefly exploring the theoretical perspective and epistemology behind adopting a specific methodology I aim to explain how it provides a context my chosen approach.

The prevailing approach to research has been the scientific method which is concerned with the measurement of sizes or associations, discovering the 'objective truth'. I briefly explored this but concluded that within the context of my study this would be unrealistic as I currently did not know what 'the truth was' in order for me to explore this with specific measurement tools. As I read further I came to recognize that I rejected the view there was an objective truth that was waiting to be discovered. In subjectivism, meaning does not "come out of" an interaction between subject and object "but is imposed on the object by the subject" (Crotty, 2007). He continues to suggest that the "object" makes "no contribution to the generation of meaning" (p9). Although this is a very simple explanation and is actually much more complex my thesis is not a philosophical exploration of epistemology, but I aim to make clear why I rejected some ideas and approaches in favour of others. Within my current subjectivist thinking I believe that knowledge about ourselves and our social world is constructed. The parents' knowledge of having a child with ambiguous genitalia is dependant upon their perception of AG coupled with their social experiences. As such personal beliefs, values and past experiences all help individuals construct their own philosophies about the world they live in (Harper & Hartman, 1997). I believe that parents' experiences are reasoned by them internally, this process does not change their perceptions of 'who they are' but perhaps coupled with interactions and their social world these experiences help them create meaning. Constructionism became an epistemological stance I wanted to explore further as Crotty (2007) explains:

> "[I] it is the view that all knowledge, and therefore all meaningful reality as such, is contingent upon human practices, being constructed in and out of interaction between human beings and their world, and developed and transmitted within an essentially social context" (p42).

Social constructivists propose that human experience is naturalist and engaging and involves continuous conscious activity and so intentionality exists (Searle,

1995). This activity is often ordered and people rely on their feelings and emotions to make meaning of their experiences. The experiences give the person a sense of self and identity but they do not exist in isolation since people grow in living webs of relationships (Gergen, 1999). These meaningful and socially-embedded orderly and disorderly experiences are reflected in people's ongoing learning and consciousness (Searle, 1995). Social constructionism does not confine reality to the mind but recognises knowledge in what peoples' experiences mean to them (Reissman, 2008). Therefore by listening to parents' descriptions or narrations of their experiences they describe experiences which are reports of how they reacted to something and thereby meaningfully constructed the experience.

Having explored epistemology I concluded that my study was rooted within social constructionism and I then began to consider how I would best be able to interpret the knowledge that I gained from parents. As constructionism and phenomology have been described as "intertwined" I explored this initially.

Phenomenology as described by Husserl is a return to the lived world, the world of experience, which he views as the starting point of all science (Sadala & Adorno, 2002). Edmund Husserl (1859-1938), was a German philosopher and is referred to as the creator of phenomenology (Annells, 1996). He introduced the concept of our common world, as experienced by us, which. Husserl called our 'life-world'. This life world provides a framework for discussing the subjective perspective and features of our way of structuring the world of which we are unaware (Annells, 1996). Phenomenology takes the intuitive experience of 'phenomena' as a starting point and tries to extract from it the essential features of experiences and the essence of what is experience. Merleau-Ponty (1908-1961) further developed phenomenological thinking expanding the concept of the body-subject by exploring the characteristic of consciousness and the ability of the human body to perceive events that are intricately intertwined. To summarise, Husserl was interested in the epistemological questions of knowing and recognized experience as the ultimate basis of knowledge with our life, his philosophy focuses on the lived experiences (Koch, 1995).

Unsure if this fitted with my thinking I explored hermeneutics. This has been defined as the practical art or process of applying understanding and interpreting the meaning of written texts (Finch, 2004). Martin Heidegger (1889-1976) was a German philosopher who in his early career began exploring the meaning of 'being'. He considered that understanding 'the meaning of being' was dependent upon the ability to consider an individual's point of view and to appreciate the cultural and social factors that may have influenced their outlook. Gadamer (1900-2002) studied with Heidegger and supported the suggestion that an individual's particular history and culture helps shape them. Heidegger contemplated that philosophy should be a description of experience, by this I believe he meant that in trying to understand experience we have to recognise that we are 'who we are' because of our relationships and experience in the world. Researchers from the hermeneutical position approach their research with some understanding of the population they are studying. Gadamer (1989) recognises that people come from different places and have different opinions, beliefs and ideas and differences in background can create a set of prejudices and bias which can give rise to intrinsic values and meanings. Koch (1998) suggests that Gadamer's philosophical hermeneutics:

"does not show us what to do, but asks us to question what is 'going on' while researching" (p1183).

For the researcher, asking what is going on while researching and having an awareness of apriori beliefs can facilitate openness and trustworthiness in the data. In summary, Heidegger's philosophy refers to the interpretation of the experience (Wilson and Hutchinson, 1991).

I wanted to try and find a perspective that would bring together these two positions as I believed that there are multiple complex factors that influence parents' lived experiences and their interpretation of these experiences. I considered that once I had achieved this I would be able to explore my methodology with greater certainty. From my reading I came across Ricoeur. Jean Paul Gustave Ricoeur (1913-2005) was a post-structuralist reflective hermeneutic French philosopher. Ricoeur believed that our self-understandings are created through the effects of the imagination during interpretation. His constant preoccupation was with a hermeneutic of the self, fundamental to

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which was the need we have for our lives to be made intelligible to us. Ricoeur developed a theoretical style and explored narrative identity within the context of 'who am I' and 'how should I live' (Roberts, 2004). He acknowledged the legitimacy of conflicting views and he believes these are explored in our search to live good lives. He suggested that people interweave varied concepts and discourses to form a synthesis discourse in which new meanings are created (Frid et al, 2000).

I considered that social construction and Ricoeur's thinking provided me with a viewpoint from which to explore parents' social worlds and the meaning they attribute to having a child with AG. Therefore I embarked on searching for a methodology that would allow me to understand how parents made sense of their experiences and in turn understand the place this knowledge had in their lives.

3.6 Narrative inquiry as a methodology

This section explores the key areas significant in respect of my methodology. I also explore both the use of and nature of 'story' and 'narrative' within the context of my study.

In accordance with Crotty's (2007) description my research methodology was the strategy that informed my research design which in turn informed my choice and use of a particular method which linked the research question to the desired outcomes. The literature review and my reflexive thinking furthered my philosophical reasoning and I recognised I would not be able to test existing hypotheses relating to parents' experiences since none were evidenced. Also since parents' accounts of their child's ambiguous genitalia and genital surgeries, specifically within the UK were unavailable, it was difficult to recognize which concepts to explore quantitatively. I therefore needed to consider a methodology that:

- Was dynamic enough to consider parents' experiences yet at the same time provide them with ways to make sense of the past;

- Allowed parents to share what they believed was important which in turn permitted me to explore with the parents unexpected emerging phenomena and follow avenues of curiosity as they occurred during data collection;

- Used a naturalistic[10] inquiry approach that was not intrusive to parents and which allowed me to explore phenomena about their 'self', life with a child with AG and their experiences;

- Took into account the active relationship parents have with their world and the influences their social world has on them;

- Allowed for a mix of both inductive and deductive reasoning. This required an in-depth approach as I desired to 'see things' from the parents' perspectives. Building on the strengths from my clinical background my chosen methodology also had to allow me to track a logical course of interpretation as well as being able to follow inductive paths which would lead to the construction of valid interpretations from parents stories;

- Gave parents the opportunity for long episodes of conversation in which they could extend their accounts of their lives and experiences in contexts that developed over the course of the researcher/participant interaction;

- Allowed for the clear identification of the detailed phenomena under study and permitted interpretation across complex and sensitive parent experiences;

- Assisted in building a 'safe inquiry space' in which a mutual understanding and trust could exist and where parents' memories were not subject to question and I was able to accept their personal experiences;

- Allowed me, where necessary, to 'check out' data so that a shared agreement existed between myself and the parents and which gave parents' freedom to restructure how they were recounting their experiences;

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[10] Within my research I use naturalistic and naturalness to describe how I aimed to engage informally and conversationally with the parents so that they could tell their stories in 'their usual ways' as much as possible.

- Took into account my explicit beliefs about AG and the notion that objectivity (neutrality) could not be attained;

- Provided a framework for reflexivity in accordance with Ricoeur;

- Provided a setting for rich data collection that was natural and unmanipulated and parents experiences could be recalled over time[11]; and

- Facilitated me to construct an interpretive account of parents' experiences over time.

Qualitative methods can be considered practice-friendly for clinicians, yet there are deeply labour intensive and time consuming elements that are necessary to create and deliver a credible study (Padgett, 2004). I acknowledged that any methodology chosen could be challenging. I considered narrative inquiry as a methodology that would, to a greater or lesser degree, meets the prerequisites as described above and also allow me to use my nursing skills within a research setting. In line with my own beliefs, as described earlier, and having read and spoken with Riessman (2008) I agree with her that stories "reveal truths about human experience". Therefore in line with others such as Riessman (2008), Ekman & Scott (2004), Frank (2002; 2004b) and Clandinin & Connelly (2000) I believe narrative inquiry is a methodology interpreting participants' experiences through questioning that develops understanding and generates knowledge.

After reading texts including those by Hydén & Överlien (2005) and Elliott, (2005) I considered that narrative inquiry gave parents the permission and opportunity to tap into their tacit knowledge embedded within their experiences while also allowing me as the researcher to learn from the process. Since narrative relies on communication and relationships, this link can facilitate connections between my research role and the parents, perhaps providing a sense of 'shared' learning. Narrative inquiry highlights these close alliances and allows for the exploration of personal truths and core values of both myself as the researcher and of the parents (Holloway & Freshwater, 2007). Narrative

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[11] Time as Ricoeur suggests is important since this allows for human experience, action and suffering to be heard. He believed that only in and through the act of telling a story can this time acquire a form

inquiry offers an in-depth understanding of the situation and meaning for those involved (Gaydos, 2005). Overall narrative inquiry provided me with the basis for both entering parents' life worlds (through their stories) and understanding socially embedded knowledge (through the interpretation and analysis of their narratives).

3.6(a) Where and when 'narrative' or 'story' within my study

The word 'narrative' originates from the Latin *gnarus* meaning knowing, while 'story' derives from the Greek and Latin *historia* which also means knowing (by inquiry as well as by account of events) (Holloway & Freshwater, 2007). Ricoeur (1981) recognised that the narrative not only describes and clarifies life but also plays a necessary element in understanding it "a life understood is one recounted". Lorem (2008) considers that using narrative as the starting point makes it possible to look for both creative and truthful possibilities to understand life through the stories we tell. Frank (2005) also offers a distinction between the concept of narrative and story. He uses the term 'narratives' when referring to the general structure which encompasses a number of perhaps similar stories and 'story' when discussing the tales people tell.

'Narrative' and 'story' are often used interchangeably within conversation. Paley & Eva (2005) are not surprised that 'narrative' and 'story' have become virtually synonymous within every day language. They do however suggest that the terms 'narrative' and 'story' should not be used interchangeably and a failure to make a distinction between the two can result in 'misconceptions' in analysis. Within the context of this study 'narrative' and 'story' will not be used interchangeably. Story will be used to reveal the parents' oral histories of their experiences, and is used within this context in the findings (see Chapters 6, 7, 8 and 9). Narrative will be used when describing 'narrative in-depth interviews' as a data collection method.

3.6(b) The nature of stories

Narrative inquiry uses stories as the 'data building blocks' for subsequent interpretation. Understanding how stories are constructed and considered as data are important within my study. Stories have two fundamental features, they have to be told (or read or observed) and listened to. Telling stories is a

basic and common means of human expression (Riessman, 2008). Recollections, reflections and memories from childhood are examples of stories which others can listen to. These stories exist in everyday life as instruments of communication. The construction of a story in literary terms displays characters, tells of events and circumstances and forecasts how characters may behave and act. Within a literary domain the study or the 'art' of the story explores the conventions of literary style, development and evidence of genres and the creativity of individual narrators (Elliott, 2005). The narrator of the story presents the setting, character, dialogue, actions and events which constitute the story (Koch, 1998). The structure of the story is the composition of knowledge while the content is the information and experiences created by the individual narrator or the "oral version of personal experience" (Labov & Waletzky, 1967 p12). People's lives are stories and they can chose to share them in order to modify them, practice telling them or create new stories. Therefore by telling stories parents provide their histories and in the process construct personal biographical 'narratives' bringing into play key features in their lives, their identity, sense of self and their values (Mason, 2004).

The constructions of stories are continuous with the progression of time and can therefore be invaluable in the research process. I considered that this temporal nature of stories would allow parents the opportunity to review and retell past events (Labov & Waletzky, 1967). Polkinghorne (1988) believes that the "temporal dimension" is crucial in stories since temporality links the effects "diverse happenings" can have on one another. Therefore in their simplest form stories are continuous and composed of connecting elements such as a beginning, middle and end (Riessman, 2008). Stories have clear structures that are instrumental in bringing about knowledge through interpretation, such as a plot and genre. They progress over time and in doing so deliver the essence of what they are 'about' (Holloway & Freshwater, 2007).

Outside of the discipline of literary studies 'narrative inquiry' has expanded to facilitate the gathering and interpretation of data across the human and social sciences in a structured approach (Sands, 2004; Mishler, 1995; Riessman, 1993). Sands (2004) suggests that, in general, qualitative researchers view stories as:

"constructions created through interpersonal, sociocultural, and historical processes. Stories or narratives constitute, rather than reflect, some aspect of a socially constructed reality" (p49).

In addition to this Koch (1998) suggests that stories can be viewed as an "interpretation and as a research product" (p1182), thereby allowing the collection of authentic information about people and situations. In my study I believe that stories have the capacity to provide a textual representation of parents' experiences following the birth of a child with AG. Interpretation of these stories would provide professionals with the opportunity to understand the previously unreported phenomena of parents' experiences of having a child with AG who subsequently underwent reconstructive genital surgery. I believed that interpretation of parents' stories would contextualise parents' own accounts of their personal development and oral histories and as such explore areas that are unknown or little known to healthcare professionals. Koch (1998) suggests that in addition to the discovery of health issues, stories can:

"show where we as healthcare professionals have gone wrong. Listening to the voices of clients may show us what to do to improve practice" (p1183).

Stories are close to parents' everyday 'lived experiences' of having a child born with ambiguous genitalia. Frank (2007) suggests that people have expectations of how things 'should be' after a series of everyday life events. Any alteration to this course of events is challenging, as perhaps demonstrated by the parents in my study. Parents may have to reassemble their lives and I anticipated they would share this through their stories. Frank (2007) suggests that people are able to "hold their own" in a world that has changed for them and their stories are evidence of how they do this in reality. My interpretation of parents' stories aimed to illustrate how they "hold their own" through both the analysis and interpretation of the content of their stories. I hoped that by understanding how parents of children "hold their own"; new knowledge would be generated which would have the capacity to increase understanding while also informing future clinical practice and service design.

3.7 Study design and methods

Lewis (2004) suggests that a "good qualitative research design clearly defines purpose" and should offer coherence between the research question and the methods. The coherence of parents' narratives and my interpretation of what Reissman (2008) calls "a related facet of trustworthiness" is made transparent in both the study design section and the analysis Chapter 5. She continues by suggesting that:

> "students can ground their claims for validity by carefully documenting the process they used to collect and interpret data" (p193).

The research aim was discussed earlier (section 3.2) as a lead into philosophical and methodological approaches and is reiterated here to add clarity to the methods section. The overall aim of the study was to explore and understand parents' experiences of their child's genital ambiguity and the reconstructive surgeries for AG that occurred in infancy and middle childhood.

The research question was 'what narratives do parents of children who have had genital surgery tell?'

The following section is divided into two subsections in order to explore the target population, recruitment approach and the sample. The first subsection explores the target population as the potential group of people that met my inclusion / exclusion criteria. The second subsection explores recruitment and the sample size.

3.7(a) Target population and inclusion/exclusion criteria

Initially I was uncertain as to the overall target population of potential participants that would be available. I sought to explore mechanisms by which I could identify parents who fitted the inclusion / exclusion characteristics. These were as follows:

Inclusion Criteria

- All parents of children under the age of eleven born with DSD, ambiguous genitalia or cloaca, which required at least more than one episode of reconstructive genital surgery.

Exclusion criteria

- Their child had experienced 'minor' genital surgery (evident from the hospital coding system)

- The child was unwell or had a life threatening illness / major reconstructive surgery (evident from the contact data on the existing hospital system, known to the researcher)

- Their child was over the age of eleven or had already progressed through puberty[12] (from parental disclosure or the child was known to the researcher)

- If the parents were foster parents (evident from hospital admissions data form)

- The parents were, themselves, acutely unwell (disclosed by the parents or attending Consultant)

- Parents of children who had a child born with ambiguous genitalia who had had surgery but later died (evident from the hospital admission data form)

- Parents of children who had been gender reassigned. There are a very small number of children in this group and thus it was felt it would be difficult to maintain parent anonymity

The coding department at the hospital retrospectively identified children who had been admitted for a broad range of reconstructive genital surgery over the last decade via the hospital data record system. Coding the surgery against the admissions data reduced the initial target population frame from 80 to 36 children. There were two reasons for the reduction in the sample. Firstly, incorrect coding had been assigned by the hospital coding department; this became evident when the coding data were compared to the admission data. On closer inspection of the coding and admission records some children had only had 'one off' minor genital surgery. Secondly, a group of children who had had surgical procedures and were correctly coded as genital surgery did not have DSD, ambiguous genitalia or cloaca, (examples included surgery for

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[12] Puberty can be an additional cause of parental concern and is a period of transition for the young person. Additionally this age group should be included in the decision-making process for surgery and I wanted to explore the parents' experiences of their child's genital surgeries during early childhood.

imperforate hymen or labial adhesions). Additionally by cross matching the coding information with the admission history data I identified those parents who were not suitable for inclusion in the study since their child had died. I purposely did not use the child's medical records to access the surgical details of the genital reconstructive surgeries as I wished to remain open to hearing the parents' experiences of events.

Once the target population was complete I constructed a sample matrix (Appendix 1). The aim in developing a sample matrix was to devise a purposive non-random sample method which aimed to recruit participants who fitted the inclusion and exclusion characteristics equally from the three DSD groups.

3.7(b) Recruitment and sample size

To achieve participation from the complete target population is unlikely within research, including narrative research (Holloway & Freshwater, 2007). I made a decision not to access the whole group but to seek a representation from the overall target population. I was unsure about the potential uptake rate from the target population and considering the sensitive nature of the topic I was aware that some parents would not wish to participate in the study. I considered the possibility that from the 36 parents included in the target population, if all were lone parents and agreed, my maximum sample size would be 36, however if all families consisted of two parents my maximum target population could be 64. Once the target population was identified, the child's lead hospital consultant was approached as they were the 'gatekeeper' (Holloway & Freshwater, 2007). The consultant was requested to provide written consent for the parents of their patients to be approached about the study. Once I had received this I would be able to approach the parents. I was guided by the ethics committee with regard to recruitment numbers; they suggested that I have no more than 15 participants in the study. This is discussed further in the ethics section in Chapter 3 (subsection 3.8); essentially the ethics committee did not wish me to exhaust the possible target population in a single study. By approaching 'families' both mothers and fathers of the same child could be considered either independently or as a couple.

Recruitment involved a single written request from the researcher to the parents asking them to consider participating in my study.

The invitation letter was addressed to both parents and emphasised the voluntary nature of participation (Appendix 2). A parent information sheet was included with the invitation letter outlining the study along with a reply slip and a pre paid envelope for those who wished to participate (Appendix 3). Again the parent information sheet emphasised the voluntary nature of participation. Recruitment data are shown in Table 3.1.

Recruitment Table 3.1

Group	Parents approached Phase 1	Parents approached Phase 2	Agreed to participate Phase 1	Agree to participate Phase 2	Non-respondents Phase 1	Non-respondents Phase 2	Parents Interviewed and number of Interviews Phase 1	Parents Interviewed and no of interviews in Phase 2	Sex of child	Age range child
DSD	11	3	4	3	7	-	3	5	3 girls 2 boys	1 < 4 years 4 between 6-11 years
AG	6	1	4	-	2	1	4	2	3 boys	3 between 7-10 years
Cloaca	3	2	2	2	1	-	2	2	3 girls	1 < 4 years 2 between 5-8 years
Total	20	6	10	5	10	1	9	9	11	

Table 3.1 identifies that a total of 26 families were approached from a total population of 36. Fifteen parents agreed to participate in my study, these 15 parents were from 11 different families.

3.8 Ethics

This section is divided into six subsections exploring the complexity of ethics within my research study. The first subsection explores those areas which focus on research ethics and includes the parents introduction to the study and the need for anonymity and pseudonyms. The second subsection includes informed consent, giving parents' time and the opportunity to discuss my study, how I sensitively approached my study participants and how they were provided with support. The third subsection explores practitioner researcher issues and the mechanisms that supported the researcher. The fourth subsection discusses the ethical issues pertaining to data management and access. This includes data collection, storage, transcription and the supervisory team's access to data and the research monitoring process. The fifth subsection explores two specific ethical challenges which were experienced during my study and how these were managed. Finally the sixth section explores ethical issues in relation to interviews and transcriptions as data.

3.8(a) Research ethics

Ethical approval for my study was sought from the university and my local research and development (R&D) committee in August 2004. The study was peer reviewed, passed local R&D and proceeded to be reviewed by the Local Research and Ethics Committee (LREC). LREC approved my study in September 2004 (Appendix 4). The University faculty Ethics Committee (FEC) also approved my study subject to receipt of a hard copy of the approval letter from the LREC, they received this in late September 2004. Fieldwork commenced in October 2004 and ran until both phases were complete in February 2006.

A discussion had taken place with the ethics committee around my proposed contact with parents. This discussion had focused on how parents should be invited to join my study. This was proposed to be either by letter or by telephone; if by telephone this would be followed up with an introductory invitation letter. The chairman of the ethics committee suggested that contact via an introductory letter while also enclosing the parent information sheet was the most suitable approach and this was agreed. Parents' postal contact details were checked against hospital admissions data and available NHS

information to ensure they were up to date and correct. Parents were invited to join my study by an introduction letter which included the parent study information (Appendix 2 & 3). Parents were assured that pseudonyms would be used throughout my study. Both my telephone contact and secure email address were included in the parent information letter to provide parents with an opportunity to discuss any details of the study. The information sheet informed parents that any stories they chose to share would be kept confidential. Confidentiality was discussed with parents at the time of consent and is discussed in greater detail in subsection 3.8(b). Additionally parents were informed that quotes from their stories would be used in future publications and presentations and that these would be anonymous and pseudonyms would be used.

3.8(b) Informed consent

If parents decided they wanted to participate in my study they returned the tear off slip in the stamped addressed envelope. Parents had a minimum of a week but more usually several weeks to read the study information and decide if they wanted to participate in my study. Once parents returned the tear off slip I made telephone contact and answered any questions they had in relation to my study. Parents suggested the location and time of the interview and parents understood they could withdraw from the study at any point.

At interview parents were approached in a sensitive manner, general introductions were made and I explained my study and discussed issues of confidentiality, anonymity and professional issues in relation to child protection. Parents were given the opportunity to ask any questions and the need for written consent was discussed. Hospital consent forms were used (Appendix 5) and written consent was taken at two time points. The Coldicott guardian had requested that written consent be taken twice. Written consent was taken at the beginning of the face-to-face interview, before parents' started to tell their stories. Written consent was taken a second time at the end of the interview after parents had had the opportunity to reflect on the stories they had shared. The second written consent was taken to confirm consent at the end of the interviews for the stories parents had told. Parents were made aware that their anonymised stories from the interviews would be cited in reports, articles and

presentations. I explained to parents that consent was a process rather than an event and they could withdraw their consent to participate at any time in my study. I reassured parents that withdrawal of consent would not effect the treatment options for their child. I explained to parents that written consent was necessary and that privacy, confidentiality and anonymity would be maintained. Confidentiality and privacy I believe are very important however, as described by Elliott (2005) it can be very difficult to "ensure participants do not become recognisable". Elliott continues even if a "few details are changed and a pseudonym used, it is likely friends and family" will recognise participants. Therefore the "possible risk to anonymity once the research was written up" (Elliott, 2005) was discussed with the parents prior to obtaining informed consent. Parents' right to withdraw from the study existed until the first paper which was published in 2007.

I explained to parents that I might need to ask questions during the interview if there were elements of their story I did not understand. I had an interview schedule that was to be used as a guide or prompt if parents were struggling to tell their stories (Appendix 6). Prior to starting the interview I acknowledged the sensitivity of the topic area and I explained that if parents were to become upset I would stop the recording. I also explained to parents that ongoing psychological support was available to them, if they wished, following their participation in the study. Arrangements were in place for a fast track parent referral to a psychologist working within our extended clinical team. This approach aimed, as Kvale (1996) suggests, to "minimising risk to the participant" (p116).

Once the interview was completed parents were given the opportunity to ask any further questions and there was a period of 'checking out'. This period was to ensure parents emotional well being prior to my leaving the interview venue (Kvale, 2007). Parents were given copies of their written consent.

3.8(c) Practitioner researcher issues

Although I am a clinical researcher the parents who had volunteered to participate in my study were not part of my active clinical caseload. Whilst the parents were directly involved in my study I ensured that I was not their direct clinical care provider although I was part of the team available to provide

parents with support if this were necessary. However, following the completion of the data collection phase of my study several parents and their children became part of my caseload. I have previously explored earlier reflexivity (in section 3.4) within the context of my study and I recognise that conventionally researchers do not admit to personal interests in their research (Schratz & Walker, 1995). However, researchers explore topics, not only on the basis of theoretical importance or originality, but also as a result of their interest in the study phenomena (Lowes & Weeks, 2006). I have acknowledged and explained my interest in my study area. The literature suggests that many nurse researchers study topics related to their work and, presumably, of interest to them. For example Campbell (1999) a lecturer in oncology explore the feelings of oncology patients nursed in isolation and Lowes et al (2005) a diabetes specialist nurse, explored parents' experiences of their child's newly diagnosed diabetes. It is perhaps the motivation that clinical researchers have for a specific practice based topic that sustains their interest, commitment and enthusiasm in their research (Singleton et al, 1988).

Throughout my study as a researcher I acted in accordance with hospital policy and the organisational, academic and professional code of ethics. As a researcher I visited parents' homes to undertake interviews and I worked within the boundaries of my own organisation's lone worker policy to ensure my safety. I acknowledged the initial 'novice' nature of my qualitative researcher skills and made sure that I had access to academic, professional and psychological supervision. An academic supervisory team based within university offered guidance, support, discussion and advice in relation to my study as well as emotional support where necessary. Professional support was from nursing leaders within my organisation as well as my medical colleagues. I also had supervision with a clinical psychologist.

3.8(d) Data management

Issues of confidentiality were addressed with parents verbally and were written into the parent information leaflet. I explained their stories would be transcribed by a professional transcriber who would be bound by a confidentiality agreement (Appendix 7). Once I had checked the transcription, the digital recording was erased. Transfer and storage of data was protected and this

process was assessed by the hospital data management lead and also the IT department. The raw transcript data and analysis was stored on a password protected hospital system and all data were anonymised. In accordance with the ethics committee and hospital policy the recruitment information and any raw paper copied data is stored in a locked and secure cupboard for ten years and will then be destroyed. Patient notes were not used in my study and therefore no information about the study is available via patient notes to other professionals or researchers.

My academic supervisors had requested access to the raw data prior to commencement of the study. The ethics committee agreed they could see anonymous data once they were CRB checked and had obtained an honorary contract within my organisation.

As with any research study my R&D department monitor the progress of research within a research governance framework. Monitoring involved an annual report submitted to the research ethics committee and a monitoring visit by the R&D lead in September 2006. Following the monitor visit I was allowed to continue with my study as all the paperwork had been in order.

3.8(e) Ethical challenges in research practice

During the design of my study I discussed the possible ethical challenges that I could face with the chairman of the ethics committee. Together we discussed the sensitivity of the subject area yet acknowledged the need to be able to understand parents' experiences of their child's AG. The invitation letter was discussed and I listened to his advice and used this approach for recruitment. During phase 1 of recruitment one parent telephoned me and was very unhappy that I had contacted (I refer to 'the parent' to try to maintain anonymity). This parents' concern had focused on the possibility that a contact letter could be read by someone else in the household or be incorrectly addressed and opened by someone other than the intended recipient. The parent was also concerned that I had read their child's notes although I was able to reassure the parent that I had not. This parent requested that I put a letter in their child's notes stating no future contact which I did. I discussed this with the chairman of the ethics

committee and he believed I had acted within the boundaries of the code of ethics.

The second ethical challenge I encountered related to a story told by one of the mothers in my study. In her story she recalled a negative experience with one support group and elaborated her grievances against them. Whilst exploring the literature I came upon a story, from the same support group, written by one of the support workers. On reading the support group workers story I was able to identify the mother from my study. I was aware that the mother had not given permission for her words to be used by the support group. I discussed this with both an academic and clinical supervisor and a decision was reached to try and contact the publisher to enquire after their consent procedure. The publisher did not reply to my attempts at contact.

Although I had tried to anticipate all eventualities, on reflection this was a little naïve. However, I believe that I managed both of these two major challenges in practice both professionally and ethically. Other ethical issues are addressed in the next section.

3.8(f) Ethical issues in relation to interviews and transcriptions as data

Within any research interview the participant has the opportunity to ask questions of the researcher. All the parents in my study knew of my clinical role and my place of employment and I speculated there would be times they would ask clinical questions or try to seek my opinion of their experiences. Oakley (1981) described how she faced similar experiences. When her participants asked questions she made a decision she felt was ethically balanced and she decided to "respond honestly" to requests for information. I made a conscious decision prior to undertaking any interviews that I would answer parents' questions to the best of my ability.

Although the aim of the study was made clear to parents from the information sheet and through our initial discussion, I was conscious that parents might not be prepared or aware of what stories they might divulge. Corbin & Morse (2003) agree with others and suggest that in a "comfortable" home atmosphere participants may share information they might not have otherwise chosen to reveal. Furthermore the margins between researcher and hospital nurse may

cross conventional trust boundaries which Corbin & Morse (2003) suggest "may entice the participant into providing information that they later regret" (p338). To try to safeguard against 'regretted stories' being included I asked parents to give written consent before and after their narrative in-depth interview.

The potentially exploitive nature of interviews has been discussed in the literature (Elliott, 2005; Holloway & Freshwater, 2007). Some authors suggest narrative interviews go a considerable way in trying to re-empower the participant and offer the participant the opportunity to reveal only what they choose (Graham, 1984; Mishler, 1986). However, some authors consider that narrative interviewing has the capacity to 'open wounds' (Lieblich 1996) perhaps leaving both the participant and the researcher vulnerable. Elliott (2005) suggests that:

> "[I] it is not necessarily harmful for research subjects to experience distress in the course of an interview, and it may in fact be therapeutic or reassuring for a respondent to be given a safe space in which to talk about an upsetting experience" (p137).

An additional consideration when undertaking interviews in parents' homes relates to what I would see around me as this "scene" (Holloway & Freshwater 2007) can impact on the interpretation of interviews. I believed that keeping fieldnotes was important. These would later help me to unpack some of the contexts in which the interviews were collected and provide an opportunity for reflection.

Stories shared through narrative interviews not only describe but constitute the parents' 'selves' and as such have the capacity to shift parents' thinking interviews have "the potential for significant transformational experience" (Holloway & Freshwater 2007). Parents were made aware that psychological support was available if they experienced any emotional challenges brought about by sharing their stories.

Interpretation and analysis of parents' narratives also has the capacity to impact either negatively or positively on the parent. Elliott (2005) discusses this in a chapter titled "ethical & political implications of using narrative". At no time did I

wish to display any "elements of dishonesty or duplicity" (Elliott, 2005) and I hoped that by sharing my interpretations I sought authenticity in my data.

The ethical dilemmas within research, including those which arise through narrative inquiry, are complicated. By exploring these prior to starting my data collection I was hopeful that I would be prepared for ethical problems as they arose in the field.

Ethical issues are also addressed as a thread throughout my study, for example the ethical dilemmas that arose during the interviews are discussed in the findings Chapter 6. The following section explores data collection.

3.9 Data collection

This section is divided into six subsections the first of which explores why I believe in-depth interviews are a good source of data. In the second subsection I explore how in-depth interviews can generate stories from participants and question if there is a difference between in-depth interviews and narrative interviews. The third subsection explores the skills that are necessary when a researcher is using narrative interview. The fourth subsection considers the dynamics of narrative interviews undertaken with mothers or fathers individually or as couples. The fifth subsection explores the four interview phases that I anticipated parents would move through during the interviews. Finally, in the sixth subsection I discuss the recording and transcription of the interviews with parents

3.9(a) Why in-depth interviews as data?

I appreciated that parents' experiences would be available as naturally occurring data in their child's medical records. I wanted to collect data in a manner that would make the research topic clear, be practical and timely whilst also being sensitive to parents' emotions. I therefore chose a generated-data collection approach. Lewis (2003) considers two approaches to generated data, 'in-depth interviews and group discussions' and proposes that generated data collection methods allow for both spontaneity and questioning and:

> "give participants a direct opportunity to convey their own meanings and interpretations through the explanations they provide, whether spontaneously or in answer to the researcher's probing" (p57).

When considering the potentially sensitive nature of data that could be shared I considered that in-depth interviews would provide safety for parents to tell their stories. Focus groups were rejected in favour of interviews based on the advice offered from the parents in my clinical caseload. These parents had suggested they would have preferred to be interviewed if they had been eligible to participate in my study. They talked about how they would feel too inhibited to discuss their child's genital ambiguity in a group. Although focus groups had strengths, it was decided that the depth of data that I hoped I would derive from one to one interviews would not be achieved within a group context as the topic was seen by parents to be extremely sensitive in nature. I agreed with Lewis's (2003) suggestion that in-depth interviews are especially helpful when exploring "very complex systems, process or experiences". She believes that in-depth interviews give:

> "depth of focus and the opportunity for clarification and detailed understanding. Similarly, understanding motivations and decisions, or exploring impacts and outcomes, generally requires the detailed personal focus in-depth interviews allow" (p58).

With reference to my study the sort of complexity that Lewis is referring to exists in relation to sex and gender and the possible secretive nature of genital ambiguity. This is further complicated by the difficulties people have when talking about bodily areas, such as genitalia, that are considered private. Therefore the choice of narrative interviews as the data collection tool was influenced by advice from my existing clinical caseload, the research question, methodology, context and content of the data to be collected. As a form of communication or talk, the in-depth interview connects 'external world events' to the "inner world of thoughts and emotions" which constitute parents' subjectivity (Nunkoosing, 2005). Riessman (2008) considers interviews as "narrative occasions" accepting that most narrative projects are based on interviews of some kind. She goes on to summarise the significance Mishler has had in relation to interviewing:

"[I] in his (now classic)[13] book, Mishler reconceptualizes research interviewing as a discursive accomplishment: the standardized protocol (where question order is invariant) gives way to conversation where interviewees can develop narrative accounts; speaker and listener/questioner render events and experiences meaningful-collaboratively" (p23).

This collaborative union can evoke an additional degree of sensitivity within the interview, which in my study I hoped would allow for the exploration of parents' emotional experiences surrounding their child's genital ambiguity and genital surgeries. In-depth interviews also afforded parents the opportunity to express themselves in a safe and secure manner, building confidence in sharing their stories with myself as the interviewer. Finally, I believed that a 'conversational intimacy' (Ramos, 1989) would exist in which parents would feel in control and comfortable 'story telling'. Parents interpret the context and significance of their experiences and share these through their stories. The co-construction of the interview between myself and parents is important as I have to understand and interpret what they are telling me rather than construct my own story about them. The authenticity of parents stories are also shared through the in-depth interview and this can challenge the argument from positivists that stories are not truths (Nunkoosing, 2005). The truth within stories and the trustworthiness of stories as data are discussed further in section 3.10. The notion that in-depth interviews generate stories brings into question whether there is a difference between in-depth interviews and narrative interviews. This is further discussed in the following section.

3.9(b) The nature of the narrative interview in my study

Nunkoosing (2005) suggests that:

"[I] interviews are the most widely used method for generating data in qualitative social research....and although the literature gives very different names to describe various suggestions for interviewing, there is considerable overlap between the many type of interviews" (p698).

Within my study I consider narrative interviews as in-depth, unstructured interactive interviews. Unstructured interactive interviews have been described

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[13] Mishler, E G (1986) *Research Interviewing: Context and narrative.* Cambridge, MA: Harvard University Press.

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in the literature as narrative interviews, a good example being offered by Corbin & Morse (2003) who suggest that:

> "[I] in unstructured interactive interviews, sometimes referred to as open-ended or narrative interviews, participants are given considerable control over the course of the interview...Participants are asked to tell their story as they see it, feel it, experience it. As such, participants determine where to begin the narrative, what topic to include or exclude the order in which topics are introduced, and the amount of detail. Although researchers play an active role in the unstructured interview process by means of focused listening, they are not the central actors" (p339).

> "[U] unstructured interactive interviews provide the opportunity for participants to construct their stories, reliving past experiences during the course of narration' (p342).

I accept that the very nature of any arranged interview infers a degree of structure since the participants are aware of the topic they have consented to talk about. The parent information sheet posted prior to the in-depth interview informed the parents of the nature of my research interest. I hoped that by keeping the interview as 'unstructured' as possible that the parents would decide which stories to tell and which to omit and that they would feel free about how to tell and sequence their story and the level of detail they would include. Therefore within the context of my study I will use 'narrative interviews' throughout because I believe that narrative interviews are in-depth, unstructured (consisting only of a very brief interview schedule) and interactive (jointly constructed between myself and the parents).

The goal in narrative interviewing suggests Reissman (2008) is to "generate detailed accounts rather than brief answers or general statements". She draws on her own experiences in the field and offers:

> "generating narrative requires longer turns at talk than are customary in ordinary conversations" (p24).

A flow of stories can ensue with one story leading to another, during this shift between stories the interviewer can explore with the participant, associations and meanings that connect stories (Reissman 2008). Narrative interviews have the capacity to consider issues of trust, empathy and sensitivity whilst also being recognized as a method for gathering diverse participant experiences (White, 1981; Weiss, 1994). Narrative interviews stay close to actual life events

rather than eliciting explanations about specific cause and effect events (Holloway and Jefferson, 2000). Narrative interviews captured the naturalness generated by participants which may have been interrupted or suppressed with a structured interview method (Elliot, 2005). The naturalness or spontaneity of participants can also direct participants' stories to "unexpected or unanticipated places" as described by Mishler (1986). Narrative interviews allowed participants to tell their powerful stories 'well' which in turn influenced my interpretation of their narratives as a researcher. My aim was to draw on all of these potential benefits to allow the parents in my study to feel safe and secure whilst exploring stories of importance to them. To be able to undertake narrative interviews I explored the skills I would need as a researcher, these are described in the following subsection.

3.9(c) Interview skills

Padgett (2004) suggests that a key skill in narrative interviewing is expert listening. Corbin & Morse (2003) believe interviewing skills develop with experience and that a good interviewer must be able to establish and develop rapport and trust. Nunkoosing (2005) advocates that the skilled interviewer makes use of the self in interviews, builds relationships, is aware of the flow of conversation and information and encourages and supports participants to deliver and end stories. Furthermore he goes on to suggest that the interviewer should always be mindful of their research question, theoretical and professional position. Reissman (2008) also believes that the interviewer has to create possibilities in the narrative interview for extended narration, which require the "investigators to give up control". My nursing abilities include expert and open listening, experience with health interviews and building relationships with children and families. Therefore all of these skills were applied within the context of narrative interviewing. My theoretical position considered both Ricoeur and social construction and was discussed earlier (section 3.5) while my professional position as a urology nurse was discussed in the earlier reflexivity section (section 3.4). Prior to actually undertaking the narrative interviews I reflected on how I would initiate and respond to the specific stories around children's genital surgeries and whether I would ask probing or clarification questions during the course of the interview. The research

committee had requested a short interview schedule (Appendix 6) which could serve as prompts during the narrative interview if necessary. An interview schedule is actually at odds with narrative interviewing as the aim is to 'follow the stories' rather than to guide, direct or prompt specific stories. However, the schedule was developed to provide the ethics review committee with an indication of the sorts of questions that might arise during the course of the interview. I made clear at the ethics committee meeting that the schedule would not be followed slavishly and only drawn on if prompting was required. Holloway & Freshwater (2007) consider that the narrative interview is restricted to a small number of questions from the researcher and interruption is avoided. A workshop presented by Riessman's (2005)[14] provided me with an early unique opportunity to critique my style of narrative interviewing. After reviewing the first three interviews I identified that occasionally I sought clarity from parents during their stories rather than waiting till the end. Discussion at the workshop allowed other students and academics to critique the first two pages of Faye's story within the workshop setting. Feedback was positive and although I had spoken while Faye shared her story of giving birth to Gabby my interruption was considered by Riessman as necessary demonstrating empathy, sensitivity and intuitiveness towards Faye while also clarifying areas of uncertainty within her story.

3.9(d) Interviews with mothers or fathers alone and interviews with couples

I considered that if parents had agreed to share their stories with me they could choose to do this on their own or with their partner at the narrative interview. Therefore if some parents chose to tell their story together, in the same way that they cared for their child and made decisions together, this was considered an important storytelling device. During my study 'parents' stories were considered the most important elements rather than whether individuals, couples, the mother or the father told the stories. Giving parents the opportunity to tell their story, as an individual did not suggest that jointly told stories were less valued or intrinsically different, all stories allowed for the construction of meaning.

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[14] Master class: 29.4.05 Swansea University – Professor Catherine Riessman. Attendees were asked to submit a section of their narrative interview for discussion in the workshop. CR chose a small segment of Faye's transcript for two reasons. Firstly to explore the narrative interview style but secondly because of her interest in the field under study.

Thus joint story telling, in this sense, was treated no differently to individual story telling. Stories were jointly told by three couples these were treated similarly to stories told by individuals. Interpretation of the three joint stories did reveal that jointly constructed stories emphasised the differences between mothers and fathers.

3.9(e) The interview phases

Corbin & Morse (2003) developed a format for 'typical' unstructured interviews in order to identify potential risk or benefit of the interview. My own narrative interviews closely matched their four step design; pre-interview, tentative, immersion and emergence phases.

<u>Pre-interview phase</u> – During this phase my aim was to explain the purpose of the interview and in turn my study. I aimed to use the parent information sheet as a starting point to explain my study and expand on the process of the interview. During this time parents had the opportunity to ask questions and discuss their beliefs in relation to the value of my research. I aimed to discuss with parents their feelings towards being audio recorded and their satisfaction with my transcription arrangements. This pre-interview phase allowed me to discuss issues of confidentiality and anonymity with parents and the use of their stories in future papers and reports.

<u>Tentative phase</u> – I anticipated that the pre-interview phase would gradually give way to the key topic of the interview. Participants, as Corbin & Morse (2003) suggest do not "just start telling intimate details" and, as reiterated by, Mishler (1999) they work up their story as they "began to trust" the researcher. My aim was to allow parents to shape their own stories as governed by their emotions. This would help them decide which stories to choose to share. I expected that some parents would build trust by "testing" me out and that the emotional intensity of stories might change as the interview progressed (Corbin & Morse, 2003).

<u>Immersion phase</u> – During this phase of the interview I anticipated that parents' stories could become emotive or more urgent. I expected that the parents'

stories might become disorderly and jumbled and I planned to make a quick note if an important story was lost half way through being told. I hoped that at this phase of the interview parents would have developed both strength and confidence in the interview relationship.

Emergence phase – Once parents had told their stories I anticipated that they would move towards this phase of the interview process. I planned to support parents to move away from the more emotive areas and to bring them back to the present. At this point I planned to give parents the opportunity to ask questions and provide them with time to reflect on their interview. We would then discuss again issues of consent and their rights to delete the interview recording at this stage.

Once the interview was complete, written consent had been obtained and any remaining issues had been discussed (for example, does the parent want to see the psychologist) I planned to leave. As soon as possible following the interview I wrote my fieldnotes. I planned to use my fieldnotes as an opportunity for reflection and critical thinking and to inform the reflexive approach which runs throughout my thesis. I anticipated that my fieldnotes would also act as an aid memoir when I read the transcription of the interviews. The recording and transcribing the interviews are discussed in the following subsection.

3.9(f) Recording and transcription

Recording, interviewing and subsequent transcription play a "major" part in representing the data that are analysed in any research study (Riessman, 2008). Reissman describes her perspective more clearly and dedicates a chapter in her most recent book to this subject:

> "[F] from a taped conversation, I constructed a written record that, like all transcripts, straddles a border between speech and writing. I transformed a complex verbal exchange into an object that would serve as a representation-my imitation on a two-dimensional page of what had been said between us. An audio recording is more selective than a video would have been, of course, but in neither case can the fluid and dynamic movement of words and gestures be captured" (p29).

The transcription process should not be trivialised (Elliott, 2005) but is a critical part of the analytical process (Silverman, 1993; Wengraf, 2001).

The dilemma of transcription is challenging and Elliott (2005) goes on to suggest:

> "[I] it is now widely accepted that it is all but impossible to produce a transcript of research interview, or any other type of conversation, which completely captures all of the meaning that was communicated in the encounter itself. Any transcription of speech must therefore be understood as a compromise" (p51).

I considered different approaches to presenting the data in order to be able to use the transcript most effectively. I explored a thematic style described by Riessman (2008) as an "unfolding sequence" this approach focuses on content and uses segments or "streams of speech" from the stories to try and convey what is being said. The transcript is usually referred to by page number and expressions of emotions can be included. A structural style focuses on how the content is organised by the speaker. The model by Labov and Waletzky (1982) provides a starting point for "beginning" researchers (Riessman, 2008). She continues, this method breaks the story into parts giving attention to the structure of the story which may otherwise be missed. This is explored further in Chapter 4 as I used Labov & Waletzky's model to check and refine the thematic style to narrative I adopted in analysis.

Therefore parents' stories were transcribed verbatim except where audio was lost in very brief sections, for example when the telephone rang. Throughout the interviews I had been able to observe parents' actions and emotional responses during their stories; I added these to my fieldnotes. I found myself in agreement with Reissman (2008) that translating dynamic talk into a written form would not be straightforward and that this would be a "deeply interpretive" process.

As a practitioner I understand that the findings of my study have potential significant value in clinical practice. I wanted the representation of the transcripts within my thesis to be accessible to both academic and professional staff. I decided that parents' stories would be presented without detailed transcription markings to ensure flow and ease reading. Transcripts that are

presented in this way are considered by Elliott (2005) as "clean" she goes on to suggest that clean transcripts are useful in research that:

> "focuses on the content of narratives told within interviews this approach to transcription may well be appropriate in that it will capture the chronology of events that are being recounted and also some of the evaluative elements, whether these are explicit statements or embedded in the precise words of the narrator" (p52).

My aim was to produce "clean" transcripts that would be represented in a manner that would allow interpretation.

By describing my study design I have explored how I planned to come into contact and engage with parents. At this point in my thesis I explore the trustworthiness in narrative inquiry.

3.10 Trustworthiness in narrative inquiry and qualitative methodology

For parents the 'truth' in their stories is embedded in their histories and perception of realities. Parents' sensitivity to their experiences is firmly rooted within their understanding of the events that they have endured over time. Truth is based on the unique perspective of the parent and their self-awareness; parents share their world with me by using stories to communicate their subjective understanding. Lorem (2008) suggests that what research participants tell the researcher about themselves and their situation is important in understanding their needs and experiences. Ricoeur (1985) suggests that in general we should recognise narratives as a characteristic property of self-understanding. Therefore as a researcher I believe that the stories parents tell me are true. While I recognise that memories could be jumbled I trust the parents in my study and will not attempt to "verify the truths of their stories" (Holloway & Freshwater, 2007 p106).

Parents tell their stories in a personal, subjective manner. All research has elements of subjectivity. Naturalist research such as this is dependent on the choices of the researcher in the focus, selection and interpretation of the data (Holloway & Freshwater, 2007). As a researcher I recognised that parents will select components of their stories in order to convey the meaning they intend to be taken away from the interview (Bailey & Tilley, 2002).

Interpretation had to remain 'true' to the parents' stories which is why at times modest segments of 'stories' are presented in the findings Chapters 7,8 and 9 .

3.11 Conclusion

Taking into account the need to demonstrate the integrity and rigour of my study, I have attempted to make explicit the thinking behind my study. I have explained my background, interest in the study phenomenon, my theoretical and epistemological beliefs and the methodological approach guiding my study.

By reflecting on and being aware of my own values and beliefs I considered I would be able to engage in narrative interviews openly with parents. Etherington (2004) explains that:

> "[R] researcher reflexivity is the capacity of the researcher to acknowledge how their own experiences and contexts (which might be fluid and changing) inform the process and outcomes of the inquiry. If we can be aware of how our own thoughts, feelings, culture, environment and social and personal history inform us as we dialogue with participants, transcribe their conversations with us and write our representations of the work, then perhaps we can come close to the rigour that is required of good qualitative research" (p31-32).

In the following Chapter I explore the organisation, analysis and interpretation of my data.

CHAPTER 4: DATA AND NARRATIVE ANALYSIS

4.1 Introduction

In this chapter I aim to make clear how I managed the parents' stories as data. This includes the approaches I used to explore the data and make sense of parents' stories. I explain the steps I took in my analysis in order to make clear my trail of thinking and analysis. I include, where appropriate, the impact the data had on my own thinking and issues of reflexivity. Therefore the following chapter is divided into three sections and, where necessary, these sections are split into subsections. The first section explains how I understood parents' stories as narrative data. The second section is divided into six subsections. These subsections explore the six steps which I used to initially manage and then analyse the data from phase 1 (and again later in phase 2). In the fourth section I explore the collection of data in phase 2 and the development of foundational, aggregate and keystones stories.

4.2 Parents' stories as narrative data

I believe that the parents' stories I collected embodied parents' experiences of having a child born with ambiguous genitalia and the child's subsequent reconstructive genital surgery. Therefore my aim was to remain as faithful as possible to the parents' thoughts and ideas as recounted within their stories without losing my own analytical stance towards them as narratives. Therefore similar to many narrative researchers (Connelly & Clandinin, 1990; Polkinghorne, 1988) I aspire to demonstrate actively and transparently the process by which I have understood parents' voices as explored through their stories.

The impact of listening to and engaging with the parents' stories as an individual, nurse or parent cannot be dismissed. The voyage of discovery I embarked on when I began to listen to the parents' stories whilst examining the transcripts was a different experience to the one I had when I first heard the stories. The impact these stories had on my own emotions and beliefs are discussed in findings Chapter 6 (section 6.3).

In this paragraph I make clear how the interviews became the data that were subsequently analysed and presented as findings in Chapters 7, 8 and 9. From each taped narrative interview I constructed a written record, a transcript, as previously discussed in more detail in Chapter 3 section 3.9(f). Translating dynamic talk into a linear written text is "never straight forward" (Riessman, 2008). She continues to explain that the transcription process is "deeply interpretive" and there are varying methods about how "talk" can be presented. Presentations of the data are dependent on the researcher's "theoretical perspective, methodological orientation and substantive interest" (Riessman, 2008 p29). Therefore when considering how to present my data I remained mindful of my own agenda as a researcher and also my theoretical thinking. I concur with Riessman (2008) that the act of storytelling "in dialogue *constitutes* the autobiographical self" (p29), which for me is how the parent wanted to be known in the interaction but also to the wider audience, for example, readers of articles, the audience at a presentation when I retell their stories. The transcripts were verbatim recordings and emotional exchanges were included in the transcripts. My fieldnotes helped me to recreate the contexts and environment in which the stories were told and the vignettes (see Chapter 5) are a representation of this.

In the following data management section I aim to make explicit the process by which the stories in their entirety were transformed into manageable data and highlight the approaches I adopted and the underlying concepts that support these.

4.3 Data management and analysis

This section is divided into six subsections in order to explore the six 'steps' I took in understanding, exploring and later interpreting data throughout my study. I handled phase 1 and phase 2 similarly and the following subsections explore how I developed my approach over the course of my study. The first subsection focuses on how I began to look at and order the data.

4.3(a) Step one: Beginning to look at the data

Initially, during phase 1 and again later at phase 2, the primary focus was to explore what parents told me through their stories. The quantity of data generated from all the transcripts amounted to 756 printed pages (interviews ranged from 36-64 pages in length). Although highly rich in detail these data initially felt unwieldy and I acknowledged that the stories would need to be ordered and managed for the data to be useful. My reading had identified a range of approaches to narrative analysis. These included thematic, content, structural, interactional, holistic and categorical, discourse and performative analysis (Holloway & Freshwater, 2007). Elliott (2005) argues that to "manage the multiplicity of technique" in narrative analysis a typology is needed. She continues that this typology has the benefit of explicating the methodology and epistemology that underpin analysis. While I wanted an approach that allowed parents to explain their experiences I anticipated I would discover something about their beliefs about 'self'. I therefore needed to consider the typology that fitted with my theoretical thinking and allowed me to order the data in a narrative structure.

4.3(b) Step two: Ordering and managing the data

The initial stage in exploring the data was to try and unpack 'what parents say' and the nature of 'how parents organise and retell' their stories. I read the transcripts several times before any narratives were 'unpacked'. The re-reading I believed was necessary to ensure that I became familiar with the transcripts, the recordings and my feelings from the interview. Fieldnotes were helpful in exploring the emotional complexities that were generated for me as a consequence of the parents' interviews.

I explored the data in several ways as I believed that each would complement the other and develop my understanding. I reflected that I needed to be able to understand the structure of narratives and where structure existed within the stories that parents had told me. Paley & Eva (2005) describe real life as "messy" and, as such, collected narratives are "less tidy" than fictional ones. I then needed to be able to explore the content of 'what' parents told me so that I had an awareness of their 'self' but also the contextual world they live

in.   I finally explored the thematic parts or themes of the narratives in order to establish where the patterns or similarities between parents existed. Although I have suggested three approaches to analysing the data I draw on the suggestion by Elliott (2005) that:

> "[I] it is unlikely that any researcher in the social sciences would examine the form or social function of a narrative without paying attention to its content" (p38).

Therefore in the following subsection I explore the content in a story.

4.3(c) Step three: The content in a story

Deconstructing a narrative into its core elements is discussed in the literature by many authors (eg; Riessman, 1993; Lieblich et al, 1998; Josselson & Lieblich, 1999; Holloway & Jefferson, 2000; Roberts, 2002; Fraser, 2004). There are different levels of agreement or disagreement depending upon the authors' background (Frid et al, 2000; Elliott, 2005; Holloway & Freshwater, 2007). Gergen (1999) explains that in telling a "proper story by Western standards" there are standards for "narrative construction". He suggests there are several significant features of a "well-formed narrative" (p68). He presents the four most prominent criteria as:

> "*A value end point:* An acceptable story must first establish a goal, an event to be explained, a state to be reached or avoided, or more informally a 'point'. This point is typically saturated with value".
> "*Events relevant to the end point:* Once an end point has been established it more or less dictates the kinds of events that can figure in the account. Specifically, an intelligent narrative is one in which events serve to make the goal more or less probable, accessible or vivid".
> "*Ordering of events:* Once a goal has been established and relevant events selected, the events are usually placed in an ordered arrangement. The most widely used convention of ordering is that of linear time".
> "*Causal linkages:* The ideal narrative provides a sense of explanation" (Gregen, 1999, p69).

Narratives consist of structured events and some authors such as Gaydos (2005) suggests narratives must have:

> "a beginning, middle and end. They have plots and sub-plots and are peopled by interesting and varied characters who move the plot along. Each narrative has themes that are both explicit and implicit" (p254).

All of these elements fit together and all are important. During phase 1 of my study I started to read and re-read my data and had started to mark on the transcripts the words 'because of'. At this point in phase 1 I explored the structure of narratives using the Labov & Waletzky (1972, 1982) framework to build my confidence in handling the data and to ensure that I did not miss important stories within my data. This aspect of my analysis is covered in more detail in the following subsection, handling the data in phase 1.

4.3(d) Step four: Handling the data in phase 1

All of the parents' stories from the first nine interviews were printed and then subsequently physically cut up into the stories that shared the same content. Each group of stories were assigned as 'story groups', there were eighty six story groups (Appendix 8). These were representative of the parents' life events and included story fragments about their child (Appendix 8). I began to explore these but found that I needed a more structured approach to the data. I decided in order to gain a better understanding I had to deconstruct the narratives. I therefore began to explore the data within a structured framework. I used Labov & Waletzky's (1972, 1982) framework as described by several authors including Hydén & Överlien (2005). Labov & Waletzky (1972) presented six formal properties that are summarised as follows;

Abstract – the summary or the substance of the narrative. [I reported abstracts at different stages in the story as described below in the reduced story column].
Orientation –the time, place, situation and characters, how they were presented to the audience and the meaning these had on the story teller.
Complicating action – the progression of events or the movement forwards. This step allowed time for the story teller to reflect and consider previous events against future outcomes perhaps a liminal period for the story teller.
Evaluation –the significance and meaning of events and actions for the story teller, their emotions and expressive analogies.
Resolution – what finally happened.
Coda – relocating the story in the present.

In the following example I demonstrate how I used the structured framework with my data (see Table 4.1). This framework not only shows how I used Labov & Waletzky's formal properties to help develop my critical thinking but also I used other devices to 'interrogate my data'. In this example I use the start of Chloe's interview where she tells a story about her complicated pregnancy:

"Yeah the first day that I arrived at (hospital name) was about, I must have been 28, 28 weeks I think. Raised blood pressure, protein in the water, I didn't know what preaclampsia was they didn't say. I didn't have a clue then. I still got the message that is was quite serious because people were coming in and asking me what was going to happen, and I was thinking, what was gonna happen? You'll have to stay in now until I have the baby they said, but obviously me blood pressure got worse and worse to the point where I wasn't even allowed to get up off me bed. In actual fact when I stayed on the bed I didn't do anything at all, it didn't come down, no, but that's neither here nor there. I was looked after very well, the midwives there were very, very good, and I was right by the nurses station, so they were obviously thinking I was going to go of like a bottle of pop, but that didn't happen I got to 30 weeks" (Chloe).

The following table is an example of the early data handling that underpinned development of my critical thinking. The table is divided into five sections headed; story segment, storied text, Labov &Waletzky framework, 'because of' and my reflective thoughts.

Table 4.1 Example of early data handling that underpinned development of my critical thinking

Story segment	Storied text	Labov & Waletzky framework	'Because of'	My reflective thoughts
Chloe 28 weeks pregnant who is aware she is early to have her baby, knows something is wrong	Yeah the first day that I arrived at (hospital name) was about, I must have been 28, 28 weeks I think.	Orientation – time, situation, place, participants	Participant predicament – ill health leading to something	Unexpected event matched against anticipation that 'faith' – cultural/social/ or trust in healthcare to make everything ok – the very very good midwife, do as told – outcome should be ok
Sense that something is seriously wrong	Raised blood pressure	Complicating action – sequence of events	Emotional response to physically cognitive thought about her predicament	Failure in the treatment for her >BP, bed rest did not 'fix' the problem
Response from those caring for her that she had a need	I still got the message that is was quite serious	Evaluation – significance and meaning of the action	Midwife cared for her at a vulnerable time – placing her where she could be seen	She got the message it was serious by the repeated asking about her health status
Making it further, lasting out another 2 weeks before the birth	I stayed on the bed I didn't do anything at all, it was it didn't come down	Resolution – what finally happened	Focus was maybe on gaining time & this was fundamental – better outcome if she could wait to have baby	Triumph at making the baby hang on for a little while, though the treatment or 'bed rest' didn't work for long enough. Worries were focused on a premature baby that's what she was expecting from her current situation.
Making it to delivery –she was able to do this - for a little while before the baby arrives	I got to 30 weeks – had a baby	Coda – return to the present – didn't get back to actual present but moved to the next story.	Inability to hold on to the baby, an event is certain to follow – baby early, will need extra help, causation – events are becoming linked,	? starting to build some anticipatory fear in the story and the achievement of being able to hang on is lost against the greater aspect of failing to hold on - loss of choice – beyond control

This example demonstrates how I began to broaden my thinking and explored the structure of Chloe's opening story. The 'story segment' related to my interpretation of segments of Chloe's story while the 'storied text' evidenced the 'actual story' its beginning, middle and end. I was able to fit this story into my table because Chloe had ordered and sequenced her story. Furthermore, the structure provided by Labov & Waletzky allowed me to think more broadly about the situation Chloe found herself and the meaning she attributed to her situation. The 'because of' column was added as I began to understand the stories and explore wider levels of thinking, as I recognised the effects of time within the story. The 'because of' column also became the place where I began to see the role of characters in Chloe's story and the influence they had on her. In order to develop my thinking further I began to write my reflective thoughts and try to explore the wider social influences in Chloe's story.

Although this approach started to open my thinking I began to struggle as more frequently parents' stories were told out of 'sequence'. This loss of sequence impacted on my ability to be able to fit the stories within my interpretation of the structure of Labov & Waletzky's framework. Mishler (1997) stresses that strict application of Labov & Waletzky's model is dependent on ordered or sequenced events and is therefore limited when participants do not provide strictly chronological accounts. However, I discovered that although I could find some sequence in data presented in this framework, I often could not then achieve this across the whole story. I recognised that by using this framework in a table I lost parents' stories of their 'self' which had been influenced by wider social contexts. One example is the nature and impact a premature birth had for Chloe, what had influenced her beliefs and the impact these beliefs held for her relationship with her new baby.

In addition the parents in my study were eager to share 'all' their stories and at times I felt the resulting 'whole story' was frenzied. Riessman (1993) supports this suggestion in that she acknowledges that a great deal of material in interviews has a story-like form but does not strictly consist of a series of event clauses. Also Labov & Waletzky's framework considers the coda as a return to the present and I struggled with applying this within the stories as the nature of the account was at times as Hayden & Overlein (2004) describe as

"historical". I decided that I would build on the 'my reflective thoughts' column as a basis to further explore how parents' emotions fitted into their stories and the influence their beliefs held in their lives.

4.3(e) Step five: Focusing on the 'thoughts section' from table 4.1

I began to focus on how parents expressed their emotions and negotiated the meaning their experiences held for them, the 'thoughts' column from Table4.1. Parents described their emotions as a result of their experiences throughout their stories and I began to explore these emotions as data and developed a 'story map' in order to visualise how parents' 'self' fitted within their narrative, Diagram 4.1. The story map is a visual representation and again I have used Chloe's story about her complicated pregnancy. However, I also included the next layer of her story which focused on the events that happened in the twenty four hours following the birth of her child. This approach enabled me to broaden my thinking and try to unravel more than the nature of the story but also the contextual detail and the beliefs that Chloe held which informed her thinking. The following Diagram 4.1 explores the time before the birth of her child; her hopes, expectations, aspirations and plans. I explore the dramatic nature of the birth event and the emotional responses that Chloe experienced; influenced by those around her and by her own beliefs about pregnancy and childbirth. I also explored the social worries that Chloe experienced, the significance placed on her own health and that of her child. In my story map I included other characters significant in Chloe's life at the time of her child's birth. Her husband (David) delivers the news about their child's ambiguous genitalia to Chloe and has to manage her response and also his own emotions. Time and information become very important for Chloe as she anticipates worrying about her child's future which, at this place in her story, is clouded by uncertainty.

Delivery date story – conflict, trying to please, wanting to wait, no control over events

Her health needs & initial admission to hospital

Matching story – placing weight on others 'normal child' stories – others worry about unimportant things –

Society issues

Getting better herself – small story

Pity story – future children?

Time – of events

Evaluation event - crisis

How do you know, are you sure its true story

Birth event – Major event delivery of a baby

Pre-realisation (Edwards AG) – fear own health, confusion where is her baby, is he alive, something's coming, fear

The news, major event – shock and confusion – loss of control and future child information delivered by David

Abandonment, separated, catastrophic event, miss match of expectations

Dramatic birth event, scary, aspect of watch events – analogy with TV shows, trying to ground her experience

Preparing for bad news, fearful, build in suspense

These could all be captured as a thriller, starting off with a steady climb, building tension, each advance slightly different, to give a more powerful story

Realisation, fear, death, shock sister seeing her, reinvestment in HCP to save her, submission to care – SURVIVOR STORY

Heroine story – she survives a dramatic turn of events

Dawning, something's not right, intuition?, issues of power and authority, truth, loss of control for parents

Time – waiting – liminal period?

All these need to fit together somehow

Diagram 4.1 Story map, Chloe's complicated pregnancy and her child's birth.

This approach to data analysis allowed me to begin to see how parents' emotions and beliefs about 'self' fitted with events and if these emotions shifted or changed over time. Presenting the data in this manner allowed me to visualise all the 'pressures' that in the example in Diagram 4.1 that Chloe was currently experiencing in her life. I started to be able to recognise how her sense of identity was being challenged and how she was managing to readdress this. I used this approach with all the data in phase 1 to explore the aspects of the stories parents told about their children's genital surgeries. At this stage of analysis I was able to identify six narratives that demonstrated parents' understanding of their child's genital ambiguity, their motives, beliefs and attitudes towards AG. The following subsection explores these six narratives from phase 1. These six narratives became significant starting points for data collection and later analysis at phase 2.

4.3(f) Step six: Six narratives from phase 1

Using both the structured and content approach to analysis (as described in the previous subsections 4.3(d) and 4.3(e)), I was able to draw together six narratives from phase 1 data. These narratives form the basis of a publication in a peer review journal (Sanders et al, 2007) which explores parents' experiences of having a child with AG and reconstructive genital surgeries. The six narratives explored at the end of phase one were:

- Temporality;
- Power and vulnerability;
- Parenting and family structure;
- Gender, medical and surgical intervention;
- Decision-making or lack of decision making; and
- Ethics and moral issues.

These narratives served as an important adjunct to my final analysis as once I had collected the final interviews they began to shape my thinking in the thematic style I described earlier, and informed my final analysis of all the narratives from both phase 1 and 2 of my study.

Temporality: Not unexpectedly the content of parents' stories identified trajectories that existed in the present time and into the child's future. The length of time parents had to wait for the child's genital surgery varied (ranging from eighteen months to three or four years). During this time parents had to 'adapt' to their child's genital ambiguity. Parents considered the medical approach took too long. They also felt that the medical approach was fixed to goals such as the child's physical size, which parents could not control. There was a dissonance for parents between the actual time taken to normalise their child and the time they 'wished' the surgery could be completed by. Once the waiting for surgery to fix their child had elapsed some parents' stories conveyed an urgency to 'complete' their child. For others the liminal waiting appeared to offer direction, predictability, reassurance and stability at a time of confusion and uncertainty.

Power and vulnerability: Parents described experiences that reflected both bewilderment and concern at having to contend with their own vulnerability while simultaneously being responsible for their children who they perceived as particularly vulnerable. Vulnerability was not portrayed as a positive concept within the parents' stories. Medical staff were a primary source of information for parents, yet parents told of poor communication, inadequate support and confusion. The stories, enacted in the course of the dialogues between medical staff and parents, reflect the parents' bewilderment, "disbelief" and initial "horror". Doctors took an early leading role in these stories and drove the subsequent discussions regarding the child's ambiguity.

Parenting and family structure: Across all parents' stories elements of parenting as an embodied experience were articulated. Their expectations about becoming parents were neither met biologically nor socially. Parents expressed negative emotions that reflected their confusion and, at times, perceptions of failure. Knowledge of the child's diagnosis and appearance of AG was often contained within the context of the family; a few parents described themselves as "good teams" focusing on achieving the "best" for their child. Parents' stories reflected that the birth of a child with ambiguous genitalia involved changes for the parents, especially in their social status. Parents believed they became

separated from the rest of their social group due to the ambiguity of their child's genitalia.

Gender, medical and surgical intervention: The consequence of the child's ambiguous genitalia appeared to be perceived across parental stories in two ways. Parents considered their child either in visual terms (having a penis or not) or in biological terms (later reproductive health). From the stories there appeared to be sparse and inconsistent 'surgical' discussion between parents and doctors and little explicit exploration of the parents' knowledge about any proposed genital surgery. Parents' stories demonstrated their passive roles and surgery continued to be considered as a solution to their child's genital ambiguity. Surgery was considered by the majority of parents as an intervention that was able to "fix" the problem. Gender influences, surgical correction and acceptable visual outcomes appeared indivisible; the parents appeared to shift the balance of each concept within the context of their decision-making. Until the child was surgically corrected parents were reminded daily of the incongruity between the child's genitalia and the socialisation and presentation of their child in their own community as either 'completely' male or female. Many parents would go to extremes to either feminise or masculinise their child, demonstrating the need to define their child's gender.

Decision-making or lack of decision-making: Parents' decisions about consenting their child to have genital surgeries emerged early within their stories and remained consistent across time. The opportunity and capacity for parents to refuse surgery for their child was only vaguely talked about. Many parents were ignorant of the controversy surrounding childhood genital surgery. Parents reorganised their lives (such as giving up work so they could attend hospital appointments) to help them solve what they perceived as the problem of their child's genital ambiguity. Generally, parents situated medical staff in a place of expert authority, accepting medical judgment and advice as they felt they knew little or nothing about AG. Parents' lack of knowledge and the disjointed medical advice they received influenced their decision-making.

Ethics and moral issues: Most healthcare professionals suggest that information about the AG should be disclosed to children by their parents. However, the balance of power and the decision regarding sharing this information remained with parents. When parents made surgical decisions their aim was to do what they believed to be 'best' for their child.

These six narratives helped me to focus phase 2 of my study. The emerging narratives were beginning to suggest that parents' surgical decision making was confounded by many factors. Parents' experiences of having a child born with AG were complex and multifaceted and were also influenced by their own beliefs. On returning to the field to collect further interviews I chose to investigate how parents constructed meaning from their experiences. I was interested in how parents' beliefs brought about cohesiveness between their experience of AG and their emotional bonds towards their child. I wanted to explore the influences AG had on family life and if parents' interpretations of AG impacted on their wider social networks. I also decided to try and further investigate the fundamental belief that lay behind parents' understanding of AG and their goal in relation to 'coping' with AG. Finally, I wanted to explore in more detail parents' relationships with healthcare professionals, including their level of engagement and how this was maximised or marginalised by either the parent or the professional.

4.4 Collecting and analysing data at phase 2

At stage two of data collection I invested in the data management software (Altlas.ti) to aid my organisation as the number of sheets of paper and mass of data were becoming unwieldy. During phase 2 I collected a further nine interviews resulting in eighteen interviews in total. The second set of nine interviews were transcribed as previously described and all eighteen interviews were checked against the recording for accuracy, at least three times and re-formatted as 'text bank data' for Atlas.ti. Holloway & Freshwater (2007) comment on how the research question and data become part of the researcher's life. They also suggest there is an "incubation period" that is critical in the process of analysis and interpretation which facilitates creativity.

Subsequent listening and reading was augmented with 'thinking time' which allowed the data to be explored intuitively and within the context of structure, content, social constructions, cooperative stories, plot and genre. Initially the very broad approach, as described earlier, was used to unpack the content and structure of these stories alongside plots and genres as described by Riessman (2005), Frank (2005) and Paley & Eva (2005). I used memos (similar to the thoughts column in Table 4.1) within Atlas.ti to track my thinking progress and I also began to group the data using 'story maps'.

During analysis in phase 2 it became evident that parts of the narrative were thematically linked as described by Roberts (2002). He suggests narratives are not drawn randomly from the text they are selected thematically and he suggests that by:

"close attention to the content of interview stories the specific narrative elements are identified" (p121).

I recognised that stories within my own data recurred throughout parents' narratives and across them. As I continued to explore these recurring stories across all the stories I also kept the narrative intact as suggested by (Riessman, 2008). I started to gather these recurring stories into groups of ideas which I called foundational stories. There are twenty foundational stories which represented the small stories and story fragments that described some of the everyday events of bringing up a child with AG. Foundational stories are the building block stories that parents told. By exploring parents' stories further I began to recognise plots as the links that were created between these foundational stories. By pulling together these foundational stories I constructed my next level of stories; aggregate stories. There are eight aggregate stories which are those stories which share similar events, story lines and confrontations. These aggregate stories then became synthesised into three keystone stories which explored the meaning AG had in parents' lives for their child, themselves and their interactions with healthcare professionals. At this point I again explored the literature on 'genre' and although this is an "analytic resource borrowed from literary studies" (Elliott, 2005) the keystone stories had the same attributes as genres. These keystone stories are the

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overarching genres within my study created by the synthesis of parents' stories. As my narrative analysis skills developed further I began to manage the further synthesis, conceptualisation and interpretation of the keystone stories. Analysis of the keystone stories revealed the core elements which support the central concept of my thesis, which is the parents' search for harmony.

4.5 Conclusion

Taking into account the need to demonstrate the integrity and rigour of my study, I have attempted to make explicit the thinking behind my analysis of parents' stories. I have explained how I believe parents' stories are data, my approaches to analysis and how I have chosen to finally present parents' stories. Foundational stories are the building blocks of my analysis. These comprise of recurring stories or fragments of stories which represent the everyday events of having a child with AG. The aggregate stories are built on parents' beliefs and also their interactions with other people. The keystone stories are the overarching genres within my study created by the synthesis of parents' stories. Finally, analysis and interpretation of the keystone stories allowed me to discover the three core elements that support the central concept of my thesis, which is the parents' search for harmony.

Prior to reporting the findings I will provide vignettes as an introduction to the parents in Chapter 5. These parents' vignettes are included to provide context and insight into the lives of parents. These vignettes have been built from my fieldnotes and are written in a 'story style'. I chose sections from my field notes that told the beginning, middle and end of my story about meeting the parents. The aim is to present the parents as people who are living their lives as well as parenting a child born with ambiguous genitalia who has experienced reconstructive genital surgeries.

CHAPTER 5: PARENTS' VIGNETTES.

5.1 Introduction

I wanted to be able to find a way in which I could share an understanding of the context of parents' lives. I chose vignettes as a style that would allow me to do this. Vignettes have been described as enabling or projecting techniques which can be used within research and are therefore not a new approach to presenting or engaging research participants (Arthur & Nazroo, 2005; Brondani et al, 2008).

My fieldnotes were helpful in recreating these vignettes. Any identifying factual data has been removed or changed to maintain the parents' anonymity. I have not included the number of surgeries that children had or their ages in order to try and maintain anonymity. All the parents were given a pseudonym and stories from all parents are included in both the findings and discussion chapters of my thesis. Vignettes of parents presented in this chapter were written to enhance understanding and insight into the characters of the parents and share important contextual and sensitive data about parents' lives. Within my study the following vignettes of parents are short descriptions, written narratively, in keeping with the essence of my thesis.

5.2 Vignettes of parents

The following section is divided into 11 subsections. Each subsection is a vignette of a mother and/or father or both a mother and father when they were interviewed together or lived together as a family. The interview number is denoted in brackets after the parents' name (pseudonym). For those parents who were interviewed twice there was on average 12 months between interviews. Some parents had experienced life changes during the intervening period and these are included in the vignettes.

5.2(a) Anne (interview number 1.1 & 1.2)

Anne opened her front door to me saying "welcome to a busy house", as she had only just got back from her part time job. On my first visit she had also been expecting the repairman and had shuffled her working week so that she could be around for the interview and later the washer to be "fixed yet again".

Her welcoming smile and eagerness to 'put the kettle on' set the scene for my two interviews with her. Prior to having to manage as a single mother of three children she had been married, but had been on her own for several years after an amicable split. Anne described her life as being "busy, busy" and she was "a typical mum" who had not been prepared for "the shock" after Barry her youngest child was born. Within ten minutes of Barry's delivery he had been noted to have ambiguous genitalia and imperforate anus. She had never expected "anything like it" as all three of her pregnancies had been managed at the local hospital and all her antenatal scans had been "perfect". Barry's birth and the subsequent surgery and uncertainty had been a stressful point in her life yet Anne was someone who seemed to always try and look on the positive side of things. Anne was delighted to have been offered the chance to tell her story and in some way "help or give support to other parents". She told me how once she had received the invitation to participate in the study she had completed and returned the slip "within the week or so". After we had had a cup of tea and biscuits she was eager to tell her story. Once during her story telling she became a little sad but she felt that she "continued to have a positive outlook and was grateful for the experience" of the interview. She remained very hopeful for her son's future and was very proud of the hurdles he had overcome and the unity between Barry's siblings and maternal family.

5.2 (b) Chloe and David (interview number 2.1 & interview 8.1)

Chloe and David were married but chose to be interviewed separately. David was very busy with "new work commitments" and his interview was a few weeks following Chloe's. Despite their individual interviews highlighting some different concerns, there was a great deal of resonance between the stories they told.

Chloe had been busy the morning of her interview walking the neighbours' dogs. She 'made me feel comfortable' in the front room and then went to make tea. She told me a story of how she met David, their travels and after how when they had "finished the house" they had "looked forward" to the birth of their first child. Chloe was disappointed she had a "troubled pregnancy" spending many weeks in hospital. She recalled fondly the rapport and support she had from the ante-natal staff during her "long stints in". She felt like a "regular" and as time went by she became more confident in the "doctors and

midwives". However, Chloe had been "terrified" when she suddenly went into labour and feared for both her unborn child's life and her own. Chloe remembers how David watched in dismay at her deterioration in health both immediately before and after their child's birth. Edward was born premature and quickly taken to the neo-natal unit where he remained for several weeks. Initially both David and Chloe thought everything was "ok" with their son until several hours later when they remember being told about Edward's ambiguous genital. Edward was "a little fighter" and over a few months he improved and left the hospital. During Chloe's interview she reflected on how she had become "obsessed" about Edward's health and she remembered the stress this placed on her and David's relationship. Chloe focused on the many positive achievements Edward had made and shared his photographs and sporting trophies. Overtime Chloe believed that her experience had given her a "great strength" to face "life's challenges" and she changed her working life to something she had always wanted to do, because she "felt stronger" because of her experiences.

David chose to be interviewed away from the family home as Edward was always around in the evenings and weekends were "busy family times". David told similar stories to Chloe adding his own depth of feeling when he recalled Chloe's pregnancy and Edward's birth. David told stories about work and how he had "escaped there" while Edward was small and that once his son had become interested in "different sports" their relationship had grown and he enjoyed Edward's "company and sense of humour".

David and Chloe had decided to have separate interviews as David hoped it would help Chloe talk more freely about her worries for her son, as he worried that "no one spoke about it anymore", which he thought caused more stress. Both Chloe and David agreed to be interviewed a second time but Edward needed "unexpected surgery". So in line with the exclusion criteria they were no longer eligible to continue to participate in contributing new data.

5.2(c) Faye (interview number 3.1 & 3.2)

It had been raining really heavily when I reached Faye's house. I had called ahead to tell her I would be a little late as the traffic was busy due to the rain. Faye had laughed and "hoped I brought my umbrella" and told me to "drive safe

so as not to miss a good story". When I arrived Faye was waiting by the back door so I did not have to stand in the rain. She joked about my journey while making the tea, she was very funny telling the "latest joke" she had heard at work. Faye was easy to listen to, she laughed as she told me about her early partner and how they had very quickly drifted apart. She described herself as being "a bad boy magnet" never having much luck with relationships. She spoke fondly of her eldest son, how he had his mother's "wicked sense of humour" and was always a good source of support for her. Faye described Gabby, her daughter as a lively and happy girl who also "thank god" had a "good sense of humour". Faye remembered her antenatal care with Gabby as "the same as the first time, with no problems". Faye recalled the story of Gabby's birth with a mixture of emotions after which she became distrustful of hospitals. She spoke about her childhood, her early family life and experiences. She smiled at the irony that she had a sibling born with mixed sex and this had prepared her for Gabby's label of "not a boy or a girl" better than "all those nurses and doctors". Her knowledge of "mixed sex babies" smoothed the progress of "going home" although going out into the community was "very hard". At times Faye remembered the "battles and fights" she had protecting Gabby from "nosey people" and how "great" her own parents had been at helping her "because they'd had the same experience". Faye spoke about how her working life had "suffered" because of all the "hospital appointments and surgery" Gabby had had. She spoke about how her experiences helped her stay "strong" and determined to do what she believed was right for Gabby. Faye was proud of how Gabby was starting to "grow up" although she was, at times, still worried about Gabby's future.

By the end of the interview it had stopped raining and Faye walked back with me to my car; a chance for a "secret smoke". She stopped to show me the new addition to the family a big grey rabbit she said laughing that Gabby "had no originality" and had called him "Thumper".

5.2 (d) Harry and Imogene (interview 4.1 & 4.2)

The first interview with Harry and Imogene was on a cold and wet autumn day in contrast to the second which fell on a hot summer's day. The warmest part of the house in winter was the kitchen so we all sat at the table with the parrot

singing in the corner until Imogene covered his cage with a blanket. Imogene offered me tea but also pointed out there's "no door on the bathroom" so I settled on a small cup. Harry's health was poor and the renovations were to make the bathroom more accessible for him since he had lost his sight. For Harry losing his sight had been especially hard, he could no longer "see his beautiful daughter" or "tickle her when she walked past him". Imogene chatted about early motherhood with Joanne and the family holidays they all enjoyed. She was "always busy" looking "after everyone now" but she loved this so much as family life was so important to her. Both Harry and Imogene laughed about Joanne still getting in their bed in the morning and being a "mischief" with her medicines. They shared a few stories about Joanne's ill health in early childhood and how they had been "taunted and questioned" by people curious about Joanne's sex. Imogene asked several health questions that related to Joanne and I answered these honestly and we also spoke about Joanne attending a clinic where she could meet other children with the same condition.

5.2(e) Kerran (interview 5.1 & 5.2)

For the first interview Kerran felt home was a place "far too busy and noisy to chat" so we met at a venue that she chose. When I arrived for the first interview I called Kerran and she came to meet me and we went to a quiet space where we would not be disturbed. She spoke about her family and her arranged marriage and how "hard this had been" especially as her husband had not spoken English, which was her first language. Sadly her husband had died unexpectedly a few years previously. Kerran told stories about Leah's late diagnosis and her own anxiety while she recalled that her husband had been more pragmatic about the diagnosis and she felt this was because of his religious beliefs. Kerran clearly expressed her frustration with doctors; she raised her voice slightly and clenched her hands as she tried to explain how she believed "they didn't listen". She spoke movingly about her son and how he was now "the head of her family" and her worries about Leah's long term future and health. As the first interview was drawing to a close she asked where she could get extra help for Leah and we agreed that I would speak to her consultant about her requests. She then spent twenty minutes telling me a story about two of her friends she believed had experienced "genital surgery" in

their childhood. She had only worked this out after having Kerran when some of the stories they had told her about their childhood began to made sense to her. The second interview Kerran wanted at home as she had arranged a day off and she felt "ok" as she had "met me before". During the time in-between interviews Leah had received the help Kerran had requested, which both of them had thought very helpful.

5.2(f) Maria (interview 6.1 & 6.2)

Maria was busy on her computer when I arrived and was "glad to stop". She chose to sit in a quiet part of the "mad house" and she laughed about the guinea pigs they had bought recently, two pregnant females that had then had seven "babies" between them. She joked about the extension to the hutch and was glad her husband was a builder. Maria had been happily married for many years and had three children, all of whom had had hospital experiences. Her own health had been a "bother for the last few years" but she was enjoying being back at work. Maria had always considered herself an "insightful mum". She had wanted to know all she could about ambiguous genitalia after Neil's birth, this included the most up to date treatments or advice and the "best doctors". She recalled vividly her shock at finding no support group to help her understand Neil's condition. Maria spoke about Neil's experiences in school and the friends she had made from their hospital visits where she had met other parents with a child born with the same condition. At both of my interviews with her Maria had remained positive about Neil's future but uncertain about how to share information with him in the future. She asked several very important questions about his long term outcome and young people's thoughts that I could not answer. She wanted what she called "proper evidence" about whether boys like Neil would grow up psychologically well adjusted and be able to have a fulfilling sex life. Although some limited evidence exists, for this long-term follow-up studies are rare and there are none in the UK. Studies that have discussed sexual outcomes with young people are also small in number and focus on different diagnostic populations (bladder exstrophy). This lack of research reflects some healthcare professionals' worries that possibly intrusive discussions with young people would affect them psychologically. Maria considered she was "very up to date" with both the current information available

from medical teams and "on-line" and she actively encouraged me to continue with future research to answer the questions I had been unable to help her with.

5.2(g) Oliver (interview 7.1)

Oliver had arranged to work from home the day of the interview, while Penny went to the shops and later went to collect Rose from school. The drive to Oliver's had been on the major roads and Oliver laughed about the number of "visitors" that had recently had speeding tickets, reminding me not to speed. Once I had met the family dog and cat, had a drink of tea, looked at the recent family holiday photographs we began the interview. Oliver told stories about Rose's birth and all the operations she had had, his worries for her in school and how shy and quiet Rose could be. He talked about how he worries "much more than he lets on" and how he manages his emotions. He spoke about his own childhood, enjoying being one of many children and how he had wanted a "bigger family" but that Penny was too scared after having Rose. Oliver told a story that was unique across the interviews, he spoke about how he had shifted his career to accommodate all of Rose's hospital visits and how "a supportive boss" was critical in keeping a job. He also spoke about the constraints that Rose's genital surgery had on "family life" and how he always had to be "the strong one". After all of Rose's hospital admissions he believed both he and Penny "knew Rose best" and he spoke with authority about how he "dealt with" doctors who just "didn't listen". Oliver was very keen for fathers' voices to be heard as he believed they have a joint role in looking after their children. Oliver was happy to give a further interview. However, Rose had major urological surgery around the time of second interviews and in line with the exclusion criteria her family was no longer eligible to continue to participate in contributing new data.

5.2(h) Sian (interview 9.1 & 9.2)

I managed to get a bit lost and had an old A to Z which did not have the new houses in her area on, so I struggled to find Sian's house, which she found rather funny. Once Sian had checked my A to Z and laughed a bit more she put the kettle on and we had tea. On my first visit a warm summer's day Tara was asleep on the sofa after a busy day playing at school. On my second visit I was

surprised to see Sian with a new baby, only a week old, she had wanted to surprise me so told me nothing about her new arrival when we had spoken on the phone. Sian was a single parent when she had Tara but had been with her current partner for several years. Sian was a bit shy at first although she was "really keen to help out" by being part of my research project. Once she started talking about Tara's birth and how supportive her parents had been she chatted very happily. She made me laugh when she described one nurse as being as "much use as a chocolate fireguard", I'd only heard the saying in relation to a chocolate teapot. She chatted about how on the whole everyone had been kind and helpful to her after she had Tara. She talked about the aspects of her life that had been "put on hold" while she had to get Tara through her surgeries. Tara was born with a cloaca and she had had many operations to her genito-urinary tract and bowel. Sian chatted confidently about how she worked out over time the food Tara couldn't eat and how comical it was that Tara "loved sprouts but they didn't agree with her". Sian was a very proud mum, she had encouraged Tara to be independent in some aspects of her care and although only young Tara was growing into a confident little girl. Sian was also very pleased that if Tara needed to come to hospital she was not afraid of theatre or meeting people and she would "take it with a pinch of salt". Sian had wanted to meet other mums with similar girls and we were able to put her in touch with someone, which she found really helpful.

5.2(i) Andrea & Brian (interview 10.1 - couple)

I had arranged to see Andrea and Brian after work one evening as they both had busy jobs. On arriving to a very busy house I was quickly roped into a story before the children, two girls and Bobby, went to bed. Once Andrea had settled them for the night, the kettle was on and we went to the kitchen. Andrea and Brian talked about Bobby's birth and the events that unfolded around them. Their local hospital had tried very hard to support them and they were very grateful to them although they wished they had had someone with "a bit more knowledge" to talk to early on. They both remembered the strain Bobby's AG had placed on them at first and how differently they handled this. Andrea and Brian worked very well as a team and although they had "different idea's" about genital surgery they were able to talk this through and had reached an

agreement. Their coping styles were very different but each accommodated the others and this was evident in how they respected each others stories. They had lots of experiences of hospitals and surgery and spoke openly about how their experiences could "have been better" and the importance of being "listened to". Andrea told a story about expecting a bounty bag after Bobby was born, but how disappointed she felt because she could not receive one since no one could decide on a boy or girl bag to give her. Andrea and Brian chatted happily about how Bobby behaved in a rough and tumble "boy" way and how very different from his sisters he was. They felt they had recently reached a point where they were more confident in making choice's about Bobby's surgery which had resulted in them declining further operations.

5.2(j) Christine (interview 11.1)

Christine was pleased to greet me when I arrived, as was the dog, who took great delight in trying to eat my shoes until he was banished to the garden. Daniel had recently had a birthday and the living room was full of his birthday cards and new toys. It was a warm day so we skipped tea and had a drink of juice and we settled down to the interview. Before recording Christine had been very chatty and confident but she became much quieter when I turned the digital recorder on. She started her story with Daniel's last operation which had been very traumatic on two counts, he was very sick afterwards and results from the samples taken had suggested he had some female tissue "inside him". Christine talked about how she felt back on a "roller coaster" and how she could not believe something else was "wrong". Christine had a small family but a wide circle of friends but she did not talk with anyone about Daniel's AG. Sometimes this made her lonely but on the whole she felt she coped and now Daniel was at school she had started work again. She was very happy that Daniel was growing up into a chatty playful lad who had lots of friends. She worried about his future and his sexuality and how she would begin to talk to him in the future about his AG and previous surgery. Christine hoped he would not "hate" her for the decisions she had made for him. A sudden bang indicated that Daniel was back from school; one of the other mums had brought him back. He ran noisily into the living room, said a loud "hello" and shot through to the kitchen to find food. Christine laughed about how he was always hungry and

how she had to get "longer trousers all the time". Daniels' arrival brought our interview to an end and once the digital recorder was turned off Christine relaxed and chatted happily to Daniel about his day at school.

5.2(k) Frank & Gina (interview 12.1 - couple)

The day I went to interview Frank and Gina there were gale force winds and a tree had blocked their road, so I had to park and walk. I arrived wind swept and was ushered through to the lounge to a waiting pot of coffee and biscuits. They had been married for many years and the family home was a busy and welcoming place. Just before we started the interview Gina explained that Frank was quite shy and would chat but "not as much" as her. They started their story from Helen's birth and her early life saving surgery; she was born with cloaca and had already had more than 20 operations. For Gina anything that meant Helen had an anaesthetic was considered as an operation, she also included "the big ones" which were operations that had taken several hours. They spoke about how they were different as a couple, Gina more "focused and rigid, living from day-to-day" while Frank "much more flexible". Gina also chatted about how she liked everything to be orderly and her expectations of having a baby which when they did not go to plan "it's a heck of a surprise". Both of them chatted about friends and family and how helpful some people were whereas others were just "nosey". They talked about Helen's future and how special their daughter was to them and how they still worried about the surgery that was still "in Helen's future". They were confident in most of the healthcare professionals that cared for Helen and had found support from another mother who had a child with cloaca very helpful. They had developed strong links with a certain member of a healthcare professional team and trusted this person, who was very important to them. Frank and Gina remained very positive about Helen's future and were striving to continue to "do their best" as parents.

5.3 Conclusion

All of the parents appeared to be happy to share their stories and believed that the experiences they had had could possibly help other people.

They all made me feel welcome and said they were happy to be approached again in future studies. By including segments from all of the parents' stories in the findings chapters I aim to make transparent the commonalities between stories and ensure that all of parents have a voice, not solely those who had the most apparently dramatic stories to share. Also by sharing segments from all of the parents I want to show the level of engagement I had with all of the parents and demonstrate that I did not "over-identify" (Holloway & Freshwater, 2007) with just a few parents or neglect the stories from others. In Chapter 6, I explore the more general results from data collection and findings from the interview process with parents. Chapter 7, 8 and 9 explore in detail the foundational stories, aggregate stories and keystone stories from which I determine the three core elements that support the central concept of my thesis, which is the parents' search for harmony.

CHAPTER 6: RESULTS FROM DATA COLLECTION AND FINDINGS FROM THE INTERVIEW PROCESS.

6.1 Introduction

In this chapter I present the results from data collection and also the findings from the interviews. This chapter is divided into three sections; the first explores the results from data collection, including the venue of interviews and duration of interviews. The second section explores the findings from the interview phases as described earlier by Corbin and Morse (2003) in section 3.9(e). Finally the last section explores reflexivity and the impact the interviews and data had on my own emotions and beliefs.

6.2 Results from data collection

In total I collected 18 interviews from 15 parents. Ten of the total number of participants were mothers from which seven of the mothers were interviewed on their own. Four of these seven mothers were single parents, one widowed and two were married. Five of the 15 participants were fathers and two were interviewed on their own, both were married. Three couples (mother and father) were interviewed together. Anne, Faye, Kerran, Maria and Sian gave two interviews and as a couple Harry and Imogene gave two interviews. Overall 16 interviews were conducted in the family home, one in a parents' work place and one in a discrete area within a hotel lobby. All of the parents chose the venue and the time of their interviews. Although all the parents from phase 1 agreed to participate in phase two three parents, from two families, were not included in the second phase of interviews. One child had major surgery and one became unwell, which shifted them into the exclusion group. These three parents agreed that their data could continue to be included in the study.

The length of time of the interviews varied but the range was one hour to two and a half hours, with more than half of the interviews lasting two hours. No parents changed their mind about consent or wished to withdraw from the study. The following section explores the findings from the interview process.

6.3 Findings from the interview process

This section is divided into two subsections the first explores the findings from the four phases of my interview as based upon the outline by Corbin & Morse (2003). The second subsection explores the interviews and performance.

6.3(a) Findings from the interview phases

The interview process worked well and during the pre-interview stage there was 'small talk' as evidenced in the vignettes in Chapter 5. The pre-interview time proved to be very important as it was the phase in which both I and the parents assessed each other and developed a level of comfort and trust. I found that I agreed with Corbin & Morse (2003) and this was the phase in which "the groundwork for reciprocity was established" (p340) between myself and parents.

During the tentative phase the interview progressed and as the parents became more comfortable different stories emerged. Corbin & Morse (2003) described this as "the layers" of participants' lives being peeled back and this process allows participants to move towards the "next phase of narration" (p342).

During the immersion phase parents' narrative interviews, as anticipated, mirrored unstructured interviews in that they did not follow an orderly linear pattern. They varied in length with some parents being more introspective and exploratory. For example Faye, Chloe, Andrea and Brian chose early in their interviews to reveal more of themselves by expressing inner thoughts and feelings. Christine tended to limit her story to facts, however with all the interviews both the parents and I gradually became immersed in the unfolding drama. This phase of the interview had the capacity to be highly emotive and all the parents were offered time to stop the interview if necessary. The apparent strength and confidence within the interview relationship gave several of the parents' security to tell what Nunkoosing (2005) describes as stories that would otherwise "remain untold".

Once parents had shared their stories they began to move toward the emergence phase which was a less intense and emotional phase. Within my narrative interviews some of the stories were still relevant to the research topic but they were less emotive. Corbin writes about her participants showing her

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their garden and I found that my interviews mirrored these with many of the parents sharing photographs, employment stories, views from their homes while making fresh tea. During this time Corbin & Morse (2003) suggest that the interviewer (if skilled) can provide information, advice or validation without impacting on the narrative flow. Harry and Imogene used this time to ask health questions about Joanne, while Chloe took this opportunity to reveal an extremely relevant story that she had delayed from sharing earlier. Reissman (2008) explains that story telling in interviews can happen at "the most unexpected times" indicating the "ubiquity of the narrative impulse". In bringing this phase to an end I offered parents the opportunity to ask any questions. This also provided them with time to reflect on their interview and consider if they were still happy for me to use their stories as data. I asked them to sign a second consent form allowing use of anonymous segments of their stories to be used in my thesis and papers.

6.3(b) The interview(s) and performance

During the interviews parents would move back and forth in events and at times appear to contradict segments of their stories. Corbin & Morse (2003) suggest this is common and acceptable as the parents are trying to make sense out of "significant events in their lives" and find "clarity". Corbin & Morse (2003) describe that at times during interviews participants may become emotional. Within my interviews Chloe, Anne and Christine were all very briefly a little tearful. At this time I stopped the interview until they regained their composure and they were happy to continue. During intense times and distressing moments Corbin & Morse (2003) suggest that the "researcher often connects with participants at a very deep level". Both I and these three mothers were caught up in their stories; however I was able to step back and provide empathy and support to these mothers whilst they worked through their very brief moment of sadness. I did not leave any parents who were apparently distressed at the end of the interviews. I found myself in a situation similar to Oakley (1981) in that several parents asked clinically specific questions or demonstrated levels of limited knowledge in relation to their child's health or birth condition. Using my clinical knowledge I answered parents' questions

honestly but also gave advice to two parents about seeking further support from their hospital.

Within the analytical process narrative interviews are acknowledged as a performance (Reissman, 1993). As such I considered the language, non-verbal communication and the audience role, which included intently listening to the performance of these stories within the interviews. As Corbin & Morse (2003) explain:

> "[P] participants are telling their story for an audience, even if at the time of the interview there is only an audience of one-the interviewer. Participants know that eventually their story will be told to others" (p 339).

I had to be patient and a skilled listener to hear parents' stories about their child's ambiguous genitalia and genital surgery. It took time for a few parents to articulate and perform their stories during their interview. Parents also included me in their narrative by using 'you' and although in-depth discourse analysis is not the analytical framework used within my study I feel this needs a brief mention. Most of the parents stories included 'you' or 'you know' when they talked about themselves. I believe this was a way in which they tried to include me in their stories. Interestingly at the Riessman (2005) workshop a discussion had taken place in relation to participants' use of 'you' in stories as a form of engagement and inclusion. I think my surprise was the level 'you' was used by some parents throughout their stories. Listening to the interviews as they were told, rereading the transcriptions and becoming immersed in the data also had an impact on my own experiences as a researcher and clinician.

6.4 Interviews, fieldnotes and reflexivity

There were occasions when parents told stories that 'tested' my response. I believed that such 'testing' helped parents decide if they wanted to share emotive stories. Parents had to develop a level of trust in my skills as an interviewer and also as a researcher. In order to explore my own interview skills I kept fieldnotes during the course of my data collection and analysis. These were written as soon as possible after the interview, for example I would stop somewhere on the way home after the interview and write my fieldnotes whilst fresh in my memory. I also added to my fieldnotes when listening to the

audio and rechecking the transcription. The transcription was always returned within a week of the interview and therefore I was able to begin to check and re read the transcript very quickly following the interview. My fieldnotes were multifarious I used them to explore the interview process, mine and the parents' emotions and actions during the interview and any problems that arose during the study. Fieldnotes were the starting point of documented interpretive thinking as the interview numbers grew and they became literary sources of evidence.

The shift from clinical practitioner to qualitative researcher resulted in my emotional integrity and skills being explored within my fieldnotes. The issues generated from fieldnotes were also taken to supervision to be unravelled and reconsidered and my own beliefs were challenged as findings began to emerge from the data.

6.5 Conclusion

A third of the parents in my study were fathers and I had an overall response rate of 58%. None of the parents withdrew their consent. The interview phases in my study mirrored those described by Corbin & Morse (2003). Fieldnotes informed my reflexivity and were important in developing my interpretive thinking as I collected further interviews. The findings from parents are examined in detail in chapters 7, 8 and 9 in which the foundational stories, aggregate stories and keystone stories are presented. In total there are twenty foundational stories, eight aggregate stories and three keystone stories.

CHAPTER 7: KEYSTONE STORY 1 – PARENTS' STORIES ABOUT THEIR CHILD

7.1 Introduction

In this chapter I illustrate the findings under the first keystone story along with the two aggregate stories which are supported by nine foundational stories. Parents' stories about their child were evident throughout all the interviews. They focused on specific key events from their own experiences and also their future expectations for their child. This chapter is divided into three sections.

The first section (7.2) describes parents' early experiences and explores how parents reflected on their responses to the unexpected events surrounding AG. This section also includes stories of the birth of their child. These early stories are considered under the aggregate story of 'beginning stories'. These beginning stories were supported by three foundational stories; before birth stories, immediately after the birth stories and an unexpected out of the blue story. The aggregate 'beginning stories' often unlocked other stories which in turn informed and developed how parents' understood and learnt about their child's AG.

The second section (7.3) describes the difficulties parents faced when they were trying to learn about AG whilst also managing the emotional shock of AG and exploring how to protect their child. These stories are considered under the aggregate story of 'stories about the difficulties in learning about AG'. This aggregate story is supported by six foundational stories which included stories about; uncertainty, the appearance of AG, sharing the news about their child's AG, danger and worry, protection and the risk in telling others and telling children about their AG.

The final section (7.4) is the keystone story and comprises of the summary of and synthesis of the foundational and aggregate stories from section 7.2 and section 7.3.

Diagram 7.1 is a visual representation of the foundational stories, aggregate stories and the keystone story and how they link together. The parents' stories are presented in the text in italic font.

Diagram 7.1 Keystone story: Parents' stories about their child

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                          ┌─────────────────────┐
                          │  Parents' stories about │
                          │      their child        │
                          └─────────────────────┘
                             ↙              ↘
         ┌──────────────┐              ┌──────────────────┐
         │  Beginning    │              │ Stories about the │
         │   stories     │              │ difficulty in learning │   ┌──────────────────┐
         └──────────────┘              │    about AG       │   │ Stories about     │
                                       └──────────────────┘   │ telling their child │
                                                              │    about AG        │
                                                              └──────────────────┘
```

- Before birth stories
- Unexpected out of the blue story
- Immediately after the birth stories

- Stories of uncertainty
- Stories about the appearance of AG
- Sharing the news stories
- Stories of danger and worry
- Protection stories and the risk in telling others
- Stories about telling their child about AG

7.2 Aggregate story (1): Beginning stories

Interestingly, all of the parents started their interview at the point at which they thought it all began. For six parents the beginning was during the antenatal period, prior to the discovery of AG. Five parents' stories started from the moment they were told about their child's AG and subsequently the uncertainty of their child's sex. Therefore, eleven parents started their story from either before their child's birth or immediately after their child's delivery. The majority of parents believed that the beginning was tied to their child's birth and the uncertainty of sex and all of these stories included their experiences of being in hospital. One mother began her story when she was told her daughter had ambiguous genitalia, yet as this was a late diagnosis she was not in the hospital at the time of discovery. The remaining three parents began their interviews with surgical stories which are discussed under the third key story theme. Although these three stories could have been considered under beginning stories I chose to include them under surgical stories as they were not directly tied to their child's birth. These three parents did talk about discovering their child had AG but this was not their beginning story.

7.2(a) Foundational stories (1): Before birth stories

Planning a pregnancy and having a baby are for the most part considered within Western society as celebratory events which follow a fairly predictable path. The anticipated outcome from a pregnancy is that a baby is delivered and the individuals involved in creating or planning on caring for this baby become parents. The process of developing into a parent involves a transition and 'parenthood' is a socially accepted concept and as such there are expectations bound up with becoming a parent. Chloe was very straightforward in describing her expectations of becoming a parent:

> "I just thought I was going to get, this baby and people just had babies and came home and I worked in a big office, and people had babies you know at the drop of a hat, and they all came in a couple of weeks later with their babies" (Chloe).

Imogene echoed Chloe's pragmatic sentiments about becoming a parent "I just thought babies were born and that was it" (Imogene). In the process of becoming parents all but four of my participants considered there was a

standard process involved in having a baby and that it was meant to be straightforward. For the parents who were aware that complications could occur during their baby's development this awareness came via two sources. Anne recalled watching a hospital TV documentary, while Sian, Maria and Faye had previous experiences of a variety of illnesses with friends or family. Anne described very clearly how after watching other people's experiences she matched these against her own apparently *"health"* pregnancy:

> *"I was mesmerised by [the TV documentary] its because there were so many problems... I used to sit here crying, just watching it and cry and cry and cry, just watching it and thinking, oh those poor kids and the parents by then I'd had all the scans and everything and [I'd been told] you've got a really healthy big baby" (Anne).*

Anne's belief that her baby was 'normal' was reflected in her understanding that a *"big baby"* equated to a healthy baby. In contrast to this story Chloe and David recalled the anxiety they experienced as a result of complications in Chloe's pregnancy with her blood pressure. Chloe told a very long story about her early admission to hospital at 28 weeks pregnant, the attempts to improve her health and safeguard her baby. Chloe felt constrained by the medical advice she was given for *"complete bed rest"* she no longer had any control or choice in her antenatal care. Although Chloe followed medical advice this did not completely manage her symptoms and she recalled the doctor discussing dates for an elective section with her. This option to choose a date gave Chloe back a degree of control *"The doctor [asked] did I have any particular strong feelings about dates in mind."* However, Chloe experienced pressure from her family; she described how her family humorously teased her about 'holding on to' her baby until their birthdays:

> *In actual fact [it was] something very funny he was actually due on the 1st of [month] and my husbands birthday is the 14th, and he would always say, oh you know, see if you can go 2 weeks over, so you can have him on the same day which you know it was just a joke......
> Me mum's birthday is the 10th [month], and my mum was saying just [a bit] longer" (Chloe).*

In the end Chloe felt she failed, as she could not 'hold on' and *"things turned for the worst"*. A medical decision was made for emergency caesarean section. Chloe recognised she would not *"just have a baby"* due to her health issues yet

she continued to believe everything was ok as she had been "*scanned all the time*". Chloe's story continues to describe the events immediately after Edward's premature birth, her own critical health condition and her recollection that, "*I thought Oh my God, I could die here*". Chloe's expectations of certainty during pregnancy and delivery were not met.

Sian related a story that bore similarities to both Anne and Chloe's stories; she recalled early antenatal scans identifying a problem with Tara, and how she had been initially pragmatic and later anxious about the scan:

> "*The doctor said 'it looks like there's something wrong with her bowel bit, but they'll send you to the hospital'...So off I toddled there and they said – 'oh no, it's the ovaries nothing to do with the bowel, it's the ovaries'.... They kept saying –'yeah, it's the ovaries, the ovaries'.....two weeks before she was due I went for a [last] scan. A black shadow was on the scan, it was 4cm by 2cm and they said it was a cyst on the ovaries. They went out of the room and didn't speak to me for about half an hour, nobody come in to see what was going on [no one] came back out and I burst into tears and they sent me back up to my room and I was crying and everything*" (Sian).

Sian had experience of ovary problems from within her family and therefore had initially not been concerned at her early scans. Until her 38 week scan Sian had confidence in her medical care and believed there was nothing "*seriously wrong*" with Tara. Up to this point Sian believed she was prepared to deliver a healthy girl with a cyst on her ovaries.

7.2(b) Foundational stories (2): Immediately after the birth stories

The birth of a baby is, for the majority of parents, friends and relatives, a celebration. However, for some parents childbirth can result in feelings of powerlessness and a loss of dignity which at times is coupled with poor professional support (Olin, 2003). All of the parents in my study had their children in a hospital environment. Parents in my study were as likely as any other parents to experience the feelings described by Olin (2003). However, they also lived through additional 'dramatic' events which either focused on their child's health or the uncertainty of their child's sex.

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Following childbirth all parents were anxious to know that their baby was alive, well and safe as Chloe describes:

> "All you want is that child to be alive, so if they say, 'oh there's something wrong'. The fact that they're still actually living and breathing, I don't know, [problems] just doesn't seem to be as important" (Chloe).

The sense of relief that the baby was alive was short lived for those parents whose children needed early 'life sustaining' surgery. Anne recalled the mismatch between what she saw "they've just given me this baby and he was all there, he was perfect", and the moment of acknowledgement that Barry was "not a well little boy". Anne recalled being so worried about her son she had to ask the doctor directly if he was "going to die?" For Frank and Gina the initial delight of a baby was matched against a fear for Helen's life, both immediately and in her future. Gina also recalled asking the doctors if Helen would "die prematurely?" Having to ask upsetting questions lead Gina to "start panicking and freaking out a little bit". If Rose, Helen, Tara and Barry had not had surgery they would have died due to the nature of their birth conditions. However, Joanne had not needed surgery to survive although she did have a serious medical condition. Her parents, Harry and Imogene recalled feeling a little "silly" at not initially having grasped an understanding of Joanne's potentially fatal medical condition shortly after delivery:

> "We knew nothing about it. I didn't even know about it. I didn't even know babies were born with it. I just thought babies were born and that was it. Not with these kind of things. Well you wouldn't would yer? (Imogene)
>
> I know it might sound a bit silly, like. (Harry)

Parents' initial concerns for their child's health formed short stories within the context of their complete narratives. Undoubtedly the dramatic nature of 'life or death' of their child for some parents was a significant experience. Once this perilous phase had passed, parents' worries about their child's genitalia became increasingly significant.

Very shortly after the birth of their child, fourteen of the fifteen parents were made aware of their child's AG or cloaca by medical staff, nurses, midwives or their partner. Eight parents recalled the 'dramatic' event of hearing about their child's AG. All fourteen parents believed that their baby would be clearly either a boy or girl in sexual appearance and genital function. This challenge, to their fundamental beliefs of a dichotomous sex actually positioned parents in a place of uncertainty. Doubt about their child's sex or function caused parents to experience a range of emotions, including disbelief, confusion and worry for their child's future. Christine's story represents the experiences of several parents:

> *"And to start with the birth in hospital, I went through that... I went to all my scans. I was monitored well because of my own condition. I thought everything was going fine. They said the baby would be on the small side and I had the name picked. There was a midwife called Maureen and she held my hand right the way through and she was lovely until the baby came out. And she said you've got a lovely... it went quiet, you've got a lovely, baby [silence and Christine became a little tearful] you don't want to know" (Christine).*

Although she was not critical of staff, she expressed how being *"put in a room away from other people so they couldn't see what was going on"* resulted in her feeling like an outcast. Also by ending her story *"you don't want to know"* this seems to suggest Christine was saving me from hearing something too awful to repeat.

Although Chloe described graphically her own poor health following Edward's delivery she described *"the worst thing"* as learning about Edwards AG. Chloe's understanding that 'uncertainty' could exist was very hard to comprehend as she explained when she said *"the realisation of what they've said, I don't think that ever dawns on you"* (Chloe).

Anne recalled hearing the news from her husband that Barry was both *"sick"* and that he had *"something wrong"* with his penis, this event came as a shock and initially she experienced feelings of disbelief:

> *"I was sitting in the bath and he came in and I could tell by his face something was wrong and he said 'he's not a well little boy' and .... I couldn't do anything, I couldn't cry, I couldn't talk, I couldn't do anything, I was just sitting in this water.... [then] I thought, well hang on, I've just seen this baby, they've just given me this baby and he*

was all there, he was perfect, and he had a mop of red hair, he was beautiful, beautiful boy he was, still is. And I thought no, he's said that wrong, he doesn't mean that" (Anne).

Anne struggled at first to reconcile the baby she had seen with the different information she was given by both her husband and the doctor.

Even when parents had experience of ill children the 'uncertainty' that accompanied AG was different. Maria's middle child had been born with a condition that was noted very soon after birth. This experience, she believed, had prepared her for any future problems she could experience with her new baby. Maria used her previous experiences and skills to enquire after the baby's health and was surprised to be told *"well we're not going there yet because we don't know what sex your child is'" (Maria).* This uncertainty surrounding her child's genitalia also challenged her confidence in the midwives as she was *"quite shocked really that the midwives didn't know how to react and what to say" (Maria).* Shock at their child's AG and the shock of how information was shared was recalled to a greater or lesser degree by all parents. Faye shared a similar experience to Maria and other parents yet her story also captured the chaotic nature of events that happened in the delivery room. Although the following excerpt is a long segment of story Faye describes several important experiences:

> *"When I gave birth, you give birth, you're 12 hours in labour, screaming your lungs out for everybody to hear, you give birth, they take your child away to the back room, obviously to clean the throat out or whatever it is they do. Then you hear this nurse at the top of her voice going; 'oh my God, what is this? Get a doctor in here quick, will somebody explain, what's going on?' kind of thing.*
>
> *I'm lying there, my feet are facing this room at the bottom with glass windows and three nurses are all there sort of peeking at my child. I'm wondering what in hell's name's is going on? So you're lying on the bed, you know with your bits and pieces hanging out for the nation to see wondering why they're all panicking in the back and you're wondering are they ever gonna come out with your child? You get all sorts going through your head at that point don't you? You know 'is your child dead, and does it have an extra leg, an extra arm or is there bits missing that should be there but aren't there?' [After what] seems like an eternity, they come out with this child and this*

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doctor lays this baby on the bed for you, opens the sheet that it's wrapped in and says, 'well the thing is, we don't know what you've got'" (Faye).

Faye's experience of childbirth up to the point of delivery was perhaps as she had anticipated. However, the behaviour of the nurse caused Faye to become uneasy, which was made worse by her feelings of exposure and loss of dignity. Her unease also led to her becoming angry as she was offered no explanation for the delay in sharing information about her child. She grew anxious about her child's health to the point where she questions if her baby was "*alive?*" The waiting, confusion and apparently chaotic events eventually peaked when Faye finally heard the news "*we don't know what you got*". Faye, like many other parents demonstrates the importance of sex in order to give the child a label of boy or girl, without this she seems to imply she just "*got*" something.

7.2(c) Foundational story (3): Unexpected out of the blue story

This story describes one mothers' experience of a 'late diagnosis'. For Kerran the first four months of Leah's life at home had been uneventful, she had fitted into family life and there were no health concerns. During this period Kerran recalled not having had any worries about her daughter's genitalia. At a routine baby clinic the attending locum doctor had questioned Kerran about her daughter's ambiguous genitalia. Kerran recalled her surprise and disbelief that her daughter's genitalia had been described as ambiguous:

> "*She went for needles, vaccinations as a baby and I think it was the third one that they actually spotted this. It was a supply doctor, it wasn't even her actual doctor, she said, 'oh has she been treated?' And she looked at me like this, very confused, cos I hadn't said anything; I said 'there's nothing wrong with the child'. She goes 'I think I want to refer you' and that's how I came to know about it... Part of the problem at that time was because we weren't given any information I would just find myself in tears all the time, I didn't know what was going on, what was going on with Leah, just that she was abnormal*" (Kerran).

Kerran clearly considered her daughter to be 'normal' and was unprepared for the doubt she experienced at a routine clinic appointment. Following the exchange with the locum, Kerran demonstrated emotions of shock, insecurity

and fear. She became increasingly anxious about her daughter and believed herself to be isolated. She also began to question her existing belief that her baby was a girl. Both how the information was delivered to her and the lack of support following the dialogue resulted in Kerran questioning medical staff skills. For Kerran, her 'trust' in medical staff, both short and long term, was significantly impacted by these events. Trust becomes a key story for Kerran, medical staff failure to notice Leah's AG post delivery, being left with little information and no *"real support"* impacted on many areas in Kerran's life, including her on going anxiety about her daughter. The impact of AG on parents' lives is discussed under the keystone story parents' stories about their experiences of being a parent in a social world (Chapter 8).

7.3 Aggregate story (2): Stories about the difficulties in learning about AG

All of the parents told stories about the difficulties they faced in trying to understand or learn about their child's AG. Parents spoke of how the uncertainty of sex and much later gender became increasingly complex[15]. Parents recognised that learning complex information about their child's AG was difficult. Medical language coupled with difficulties in conceptualising information was complicated for parents.

A parent believed in an image of biological sex as either a boy or girl and uncertainty was considered a dilemma. Once parents had heard that their child had AG a whole new set of stories emerged. These focused on how they dealt with the initial situation and how they began to understand what AG meant to them. Parents spoke about how AG had impacted on so many 'taken for granted' assumptions. They acknowledged social models of male and female such as 'blue for boys' and 'pink for girls'. Parents also had to look at their child's genitalia and deal with any feelings this had for them. Some parents wanted to share the news of their child's birth but were hesitant about how and what information to disclose and to whom.

Parents forced themselves to explore how best to protect their child. They also explored how and when they would share information with their child about the AG and surgeries. The AG caused parents anxiety, they would talk

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[15] Sex and gender are explored in greater detail in Chapter 2 (section 2.6). Within the context of my thesis sex is considered to relate to the genital appearance as either a boy or girl and gender is the model of behaviour and social influence of either male or female characteristics.

about their worries for their child's future, intimate relationships and future paternity issues. The following six foundational stories illustrate parents' experiences that occurred following the birth of their child with AG.

7.3(a) Foundational story (1): Stories of uncertainty

Four parents recalled changing the name of their baby from male to female or visa versa due to the uncertainty. Imogene remembered *"it was like backwards and forwards to begin with"* although to her Joanne *"looked like a girl to me but to other people she didn't, it was mixed"*. Hesitancy in assigning a sex or changing the child's sex impacted upon the colour of the child's name band. The complexity of 'labelling' of pink for girls and blue for boys is not new in relation to children born with ambiguous genitalia (Preves, 2003). In my study Faye was one of several parents who focused on social cues to identify sex *"if it's a boy, it's got to wear blue, if it's a girl it's got to be in pink" (Faye)*. Four parents considered their experience of being asked about pink or blue photographs in hospital as stressful as they felt unprepared as to how to respond to the photographer. However, Maria clearly retold how she felt and the tactic she employed to constructively manage the situation:

> *"There was a woman in the hospital who was taking photographs, you know they come and do the nice little picture. It's a wonder I didn't strangulate her. She kept on 'pink or blue?' And she just looked at me and I just said 'no we're not having photographs done' as I couldn't say well he's was a boy or he's a girl or I'll have pink or I'll have blue" (Maria).*

Although Maria demonstrated a level of adaptation to the circumstances in which she found herself she later expressed a level of sadness at not having these pictures in the family album. Andrea and Brian recalled having a third option, white and felt that staff were trying and failing, to offer a solution to their child's ambiguity:

> *"On the cots in the maternity it's blue or pink, he had white" (Andrea)*
> *"He didn't even have a proper card, did he it was just" (Brian)*
> *"No, it was a piece of paper" (Andrea)*
> *"It was just baby [surname] that was all it said on it. There wasn't even a weight or a time or anything like that, was there, because I don't think the nursing staff knew how to deal with it" (Brian).*

The example of healthcare professionals not having a 'colour' for the baby, and using a piece of paper reinforced for Andrea and Brian that their child was something 'different'. More cautiously, I reflected on whether the language in their story, such as the lack of a 'proper' card and omission of important 'birth information', hinted at deeper concerns of the status of their child. Also if Andrea and Brian consider that Bobby is 'different' whilst he is a baby do their roles as parents change? For example do they treat Bobby differently to his siblings and are they perhaps more protective.

7.3(b) Foundational story (2): Stories about the appearance of AG

Given the significance of their child's uncertain sex, seven parents spoke about their interpretation of the appearance of their child's ambiguous genitalia. These very short stories were predominantly descriptive but at no time did parents express a fear or revulsion towards their child's genitalia. Kerran had considered Leah's genitalia to be normal for four months yet even after the suggestion Leah had AG Kerran believed *"there was nothing there to see really"*. For Anne, Barry's penis was perhaps something she only noticed because she had been told it was ambiguous. She was not unduly worried about his penis, because he could easily pass urine:

> *"I had [seen his willy], and the only thing I could, I mean it was fine, it was just, it was sort of bent really, it wasn't straight but it looked okay…but I thought he's weeing so he must be okay" (Anne).*

Chloe also experienced similar emotions to Anne except that she considered Edward's genitalia were not quite right because *"wee wasn't coming out of the right place"* and *"it didn't look right"*. David recalled thinking early on that Edward's penis was *"strange"*. Maria recognised that Neil's penis *"didn't look as it should of done"* but she coped with this for the majority of the time. Harry and Imogene very briefly and pragmatically, explained that they felt the appearance of Joanne's genitalia was perhaps a little masculine but this they believed did not stop her from being a girl:

> *"The sister said 'they're not sure if it's a boy or a girl'. If you ever seen her, you know she's a little girl. All that was up with her was she had a protruding clitoris. It looked like a little willy at the time" (Imogene).*

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Parents' stories relating their visual interpretation of the appearance of their child's ambiguous genitalia were surprisingly composed. None of the parents described being frantic upon looking at their child's genitalia nor did they describe their child's genitalia with revulsion. Although hearing the news of their child's uncertain sex shocked parents and resulted in them becoming anxious about their child's uncertainty the actualisation of looking at their child's AG appeared to be recalled pragmatically. The initial disclosure of AG by the healthcare professional appeared to cause the shock and subsequent anxiety for parents rather than when they looked at their child's genitalia. Parents based severity of AG against their child's bodily functionality, such as being able to pass urine.

For five parents the appearance and behaviour of their whole child were the factors that motivated them to affirm their child's sex and gender. Andrea and Brian recall balancing the medical input against the fact they *"looked at him"* and decided that he was *"a boy, he's a thug"*. Maria told a story about how her mother had reassured her Neil was a boy because he had *"boys' feet"*. Faye told a story about how the changing colour of Gabby's hair, face and behaviour helped her decided that *"this really wasn't a boy at all"*.

7.3(c) Foundational story (3): Sharing the news stories

Once parents had internalised their initial reaction to the news of their child's AG they began to consider how they would share their story with their family and friends. All the parents had told at least one person that they had either a boy or girl, even when medical staff had suggested that they wait until a sex had been decided. Whilst waiting for test results parents were uneasy due to the possible complications that could arise from having made the wrong decision about their child's sex. This had two implications for parents; firstly their confidence in their own ability to make decisions was questioned and secondly, the consequences of being wrong, as Chloe explains:

> *"We'd told everybody we've got a boy, imagine now having to tell everybody actually we haven't got a boy, we've got a girl. And I thought how awful is that for like, for him? I mean, apart from us"* (Chloe).

David recalled experiencing similar feelings to his wife and was worried about how he would be able to *"go back and tell everyone"*. They believed having to change their child's sex would be *"awful"* because everyone would then know about the uncertainty surrounding their child's genitalia. Tentatively, I reflected whether their story intimated feelings of shame. However, interestingly, none of the parents used the word shame when referring to AG. Faye, Oliver and Imogene spoke fervently about how they were *"not ashamed of it"* (it being either AG or cloaca).

Sharing news with immediate family when they came to visit the 'new baby' was difficult. Maria felt she lacked the skill and expertise to know what to say to her family:

> *"It was horrendous, the worst day I think of me life because everyone came and everyone was upset, no one knew what to say and the staff hadn't helped and you'd think they'd have some sort of training you know to deal with this" (Maria).*

Asking people not to visit was hard for parents as Chloe recalled:

> *"I didn't know really if I want people going to see him...we were just demented, and of course we'd got our families who wanted to come and see this baby and for 4 or 5 days or whatever it was, when we didn't know what he was, it was just terrible, and, and it must have terrible for them. How they sort of just passed it off I don't know" (Chloe).*

All of the parents either spoke to each other or their own parents as a source of support. Chloe told her parents and although she couldn't recall their reaction she remembered they *"were trying to keep everything calm"*. Some months later Chloe recalled her sister being shocked and upset when she accidentally discovered that Edward had AG. Chloe immediately regretted that her sister found out about Edward:

> *"I told her in a weak moment the whole story, and then I was sorry because I thought, although you trust people and I do trust her, I don't want people telling anybody else, and I thought I don't want people knowing" (Chloe).*

Sadly for Faye the moment of sharing the news was taken away from her as the midwife spoke to Gabby's father in the waiting room and said:

"That I'd got a boy and then went back in the room within a minute and told them I'd got a girl and then at that point they thought I'd got twins" (Faye).

This one moment had long lasting negative repercussions for Faye. The news of a 'twin birth' filtered through to the community and when it became apparent she had one baby the uncertainty of the child's sex became a curiosity to the local community. Not all parents were apprehensive about disclosing information to friends and family. Kerran had always had support from her family and did not consider not telling them:

"We're a very close family, I've got two sisters, a lot of very close friends, parents and brothers... I do tell my sisters and my mum" (Kerran).

Although Kerran received a good deal of support from her family she still believed that they were *"all curious"* about Leah's AG. Oliver recalled sharing very little information about Rose with family or friends. He used his mother's forgetfulness as a positive method for avoiding questions about Rose:

"I tell my mum as much as I can tell her and she takes in, but she's very forgetful which is convenient. Now if I was to tell a couple of other members of the older family, it would be Chinese whispers and been blown out of all proportion....All they need to know, is [that] she's okay or not, as far as I'm concerned" (Oliver).

Parents were selective about the information they chose to share. Their ability to 'tell others' was based on both their understanding and knowledge of AG. Half of the parents talked about how hard it was to 'tell'. Christine demonstrated this when she recalled how she tried to tell her mother but in the end *"she was as much in the dark as I was"*. Gina and Frank recalled wanting to tell people more but they struggled because:

"You couldn't explain it because you didn't really know. It was just something that was totally alien to us, you know. We didn't understand the word, we didn't understand anything about it" (Gina).

Even when parents had gained some understanding of AG and they no longer had any 'uncertainty' about their child's sex, the decision to not share information, for some, remained resolute. Both David and Chloe agreed that

sharing the news about AG would have been hard, so they chose not to. Not sharing news with friends or extended family was, for some parents, a method of protection and conflict arose when parents had different opinions. Maria very clearly told a story about the differences between herself and her husband. This segment summarises the differences and her belief in the value of 'talking':

> "If it was my husband, he wouldn't have told a soul, no one would have known. It would have been a secret, nothing mentioned because it is genitalia and it still is looked at as something that's taboo. But my personality and my outlook on life is well, could be dead tomorrow and I didn't worry about who knew and wasn't embarrassed by it at all and I think that's what help me" (Maria).

The difference between Maria and her husband continued over the years. She felt that he was able to "block a lot of it out, like it's not happened and we don't discuss it". The need to talk and share information inside and outside of the 'parent unit' was not reserved to mothers. Brian, rather than Andrea, took the lead in talking about Bobby's AG:

> "I don't stop myself from talking to people about it and his condition, it's the case of I found it easier talking to people about it" (Brian).

Some parents had chosen to tell family and friends whereas others had not. For a few of those who did share information the level of disclosure was kept to a minimum in order to avoid having to answer difficult questions. The reluctance for parents to be open was driven by a belief that other people would not understand AG. A lack of understanding is a major contributing factor to not tolerating difference. Parents considered that AG made their child different and was perhaps a stigma although they did not attach the concept of shame to stigma. Parents tied difference to a need to protect their child from harm. Parents' worries about disclosure were motivated by fear for their child's physical and emotional well being, both presently and in the future. All parents wanted to safeguard their child to the best of their ability in the complex situation which arose as a result of AG. Parents perceived there were potentially dangerous consequences with sharing information about their child. Therefore, the majority of parents wanted to keep control over the information they disclosed in order to protect their child from harm. The consequence of parents loss of control of information within their community is explored as

stories of danger and worry in foundational story 7.3(d). While parents perception of risk and their worries about how to protect their child are described in foundational story 7.3(e).

<u>7.3(d) Foundational story (4): Stories of danger and worry</u>
Communities are an integral aspect of society and as such impact on people's lives. All of the parents' communities, friends or family had the capacity to directly affect them either positively or negatively. Three parents experienced encounters they believed to be negative and were a direct result of their child's AG. Harry, Imogene and Faye, were shocked by what they believed to be the audacity of people within their community to ask personal and upsetting questions about their child. For Harry and Imogene a family member who had happened to be present at Joanne's birth had shared information about Joanne's AG with her friends, without either Harry or Imogene's knowledge or consent. This resulted in Imogene and Harry being shocked and ill prepared when they were asked questions about Joanne when they were out socialising with friends. They felt the questions about Joanne were *"just like a knife all the time"* and they were afraid for her future as *"unfortunately for Joanne everybody knows about her" (Imogene)*. For Faye the local community expressed a cruel interest in her baby:

> *"When you've got people stopping you in the street, pulling the covers back off you're baby and it's like, 'oh you'd never know would ya'? Never know what, 'you know that your child's basically a freak', you know. Oh I had one woman ask me, 'what it was like to have a child that was born with a gaping hole between its legs'. Yeah. I actually got beaten up. Outside a shop with both my kids with me because this woman pulled me up, 'Oh you're her that's had that freak for a baby' (Faye).*

Faye had not initially considered how she had expected friends and strangers in her community to react to Gabby. She felt she had little choice in how to control the information about Gabby's AG because of the existing rumours and gossip within her local community. Faye clearly found the level of curiosity about Gabby's AG very intrusive. Faye was clearly concerned about the images that people had fabricated, describing Gabby as a freak. The use of the word 'freak' I would suggest described Faye's fear that her daughter was considered as not

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human by some people. This dehumanisation in some way left Gabby 'open' to inspection and scrutiny as she was a source of curiosity. The majority of parents did not share Faye's or Imogene and Harry's experiences yet Oliver describes how he was fearful that:

> "If the community knew, it would possibly have a detrimental effect to Rose's wellbeing" (Oliver).

How communities could find out about their child was a concern for parents. Parents described the post-natal ward, baby clinic and changing their child in public places as possible settings in which the AG could be noticed. Post-natal wards were often difficult environments for mothers as they described feelings of being alone, afraid and angry. Chloe recalled why she had considered the post-natal ward a testing environment where she had to be ready to think quickly:

> "[I was] in this room where there was six beds, they'd come up to tell us [the doctors], you know, 'oh we haven't had the results back of the chromosome test' and all these mothers are 'ugh'? I remember a mother in the next bed to me said, 'what's that test'? 'Do we all have that'? And I was thinking 'no'! And I thought 'oh my God, I'm not going to be saying to you, we don't know whether my baby is a girl or a boy, I'm not saying that!' And then I'm just saying 'no it's a, we don't really know what it is', and I'm like trying to make up some silly story in the ward"
> (Chloe).

The need to be alert and not share information unwittingly was an additional burden for Chloe as she clearly did not wish to disclose information about Edward. I reflected on the language in the last sentence and explored if the word 'silly' was an expression of Chloe's frustration. She was anxious to conceal the dangerous information about Edwards AG and I believe she was also discouraged by her 'silly' or perhaps vain attempts at creating a 'cover' story.

Often new social links are made in post-natal wards; mothers especially will see other mothers who attended the same ante-natal clinics and they may also meet at baby clinic. These stories made me reflect on whether parents underwent a feeling of loss for the events they hoped would happen when they had a baby. Parents were likely to use their interpretation of AG as an

indication of how other people would view their baby. Maria reflected on her worries about other people's reactions to the appearance of Neil's genitalia at baby clinic:

> "The hardest thing was going to the clinic and having him weighed. Get the prams out, everyone's got the new prams and everything's matching and you all march down to the clinic. It was like everyone's gonna see him undressed, and this is like horrendous" (Maria).

Parents adapted their behaviour as a result of this type of anxiety. Some mothers attended baby clinics whereas others avoided any contact where they might have to change their baby in front of other people. Chloe was not alone in that she "wouldn't let people change my baby, because I hadn't told people". Avoidance or withdrawal was a type of coping strategy adopted by several parents which resulted in them becoming the sole carers for their child. Coping without help from family or friends resulted in some parents becoming isolated or as Chloe recollected she became "this obsessive psycho mother". For those parents who wished to engage in mother and baby groups and sharing the care of their child with family there was the dilemma between a desire to be open and the reality of the implications and consequences of such openness.

7.3(e) Foundational story (5): Protection stories and the risk in telling others

Maria had been open in sharing information about Neil with her family and friends and she told a story about trying to find a balance between sharing information and maintaining privacy for Neil in the future:

> "We knew that whoever was told would know for his full life and hopefully you know they wouldn't make an issue of it later on cos you have to think of things with genital surgery because he could be 16 and people who know him now obviously knew him then that it would be embarrassing for him. No, we didn't keep any of it a secret everybody knew, family, friends. I didn't have a problem with speaking about it or discussing it, but people did say to me 'well we'll know him when he's 16 so, you know, so don't say too much' and I suppose that's right in a way because of the children that he was born with, he's gonna go all the way through school with them children and them parents will always know and I didn't want anyone to look at Neil as though to say, 'Oh he's the one that we didn't quite know whether he's a boy or a girl'. And I didn't want that to stick" (Maria).

All of the parents to either a greater or lesser degree told stories similar to Maria. Parents felt the need to protect their child and maintain their future privacy thereby saving their child from "*embarrassment*". The dilemma of sharing information conflicted with the parent's desire to shield their child from being teased in the future. David described his fears for Edward:

> "*I'm just trying to protect him, when he's sixteen and all the hormones start going he's only got to try and go with a girl. Next thing you know it's round the school, I can see it coming*" (David).

Although AG was a problem that parents could not control, the disclosure of information for the majority of parents was governed by them. Therefore deciding on the level of disclosure became a protective mechanism and a coping strategy for parents. Some parents considered themselves to be ill prepared to answer questions about AG. Disclosure could also open an area that parents believed to be unhelpful as Chloe explained she did not want other people "*feeling sorry for me*".

Challenges also arose for some parents when their child decided to share information about their own body with other children. Sian considered Tara's decision to share information as positive and not something "*she's got to hide*". Many of the parents thought that their child had to have some understanding of their condition to be able to share information about themselves. All of the parents believed that their child also had to have some understanding of what had happened to them surgically. All of the parents told stories of how they had already shared information with their child about their birth conditions or surgery.

7.3(f) Foundational story (6): Stories about telling their child about AG

All of the children in this study were under the age of eleven at the beginning of data collection with the youngest at the time being 3 years 9 months. Parents recalled several reasons which influenced the information they shared with their child. These were the child's age and level of understanding, their own ability to know 'what' information to share and 'how' to do this and the impact they anticipated that sharing information would have on their child emotionally.

Most parents worked on the principle that when their child started asking questions about their own bodies they should answer them *"honestly"*. Christine had begun to tell Daniel information but felt whilst he was in junior school it was still too soon to *"explain everything to him"*. Maria wondered about when she would know it was time to talk with Neil as she explained:

> *"I keep thinking, would there be a right age to sit down and talk to him about it all and. Is there a right age?" (Maria).*

For some parents the decision of when to tell their child was prompted by their child's experiences. Anne recalled Barry coming home from school asking about his penis because *"a little boy had noticed it was different"*. Anne explained how she normalised the information she shared with Barry and proceeded to tell him about his surgery:

> *"I actually told him every little boy is different, you'll never see two little boys the same. He asked why he hadn't 'got skin' that came to a point at the end of his. I said 'well that's because when you had your operation you had a piece of skin taken away, to make you better' and he said, 'oh right', I said and 'it's nothing to worry about'. He asked 'why did I need the operation', so I said 'well because the hole that you wee out of wasn't on the end of your willy it was just underneath so, they moved the piece of skin and made it so that the hole was at the end of your willy and that you can wee in a straight line' and he said, 'oh right' (Anne).*

Although Maria had been anxious about what age to talk with Neil she recognized that Neil was worrying about something, which she believed to be his genitalia, since he would no longer change at the swimming baths. She had tried to talk with him but *"he wouldn't talk about it, he got very, very upset and very agitated"*. This experience left Maria unsure about how to talk with Neil. Similar to all of the other parents, Maria did not believe in keeping previous surgery secret from Neil. The majority of parents were thankful their child *"didn't remember"* early surgical experiences but they did not use this as a reason to *"not talk about"* previous surgery with their child. Genital surgery was an important aspect for many parents as this allowed them to increase clarity about their child's sex. (The place surgery had in parents' lives is discussed in much greater detail in Chapter 8). However, for some parents' inconsistency existed in relation to being open and what they would tell their child about AG in

the future. Chloe, Imogene, Maria, Anne and Christine all offered their opinion of what their child should not know about themselves. Chloe's story illustrated this well:

> "You know, don't hide it or whatever, you say what it is, and really he should understand what's happened. I haven't told him, obviously, we didn't know when he was born whether he was a boy or girl, I wouldn't tell him that, because I can't imagine what that would do to him. And I don't think he needs to know that because, what is the point of that?" (Chloe).

Chloe's reticence was perhaps a form of protection although her avoidance to disclose all available information to Edward might be considered a negative coping strategy. Reflecting on this, I wonder if this approach allowed parents to actively avoid thinking about the complexity of 'uncertainty' thereby facilitating their coping on a day-to-day basis. Also if the 'uncertainty' had been resolved following ultrasound and chromosome tests then the parents seemed to consider their child either completely male or female. Parents also feared they could confuse their child by telling them about the 'uncertainty of their sex' at birth. For Maria, labelling Neil's genitalia as "not right" could result in him thinking "it's not right" and this could mean "it'll never work properly". So some parents decided to tell their child what they felt they needed to know thereby controlling the level of disclosure. This was based on the belief that if the child did not remember the dilemma of uncertainty it would not worry them or make them think they were different. Faye, in particular, felt very strongly about parents lying to or hiding information from children. She explained why she believed children had a 'right' to information:

> "You know, how would you feel if your mum kept information away from you about your medical health, that could, say for instance affect your fertility in the future, whether you do or do not have children, whether it will affect your development, you know, if you knew there was something nagging at the back of your head and you couldn't put your finger on it and I could give you the answer to it, would you want to know? These kids are gonna go, 'yes I'd want to know'.

> "I wouldn't like that kept from me. How can these parents sleep at night knowing that they're lying? It's not that they're keeping it from their child, they're lying. Because they're lying to themselves

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therefore they're lying to the child. It infuriates me, it really does annoy me" (Faye).

Faye reflected on how she believed *"you have to put yourself, as hard as it is, as silly as it sounds you have to into Gabby's position".* She also used this as her rationale for genital surgery and she described the socialisation of gender *"you're trained all them years aren't you, to be male or to be female".* The clarity offered by a clear sex and gender allowed Faye to relate to Gabby as a girl and develop a relationship in which she could *"be open and honest"* with her. Once Faye had a relationship with her daughter she considered any omission or evasion away from the *"truth"* as being a *"lie".* Faye very clearly recognised that AG and the complex nature of initial uncertainty about Gabby was *"a lot to think about"* summing up that *"it wasn't easy".*

The following section is the keystone story and is both a summary and synthesis of the foundational and aggregate stories described in section 7.2 and 7.3.

7.4 Summary and synthesis of keystone story 1: Parents' stories about their child

Parents very clearly told stories about their child and the complex nature of AG and uncertainty. Most parents thought that *"people had babies you know at the drop of a hat" (Chloe)* and that childbirth resulted in a boy or girl. The majority of parents were unaware that doubt over a child's sex could exist at birth. Having a child with ambiguous genitalia was clearly shocking to parents since almost all them were surprised that AG could exist. Several parents, but Sian especially, were stunned that their ante-natal scans had not detected their child's AG. The doubt or uncertainty of their child's sex challenged their beliefs about a dichotomous sex and left parents unsure of their child's future. As Faye described certainty of sex is expected *"society expects male or it expects female and it gets confused when there is confusion".* Parents appeared to value a clear biological sex as essential in humanising their child. I would suggest that parents' anticipated experience of having a baby was temporarily lost because of the AG. Parents' experiences of uncertainty as a consequence of AG started from the point where medical staff identified that there was

something wrong with their child's genitalia. Events immediately after their child's birth were described as *"confusing"* and *"chaotic"*. They had expected doctors, nurses and midwives, as professionals, to *"know how to react and what to say"*. Healthcare professional's vagueness surrounding both AG and how to approach parents were lasting memories for both mothers and fathers as Maria summarised:

> *"The worst part was the first day when no one knew what to say or how to handle what was wrong and the shock of it all really" (Maria).*

Parents had anticipated medical staff would be *"trained"* in managing *"out of the ordinary births"*. Anne recalled that *"they didn't explain anything to us"* and *"they're not clued up"*. Confidence in healthcare professionals was shattered for some parents and seemed to contribute to their overall confusion. On reflection, my sense is that for some parents the language used by medical staff to reveal the uncertainty, such as *"we don't know what you got"* or *"what it is"* resulted in worry, fear and concern for their child. Both *"it"* and *"got"* were words parents saw negatively inferring that something was *"wrong"* with their child and therefore framing their child as a problem. It was not clear how this 'negative' dialogue impacted on parents' feelings towards their child or how they viewed healthcare professionals. Anne remembered the doctor *"trying to say to me what was wrong and I couldn't understand what she was saying or anything"*. David recalled feeling *"angry at how he [the doctor] was telling me"*. Parents also had to match what they believed about their child's genitalia *"everything was there, there was nothing missing" (Anne)* with what they were being told.

Parents' stories reflected a range of emotions associated with a variety of feelings towards their child and also as a consequence of their experiences. For the majority of parents it came as a shock to discover that boy or girl sex might not be straightforward and resulted in feelings of disbelief and bewilderment. Shock was also described by Anne both physically and emotionally. Shock resulted in her being unable to experience feelings immediately, she *"couldn't cry....talk....do anything"*. For some parents shock, was tied to confusion and a loss of orientation. Parents had to try and move

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forward from their experience of shock. They sought to find a sense of harmony between their emotions and understanding about their child's AG.

Parents also experienced anxiety as an emotion as evident in their feelings of fear, apprehension and worry. Kerran, Faye, Christine, Sian and Gina all spoke about *"crying"* as their early response to anxiety. Five families also experienced anxiety as a consequence of their children's life threatening health conditions[16]. For most parents their initial anxiety lessened once the initial stressors of uncertainty had been overcome and their child was 'sexed' as either a boy or girl. Another factor which influenced anxiety was surgery (for greater detail see Chapter 9). Like several parents Oliver described initially wanting *"to know everything"* wanting to *"know it now"* but this *"information overload"* increased his anxiety and made coping difficult for him and his wife. He adapted his thinking and believed:

> *"If you get too much info, you can worry about things that might never happen, and you can worry about things that you don't need to worry about. You'd just worry yourself stupid" (Oliver).*

For parents there were difficulties including understanding complex information, deciding who to tell and being certain of what information they wished to share with others. Therefore they faced the obstacles of both understanding and remembering the information they had been given about their child's AG. Kerran reflected she *"never understood the importance of it..to be honest for the first few years".* Also parents often *"couldn't understand why it had happened"* (Chloe) or *"it's too difficult, I don't understand it"* (Oliver) and often were left *"panic stricken"* when they did not *"understand a word"* (Gina and Frank). Clearly, parents did need information that was delivered in a meaningful manner to allow them to make decisions. Their need for information and their subsequent understanding are explored in much greater detail in Chapter 9. In essence some parents wanted *"what they needed to know"* while others wanted much more information as Andrea explained:

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[16] Although the stories that parents told and the anxieties they experienced in relation to their child's life threatening health condition were both interesting and informative, these have been set aside as they were not the focus of my thesis. Where these stories are linked to AG, they have been considered.

> "*Some people might see it as too much information but I've always found that a little bit of knowledge is a bad thing, I'd rather know the bells and whistles*" (Andrea).

My sense is that using the terms "bells and whistles" within this context refers to Andrea wanting to know everything about Bobby's condition, regardless of how 'easy' or 'hard' it would be to understand or how it would make her feel.

Parents used the information from healthcare professionals to help them decide what to *"tell other people"*. However like healthcare professionals, parents' were not equipped with a language to address their child's sex uncertainty. All parents found it impossible not to refer to their child as he or she even though *"[the doctor] told us that we couldn't tell anyone he was a boy or he was a girl"* (Maria). On a micro-level of social expectations and roles parents had wanted to announce the birth of their son or daughter to friends and family. Yet on a macro-level the sex uncertainty resulted in a challenge to their belief system and they became anxious about the consequence of sharing information within 'society'. Parents struggled in deciding who to *"trust"* as they were fearful people may *"gossip"* about their child or *"treat them different"*. My findings indicate that parents were anxious to keep control of the information they disclosed about their child. Maria, Harry, Andrea, Brian and Anne, did not want their children *"laughed at"* for being different and they feared this could happen because children can be *"cruel"*. Others, Sian, Faye, Christine and David were anxious that they did not want their children *"bullied"* in school as a consequence of their AG. Therefore, parents had to match disclosure of information against risk. They had to assess the danger in sharing information against possible harm towards their child in the future and they wanted to protect them, For Maria, who was one of the most open parents in my group of participants, she still believed *"no one else really understands"* what it was like to have a child with AG. Parents worry about sharing information and their resistance to this could be construed as 'keeping secrets'. Reflecting on this made me wonder if 'secrets' were a type of armour that parents employed to protect their child from harm, as Imogene very pragmatically suggested:

> "*Well it would be secretive wouldn't it? Because people not like people to know, about things like that*" (Imogene).

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Interestingly parents did not want to keep secrets from their child about their previous surgery. Retelling their stories to their children opened up a dilemma for some parents as a few had decided that they *"wouldn't tell"* their child about their initial uncertainty. The decision of what information to disclose to their child, when and how rested with parents, yet parents stories demonstrated they received little or no support in managing this. For the parents disclosure was both complex and also worrisome, once the child had some understanding they could share the information with others. This could be done unwittingly by the child and as a consequence the parent would lose the control they had in managing information about AG. For parents there was always the potential consequence that their child could be unhappy with the surgical decisions made on their behalf in childhood. Sian, like many others, described how she hoped she had done *"the right thing"* whereas Christine, like Andrea, was troubled *"it has crossed my mind loads of times, was I doing the right thing"*.

Parents had to actively address their concerns in the complex situation which resulted from AG. They had to try and find a sense of harmony from their experiences and tackle their lack of knowledge and understanding about a phenomenon that challenged their basic belief of a clear and synonymous sex and gender. All of the parents believed they were learning to cope with AG and they had managed this by bringing together complex reactions and emotions thereby achieving a sense harmony. They managed their emotional responses to AG, shock and worry, against their desire to protect and safe guard their child. They managed to cope and bring harmony to the discordance between uncertainty and sex by taking control of sharing information with others. They recognised the value in enabling their child to understand their own AG in the future.

CHAPTER 8: KEYSTONE STORY 2 – PARENTS' STORIES ABOUT THEIR EXPERIENCES OF BEING A PARENT OF A CHILD WITH AG IN A SOCIAL WORLD.

8.1 Introduction

In this chapter I illustrate the findings under the second keystone story along with the three aggregate stories which are supported by six foundational stories. Parents' stories about themselves and their interaction with their social world were evident throughout all the interviews. They focused on specific key events from their experiences of having a child with AG, their beliefs about being a parent and also their social interactions. This chapter is divided into four sections.

The first section (8.2) is the aggregate story which focuses on parents' stories about the importance of their child's gender. In the three foundational stories that support this aggregate story parents' stories revealed the significance of a clear visual gender in order to establish and reinforce appropriate gender behaviour. Parents also described their concerns about their child's sex uncertainty within their understanding of a social construction of gender. In both of these foundational stories parents explored both why and how they currently protected their child and how they aimed to protect them in the future. The final foundational story describes the differences between mothers and fathers, gender and gender roles.

The second section (8.3) is the aggregate story that explores the value parents placed on early reconstructive genital surgery. The foundational story which supports this aggregate story is built on the early surgical intervention and childhood memories including the reasons why parents considered early surgery was important.

The third section (8.4) is the aggregate story which encompasses how parents understood parenting a child with AG. The two foundational stories which support this aggregate story describe how parents searched for normality and

why some of the parents in my research struggled to build emotional bonds with their child.

The final section (8.5) is the keystone story and comprises of the summary of and synthesis of the foundational and aggregate stories from section 8.2, 8.3 and section 8.4.

Diagram 8.1 is a visual representation of the foundational stories, aggregate and keystone stories brought together. The parents' stories are presented in the text in italic font.

Diagram 8.1 Keystone story: Parents' stories about their experiences of being a parent of a child with AG in a social world

Parents' stories about their experiences of being a parent of a child with AG in a social world

Parents' stories about the importance of their child's gender

Parenting a child with ambiguous genitalia stories

Stories about visual gender and typical gender behaviour

Difference between mothers and fathers

Searching for normality stories

Stories about the social complexities of gender

Stories about the place early surgery had in parents' lives

Stories about parents' emotional bonds to their child

Early surgery and childhood memories

8.2 Aggregate story (1): Parents' stories about the importance of their child's gender.

Once parents had started to overcome the shock of AG as described in chapter 7 they began to consider how both the child's sex and gender fitted together. Parents began to explore 'everyday' events and experiences in which sex and gender were taken for granted, such as visiting the girls or boys toilets. When parents actively sought to protect their child, for example by using a disabled toilet to change, they became conscious of intolerance or direct challenges from other people. The entire parent group highlighted the challenges they faced in relation to their child's gender typical behaviour. Also parents talked about how they encouraged their child to pursue gender typical hobbies or sports. When concerns arose about their child's emerging gender behaviour parents would reinforce gender typical activities. The following three foundational stories illustrate the significance of gender to parents and how this was actualised in their world. Also the differences between mothers' and fathers' views and understanding of gender and gender roles are explored.

8.2(a) Foundational story (1): Stories about visual gender and typical gender behaviour

Parents told stories about how their child's sex and gender came together from blood tests and ultrasound scans as either a boy or girl. Parents believed that sex certainty was important in society. They considered sex as visual for example, a boy has a penis and a girl does not. Parents also hoped that biological and sexual functions would be synonymous. For the parents their child's sex informed their gender and this in turn impacted on how they were dressed and encouraged to participate in gender typical roles. In effect, the child progressed from being given a 'sex' to becoming classified in terms of gender as they grew and developed. The visual image of the child's gender was important for parents as it helped determined how they saw their child (as either a boy or a girl). Six parents considered appearance important since this brought some harmony to the visual idea of sex and the uncertainty of AG. For both Andrea and Brian, Bobby's stature and facial features matched other little boys they knew which reinforced for them that *"he didn't look female in any way"*. For

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Imogene and Harry, Joanne *"just looked like a little girl"* this was reinforced for them following her scans *"she was all girl inside, which I was made up cos to me she was always a girl" (Imogene).* The colour of their child's hair was cited by several parents yet it was not consistent across parents. For Imogene, Joanne was *"a girl to me, with red hair"* while Maria believed Neil *"looked like a boy, he wasn't pretty at all but he had the brightest reddest hair".* The change in the colour of Gabby's hair was one of several events that cemented her gender for Faye:

> *"What was really weird right was when I gave birth and I looked at this child's face with jet black hair and everything, it was a stocky little boy...but when his hair started lightening up and suddenly went blonde I thought, this is a girl" (Faye).*

Faye described that validating Gabby's gender on hair colour was *"shifty"* but she believed that appearance including *"hair colour had a lot to do with it".*

Parents considered gender typical behaviour for their child to be important and also socially and culturally determined. Their child's choice of toys and approach to play either reinforced that they had the correct sex and gender for their child or became a cause for concern. Kerran spoke of Leah's love of *"barbie doll images and wanting to wear skirts and short dresses".* Both Imogene and Harry were worried that Joanne was *"confused"* as she preferred to *"play with the lads"* and had lots of *"cars and a garage".* Andrea and Brian were pleased that Bobby demonstrated a clear gender typical behaviour in his play. He preferred *"cars and football"* which they felt had not been either a *"conscious decision on their part"* or actively reinforced in the family home. They believed Bobby had made a choice about his gender preference, choosing not to play with his *"sister's toys".* For those children sexed as male, boisterous play was viewed as typically male behaviour, Andrea and Brian chatted jovially about Bobby's approach to playing with dolls:

> *"He doesn't want to play with dolls or anything like that, he'd rather rip the dolls head off" (Brian)*
> *"Yeah, he likes to take their heads off, he undresses them and takes their heads off" (Andrea).*

This display of 'maleness' reassured them and they said "we've no worries" about his gender.

Parents of slightly older boys encouraged them in gender typical sports and hobbies. Chloe and David told separate stories focusing on how they "sort of pushed" Edward into joining sporting clubs. These included "football and cricket and tennis" and that they would also send him to "football camp". Anne told several stories about Barry and his sporting activities and how proud she was of him:

> "He plays football; he's the only 7 years old, [he's also] in the life saving class in swimming. He gets man of the match playing football and he's fantastic" (Anne).

Later in her story Anne explored other consequences of playing in a team, such as shared showers. She spoke about the future importance of Barry's sexual appearance being the same as his gender:

> "Changing at football and rugby and things like that I think as they get older, it's better for them if everything is in the right place and you know working" (Anne).

Anne clearly believed that Barry would be embarrassed if he recognised he was different from his peers. Maria's story reiterates Anne's story, as she spoke positively about Neil's football achievements. She also continued the theme of matching sex to gender by exploring conformity in genital appearance and function. She believed Neil's genitalia had to be fixed so when "they were all out playing footie" he would be able to "have a wee behind the tree" like "all the lads". Parents of girls did not discuss physical activities as integral to their daughter's gender.

These stories reflect the parents search to affirm the sex of their child despite initial uncertainty and the importance of synonymous sex and gender. The appearance of a clear sex and associated gender roles and behaviour was important for parents. The potential that their child could be considered as different because of their AG was a worry for parents. They encouraged their children to participate in roles that maximised typical gender behaviours as a form of protection. However, several

parents spoke of the difficulty in matching sex and gender in day-to-day life. The challenges they faced were, at times, a reflection of their social understanding of male or female and this is discussed in greater detail in the following section.

8.2(b) Foundational story (2): Stories about the social complexities of gender

For all of the parents to be successful they had to be capable of following the social rules of presenting their child as either male or female when interacting with others. A few parents were conscious of how they dressed their daughters' to accentuate their gender. Imogene *"always had"* Joanne *"in pretty dresses"* to avoid any unnecessary uncertainty. Faye's anxiety to avoid further curiosity in her community led her to intentionally avoid non-gendered colours which she described as *"mint green"* instead she dressed Gabby in *"pink frilly dresses"*.

As well as appearance, the need to conform socially in day-to-day activities, such as how to void was considered important by parents. Christine, Maria and Anne all believed it was important for Daniel, Neil and Barry to be able to void standing like their peers and use a urinal. For Harry, girls did not have a penis and would therefore sit to void. This they believed also helped Joanne understand she was a girl:

> *"If you haven't got a willy you sit down to have a wee. And that tells you what you are, boy or girl" (Harry).*

For some parents' their experiences of other people's curiosity seemed to be a reflection of 'societies' intolerance towards sex uncertainty. Faye's story of curiosity and cruelty was told in chapter 7, Brian recalls a similar experience of when Bobby was in the *"paddling pool, starkers and one of the neighbours said, 'oh what's wrong with him?'"*. The story about the paddling pool prompted Andrea to share her thoughts about social acceptance and the implications this has for her family and Bobby in his future. She believed that society did not demonstrate leniency when individuals were different and this saddened her:

> *"If people accepted that everybody's different, it isn't a problem. But people don't accept that everybody's different. You have to conform to the way that the public says you should be. Bobby, unfortunately for him, is never going to conform, so he's always going to be the*

*centre of attention, he's always going to bring attention to himself so I
suppose to have him anatomically correct is easier. If he feels right in
himself and I'm hoping that he does because of what, what's
happened, what's been done, what he's gone through, then life's
going to be easier for him. But it's difficult because the public
perception of things is that he must be weird, there must be
something wrong with him and **no** there isn't"* (Andrea).

To some degree all of the parents in my study shared similar stories about their
anxiety in relation to social perceptions of sex and gender. From reflecting on
their own experiences parents speculated about how other people in society
would think and react towards their child. They appeared to worry about the
lack of knowledge of AG and DSD conditions amongst other people in society.
Parents were also anxious that other people in society would consider sex
uncertainty synonymously with sexual variance or homosexuality. Faye
pondered on her own experiences of sexual variation within her early peer
group. One of her friends had not *"conformed"* and Faye recalled a story of
exclusion:

*"There was a girl at school that was gay and you know there weren't
many people that accepted her the way she was, she was only
young and she actually told people. There were only a couple of us
that were friends with her, other people just sort of stood back and it
was like 'Oh that's disgusting!' Well it isn't disgusting, it's life, it's the
way you are, you are what you are that's just the way life is and I
think maybe I'm just open minded"* (Faye).

Faye uses this example to try to describe her open minded approach to life
whilst describing other people's reactions to something they did not understand.
Faye and Andrea both told stories that emphasised the need for acceptance of
variance in society. On reflection I thought that parents believed other people
would either struggle to understand AG or that the idea of AG would leave them
feeling uncomfortable. If other people were left feeling awkward after learning
about AG, the parents inferred that this would impact on how people behaved
towards their child.

The way in which mothers and fathers talked about gender was slightly
different and is explored in more detail in the following section.

8.2(c) Foundational story (3): Stories about differences between mothers' and fathers' understanding of AG

Although there were fewer fathers in my study they made a substantial contribution. Where possible I have explored both the differences and similarities between mothers' and fathers' perspectives. The need to understand information was important for both parents. Often fathers demonstrated they did this much more quickly than their partners. Gina believed that Frank was more able to "*take things in*" and he became "*more knowledgeable*" because he "*grasped things a lot quicker*". Frank often found himself "*explaining again*" so Gina could understand. Frank's ability to understand much more quickly meant he could repeat information to Gina. Once confident in her understanding Gina would then lead the consultations with medical staff.

Fathers also reflected that they contained their own emotions in order to be supportive. Oliver summed the difference between how he and his wife managed their emotions "*she just let its out which is not a bad thing, whereas I'll store it up*". He believed that his role was one of protection towards his wife making "*sure she was ok*" achieved by "*bottling up*" his emotions.

Brian worried about staying in hospital to look after Bobby after his genital surgery as mothers "*might be uneasy about a bloke stopping over with their kid*". Both Andrea and Brian shared the responsibility of caring for their children at home and in hospital. The only other father who would stay with his child in hospital was Oliver, so mothers were often the main carers for their child in hospital post surgery. However, this did not stop fathers from worrying about their child's health. Harry recalled that Joanne would be in hospital "*what felt like every February*" and he was "*always worried*" when she "*got an infection*".

Often fathers were more pragmatic than mothers and considered each situation or new piece of news as a separate event which could be managed sequentially, as Oliver suggested "*each step at a time*". Although fathers initially found themselves angry, this emotion was always directed towards the "*incompetence of staff*" rather than their child's AG. Like mothers, fathers described feelings of shock. However, for fathers this emotion quickly passed as Oliver explained "*it doesn't matter how traumatic it is for the parent, you've*

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got to get over that". Fathers appeared to keep their experiences grounded in every day events. David recalled the events on the evening Chloe went for an emergency caesarean section. He recalled his experiences and the chance meeting with the same man he had played tennis with earlier that evening in the delivery waiting room. His story below includes several elements of everyday life that seem to ground his story:

> *"So one night I got the call 'you better get here near now'. It was strange really because I had played tennis that evening, Chloe used to go mad when I was out enjoying me self and she was stuck in hospital, 'what do you want me to do mope?' So after the game finished the guy from the club just went and I'm thinking 'miserable sod' could have stayed for a pint or whatever and then I got a phone call myself about an hour later and went to hospital and was stuck in a waiting room. She was rushed out, she was already on her way down by the time I got there so I met her in the corridor. So I went to wait in the waiting room and that's when I spotted the other fella, the 'miserable sod' sitting on the other side and thought that's a bit of a coincidence" (David).*

The language he used, *"mope"* and *"miserable"* seemed to be a reflection of unhappy feelings he may have been trying to avoid in relation to Chloe's pregnancy being problematic.

Fathers like mothers worried about their child's future. David was concerned about his son's voiding *"he can't keep going to the toilet and pulling his pants down"* and the implication this had for singling him out. For David being singled out reinforced Edward's difference and put him at risk, which reinforced David's need to protect his son. Brian described how he would *"move heaven and earth"* to protect Bobby and if for some reason in the future *"he started getting terrible torment, we'd up sticks and go, we wouldn't hesitate".* Tentatively, I recognised that although fathers worried about their child's future and their need for information about AG, none of their stories gave in-depth accounts of sharing information like the mothers' stories did.

Fathers often used humour as a strategy to help themselves and their children cope, as David described pre genital surgery *"we used to joke about him needing a puncture repair kit".* Fathers' stories also included elements of highs and lows as Oliver suggested *"you have your good times, you have your bad times, but you just get on with it".* Fathers told how necessary they

considered the skill of being 'level-headed' was in managing day-to-day life. Fathers' pragmatic attitude appeared to help them manage the complexity of their child's AG.

Stories about gender and gender typical behaviour prompted parents to talk about their child's early genital surgery. All the mothers and fathers in my study had agreed to genital reconstructive surgeries for their child.

8.3 Aggregate story (2): Stories about the place early surgery had in parents' lives

Early surgery played a significant and important part in helping parents cope with their child's initial diagnosis of AG. Parents appeared to believe that early surgery would not be remembered by their child and I wondered if this served as a form of protection. Genital surgery was also important as it substituted the uncertainty parents' experienced at their child's birth with a firm sex and gender for their child which for some parents helped them relate to their child.

8.3(a) Foundational story (1): Stories about early surgical intervention and childhood memories

Surgery was discussed by all parents as this was the initial focus of my study. As described earlier Rose, Helen, Tara and Barry would have died if they had not had surgery. All of the children had had genital reconstructive surgeries that have previously been described as 'cosmetic'. A hugely significant event for all of the parents was the concern about their child surviving surgery. However, Gina recalled surgery as something that "had to happen". Gina also considered that early surgery was "a lot easier" when Helen was a baby because she "just brought her in" and Helen did not cause a fuss about "coming into hospital". Gina's fear was not the surgery itself but the risk that she saw accompanying it. She described that she "just couldn't cope with ever losing her" and the distress of Helen being upset about "going to theatre". Sian recalled that surgical events happened so quickly that she had "little time to think". Like several parents Chloe considered that "it was not good [for Edward] to look different" and she needed "some clever surgeon to fix it". Other parents also used the word "fix" to describe their interpretation of the role of surgery. All of the parents accepted or

believed in the importance of the initial surgery. For some parents early surgery resulted in a sense of relief from the anxiety of sex uncertainty at birth. However, some of the subsequent or multiple surgeries were questioned by a couple of parents. Andrea's view of Bobby's surgery changed over time as she learnt more about his condition and as he began to voice his own opinion. This resulted in Andrea and Brian having opposing views on both the timing and multiplicity of surgeries for Bobby. Andrea viewed multiple surgeries as "*cosmetic*" and therefore these could wait while Brian considered that surgery was "*corrective*" and needed to be completed.

Many parents remembered being told of the importance of early surgery. Sian agreed with medical staff, reconstructive surgery was best "*tried early*" so that Tara would be viewed as "*normal*". However, once parents had made a decision for surgery, waiting for up to 18-36 months or surgery being cancelled resulted in them feeling frustrated. I sensed that this conflict between what they had been told, and what actually happened, was an ongoing source of anxiety for parents.

Several parents wanted their child to achieve their development milestones, such as potty training, in line with their peers. Christine recalled being asked directly when Daniel was very small if she wanted him to "*wee from the end of his willy*", this she linked to him being able to stand to void and it became her motivating factor for early surgery. It seems that when surgery did not happen at the time that was initially suggested parents began to feel out of control of the situation and some became distressed. As Chloe reiterated she both wanted and had agreed to early surgery and felt irritated at having to wait for something she "*couldn't fix*". Waiting for Maria also put "*an awful strain on my relationship with my husband*" while for others waiting restricted family events such as holidays.

All the parents' stories to a greater or lesser degree considered that early surgery resulted in their child not remembering traumatic experiences as Faye described very well:

> "*You just took her in, you had the surgery, you dealt with it, you take them home. They're 1, they're 2, they're 3 years old, they don't know any different*" (Faye).

Anne's story is similar to Faye's and other parents but she also focused on the social aspect of early surgery and how this would allow Barry to go to school and no one would know his history of AG:

> "I just felt it needed to be repaired he was going to the toilet and everything so I suppose it was more cosmetic. I knew it [ambiguous genitalia] wasn't having an effect on his health, apart from the reflux he had. But cosmetically he wasn't weeing from the right place, and I suppose in the long term I was thinking well when he starts going to the loo, not in his nappy you know. And going to nursery and things like that, that is when it would be noticeable. So I was quite glad that he had it done when he did. Because by the time that he did come out of nappies and started going to the loo, joined nursery and school he was standing there like the others" (Anne).

Many parents believed early surgery would allow their child to integrate more easily at school as they would appear physically the same as their peers. Surgery also reduced the difference between AG and what parents believed were 'normal genitalia' and as suggested previously being different was something parents wished to protect their child from. Therefore some parents had a sense of urgency towards genital surgery and changes to the timing of surgery reinforced the parents' sense that they were not in control. This was difficult for some parents to tolerate. Early surgery also had the perceived benefit of the child being less likely to remember the possibly traumatic or unhappy experience of an operation. Parents believed that the surgery made their child's genitalia "look normal" and I wondered if the early surgery fulfilled the promise offered by surgeons as Anne described "they said it'll just look like a normal little boy". Early surgery seemed to be essential for some parents who were reminded about their child's AG when they were bathing their child or changing their child's nappy. Parenting a child with AG was described as a complex experience by parents as they have emotional challenges to face and address. Against their own emotions, such as shock and anxiety, they also had to deal with other people's understanding of AG built on their own social interpretation of sex and gender. Parents had to develop a parenting style and attitude that they believed would safeguard their child both in the present and future. Parents had to begin to explore ways in which they could bring all these elements together in a harmonious way which would facilitate coping and

adaptation. How parents tried to achieve this is described in greater detail in the following aggregate story and foundational stories.

8.4 Aggregate story (3): Parenting a child with ambiguous genitalia stories

The transition to parenthood necessitates changes in both life and working patterns for the majority of people. Most people who have children will experience a range of emotions in relation to their child and their likely future together. Nine of the parents were new mothers or fathers whilst the remaining six had had one or more other children. Regardless of whether they were a first time or an experienced parent, all mothers and fathers had to reconcile the challenge to their fundamental belief that a child is either a boy or girl.

Four mothers questioned whether their actions in pregnancy had caused their child's AG. As Anne remembered *"saying to somebody in the hospital, 'is it [AG] anything I've done?'"*. These thoughts were only considered fleetingly as Anne described you ask *"stupid questions"* when you are under pressure or tired. Likewise, Sian considered herself *"quite healthy"* during her pregnancy which resulted in her briefly thinking that Tara's cloaca was her being *"punished"* for being *"bad somewhere"*. All of the parents initially wanted a reason for why their child had AG. This is discussed in chapter 9.4 in relation to understanding and decision making.

All parents had testing experiences of regular hospital check-ups and surgical admissions. This resulted in them learning to become *"experts"* in knowing when their child was sick or when something *"wasn't right"*, which they referred to as their *"intuition"*.

8.4(a) Foundational story (1): Searching for normality stories

Parents described intense reactions immediately following the discovery of their child's AG. These reactions, described by all the parents, included shock and worry. Parents' emotional reactions were perceived initially by them as negative and they heightened their anxiety which in turn intensified their loss of normality. Imogene felt *"blown away"*, while for Oliver *"it was like a bomb had gone off"*. The use of such words and phrases seemed to be powerful ways in which the parents were trying to communicate the shock and devastation they felt initially. In my study parents encountered a significant deviation from what

they anticipated were 'normal' events following the birth of a child. Christine talked about her desire for a perfect child, whilst pointing out that she did not love Daniel any less:

> "Well everyone wants their, 'oh this sounds awful saying this because I know he's mine and I love him' a perfect child, don't they? They don't want all the complications that go with something like this" (Christine).

Chloe like several other parents found being a parent of a child with AG hard. She made a conscious decision to hide her feelings from herself and society, thinking about how she could "make it not as bad as it is". Appearing to cope was very important as Maria, Anne and Faye described "how they put a brave face on". Sometimes parents found strength from their child as Sian explained:

> "I just had to cope with it [cloaca] for her sake. There were many times I want to lock myself in the bedroom and not come out at all. But I have to do it for Tara, and then when I see her I get days where I'm like, I can't cope, I can't cope. Then I'll see her smiling or she does something, she just makes me laugh and I think if she can cope with it, what am I worrying for" (Sian).

For some parents coping was incredibly difficult and impacted on thoughts, feelings and ultimately their own behaviour. Chloe talked negatively about the "panic attacks" she believed were a result of the "trauma" of both Edward's AG and his early health needs. She continued talking about how her anxiety impacted on her parenting; she recalled how "possessive" she became towards Edward during his infancy and that she "wouldn't let anybody near him".

Although parents were coping with AG, ordinary day-to-day challenges continued. In order to maintain the sense of normality they encouraged their child to engage in every day activities, such as swimming, sports, cubs, piano and other out of school activities. When the child's activity for example swimming, became compromised by their genital ambiguity parents would try to devise strategies to help their children disguise their differences. David, Maria and Anne all told stories about how they protected their son's "embarrassment" when getting changed for swimming lessons. Kerran explained that she was becoming aware that Leah's large clitoris was "just noticeable" which worried her as other people may notice or comment. Therefore 'normal' social activities

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changed for some families as a result of their child's awareness of their genital ambiguity. Parents had to find ways in which they could adapt and bring together their initially intense emotional reactions about AG and day-to-day life. The three emotional elements of shock, protection and anxiety were consistent across parents' stories. Parents tried different strategies to alleviate the intensity of these core emotional elements which in turn allowed them to cope with AG.

As parents began to adapt to having a child with AG and they felt they were coping they considered that pity was unhelpful. Pity reminded them of the negative aspects of having a child with AG rather than considering the child as an individual. Sadly for Oliver it was the district nurse who, through her own feelings about AG, questioned his coping in a manner he believed was akin to pity:

> *"The district nurse would go, 'Oh, I don't know how you cope?' I said, 'Sorry...No, I want the support, not how do you cope, if you're going to say that, don't come and visit'. I can do without that. I don't want [them] to come and visit for half an hour and bring me down on the floor, when we're nice and on an even keel" (Oliver).*

Parents believed that regular encounters with healthcare professionals and visits to hospital for a child were not a 'normal' part of childhood. They experienced regular hospital appointments, admissions and surgeries for their child an additional challenge to their coping. Several parents considered hospital attendances as disruptive and at times unhelpful. Kerran described the negative impact of hospital appointments on family and work life;

> *"A doctor's appointment means I have to take time off work to go then I'm spending time after work catching up, it's like where do you fit everything in" (Kerran).*

Professionals' attitudes challenged parent's ability to cope. The relationship between parents and professionals is discussed in greater detail in chapter 9.

8.4(b) Foundational story (2): Stories about parents' emotional bonds to their child

Eight parents spoke about the impact their child's ambiguous genitalia had on their relationship and emotional bond with their child. Faye talked about how Gabby's AG and its management were the significant factor that hindered her bonding with Gabby:

> "I never ever felt maternal with Gabby I never felt like I'd had a baby, it was only after the surgery that I felt like I'd wake up and she was 7 months old. The first six months of her life it was like you were, it was like looking after somebody else's child cos you can't form a bond a proper bond with a baby that people are just fiddling 'down there with' constantly" (Faye).

"Fiddling with" was a phrase adopted by a few parents to describe 'what people did' to their child's genitalia. Clinical inspection of their child's genitalia by healthcare professionals reinforced the child's AG for parents and seemed to make them feel vulnerable. I speculate that this amplifies the "freak" aspect of AG which in turn impacts on how they felt towards their child. Other issues also complicated bonding, for Chloe it was Edward's prematurity he was nursed in an incubator which limited the early physical contact she had with him. Both separation and bonding were remembered by Sian as upsetting because she did not "get to hold Tara" as she was taken straight to special care. Anne had held Barry briefly but he then went to special care. She found this difficult as she had hoped to keep him with her and breastfeed, which she felt she could not do since he was initially not allowed to take any oral fluids. Imogene talked about developing an emotional bond with Joanne very quickly and the bond was "very, very strong". Joanne's ambiguity at birth and the initial sex assignment of male did not influence Imogene's bonding because she "always knew she was a girl". Gina told a story and Frank agreed that this captured their growing bond towards Helen as well as Helen's evolving awareness towards them as her parents:

> "I suppose although you are bonding [when they're born] and you are close it's totally different. When I think how I felt when she was six months old having surgery to how I would feel now because she's got her personality, her character. We know her. We would find surgery much more difficult now" (Gina).

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All parents recognised they developed an increased sense of protectiveness towards their child. Imogene talked about how she was "really protective of her" in both an emotional and physical sense. She thought that protectiveness was "a very good word" that described some of her ongoing actions, such as "not telling people" about Joanne's AG and "keeping her close by". Chloe spoke about how her maternal feelings made her protection of Edward go into "overdrive". Anne, Faye, Kerran and Maria all recalled stories in which their other children also became "very protective" and "looked out for" the child born with ambiguous genitalia, although they were unaware of their parents' concerns about their siblings sex or gender. Parents paid attention to events where they felt their child was extremely vulnerable and sought to protect their child from them if possible. These events were disclosure of AG to other people or circumstances where their infant or young child could be undressed, such as going to stay with relatives or friends. Sometimes parents questioned themselves about what was 'best' for their child. David talked very movingly about the struggle he faced in relation to his "just trying to protect him" and not knowing "how do I protect Edward, really". Anne talked about surgery as a form of protection, which was echoed to a greater or lesser degree by all of the parents. She believed that if Barry did not have surgery he was at risk:

> "It's when they get older with their friends in school that they do compare and I didn't want him to stand out from the crowd or be embarrassed or feel that he was different" (Anne).

She continued to talk about why surgery was an important means by which she protected Barry and aimed to ensure some degree of conformity:

> "Yes, you protect your children don't you and you don't want them to be picked on for having something wrong with them. I mean, physically to look at him, you would never ever think he had anything. There are many people that don't know what he's gone through. Because he's always out playing football and he's scared of nothing he will do everything, and have a go at every activity.
> I wouldn't want him to be singled out as being different, just because his willy doesn't look right. That can start the bullying thing, so I suppose in a way, you just want them to be the same as everyone else and you're protecting them, really" (Anne).

Clearly Anne was very proud of Barry's achievements and his confidence. Her decision not to disclose information in her opinion had a positive outcome. She also believed that genital surgery had helped her to protect Barry by establishing his genitalia as male. He was therefore able to join in gender typical activities which resulted in him not being *"singled out"*. It was not clear whether she attributed some of his confidence and his ability to participate in activities to his genitalia having a similar appearance to his friends.

The emotional bonds that parents built with their child were, for some, affected by AG. Reconstructive genital surgeries had the capacity to be able to positively influence parents' perceptions of their child's genitalia. This resulted in some parents being able to build stronger relationships with their child. In the following section keystone story section I will both summarise and synthesises the aggregate and foundational stories from sections 8.2, 8.3 and 8.4.

8.5 Summary and synthesis of keystone story 2: Parents' stories about their experiences of being a parent of a child with AG in a social world

Parents talked empathically about the importance of having a clear biological sex for their child that matched gender. Blood tests which aimed to identify chromosomes were keenly awaited by five parents although none of the other parents spoke about chromosome testing. Matching external genitalia to any internal reproductive organs was important for all parents. Three parents recalled being told that their child had internal structures that were to some degree both female and male. This caused additional uncertainty and anxiety for one mother as she described the news as a *"double whammy"*. For all parents surgery aligned their child's genitalia (sex) and gender by providing what Maria described as a *"clear visual sex"*. The use of repeated language such as *"same"* and *"like their friends"* highlights the importance parents place on the need for their child's genitalia to conform to either male or female. The socially and morally complex issue of gender was closely tied to parent's desire to do what they perceived as right for their child, which included genital surgery.

As parents became more informed and they began to adjust to AG they focused on giving other people very clear gender signals about their child. How the child

was dressed and gender typical behaviour became very important in uniting and strengthening the child's sex and gender. Once parents were clear about their child's sex they adopted common gender typical patterns of parenting. They encouraged their child to take part in common gender typical hobbies; this further secured their child's sex and gender which reduced uncertainty in parents. Often parents were pragmatic about coping and how they managed to achieve this. When they were able to cope this was viewed as a positive experience by all parents; some felt they managed to achieve this in a short period of time while for others adjustment took longer.

Some parents talked about how they *"were a good team"* supporting each other and finding a balance between them. This was also true for parents who gained support from a family member or close friend. Although there were times when some parents did not agree they managed to negotiate this between themselves. Parents were often very proud of their child; and this could be seen in their pride in achievements in hobbies such as swimming and football. Parents tried, where possible for day-to-day life to continue although there were occasional challenges such as sleeping out at friends or relatives. For Kerran she could not get Leah to go and *"even sleep at my sister's house"* which Kerran considered was *"just ridiculous"*. She believed this was due to her very close bond to Leah which she described as *"the umbilical cord was never cut with us"*. Evidently some parents developed very strong emotional bonds with their child yet for other parents bonding took sometime to strengthen as a result of the AG.

Clearly parents made decisions about how they chose to live with their child's AG. Their stories about being a parent of a child with AG gave me insight into the social and cultural foundations on which parents based their strong protective emotions. Positive experiences of having a child with AG happened for some parents as Anne recalled how her experience with Barry had *"enriched her life somehow"*.

Parents had to actively address their concerns in the actual and potentially complex social situations which resulted from AG. Parents had to bring

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together their emotional reactions to AG, which were shock and anxiety and their desire to protect their child. These three core elements had to be built into their relationships with their child and also their interactions within their social world. They believed they had to try and find strength from themselves, and as parents, they would then be able to support their child's gender and meet their child's health and emotional needs. Parents believed that typical boy or girl gender socialisation would support their child's development and also limit uncertainty. Therefore, gender typical activities and behaviour were positively reinforced. Parents had to overcome the dissonance between AG and a clear single sex and gender for their child. Therefore parents had to bring together, in a harmonious way, their belief in their child's gender and how this could fit in their social world. They developed bonds with their child despite the uncertainty of AG. Genital surgeries offered the prospect of achieving a sense of harmony between their child's sex and gender, which in turn upheld parents' protective instincts.

CHAPTER 9: KEYSTONE STORY 3 - PARENTS' STORIES ABOUT HEALTHCARE PROFESSIONALS

9.1 Introduction

In this chapter I illustrate the findings under the third keystone story along with the three aggregate stories which are supported by five foundational stories Parents' stories about their interaction with healthcare professionals were evident throughout all their interviews. They focused on specific key events following their experiences of having a child with AG, their need for information and the importance of good communication. Parents also explored how they developed trust in healthcare professionals and the significance of this, and finally their role in decision making. This chapter is divided into four sections.

The first section (9.2) is the aggregate story of information, understanding and communication. This aggregate story comprises of two foundational stories. The first focused on how information was shared with parents and how their information needs changed over time. Parents also described the difficulties they faced in developing understanding and the consequences this had for them and their child. The second foundational story explores the stories parents told about how they communicated with healthcare professionals. As parents' confidence grew they began to engage in more directive and questioning communication with healthcare professionals about their child's AG and surgeries. Stories of learning about AG as a condition and the usefulness of the information they received focused parents' stories on their relationships with healthcare professionals.

The second section (9.3) is the aggregate story of trust and healthcare professionals. The foundational story that supports this aggregate story focuses on the reciprocal nature of trust between parents and healthcare professionals and the influence and impact this has on parents' decision making.

The final aggregate story (9.4) focuses on parents' decision making in relation to their child's reconstructive genital surgeries. This aggregate story comprises

of two foundational stories. The first describes parents' roles in decision making and how this fitted with their responsibilities of being a parent of a child with AG. The impact of genital reconstructive surgery is discussed together with the considerable emotional strain of surgery. The second foundational story explores parents' search for a balance between understanding, surgical action and their ownership of decision making in relation to their child's future.

The final section (9.5) is the keystone story and comprises of the summary of and synthesis of the foundational and aggregate stories from section 9.2, 9.3 and section 9.4.

Diagram 9.1 is a visual representation of the foundational stories, aggregate and keystone stories brought together. The parents' stories are presented in the text in italic font.

Diagram 9.1 Keystone story: Parents' stories about healthcare professionals

Parents' stories about healthcare professionals

Stories about information, understanding and communication

Stories about decision making and surgery

Stories about information about AG

Stories about understanding surgeries

Stories about communication with healthcare professionals

Stories about trust and healthcare professionals

Decision making and responsibility stories

Stories about the reciprocal nature of trust between parents and healthcare professionals

9.2 Aggregate story (1): Stories about information, understanding and communication

All parents regardless of their previous experience or knowledge had a need for information about AG. They wanted, to a greater or lesser degree, to gather and process information so that their experiences were meaningful and they understood the consequences of AG. Every parent had access to doctors, nurses or midwives and for the majority this access was available quickly after their child's birth. Parents sought guidance and expert opinion from these teams which resulted in an initial reliance on healthcare professionals as the main source of information. Some parents had difficulties with the language healthcare professionals used and in turn they struggled to conceptualise the information in a manner which made sense to them. For many parents remembering and recalling the complex information they had been told was difficult. There were situations in which parents talked about not having enough information and this directly impacted on what they understood. Occasionally parents spoke about how they struggled when healthcare professionals gave contradictory information. Finally a few parents had given some thought to how they could become empowered when communicating with healthcare professionals thereby making sure they felt they were being listened to.

9.2(a) Foundational story (1): Stories about information about AG

The environment in which information was delivered had the potential to impact on parents understanding. Both Christine and Faye talked about how being given information when they were alone increased their anxiety. They spoke about being *"put away in"* or *"shoved"* in a side room, although they graphically described being moved away and isolated they did not talk about feeling ashamed of themselves or their child. However, for Maria and Chloe being nursed in a busy environment with new mothers and babies caused additional anxiety. Maria had wanted *"some sort of privacy"* but it was also essential that you had *"someone to come and just give a little bit of support"*. For Chloe it was the other babies' crying that upset her as Edward was nursed in a special care unit and the questions from *"the mother in the next bed"* which she did not want to answer.

Parents wanted information. Anne spoke about how it was *"so important to know and be explained to"* in relation to AG. Yet all the parents in some aspect agreed with Christine's comments that in the beginning *"it was just so complicated, it didn't make sense at all"*.

Once parents knew their child would survive they wanted to see an experienced professional. Maria recalled a story in which she felt fortunate to see someone very quickly who specialized in managing AG:

> *"Luckily the doctor was at the clinic that day in our hospital, because I don't know how long we would have waited to actually see someone" (Maria).*

Healthcare professionals were, to begin with, the main source of information. Parents had an expectation of how they should be given information about AG once the initial shock had settled. Faye talked about how *"nothing was explained"* by healthcare professionals no one asked her if she *"was alright"*; the sex uncertainty, confusion and a lack of information lessened her *"maternal feelings"* towards Gabby. For Maria it was very important that any HCP she engaged with demonstrated sensitivity as she wanted to be *"treat[ed] with kid gloves in the beginning"*. David talked about how one doctor shared sensitive information about Edward as he *"walked down the corridor"*. This memory encouraged David to talk about his feelings and how *"angry"* he was that information had been given, in what he believed, was an unacceptable setting. Although he acknowledged that the doctor had seen the chance meeting in the corridor as an opportunity to share information, David believed the doctor gave no thought to the family's privacy. David was one of six parents who spoke at some length about the importance of privacy. Harry, Imogene, Maria, Gina and Chloe also considered that *"privacy"* was essential when healthcare professionals were communicating information. A poor attempt to achieve privacy was described by Chloe:

> *"In this room where there was six beds, they'd come up to tell me [the doctor], you know, and were like 'oh we haven't had the results back of the chromosome test' and all these mothers are [wondering what's going on]. Why bother pulling the curtains round?" (Chloe).*

Information shared early on with parents was governed by when the healthcare professionals could visit them on the ward. By giving information in an unplanned manner healthcare professionals failed to recognise that parents needed time to prepare for what they might hear. This reinforced the parents perceived lack of control they had in relation to caring for their child and seemed to leave them feeling vulnerable. A feeling of intimidation was also emphasized in Andrea's and Brian's story when they talked about a particular clinic visit. Brian described how they had *"gone to clinic and then there was about 8 or 9 doctors or whatever sitting in a circle"*; this experience became *"the only thing I found daunting throughout the whole lot"*. For Brian this negative experience had a profound impact on his relationship towards one doctor. Brian believed that this doctor should have limited the number of staff in clinic to ensure that Brian was not left feeling threatened or vulnerable.

The degree of information required by parents was variable and for a few this changed over time. In particular Anne was eager to learn *"everything"* she could. Others, like Oliver, wanted information on a *"need to know"* basis. Although he went on to say *"the wheel starts rolling, it gets faster and faster, and you think, hang on"*. It seems that for some parents the speed at which they heard new information became overwhelming and could have been more manageable when taken in smaller sections or, as Oliver suggested, when people *"get to that stage"*. Furthermore based on my own reflections on this, I wonder if for Oliver and for the other parents increasing amounts of information meant they were confronted with the limits of their own knowledge. Even when parents had a greater degree of information and knowledge this did not necessarily impart a greater ability to predict or control their child's future. Although Maria recognised that some parents wanted more information than others she linked the notion of an informed parent being a *"happy"* parent:

> *"I know some parents are on a 'need to know basis' and don't want to know but surely there's a lot more like me who want to know? The what, where, when, how, you know, what's gonna happen and I think if then parents are happy and they can support a child better"*
> *(Maria)*.

My understanding of the way in which Maria used happy she was also describing parents' adjustment to the AG which was made possible by sharing information. Information allowed parents to develop understanding and was therefore valuable.

For Kerran there was a period of transition where her usual information seeking behaviour was suspended. She considered that her lack of knowledge initially *"didn't really bother"* her for *"the first couple of years"*. Kerran felt she came to a point where she felt comfortable to *"see in the next few years what happens"* with Leah. During these years of transition Kerran acknowledged that her thoughts about Leah's future changed *"ask me in 6 months time I'm sure it'll be different, ask again in a year's time I'm sure [my thinking] will be different again"*. Kerran was hopeful that the events that happened during this transitional time would help her to move forward to a point where she would be supporting Leah in making decisions *"for herself"*.

The language used by healthcare professionals to share information was very important as this apparently either hindered or enhanced parents' understanding. If parents believed that healthcare professionals could not *"explain anything"* then they lost confidence in them as professionals. The use of complex and difficult medical terms was hard for parents as Kerran described when she said she felt that Leah's doctor *"just comes out with medical jargon continuously"*. For Anne, Chloe, Gina and Sian the doctor *"drew diagrams"* which were helpful, although these related to the surgical aspects of AG.

Recalling and remembering information was difficult for some parents. Faye spoke about her experience in the out-patients department; these appointments were the only opportunity in which she had the chance to meet with healthcare professionals who understood AG. She felt *"bombarded with an awful lot of stuff in a very short space of time"* and this impacted on her ability to recall information as she explained *"someone would say 'oh how did you go on'? and I would have to say I don't know"*. Faye recalled information slowly *"for weeks after, you keep remembering little bits of the conversation, you know"*. She had even considered alternative methods to help her remember in the future:

*"I should take a take a tape recorder with me really to these places
and get somebody to explain it to me when I come home" (Faye).*

This demonstrates the complexity of how healthcare professionals give
information but may not check parents' understanding. Parents face enormous
difficulties at first in taking the information away and often have to *"get
someone"* to help them understand. This is often complicated by the fact
parents have not disclosed AG to other people. All the parents talked about
how they eventually found someone who *"explained quite well"* but for some this
was several years after their child was born.

For some parents healthcare professionals did not provide enough
information. Support groups and also the internet were another area from
which they gained information. Harry and Imogene described the
importance of access to information via the internet while Sian considered
that information *"learnt from other [parents] going through a similar
situation"* had been helpful.

Sharing information was considered by several parents as a two way
process. Over time Andrea and Brian gathered more information about
Bobby's AG, from the internet and global contacts with other parents and
they were happy to share this with any healthcare professionals that were
interested. Faye talked about how her family experience of AG gave her
insight into Gabby's AG *"I was trying to explain to this doctor"*, however the
doctor dismissed her saying *"no, no you're wrong"* which left Faye feeling
angry. Being listened to by a healthcare professional was very important
for all parents.

The parents in my study undoubtedly had to gather, process, organise and
interpret the information they were given or sought for themselves. Parents had
to be able to develop an ongoing dialogue with a healthcare professional at first
this was predominantly a medical doctor or surgeon. The importance of
communication between healthcare professionals was also essential as many
of the children with AG had more than one doctor. When parents discovered
something had been communicated in a manner they were dissatisfied with this

caused additional anxiety. Christine describes the early miscommunication between nursing and medical staff:

"We were waiting for the tests to come back and one of the nurses said, 'have you got a name for her' and I said 'we can't name her yet because it don't'. So she goes, 'yes, it's a girl, you've got a girl', puts the pink band on and name. I phoned me mum up to say it was a girl" (Christine).

However, the chromosome tests were not back and the nurse had incorrectly informed Christine she had a girl when in fact the results received a day later suggested that she had a boy. David talked about how in clinic Edward "used to get different guys, doctors" and that they "changed a few times" and this caused some confusion and David and Chloe had to repeat their story, which was difficult for them. Faye described how different doctors suggested different ages for Gabby's surgery:

"I don't remember which doctors have said what to me, but I remember bits of conversations where they've said, 'right between 8 and 9, they'll do it between 8 and 9' and then it was 'between 9 and 11' and then it was 'between 10 and 13'. I can't remember who it was, you know I've had conversations with so many doctors I couldn't tell" (Faye).

She was frustrated that the communication within the surgical team and then with her was not consistent. Communication between healthcare professionals and parents is important yet often problematic. Despite this parents need to feel confident asking questions about their child's AG and surgeries. The following foundational story explores the communication approaches used by healthcare professionals. Also this foundational story explores the challenges that parents described in relation to communication, understanding and questioning healthcare professionals.

9.2(b) Foundational story (2): Stories about communication with healthcare professionals

How parents and healthcare professionals communicated was predominantly verbal but as mentioned earlier some doctors drew diagrams. At times parents worried that they were asking too many questions, Anne became concerned that the doctors would become "fed up with" her as she "wanted to know

everything" and asked "question after question". Some parents, like Christine, did not "know where to start" when asking questions, while Sian struggled to marry "what I believe and what I think I've heard consultants say". Faye talked about how she had to "stay alert" when listening to doctors because she really had to think "about what they're saying". Sometimes healthcare professionals were very good at communicating verbally with parents as Sian explained "he's always talking to me, he doesn't overlook me and he listens".

For a couple of parents written communication reinforced their knowledge and understanding about their child's AG, which for Kerran was a positive experience. Kerran had seen of a copy of a letter to her GP about Leah on which was "the actual statement of what her condition was, I actually never knew until that letter". For Chloe a healthcare professional had written in Edwards infant development book and she spoke about how intrusive she found this:

> "I don't let him see this book it's most unpleasant. She wrote it in, I wasn't very happy about it, but I thought well maybe they have to. Premature, ambiguous genitalia, it's written on that page there, but it's also written actually in here as well. Well yeah, I really should rip the page out get rid of it" (Chloe).

Sadly for Edward there were many positive entries in the book that he would be unable to see unless she destroyed the pages that specifically mentioned his AG. Chloe was angry that she had not been asked if AG could be written in his book. Also she believed she was expected to share the book with other healthcare professionals which resulted in Chloe not being able to stop Edwards' AG being disclosed.

Sian recalled one healthcare professional "draw[ing] me a picture and say[ing] that's 'what it is', and I said 'oh' I get it now". None of the parents spoke about being given an information book or pictures or diagrams after the discovery of AG.

The importance of non-verbal communication in establishing a relaxed relationship between the healthcare professional and the parent was mentioned

by a few parents. Gina described more generally how she just *"took to"* the doctor who usually looked after Helen because of his *"nice manner"* towards her. Good communication helped parents feel at ease and lessened their anxiety. However, not all parents told stories about positive experiences. Anne recalled one healthcare professionals's *"bed side manner [as] atrocious"* as she had felt the professional *"couldn't wait to get away"*. Faye recalled the *"door flying open"* constantly as healthcare professionals came in and out, carrying out tests on Gabby. Kerran spoke about being *"watched"* in clinic and how this made her feel *"intimidated"*. Gina recalled how one healthcare professional who was standing in for her usual doctor would undress Helen, even when Helen had said *"no"*. This professional would not look at either Gina or Helen and would carry on *"no matter what"*. Oliver talked about Rose being in hospital for surgery and how he was not proud when he had lost lost his temper because *"no one was listening to him"* and he *"had to be removed from the cubicle"*. When poor communication did exist some parents would find excuses for healthcare professionals such as they *"just didn't have a clue"* or were *"overworked"* or they did not have *"enough time to sit down"*. On reflection I wondered why some parents were more tolerant than other parents of poor communication from healthcare professionals. Perhaps their leniency related to their belief that circumstances existed beyond the healthcare professionals' control, such as not being a skilled specialist in AG and these had led to poor communication.

Sometimes the child's test or biopsy results were a trigger for communication between parents and healthcare professionals. Three parents had the opportunity to watch a video clip of their child's surgeries. These clips were chosen by the healthcare professional to enable parents to visualise the nature of their child's surgical procedure. Andrea watched a short clip of Bobby's surgery and only at this point did she become aware that 'female like' tissue had been removed, this filled her with sadness and regret. She talked about how *"medical science and the way it's progressing"* could have offered Bobby a different choice in the future in relation to managing his mix of male and female. Watching the video had helped her understand very clearly the complex nature of Bobby's AG, she *"thought it was a malformed undescended testicle"*. Andrea

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did not recall *"having the conversation"* about the possibility of female-like tissue before surgery; hence she was not prepared and the news came as a shock although she was not critical of the healthcare professional.

Seven of the parents supplemented their knowledge with information via the internet with varying degrees of success. Kerran made a conscious decision to try to find more about Leah's condition from web based resources and online self help groups and this helped her eventually begin to *"understand"* Leah's condition. Sian *"tried looking on the internet"* for written information to expand her understanding. She had wanted to be clear about *"what they're gonna do"* to Tara and also consider how she and Tara were *"gonna feel"* after surgery but *"there was nothing"* that she could find. Harry and Imogene got the diagnosis *"written down and tried to find out about it but then there was hardly anything about it"*. Maria found American information on the internet but this did not meet her needs; this inadequacy led her to use the internet as a vehicle to seek and *"share information"* with other parents.

Verbal and non verbal communications were important for parents since these were the dominant approaches utilized by healthcare professionals to share information. Parents' understanding was only explored by healthcare professionals when they asked specific questions about their child's AG. None of the parents spoke about talking with a psychologist as part of the HCP team that cared for their child. Many parents spoke about the significance of their trust in healthcare professionals and how they had to find this trust for themselves. The following aggregate story explores parents' trust and honesty in healthcare professionals.

9.3 Aggregate story (2): Stories about trust and healthcare professionals

The degree to which parents trusted healthcare professionals was a measure of their belief in the professionals' honesty and clinical competence. For some parents there was an immediate need to trust the actions of healthcare professionals in order to save their child's life. This resulted in a voluntary transfer of the responsibility for the child from the parents to the healthcare professionals. This was possible because parents trusted healthcare

professionals to deliver the care needed to save their child's life. However, for some parents a time lag existed between the immediate need to trust healthcare professionals and parents demonstrating trusting behaviour towards them.

9.3(a) Foundational story (1): Stories about the reciprocal nature of trust between parents and healthcare professionals.

Parents considered that for trust to be established and nurtured honest information needed to be communicated clearly by healthcare professionals. Approachability, consistency and sincerity were characteristics which parents described as integral to a supportive parent / professional relationship. Gina explained how hard making a decision to trust a doctor was after only meeting him briefly:

> "I found him very nice when I first met him it was still very difficult to actually, you were thinking, I'm trusting you with my daughter. I don't know you, you know, this is very difficult" (Gina).

Oliver, like several other parents spoke highly about the "*regular*" team that cared for Rose. He felt "*they're all fantastically approachable*", although he recognised that this was not necessarily true of all healthcare professionals as "*some are less approachable*". He was not critical of the skills of the less approachable healthcare professionals but considered that "*it's just not in their nature*". On reflection Oliver did not talk about the possibility that healthcare professionals could change and develop skills. He perhaps believed that since it was "*not in their nature*" they would not be able to develop skills that made them more approachable.

The parents in my study fell broadly into two groups, those who trusted healthcare professionals immediately and built on that trust and those who initially felt disappointed by healthcare professionals and had to develop trust over time. Healthcare professionals had to demonstrate behaviour that allowed this second group of parents to learn to trust them. Twelve of the parents held an opinion which was similar to Sian's. She talked about trusting her regular surgical healthcare professional team as something she had not found particularly difficult:

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"I haven't found it too hard. I know no different, if that makes sense. So I've had to trust them so much, I've always had faith in (the surgeon) cos he hasn't let me down once. He's said it how it was, if you trust somebody like that you're not thinking 'oh', I'll ask somebody else" (Sian).

Anne likened trust to being safe, reducing her anxiety and building hope. She relied on healthcare professionals to be both honest and competent:

"You put your trust in, it's like when you do 'trust falls' you say you'll be fine, just fall. You're putting everything that you have in the world into someone's arms. You want 100% reassurance that 2 hours later they're gonna open that door and give you him back just slightly improved than when he went in" (Anne).

Christine was one of the three parents that assumed *"you should trust them"* but questioned *"can you?"* Her story continued:

"Well when they went after that hernia when he was nine, ten months old. The doctor examined him and said he had a little hernia on his left side. I signed the consent forms they were doing this hernia but they found something that looked abnormal which would have caused him problems later on in life and removed it. I signed a consent form for a hernia and he came back minus his testicle. Now was I doing the right thing? do you trust doctors?" (Christine).

As described earlier Maria had felt *"lucky"* to see an expert healthcare professional early however her first impression increased her anxiety as his non verbal communication had been *"atrocious"*. Despite his dreadful manner she learnt of his *"fantastic"* surgical reputation from talking with other healthcare professionals. Maria described how she went *"back with my tail between my legs"* and began to build trust, the measure of which was that she allowed him to *"operate on her son"*. Clearly Maria's story highlighted that she judged the healthcare professionals' character as an indicator as to whether to trust him. Clearly this highlights the distinction between clinical competence and moral competence and the value Maria placed in surgical excellence. Maria was willing to give the professional a second chance. She compromised her initial emotional reaction towards the professional, allowed surgery and adjusted to what she believed were the professional's social skill deficits. Several other parents also considered non verbal communication as a reflection of the

healthcare professionals character and consequently their ability to trust them, without trust as Oliver explained he *"wouldn't let them touch Rose"*.

For Kerran trust was coupled with feeling secure as she did not want to *"sound stupid"* when talking to Leah's doctor. She made a judgement about Leah's doctor whom she saw as *"an educated man"* and as such she did not want to be considered foolish when asking questions. She wanted to feel safe and believe that he considered her an intelligent woman; however she was uncertain about the rapport between them. This resulted in Kerran feeling uneasy and anxious around Leah's doctor which impacted more generally on her confidence with the healthcare professional team caring for Leah. However, Anne experienced feeling valued and respected by her healthcare professional team. She recalled, with pride, her confidence in becoming a recognised expert in caring for Barry:

> *"What was also nice was that a few times when they came, when the doctors came, they had students with them and the students asked me so many things and I was telling them and they'd say 'well this is brilliant, you know so much'" (Anne).*

A rapport with healthcare professionals built parents' confidence in their abilities and they were able to develop and demonstrate new skills in caring for their child which included asking questions as Anne recalled:

> *"A couple of times I said 'I'm sorry but I just need to know this', they said 'don't apologise, we'd rather you ask' and then I think if you ask the questions and get to know as much as you can about what they're going to do you're not questioning them, you're not questioning their skill, you're not sort of saying, 'well do you know what you're doing', I trusted him completely but I just wanted to know what he was going to do" (Anne).*

Parents also thought about trust when they were making decision about their child's surgeries. As Oliver described, parents wanted surgical teams to use the *"tried and trusted"* surgical methods rather than *"trying new techniques"*. Parents trusted healthcare professionals to have a strong evidence base for the surgical approaches they used on their child. Belief in healthcare professionals' clinical skills informed parents' decision making and reduced their anxiety. This is discussed in the following aggregate story.

9.4 Aggregate story (3): Stories about decision making and surgery

Many of the parents felt there was no decision to be made about reconstructive genital surgery. Andrea reflected on surgical decision making and concluded that:

> "I don't think we ever came to a decision as to whether or not we would have the surgery, we were led into it" (Andrea).

For all of the parents their child's AG was something that just had to be "fixed". Whether this need to 'fix' reflected how healthcare professionals had discussed surgical options or if parents' need for a synonymous sex and gender meant it was impossible to accept there was a choice was initially difficult to ascertain. However the concept of parental choice and decision making are reported in the following two foundational stories.

9.4(a) Foundational story (1): Decision making and responsibility stories

Parents' decision making became focused on either "cosmetic / corrective" surgery versus "health / normalcy" surgery for their child. Brian, David and Harry had all deliberated on the value of cosmetic surgery and concluded pragmatically that:

> "my major concern was thinking about when he's going to school and things like that, kids being cruel, do you know what I mean, because what stuck in the back of my mind was being normal" (Brian).

There was no doubt about surgery when parents had been told that it was needed to promote their child's health. For Gina this was minimising "the infection side of things". A bonus for Gina was that surgery would also make sure that Helen "is as normal as possible for her future life".

When parents had made a decision they still faced uncertainty as to whether they had made the right decision or not. For parents the right decision was considered in terms of conferring benefits on their child for the future. Although both of Bobby's parents had agreed initially to allow Bobby to have surgery, their thinking changed over time. Andrea questioned her decision making in relation to taking away Bobby's future choices:

> *"Whilst I appreciate that [the female tissue] really had to go because it could cause him a problem in the future, it was a case of well, that's the last chance he's got of making a decision for himself. Because we've taken that away from him and I sometimes think well maybe if [the surgeon] had said to us halfway through the operation, 'listen what we've found is not what we thought it was what would you like us to do?' I don't, I honestly don't know what I would have done, given the situation but I did think when he said that to us that it was a case of cut and dried, that's it now, no going back, no change. If Bobby changes his mind at any point in his life [about his gender] he doesn't have the same options anymore" (Andrea).*

Brian believed Bobby needed further *"corrective surgery"* while Andrea argued that more *"cosmetic"* surgery would impact negatively on Bobby's mental health. Andrea talked about how *"unhappy"* Bobby had been following surgery, with the appearance of his penis and also the discomfort he experienced. Bringing together the decisions they had already made and the 'right' next decision for Bobby became much more complicated:

> *"There's a lot of pros and cons to it [more surgery], I mean certainly from going on the websites, there's a lot of people who have been through hell and back over decisions and choices that they've made and whilst they don't say to you 'don't do it' or 'do it', you read some of the stories and you think, you got that wrong, but then did they?"(Andrea).*

Faye believed she did not have a decision to make about Gabby's surgery but she had a role in deciding when Gabby had surgery:

> *"Nobody said to me, 'would you like your daughter to have this done'. It was, 'your daughter's got to have this done'. And when the ages went up and up and up and then I kept thinking it's a long time to wait... I didn't decide for her to have it done, I would never have known what a vaginoplasty was; I would not have had a clue. It wasn't a choice I made, I agreed to it. (Faye)*

Faye agreed to surgery since this would remove uncertainty and promote normalcy and she wanted Gabby to:

> *"Develop as much as it's possible at the same speed as everybody else and at the same rate whereas she seemed to have been sort of stuck in limbo, same shape doing nothing and nothing's happening" (Faye).*

Maria held very strong beliefs about Neil's surgery and the decision she believed was owned by both herself and her husband:

> "There's no way in my mind that we've done the wrong thing and that I ever think 'I wish we'd left him to make his own decision when he was older'. Definitely not. I watched a documentary on Channel Four just after Neil was born and it was an American one. They were lobbying Parliament to stop parents making children have genital surgery and we just said 'how ridiculous'. We both together agreed that it was ridiculous" (Anne).

Similar to Faye and Maria's stories, Oliver considered that it was reasonable for surgical and medical teams to guide and support parents "*in a particular direction where you make the decisions*". Oliver's opinion was "*you're given the facts, and there is no decision to make*".

A few parents worried about what their child might think about their decisions when they were older, Gina hoped she was making "*the right decision for*" Helen and it was what Helen "*would have wanted her to do*". Several parents looked forward to the 'time' when their child would take over the responsibility of decision making. Once their child achieved a degree of autonomy parents believed they would feel less anxious as parents about making surgical decisions. However, whilst waiting for their children to become autonomous parents were hopeful they would stay in a phase as described by Oliver "*where everything's going alright*". Oliver's story was consistent with many other parents in that he knew "*we've got surgery to come*" but as Rose was growing up the "*decision now*" was up to her.

Kerran felt she had no choice in the decision about Leah's clitoral surgery. Initially Kerran had wanted a clitoral reduction for Leah but at Leah's out-patient appointment she felt that the doctors "*were like trying to put me off having this operation*" which sadly left Kerran feeling "*bullied*". Kerran was no different to the majority of parents as she worried about Leah's future especially her "*sex life*" and "*relationships*". She accepted the healthcare professionals' reasons for not having surgery:

"The implications of having an operation would obviously ruin her sex life or could because of the nerve endings being cut off" (Kerran).

She carried on talking about how worried she was that the HCP were not listening to her experiences of living with Leah's AG and also her concerns about Leah's future:

"If she doesn't have it [clitoral reduction] then how's she gonna have a relationship in the first place or, you know, will there even be a relationship in the first place?" (Kerran).

In the main parents felt there had been no decision to make. Generally parents believed there would be a time when their child would make choices and decisions independently. For many parents AG was considered a problem and surgery was adapted as a problem solving strategy to limit uncertainty. Even when decisions had been made and certainty of sex had been established, uncertainty still existed regarding whether the decision taken was right or not. Parents also had worries about the risk of their child not having reconstructive surgeries and they were anxious about surgery and their child's recovery.

9.4(b) Foundational story (2): Stories about understanding surgeries
Although the genital surgeries were explained to parents prior to the operation the actual event of hospital admission and after care were significant worries for all but one of the parents. The genital aspect of the surgery was considered as a very small part of the child's overall surgical event. Parents worried predominantly about *"taking their child to theatre"*, *"pain"* and *"getting blood"*. Prior to having a child with AG most parents had little experience of regular hospital visits. Perhaps because Maria was familiar with hospitals her surgical experience stories were brief and very pragmatic *"[the surgeon] did the operation and we were very happy with the result"*. Chloe described her surgical hospital experiences with Edward as *"just horrific"* and she would hide herself away because she *"just didn't want to see anything"* whilst she was in the hospital. Hospital administration failures such as those described by Chloe included *"the usual trauma of turning up for operations and no beds, being sent home"*; these caused additional anxiety and stress for the parents.

Post operative care was often talked about as traumatic and unexpected. David, Chloe, Anne, Brian, Andrea and Christine all considered the management of catheters and wound dressings after surgery as an appreciably negative experience. Anne's comments are typical of the parents' experiences:

> "[The doctor had] explained it so well that I was fine with that. He went down and had it done, but the biggest shock was the bandages, like a plumber's tube, and I thought my God, and I really thought surely what on earth have they done. It did sort of take me back a bit, he said he'd have to have a catheter for at least ten days and I hadn't realised that" (Anne).

Gabby's first surgical experience had a profound effect on Faye (and this was unlike the stories told by other parents):

> "It was awful, probably one of the most worst experiences I've ever had in my life, because, you know an ambulance turned up for me that morning, I remember, about 5 o'clock that morning and we got into the hospital and they put us in this little side room and the nurse come in explaining everything. Lovely, really nice, she sat there with me for about an hour and a half just talking to me and generally keeping me calm I think, although I didn't look like I was worried, but I must have kind of look like I was worried".

> "The next thing they took Gabby off and it's like they took one child away and fetched me another one back in it's place although it was the same child but to me it wasn't, it was different. It was like, it really was like going to a funeral because it's like you go in and you bury your child but without the funeral and then suddenly somebody presents you with another one and it's like getting use to this child all over again, it's just like giving birth and starting to be a parent again". (Faye).

None of the other parents explicitly talked about losing a child through surgical fixing. Faye clearly talked about the loss of the child as a bereavement. For Faye one child was replaced with another and although she had the memory of that child the surgery had ensured that this child could not be brought back. Faye had consented to Gabby's surgery yet she found the experience shocking. She did not anticipate she would grieve for the child she lost and this feeling initially challenged her belief she had made the right decision for Gabby's future.

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In the following section, the keystone story, I will both summarise and synthesise the aggregate and foundational stories from sections 9.2, 9.3 and 9.4.

9.5 Summary and synthesis of keystone story 3: Parents' stories about healthcare professionals

Parents spoke about the need for information and how having *"someone there"* to help them understand would have been very helpful, especially in the early days. There was reliance on healthcare professionals to deliver information and for Anne, Barry's surgeon was especially remarkable:

> *"The surgeon explained, you know, if you asked him anything he'd have an answer, he was not one of these, well I'm not sure, he just knew. I mean he's just amazing really, what amazed me about him was the knowledge they've got to put these babies right" (Anne).*

Parents had to pull together the information they received from healthcare professionals and try to make sense of AG and surgery. Very importantly parents wanted to feel a valued part of the healthcare professionals / parent relationship. They were dependent upon healthcare professionals to foster a secure relationship in which they felt safe and were able to ask questions. Safe relationships helped reduce parental anxiety and allowed them to process information and build their own understanding. Too much information resulted in what Brian described as *"too many blurs"* which overwhelmed parents and caused additional anxiety. Therefore, parents had to balance how much information they felt comfortable with and could cope with against when to ask healthcare professionals for additional information. If healthcare professionals were overly keen to deliver information 'at parents' a dissonance developed between what parents wanted to know and what they believed they needed to understand to be able to support and protect their child. Parents were left trying to bring together in a harmonious way their own understanding of AG and the need for information to support their growing knowledge.

By communicating different aspects of the child's needs (such as test results, medication, surgeries) and by using different approaches (such as drawings

and video clips) a few parents built a much clearer understanding of their child's physical anatomy and Andrea was *"grateful to see that"*. Visual images helped some parents, like Frank, to create a *"mental image"* which helped them understand the complex nature of AG and the purpose of surgery. Parents learnt in different ways and therefore different communication styles suited different parents. Sharing information with open communication helped parents to harmonise the shocking event of having a child with AG. On the occasions when a lack of, poor or complex communication with healthcare professionals existed parents did not have enough information to be able to understand AG. The consequence of limited or poor communication heightened parents' anxiety which in turn reduced their confidence. Mis-communication often resulted in parents feeling angry which could also result in a loss of trust in the healthcare professional. Parents also found it especially *"difficult"* when there was a delay from healthcare professionals in sharing information. Parents did not appear to be offered routine psychological support. Eventually parents managed to find a healthcare professional they could understand through a process of trial and error. Finding a professional they could trust who was able to share information and answer questions in a meaningful way while also being clinically competent was highly valued by parents. The professional relationship they were able to develop with this individual or team was an enormous support at difficult or challenging times.

The goals from surgeries were very clear in parents' minds. Genital surgeries were a problem-solving approach that aimed to bring back order and establish clarity about their child's sex. Reconstructive genital surgeries were also for many parents a method of protection since the AG could be minimised and the risk to their child reduced. Parents' experiences of their child's reconstructive genital surgery focused on specific aspects as Imogene explained how she *"couldn't take Joanne in"* to the anaesthetic room. After surgery post operative wound care or catheter care troubled parents and impacted on daily life and as Anne described *"I don't think I slept for a week when he had that catheter in"*. Surgery was a shocking event for parents and their expectations and understanding were limited as they described being unprepared for post operative *"bandages"* or *"catheters"*. They had to learn to manage events they

believed were shocking and bring these into harmony with what they believed was the right course of action for their child's future. Parents were not naive enough to believe that surgery was curative and an 'end point' of their child's AG. All the parents recognised that in reality there was no 'end point' and that issues associated with the AG would continue to arise in the future. However, parents did believe that a stable point would be reached and they had to learn to manage the consequences of their child's AG within their structure of their daily lives. Parents also recognised that there would be a shift in the future too with their child taking responsibility and perhaps ownership of their AG. Eventually, decisions would no longer be the sole responsibility of parents. However, parents described how they would continue to be anxious about their child's future and possibly the early decisions they had made for their child.

In the three keystone story chapters I have demonstrated how the three core elements of shock, protection and anxiety are evident to a lesser or greater degree in the stories that parents told. Therefore in the following chapter I bring together the keystone stories and the core elements. In Chapter 10, I present my discussion of the central concept of my thesis, which is the parents' search for harmony. This search for harmony is underpinned by the three core elements of shock, protection and anxiety. In presenting these elements and this central concept I clearly demonstrate how this constitutes a contribution to new knowledge both conceptually and in its potential for guiding practice. I also link through to the existing relevant literature to place my discussion and synthesis in context.

CHAPTER 10: PARENTS' SEARCH FOR HARMONY

10.1 Introduction

The parents in my study, like the majority of people in society, have a socially constructed conception of sex and gender as binary; a child is either a boy or girl. For the parents in this study the inconsistency and uncertainty they experienced between their child's sex and gender produced tensions[17] in their reactions, emotions and behaviours. These tensions acted to motivate parents to seek out ways in which they could work towards trying to regain some order and control in their lives. In exploring and interpreting my findings I discovered that the essence of the tensions that were interwoven throughout parents' stories were shock, protection and anxiety; which I labelled core elements. Uncertainty about their child's sex caused parents to react with shock. The conflict parents experienced between their expectations of having a child with a 'clear sex' against the reality of their child's AG were told as shocking stories. Following these shocking experiences the parents' became conscious of a responsibility to protect their child from the perceived consequences of other people finding out and knowing about their child's AG. As a result of their experiences the parents had to deal with the inner turmoil and anxiety they experienced in responding to and coping with their child's AG. The disruptions their child's AG caused the parents were experienced as tensions within and between the core elements. This forced them to step outside of their previously held beliefs about sex and gender and to ask significant questions about both the immediate and life long repercussions of having a child with AG. In trying to combat and manage tensions parents actively engaged with and faced the core elements that had disordered their lives. In doing this, I argue, they were searching for harmony.

The following chapter is divided into three sections. Firstly, I describe and discuss the tensions parents experienced within and between the core elements of shock, protection and anxiety within the context of my study. In the second

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[17] Tensions experienced by the parents are twofold. Within the context of my syntheses 'a tension' happens as a result of parents' beliefs being stretched. Tension also describes the contradictory emotions parents experienced as a result of their child's AG. For example being happy they had a new baby whilst also being sad that the child's sex was uncertain; making right or wrong choices for their child.

section I explore the concept of harmony and its place within the context of my study. Finally, in the conclusion section I examine my concept of harmony.

Diagram 10.1 is a visual representation of the three keystone stories, the core elements and the central concept of harmony.

Diagram 10.1 A visual representation of the three keystone stories, the core elements and the central concept of harmony

Parents' stories about their experiences of being a parent of a child with AG in a social world

Shock

Search for Harmony

Protection

Anxiety

Parents' stories about their child

Parents' stories about HCPs
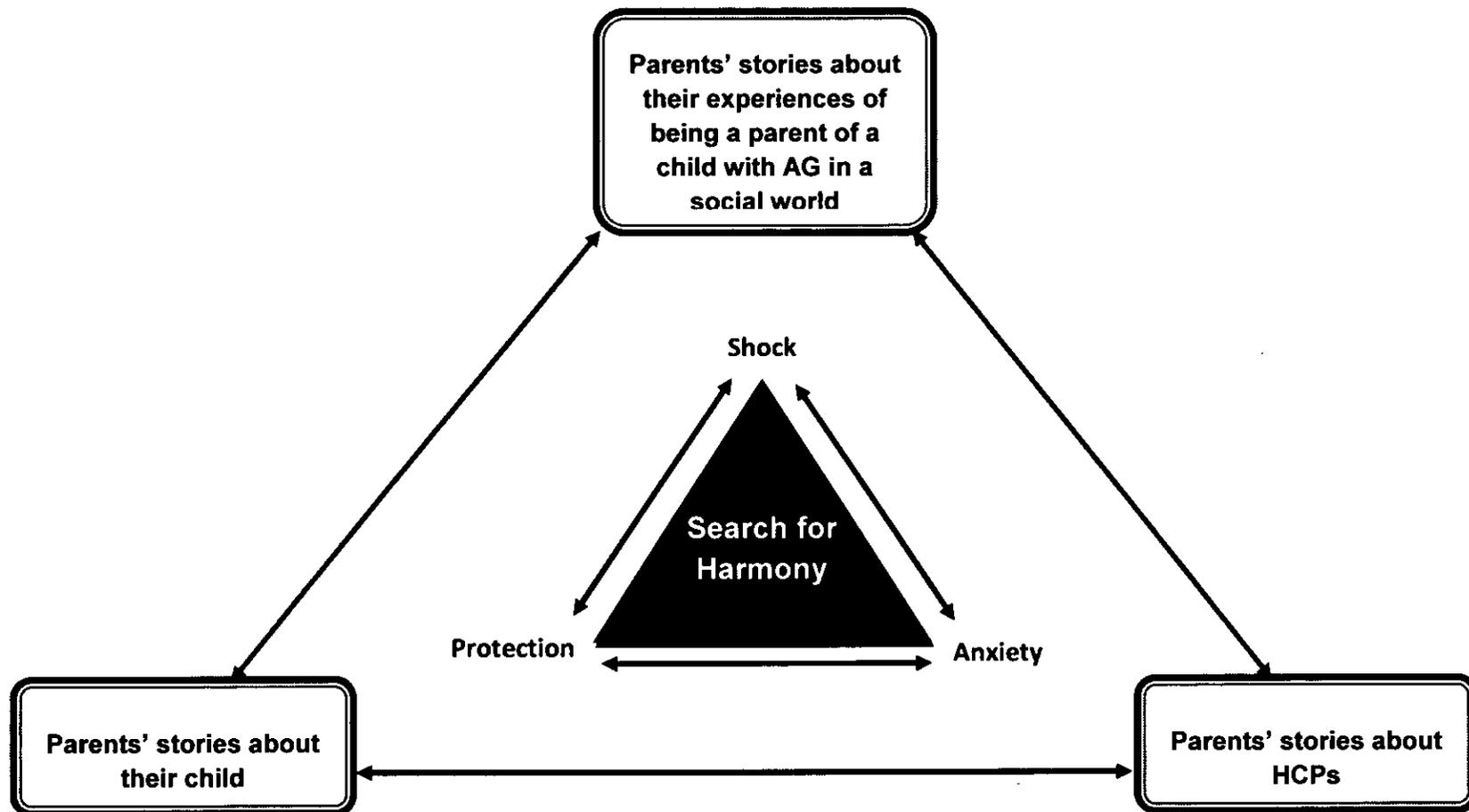
10.2 Core elements: shock, protection and anxiety

Parents struggled with many competing tensions between the core elements which constantly demanded their attention. The following section is divided into three subsections shock, protection and anxiety. Although all three are entwined there are areas where the tensions are different.

10.2(a) Parents' reactions to the shock of AG and shocking events

The initial reactions of shock that created tensions for the parents were triggered by their child's sex uncertainty and/or the immediate risk to their child's health. Shock was a multifaceted experience for the parents and compelled them to renegotiate and subsequently compromise the "socially constructed character" (Gregen, 1999) of what it was to be the parent of a 'healthy' baby girl or boy. Similar to Göllü et al's (2007) findings, the parents in my study were shocked generally that uncertainty and imbalance between sex and gender could actually occur and that more specifically it had occurred in their child. For the parents the immediate shock of their child's AG resulted in an internal tension and this created discord between some parents and professionals. The shock of the incongruity of the child's sex and gender resulted in a temporary loss of equanimity about relationships for parents and professionals. Whilst parents' experiences of shock following the birth of a child with AG have been reported previously (Slijper et al 2000, Le Maréchal 2001, Duguid et al, 2007 and Gough et al, 2007) my findings go further. Although both Slijper et al (2000) and Durgid et al (2007) suggest parents experienced shock they neither discuss the significance of shocking events in parents' lives nor do they explain how or why parents needed to navigate and understand their shocking experiences. The stories told by the parents in my study reveal how they actively sought to overcome the strife that the shock of their child's AG brought into their lives. This was particularly clear during the liminal period, which occurred following the birth of their child with AG, the parents tried to reach a sense of agreement between their ideal (a girl or boy) and their reality (a child with AG). Shock seemed to be a precipitating factor which pushed parents to try and make sense of their child's AG and this is also seen in other studies (Le Maréchal 2001 and Gough et al 2007). Gough et al (2007) discuss their interpretation of how parents' understood their child's AG

and describe the "quest" parents took on in order to "negotiate a clear sex identity" (p12) for their child. They concluded that the disruption to the certainty of sex left parents hunting for their child's true sex, with the goal being the driving out of uncertainty (Gough et al 2007). Interestingly, the quest metaphor is less helpful within the context of my study as parents did not believe that they could banish or drive out the uncertainty which resulted from their child's AG. The parents sought to actively engage with the experiences that had caused tensions in their beliefs, understanding and emotions and this active engagement was aimed at restoring congruity to their beliefs which were built on their social construction of sex and gender. The stories showed how the parents were motivated to act and work in ways which they believed allowed them to regain a sense of order in their lives.

10.2(b) Parents' protective behaviours

The parents in my study worked towards trying to protect their child from the risks and dangers they believed were a consequence of AG. For the parents the safety of their child was of paramount importance and this desire to keep their child safe was focused both on the present as well as the future. Being a parent brought about a sense of responsibility and duty for all the parents in my study and Brian captured this when he said *"your child is the most important thing to you in your life"*. The parents' initial dominant need to manage their child's safety is similar to that described by Maslow (1999). However, importantly the parents in my study told stories about searching for a predictable, orderly understanding of their experiences in which inconsistency could be negotiated (rather than completely controlled). The parents were unable to completely satisfy their needs for order because they were unable to reach a sense of agreement between what they anticipated their community's social construction of sex and gender would be and their child's AG. Although AG is invisible to the majority of people there are some situations in which the child's ambiguity could potentially be observed by others within the parents' family, friendship and community groups. In the initial days, the parents believed they had to help construct a future social identity for their child and this is in line with the way in which Goffman (1968) describes how first impressions in social encounters are important since these help construct social

identity. The parents worried that the incongruity between their child's expected genital appearance and their actual ambiguous genitalia would make a lasting impression on other people, just as it had done on them. Therefore the parents endeavoured to protect their child from situations in which their AG could be judged as a visible stigma, which in turn could affect the child's future ability to "pass" (Goffman, 1968). The parents' desire to protect their child motivated them to build a sense of social congruence in their lives and this took time. This is also seen in the work of Carmichael & Alderson (2004). As in other studies, the parents were apprehensive about sharing information with friends and family (Warne, 2005; Duguid et al, 2007) and needed to reach a sense of agreement about what information to share (Ogilvy-Stuart & Brain, 2004). The parents were especially vigilant and protective; wanting to keep their child *"safe"* until such time as their child could understand their own AG and make choices about their own future and disclosure of information. This is similar to the concept of "alert assistants" as described by Williams (2000) in that many of the parents consciously recognised the "invisible work" they directed towards others in order to protect their child (Williams, 2000). The parents sought to shield their child by controlling the information they disclosed, thereby the parents' in my study assumed the role of "information controllers" (Goffman, 1968). However, being the "information controller" created tensions for parents'. This approach limited their opportunity to share what they saw as 'truthful' information about their child. By keeping information about their child secret, the parents limited the emotional support they could receive from others. Concealing information from family and friends was not a natural act for parents and the tension this created was balanced against the possible risk of their child being stigmatised.

The visual information about AG their children could inadvertently "convey through their bodies" (Goffman, 1968) prior to surgery worried parents. Reconstructive genital surgeries lessened the child's genital incongruity and limited the visual uncertainty and the child's body no longer gave them away as easily. Parents had to balance the risk of the surgery and surgical complications against the risks they perceived to their child's social identity, future self-esteem and safety. For the parents reconstructive genital surgeries

in infancy and early childhood reduced the risk of stigmatisation but did not eliminate it. Parents acknowledged that reconstructive surgeries would not achieve what Li (2006) would describe as complete "sameness" akin to a child without AG since they recognised this was impossible. However, reconstructive genital surgeries did facilitate a degree of unity between sex, gender and physical function that was impossible without surgery (see also Dayner et al 2004; Jürgensen et al 2006; Rebelo et al 2008). Furthermore reconstructive surgeries were described in terms of being a necessary compromise that allowed parents to seek harmony between their child's sexual future and their later sexual intimacy with a significant other.

The work parents did in order to protect their child's future was visible when it involved dialogues with their child. Parents were consciously aware that they did not want their child to become an adult with little or no knowledge about their condition. This need to forewarn and inform is also seen in work by Wisniewski et al (2000) and again by Brinkmann et al (2007). Therefore parents did not keep reconstructive surgical secrets about AG from their child and this has also been previously reported in the literature (Creighton & Minto 2001; Hester, 2004). The parents believed that if their child were to remain ignorant of their reconstructive surgeries this could harm their child's emotional well being in the future. In trying to move away from becoming trapped in a state of persistent discord and disagreement about their child's uncertainty; the parents actively engaged with the tensions that caused them anxiety. This was a dynamic and evolving process.

10.2(c) Parents' anxiety about their child's uncertainty

Anxiety had the capacity to challenge parents' fundamental perceptions and beliefs about themselves and this threw them off balance. For some parents anxiety and worry were tied to achieving what they believed were the best outcomes for their child both in the present and the future. Several authors have suggested that parents' distress and anxiety at the discovery of their child's AG is debilitating and early reconstructive surgeries are helpful in allowing them to move towards combating the uncertainty of sex and gender (Melton 2001; Abassi, 2003 and Thomas 2004). However, Creighton & Liao

(2004) and Morland (2004) suggest there is no evidence that genital surgery relieves parental anxiety and that the surgeries cause long-term psychological and physiological problems for the child once they become an adult. Berenbaum et al's study (2004) contradicts Creighton & Liao (2004) by demonstrating that adult women were not psychologically impaired as a consequence of surgical treatment in childhood. I did not explore the psychological implications of early surgery on either the parents or the children in my study and am therefore unable to add to this specific discussion. However, in my study the parents did worry about their abilities to discuss their child's incongruity and in turn this influenced their decision making and interactions with healthcare professionals. Most of the parents questioned whether they had genuinely been included in the surgical decision making process although they did not regret initially agreeing with healthcare professionals' decisions that resulted in their child having reconstructive genital surgeries. The tensions parents experienced in respect of always making the right choices for their child were similar to those suggested by Gough et al (2007). Some of the parents in my study recognised that in trying to find unity there was no consistent "right course of action" (Li, p599) in relation to reconstructive surgeries. The parents were hopeful that in the future their child would be happy with the choices they had made on their behalf during their childhood.

The parents were generically anxious about their child's surgical experience, the hospital admission, going to theatre and the recovery period. All of the parents' talked about the anxiety of *"hospital routines"*, *"blood test"* and their experiences of *"days that lasted forever"*. Leaving hospital and caring for their child post-surgery also evoked feelings of anxiety and was described as *"awful"* and *"terrible"*. Parents worried about their child *"being in a lot of pain"*.

The parents were concerned for their child's well being (as can be seen in Maslow's (1999) description of the hierarchy of needs). They worried about their child's future and this added to their own inner turmoil about how they were responding to and coping with their child's AG. Parents believed they needed to sustain a sense of balance in their social lives, which included family and

friends and work, as well as managing their fears and anxieties about the present and future needs of their children. At times parents showed concern in non-typical gender behaviours in their child since they were anxious that such behaviours would restrict the child's "passing" effectively (Goffman, 1968). This in turn created a tension for the parents and they worried that their child would be rejected by others. The parents agreed to cosmetic reconstructive genital surgeries to build a congruity between visual sex and gender in order to help their child avoid future experiences of "rejection" similar to the cleft lip experiences described by Aspinall (2006).

Communication with healthcare professionals had the potential to heighten parents' anxiety and this resulted in tensions for the parents. The imbalance of knowledge about AG between parents and healthcare professionals limited some parents' sense of order, risk and understanding which in turn for a small number of parents compromised their child's safety (for example understanding and administration of medication). Gathering and understanding information could either offer reassurance or cause additional anxiety and parents need for information changed over time. Oliver described at first he had *"wanted to know the worst scenario and best scenario"* but over the years he had come *"a long way"* and now *"just wants to know what I need to know"*. Parents wanted to unify what they believed they 'needed' to know against what they believed they 'should know' in relation to their child's AG and reconstructive surgeries. This was not about balancing being informed or not informed. By gathering information on a 'need to know basis' the parents reached a place within themselves where they felt comfortable and confident with their own understanding and the meaning that their child's AG held in their lives. All of the parents experienced events and interactions which brought about emotions that resulted in worry and anxiety. Parents were continually learning and this resulted in them striving towards a new state of mind which they felt comfortable with. Parents negotiated, compromised and overcame challenges and this helped them create new states of understanding and navigated towards harmony.

Although parents did not explicitly use the word 'harmony' in their stories my interpretation is that they were searching for a sense of harmony in their lives.

10.3 The concept of harmony and its place in my study

Ambiguous genitalia were talked about both implicitly (and less often) explicitly as an 'abnormality' by parents throughout their stories. The incongruity between the visual and functional ability of their child's genitalia created tensions for parents. These tensions or conflicts acted to motivate parents' to search for a sense of harmony relative to how they felt about their child's uncertain sex/gender. Parents' social construction of sex and gender were irrevocably challenged. The physical appearance of their child's body was important since the visual body and the social values associated with it are socially constructed and valued (Howson, 2004). Looy & Bouma (2005) suggest that within Western society:

> "most of us take for granted a harmony between our biological sex and our psychological experience of being female or male, our gender identity" (p166).

Disharmony ensues following the birth of a child with AG and I will later argue that disharmony is not necessarily negative or everlasting although I acknowledge that it is challenging. The inconsistency brought about by the child's sex and gender uncertainty produced tensions which worked to encourage parents to try and achieve a sense of harmony. Harmony is the conceptual construct that best describes how the parents' experiences influenced the work they did in order to be able to give meaning to their experiences. In trying to achieve harmony I recognise the influences of Goffman (1968) and Maslow (1999), however I believe that harmony is a broader more encompassing concept which includes stigma and recognises needs but also offers something else to parents. The parents had to align their prior understanding of sex and gender with their experiences of having and parenting a child with AG. This required them to bring the tensions they experienced in their beliefs, values, understanding and knowledge into alignment, creating a congruent sense of their child.

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Harmony and harmonising are not new concepts and although they do not have a strong basis within Western cultures they are described by Li (2006) as "probably the most cherished ideal in Chinese culture". Li (2006) describes harmony as resulting from the "continuous interaction of opposing forces" and this definition has resonance with what I saw occurring in the parents' stories. From interpreting their stories I identified there was a great deal of work that parents did in relation to finding a sense of give and take and degrees of compromise. This renegotiation helped parents to try and find a sense of harmony between the tensions in their beliefs, reactions, behaviours and emotions.

Harmony is a complex concept. It is often seen as a binary concept (harmony/disharmony); linked to well being as a balance to be achieved between the body, mind or spirit. Within the literature harmony is approached in different ways depending on the context and harmony has been explored in relation to nursing theory, health promotion, chronic illness, responsive cohesion, as a concept analysis, and within Confucian philosophy. These aspects are now briefly described although more attention is paid to Li's (2006; 2008) work.

Watson (1985) describes harmony within the context of the nursing care of the body, mind and spirit and associates harmony with health while disharmony is linked with illness. Saylor (2004) explores balance and harmony within the practice of health promotion. Harmony is described as an optimal way in which to live, physically, mentally and spiritually. Any actions that distance people from harmony (such as smoking) are potentially damaging and can have negative repercussions on health and wellbeing. Both Watson (1985) and Saylor (2004) use a binary connotation of harmony with one aspect being sought while the other is to be avoided. Within the context of my study disharmony was not perceived to be totally negative; but a point from which parents were able to acknowledge the tensions and conflicts they experienced and in so doing begin to work towards unity. Harmony within the context of coping is described by Delmar et al (2005). Achieving harmony is discussed in relation to chronic illness and the ability to cope and capacity of the individual

for hope. Harmony in the context of the work by Delmar et al (2005) is linked to acceptance. This is different to the parents in my study since their work is not to do with accepting but it focuses on changing in order to move towards a sense of harmony. Haley and Ratliffe (2006) describe harmony in relation to their work with ventilated children in the community. They describe harmony as:

> "being open to the present moment with a sense of presence that is all-pervasive and without limitations......The presence of balance or harmony is not dependent upon a particular set of external circumstances. Harmony/balance is not the mending of exterior forces, for they can never be totally "fixed." A person in a state of balance or harmony has energy and personal resources available that would otherwise be squandered fighting *what is*" (Haley & Ratliffe, 2006 p57).

Haley and Ratliffe (2006) were drawn towards a more Confucian model of harmony. However, their definition includes elements that are more commonly affiliated to coping. They also use balance and harmony as interchangeable concepts. I believe they are not the same; balance can be a component of harmony but it is not identical to harmony. Haley and Ratliffe (2006) explored adjustment and adaptation within their harmony framework. Although important models I do not believe that adaptation and coping are the same as harmony. Coping and adaptation are concepts that were described within my study; however parents sought more than being able to cope or adapt. The concepts of coping and adjustments do not capture all the elements within the parents' stories, whereas harmony does. Within my study the parents position coping as a mechanism that they use in their search for harmony. I also argue that when the parents in my study are moving towards a sense of harmony they have the same sense of determination as those that Haley & Ratliffe (2006) describe as achieving "a state of balance or harmony". Fox (2006) describes a model of responsive cohesion which shares some similarities to harmony. For Fox (2006) there is a continuum between cohesion and discohesion and suggests that "responsive cohesion" exists by virtue of the "mutual responsiveness" of the elements that constitute any interaction. The similarities this holds with my study relate to the tensions that the parents experienced and how parents interact and work with these tensions. Easley (2007) undertook a concept analysis and reviewed the literature over a five year period ending in 2003. This

resulted in the analysis (using Wilson's framework) of 29 papers. Easley (2007) identified several areas of harmony including: harmony within the environment; as a balance of self and the outside world and harmony in clinical interactions, relationships and interventions. Easley's paper was not particularly helpful in my exploration of harmony. Although the author had undertaken a concept analysis the language used to describe harmony remained vague. Easley's (2007) paper drew on non-academic references and internet based references which reflect the current lack of academic discourse in relation to defining the concept of harmony.

Parents' stories conveyed that they recognised the need to manage "a series of ups and downs" and at times the "struggles" (Gergen, 1999) AG brought to their lives and those of their family. Li (2008) discusses the idea that "the family consists in persons and relationships" and harmony in the family is built on managing ups and downs with a "give-and-take" (pragmatic) approach. The parents in my study were trying to reach a sense of agreement about their child and were willing and able to change in order to achieve this. Parents' experiences challenged their beliefs and as a consequence this led them onto a path towards harmony. The concept of harmony that most closely resembled my interpretation from the parents' stories was the Confucian notion of harmony as described by Li (2006), who states:

> "Confucians see harmony coming out from continuous interplay of opposing forces. Through such interplay various parts of the world "negotiate" with one another in order to strike a balance, not from a predestined principle but through some kind of compromise, some kind of give-and-take" (p594).

The parents in my study experienced tensions which were the result of having a child with AG. In the Confusion view "harmony does not require the elimination of difference" (Li, 2008), but is a "proactive" move towards understanding, acceptance, and tolerance rather than the "Daoists argument" of "just letting things be". Parents wanted to be proactive and engage with the experiences that gave meaning to their lives following having a child with AG. Many parents developed a pragmatic attitude when striving towards degrees of compromise about their child's ambiguity. Therefore moving towards harmony for the

parents became a dynamic and evolving process (in which they changed in order to shift towards finding harmony) and which responded to new information and experiences. As their child grew and developed and adopted more socially congruent sex and gender roles this further reinforced parents' sense of movement towards harmony. When new experiences had to be negotiated, such as the child starting school, the possibilities of new tensions were evident in parents' stories. Once again parents acknowledged these tensions and undertook work in order to find a renewed sense of give and take and sense of compromise. This renegotiation helped parents move, once more, towards a sense of harmony between the tensions in their reactions, behaviours and emotions.

Within an academic context my concept of harmony shifts the focus from a binary model of harmony or disharmony to a concept that embraces life experiences. In order for the parents in my study to shift towards the life they were hopeful for prior to having a child with AG they needed to "harmonise various aspects of life towards a life of wholeness" (Li, 2008). Such harmony was not merely achieving congruity for their child but also related to how they managed the core elements; especially anxiety. Li (2008) suggests anxiety is "contrary to the principle of equilibrium and harmony" (p429) and is a sign of disharmony. I argue that the parents accommodated and learnt from their anxieties and did not polarise these as always "disharmonious". An example from Anne's story is when she reflects on her experiences and describes being *"glad of the experience"* although she *"wouldn't have wished for it"*. She describes her own growth, resilience and abilities that developed as a parent following her experience of having a child with AG. Harmony is "reflective" and is "generated through conscious and persistent efforts" (Li, 2008). By seeking harmony parents develop an internal strength which allows them to create order from what they initially believed was a chaotic experience. Parents' stories described how they viewed themselves as 'sensible' and as such they were "able to respect different opinions" (Li, 2008). However, there were limitations in how parents and professionals worked together in a harmonious way.

The value of harmony and the challenges to embracing harmony in clinical practice are evident in the keystone stories. Healthcare professionals' have responsibilities and duties which are focused towards the child and parent. Healthcare professionals need to realise how their own "nature" (Li, 2008) towards AG influences the start of the parents' journey towards harmony. When parents are supported earlier in their journey they find strength to face the tensions they experience.

Within a broader context harmony has the potential to influence larger social systems and challenge the social constructions of sex and gender. If parents and healthcare professionals work harmoniously a political and social debate surrounding AG and the social integration of diversity could grow. Li (2008) describes individual harmony as "comprehensive" but also suggests that:

> "[H] harmony can take place between individuals at the level of family, the community, the nation and the world" (pg427)

Harmony as a concept has the capacity to explain how parents' understand their experiences while also being a goal of parent and professional relationship.

10.4 Conclusion

Harmony is a worthy concept to explain the experiences of the parents participating in this study as it has value in an academic and clinical context. Within an academic context my concept of harmony shifts the focus from a binary model of harmony or disharmony to a concept that embraces life experiences. Episodes of disharmony are not necessarily considered to be destructive but have the potential to be enlightening and instructive. Harmony as a holistic concept is a part of life and is a goal in itself that the parents in my study strive towards. Harmony is all embracing and all encompassing and is at the core of parent's beliefs and helps them accommodate and understand life's experiences. The value of harmony in clinical practice relates more pragmatically to the keystone stories parents described. Harmony as a concept has the capacity to shape moral values both for individuals and at a wider social level. As a researcher-practitioner wanting to contribute to new knowledge and

develop practice I needed to develop a conceptual synthesis that could guide practice and help support parents of children with AG to move towards harmony more readily. The following epilogue explores implications for practice and my concept of harmony within the context of clinical practice.

CHAPTER 11: EPILOGUE

11.1 Introduction

In this chapter I aim to suggest ideas for further areas of research and explore how the findings from my study have the capacity to inform clinical practice. The epilogue is divided into three sections and draws together the limitations from my study and frames these against future research and implications for practice.

11.2 Limitations

Parents' experiences of ambiguous genitalia or DSD are under researched. My study focused on parents' experiences and discovered the importance in their moving towards a sense of harmony. The time it took parents to gather a sense of harmony was variable. I did not explore the influencing factors in respect of how quickly parents achieved harmony. I am unable to offer insight into which parent and professional relationships fostered a greater sense of harmony for parents. I did not explore the experiences of healthcare professionals in my study and am therefore unable to discuss the influences professionals believe they have on parents.

My study was retrospective and focused on parents whose children had had reconstructive surgeries and I had no comparison group. Furthermore I did not explore the experiences of parents who decided their child would not have reconstructive genital surgeries. I chose to recruit from parents of children that had a range of DSD conditions. This was purposeful since the services delivered by healthcare professionals have to cater for all DSD conditions. Some parents' experiences were specific to certain diagnosis, for example; life long medication for girls with CAH. Perhaps the influences of chronic health issues need to be explored further with this group in comparison to the 'well' AG group. The nature of the DSD for several children resulted in life-threatening conditions which parents had to experience. I did not directly compare these two groups but I did recognise they told the same stories as parents of children without a life threatening condition. All the children in my study were either pre-school or primary school children and I did not explore adolescence.

Only a third of the parents in my study were fathers. There were difference between mothers and fathers however to compare this further more fathers would need to be included. It could be that fathers found it to difficult to talk about their child or to sensitive to talk about their experiences in-depth. Fathers' may have had a smaller role in caring for their child's healthcare needs which may in turn have limited their desire to be included in my study. There may be ways of recruiting fathers that would make it easier for them to take part in studies such as this. This would need to be researched in the existing literature. The trustworthiness and credibility of my data could have been further enhanced if I had included the informal discussions I had with my active clinical caseload about the findings from my study as part of my research. However, these informal discussions were not an official and ethically approved element of the study so they have not been included in any depth within the thesis. Acknowledging they occurred reflects the way in which the boundaries between research and practice can blur.

I did not explore with parents the wider social influences they could have on the political drivers for acceptance of DSD. Nor did I explore parents' experiences in developing and using support groups although one parent did talk about this in some detail.

11.3 Future research

There are several areas that would benefit from further investigation. Future research could expand on the findings in my study by exploring the following areas:

- Research could be undertaken into ways in which parents can be better supported within the first few days and weeks following the birth of a child with AG, including the interaction between doctors and parents in clinical encounters. A descriptive ethnography study could explore the clinical as a 'society' and explore the interactions and environment between parents and professionals.

- In my study it was clear that information was important and it helped parents build understanding although it did not always initially help them to move towards harmony. Therefore, how and what information is shared with parents including the value of written information, verbal information, the use of multimedia and parents' support networks needs further study. Future research should focus on how to ascertain whether parents understand the information they have been given. For example, research into the use of the internet as an information source, how parents utilise this and how they in turn share information with their child. I am interested in exploring this further. In clinical practice I believe that parents have limited understanding of some issues and I am keen to explore the value in parents acting in a peer support role for others in the similar situations. Another research question relates to how parents' information needs change during their child's transition to adolescence and young adulthood.
- The emotional impact of AG and how children and young people with AG cope remains unknown. How do young people with AG find a sense of harmony and are there situations or times when this is particularly challenging. How and when do young people begin to take ownership about their AG and create a sense of harmony about themselves and manage any disharmony. Exploring young people's experiences and thoughts is one of my particular interests especially since as a clinical practitioner I am faced with the challenge of how best to support young people and their parents on a regular basis.
- The experiences of children with AG (and their parents) who have not had genital reconstructive surgeries is an area that should be explored to see if they search for a sense of harmony without surgical intervention. This area is influenced by my clinical practice as I have recently experienced a primary school age girl requesting clitoral reduction for AG. Her parents describe their struggled to find someone to listen to their concerns and I wonder whether they are isolated in this experience.
- A longitudinal study of parents' reactions could provide further insight into the critical points for creating disharmony. This may lead to healthcare

professionals being better placed to offer timely interventions and support for children and their families.

- Harmony was the conceptual idea that enabled me to understand parents' interactions with the core elements in their lives. The interplay between these elements was important in helping parents live their lives and understand their experiences. Future research needs to explore the concept of harmony within a wider age range of children with DSD.

- The concept of harmony also has the potential to offer a new way of thinking about other health conditions which have previously been explored within conceptual frameworks such as adaptation or coping. In this way further research using harmony as the underpinning concept could explore, for example, the lives of children with disability, those with a chronic illness and lives of parents bringing up a technologically dependent child.

- The 'concept of harmony' could also be explored from a phenomenological perspective.

11.4 Implications for practice

The implications for practice and the significance of harmony are related to the findings about parents' experiences of AG and reconstructive genital surgery. These are discussed under six subsections; information, support, engagement, involvement, and surgical experience. The final subsection considers the wider application of the concept of harmony.

11.4(a) Information

Parents frequently experienced a lack of clear information about their child's AG. They were not provided with information about how to develop stories they could share with others about their child. They did not have access to information that allowed them to 'hold off' telling others of the sex uncertainty. Healthcare professionals' lack of knowledge was viewed negatively and parents quickly lost confidence in inexperienced practitioners. Healthcare professionals working with this patient and parent population need to have a sound understanding about AG. Providing information in an appropriate manner for

each parent is crucial in establishing understanding and trust, both of which can help to create a sense of harmony for the parent. Healthcare professionals need to feel confident asking parents what they believe they need to know and what they feel they are ready to know. Forceful or overwhelming amounts of information are difficult for parents to manage as this can add to the shock they experience. Healthcare professionals should aim to understand what the AG experience is for each parent and what they think that this will mean for their lives and their child's life in the future.

Parents can sometimes adopt medical language when talking to healthcare professionals, and this can be misinterpreted by healthcare professionals who may believe that parents have a very sound knowledge. Being able to use medical language does not necessarily mean understanding and healthcare professionals need to check this out with parents.

Different information strategies need to be explored. Some parents want written information, others want to talk and some may find that diagrams are helpful. Open discussion about the internet is important as this can be a cause of additional anxiety if, for example, frightening information is discovered; providing good links to trustworthy sites may be helpful.

Healthcare professionals need to take time to listen, without interruptions from telephone calls or pagers, to parents as information can be very difficult for parents to process in the beginning. Professionals must respect parents' privacy and talk somewhere that parents feel safe. It is critical that health professionals prepare parents for hearing what could be considered 'bad' or 'shocking' news. If sharing information and news with others about their child is not managed by parents they experience a loss of control which can heighten anxiety and bring about disharmony. Parents should not be pressurised into sharing information with friends or family. Keeping news secret from family members may not be a natural act for some parents but they have to balance this against protecting their child and regaining some control.

Information needs to be given consistently and accurately as conflicting information causes confusion and heightens anxiety. Sharing information is something that is an ongoing process and parents should have access to the same group of healthcare professionals where possible, since seeing a different person in clinic or large numbers of healthcare professionals in clinic is overwhelming for some parents. As their anxiety increases they are less able to listen at the consultation and a tension is created as they experience a loss of control. Summarising and writing down the information that was discussed with parents and providing them with a copy of this are also helpful.

11.4(b) Support

Parents reported a range of feelings with regard to their emotional well being these included being isolated, alone or supported. Feelings of being alone and/or unsupported were unhelpful and resulted in feelings of hopelessness which increased their anxiety and did not help parents move towards creating a sense of harmony. Healthcare professionals are able to support parents by being approachable, understanding, reassuring, patient and by giving them the time in which to tell their story and express their fears and worries. Exploring the range of emotions described in all studies about AG with parents is helpful especially if discussed in the context of 'other parents have experienced similar feelings'. This could help some parents to articulate the emotions they are experiencing but believe they are supposed to keep hidden. Giving parents' permission and support to explore their emotions and feelings is important as they may have a limited network with whom they talk about their child's AG.

Psychological services can offer support to parents and this is suggested by the BAPS guidelines (Rangecroft, 2003). However, some parents may be anxious about seeing a psychologist as there can sometimes be a stigma attached to seeking this type of support. Parents in my study were not routinely referred to psychology and this should be considered within practice. Parents needed to find a healthcare professional whom they could trust and be honest with that they could contact easily when they needed additional support.

11.4(c) Engagement

Communication between parents and healthcare professionals is important. For most parents this happens in the busy out-patient clinic setting. Some parents recalled feeling intimidated in the out-patient setting and this impacted on their engagement and communication with healthcare professionals. Seeing a different doctor each time they were in clinic was unhelpful and resulted in disengagement for some parents. Clearly healthcare professionals need to establish and maintain good communication with parents in order to give parents a sense of harmony about their child's care and treatment.

Waiting for test results and failure to communicate the correct information to parents was distressing and clearly this is not acceptable practice. Helpful communication needs to be consistent and parents should not feel at any time that information is being withheld from them. Parents should not have to resort to threats or become overly distressed while waiting for the information they were promised to be delivered. When parents experienced difficulties with healthcare professionals this resulted in increased anxiety which had the capacity to create or increase their sense of disharmony.

11.4(d) Involvement

Feeling involved and skilled in caring for their child empowered parents and helped them achieve a sense of harmony. This gave them a sense of inclusion and helped them gain some control. When parents were able to teach or share information with junior staff or students they realised a sense of achievement which was very positive. This also had an impact on harmonising their emotions and actions with being a good parent while also positively enhancing their self-esteem. If healthcare professionals are able to recognise parents as the experts in knowing, understanding and caring for their child parents' experience an increase in their confidence which helps them create a sense of harmony. There is a balance between parents feeling abandoned in the care of their child and still needing encouragement and support. This needs to be managed effectively by healthcare professionals by maintaining good lines of communication with parents.

Involvement in decision making needs to be much more explicit and options of surgery versus no surgery need to be discussed with parents on more than one occasion. Often decisions made at a certain point in time were not questioned again and one mother felt unable to make 'surgery stop'. When parents were included in decision making they felt valued.

Training opportunities exist for teams who care for children born with AG and these should be considered as integral to service delivery. Many localities have a multi-professional team caring for children and families. These teams should come together nationally to explore how practice can be developed and research opportunities explored.

11.4(e) Surgical experience

Waiting for surgery was an experience parents became resigned to. However if surgery was cancelled this became very upsetting and heightened parents anxiety which shifted parents away from a sense of harmony that that episode of surgery was over. Surgery should be planned and pre-operative education and support should be provided to parents in order for them to be involved in caring for their child. Surgical outcome should be discussed on more than one occasion and parents should be prepared for the appearance of their child's genitalia post-operatively.

11.5(f) Widening the application of harmony

Harmony, although contextual and relational does not exit as a concept in isolation. For any person to seek harmony they have to choose between one or more things; in my study these choices were around the core elements of shock, protection and anxiety but it could also include as yet unknown areas. Harmony is dependent upon the interplay and energy generated between elements and it is for this reason that harmony has applicability to a wider area of research and clinical practice. I believe that harmony has a place within chronic health research and also clinical practice for people with long-term health conditions. Harmony is a concept that although associated with coping or not coping or adjustment or maladjustment, does not burden itself with the negativity that some of these processes imply. In seeking harmony people are constantly engaging with and accommodating the events that have disrupted

their lives. This accommodation gives way to emancipation and to opportunities to creatively constructing alternate futures for themselves. Future work could usefully consider exploring the links between harmony and concordance in relation to healthcare. I believe that all of the recommendations I have suggested in the earlier sections of the epilogue hold resonance with other areas of clinical practice within the field of children's nursing and beyond. I hope that I have generated interest and curiosity around the concept of harmony for further research.

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APPENDICES

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Matrix for purposively selecting parents for the study

The matrix has been developed to try and recruit a purposive sample to the study and to guide the researcher in identifying prospective participants

<u>Primary Elements</u>
- Child has to have experienced at least one, major 'genital surgery' episode.
- Child has to fit one of the sub groups identified for this study.
- The child is less than eleven years old at recruitment into the study. This aims to limit issues of puberty within this study.
- Parents' will be viewed as mother/father (or those that take that role within a family situation, a step parent may have been within the family since surgery and have stories that equally need to be told). To avoid confusion foster families will not be included as they may not have been present at major life events or remain consistent carers for the child.

<u>Secondary Elements</u>
- Representative across gender and age group
- Aim to try to get an even recruitment of mothers and fathers in the study

<u>Further Elements</u>
- Demographic data may be collected to later examine spread of families and also family history, if this was thought to be important.
- Local statistics are kept by the medical teams that care for these children and average birth rates are reported nationally and from this data is likely with the birth rates in the Northwest, served by our hospital, will yield approximately 60 children who are under the age of eleven years. Recruitment of 15 parent sets will represent 25% of the possible total population available.
- No gender-reassigned children will be included in the study (this group includes children who have a clear genetic male type but have been surgically reassigned as female).

Groups	Pre-School 0-5yrs	Primary 5-10yrs	Total (sets)	Mothers / fathers	Ratio	Geographic location	Ethnicity
Ambiguous genitalia	2	2	4	8	1 girl, 3 boys	Where possible within the catchment area of the service. Only participants cared for by present organisation are to be recruited	No limited by the study, however it is recognised that there maybe limited parents from within ethnic minority groups due to these phenomena being increasing rare in these groups.
Urogenital issues	2	2	4	16	4 girls		
Intersex	2	2	4	24	4 Intersex (2 raised girl: 2 raised male)		
One of the above as identified after preliminary analysis	2	1	3	30	1 girl, 2 boys (depending on group identified from analysis)		
		Total mother or father recruited	15 (pairs)	30	8 girls: 7 boys		

Key to terms:
Children born with ambiguous genitalia are those where initial examination of the genital area questions the sex of the child, but chromosome testing reveals a clear male or female. Girls born with urogenital conditions can have common openings for urine, stool and vagina (this group can only be female). Intersex children can have mixed gender at physical, chromosome or endocrine level; those in this group often have a gender of rearing.

Appendix 1
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Recruitment Matrix
Caroline Sanders
Qualitative study: Listening to the narratives parents tell about their child's genital surgery

Appendix 2
Invitation letter send to parents

XXth September 2004

Dear Parent [name of Mr & Mrs XXXXXX]

Thank you for taking the time to read this letter. I would like to introduce myself. My name is Caroline Sanders and I am the Clinical Nurse Specialist in urology working between the [hospital name] and the University of Central Lancashire.

Throughout my clinical experience in [hospital name] and other settings I have developed an interest in understanding parents' experiences of their children's genital surgery.

I would like you to consider being involved in a piece of research that is currently being undertaken in the hospital. I have included with this letter a detailed parent information leaflet for you to read. Please take time to read this and consider whether you are happy to be involved in the research.

There is a tear off slip for you to return in the prepaid envelope if you decide you would like to be part of this study. Your children are not involved in this study.

Permission has been sought from the hospital doctor to make this initial contact with you.

Yours sincerely,

Caroline Sanders
Urology Clinical Nurse Specialist

Parents Information

Title of Study: **An exploratory study to explore the experiences of parents whose child has had genital surgery.**

What is the study about?

The study is about listening to the stories that parents have to tell about their experiences of their child's genital surgery.

Why do we need to do this study?

We want to try to understand the experience and what it means to the family. We hope that this can then help develop further understanding in medicine about the impact genital surgery may have for parents.

What will be involved?

There is a tear off slip and return envelope included with this information, should you decide to take part in the study you will be contacted by the researcher and an interview will be arranged either at your home, or in a venue or your choice, at a time that is convenient for you. The researcher is interested in understanding both mothers and fathers experiences and would invite you for individual interviews. The length of the interview should be no more than an hour, it is recognised that some parents may need more time and this will be arranged as needed. The interview will be tape recorded to aid later analysis.

The information you share will be kept confidential and will be used to inform future articles and presentations. The researcher may wish to use a quote from your story in future publications; these will be anonymous. Before any quote is used from your story you would be asked to sign a second consent form agreeing to let the researcher use your words.

Should you choose not to return the slip you will not be contacted again.

Which children will be asked to be involved in the study?

No children will be asked to be included in the study

What benefits are there for children?

There are none. It is fair to say, however, that future generations of parents and children may gain benefit as a result of the research.

Are there any risks to my child taking part?
There are no known additional risks

What information will be collected and will this be kept confidential?
It will be kept confidential but not in your child's hospital notes, only those conducting the study will have access to information. The information will be stored safely by the research nurse on a computer requiring a password to gain access and made anonymous. All tapes will be destroyed once the information is complete. The researcher will also sign the consent form to ensure participant confidentiality.

Do I have to take part in the study?
No and you do not have to give a reason why you chose not to take part in the study. You are assured that if you do no want to be associated with the study your child will still receive the best treatment available. Furthermore, if after an original decision to take part, you withdraw your consent during the trial period, you do not have to offer a reason and the assurance of receiving the best possible treatment still applies.

Can further information be supplied?

Yes. You can make further enquiries at any time during the study to the research nurse on ************, the urology nurse, Caroline Sanders.
Email (secure address) ************ please use for questions and not to opt into the study.

I have received the information and would like to take part in the study.
Please complete the following information so the researcher can contact you directly:
Name :...
Contact telephone / address /
....
....
....

Appendix 4. Approval letter ethics committee

Liverpool Paediatric Research Ethics Committee
Hamilton House
24 Pall Mall
Liverpool
L3 6AL
Tel 0151 285 2408
Fax 0151 236 4493

17 September 2004

PRIVATE & CONFIDENTIAL
Mrs Caroline Sanders
Urology Nurse Specialist
XXXXXXXXXXXXXX
XXXXXXXXXXXX
XXXXXXXXXXX

Dear Mrs Sanders

Full title of study: What narratives do parents of children who have had genital surgery tell?
REC reference number: 04/Q1502/69

Thank you for your correspondence of 15 September 2004, responding to the Committee's request for further information on the above research.

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

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.

The favourable opinion applies to the following research site:

Site: XXXXXXXXXXXXXXX
Principal Investigator: Mrs Caroline Sanders

Conditions of approval

The favourable opinion is given provided that you comply with the conditions set out in the attached document. You are advised to study the conditions carefully.

Approved documents

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

Document Type: Application
Version: 3.0 Dated: 23/08/2004
Date Received: 01/09/2004

Document Type: Investigator CV

Appendix 4. Approval letter ethics committee

Date Received: 01/09/2004
Document Type: Protocol
Dated: 23/08/2004
Date Received: 01/09/2004

Document Type: Interview Schedules/Topic Guides
Date Received: 01/09/2004

Document Type: Interview Schedules/Topic Guides
Version: Appendix 7 Undated
Date Received: 01/09/2004

Document Type: Participant Information Sheet
Date Received: 01/09/2004

Document Type: Participant Consent Form
Version: Appendix 4
Date Received: 01/09/2004

Document Type: Participant Consent Form
Version: Appendix 4 (i) undated
Date Received: 01/09/2004

Document Type: Investigator's Brochure
Undated
Date Received: 01/09/2004

Document Type: Transcription Agreement
Version: Appendix 6   undated
Date Received: 01/09/2004

Document Type: Supervisor C V – Professor .B Carter
Undated
Date Received: 01/09/2004

Document Type: Letter – Head of Nursing
Dated: 18/05/2004
Date Received: 01/09/2004

Document Type: Letter – Consultant 1
Dated: 28/04/2004
Date Received: 01/09/2004

Document Type: Letter – Consultant 2
Dated: 21/04/2004
Date Received: 01/09/2004

Document Type: Letter – Consultant 3
Dated: 30/04/2004
Date Received: 01/09/2004

Document Type: Letter – Consultant 4
Dated: 05/05/2004
Date Received: 01/09/2004

Appendix 4. Approval letter ethics committee

Document Type: Studies Report
Undated
Date Received: 01/09/2004

Document Type: Recruitment Matrix
Undated
Date Received: 01/09/2004

Document Type: Response to Request for Further Information
Version: Appendix 3(i)
Dated: 17/09/2004
Date Received: 17/09/2004

Document Type: Response to Request for Further Information
Version: Amendments to Questions A48 and A49 of application form
Dated: 17/09/2004
Date Received: 17/09/2004

Management approval

The study may not commence until final management approval has been confirmed by the organisation hosting the research.

All researchers and research collaborators who will be participating in the research must obtain management approval from the relevant host organisation before commencing any research procedures. Where a substantive contract is not held with the host organisation, it may be necessary for an honorary contract to be issued before approval for the research can be given.

Notification of other bodies

We shall notify the research sponsor, [name organisation] and the Medicines and Health-Care Products Regulatory Agency that the study has a favourable ethical opinion.

Statement of compliance

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

REC reference number: 04/Q1502/69   **Please quote this number on all correspondence**

Yours sincerely

Peter Rogan
Chairman
Enclosures   List of names and professions of members who were present at the meeting and those who submitted written comments available

Standard approval conditions

RESEARCH IN HUMAN SUBJECTS OTHER THAN CLINICAL TRIALS OF INVESTIGATIONAL MEDICINAL PRODUCTS

Standard conditions of approval by Research Ethics Committees

1.   Further communications with the Research Ethics Committee

1.1   Further communications during the trial with the Research Ethics Committee that gave the favourable ethical opinion (hereafter referred to in this document as "the Committee") are the personal responsibility of the Chief Investigator.

2.   *Commencement of the research*

2.1   It is assumed that the research will commence within 12 months of the date of the favourable ethical opinion.

2.2   In the case of multi-site research requiring site-specific assessment, the research may not commence at any site until the Committee has notified the Chief Investigator that there is no objection from the relevant Local Research Ethics Committee or other approved local assessor.

2.3   *The research may not commence at any site until the local Principal Investigator or research collaborator has obtained management approval from the relevant host organisation.*

2.4   Should the research not commence within 12 months, the Chief Investigator should give a written explanation for the delay. It is open to the Committee to allow a further period of 12 months within which the research must commence.

2.5   Should the research not commence within 24 months, the favourable opinion will be suspended and the application would need to be re-submitted for ethical review.

3.   Duration of ethical approval

3.1   The favourable ethical opinion for the research applies for the expected duration of the research as specified in the application form. If it is proposed to extend the duration of the study, this should be submitted for approval as a substantial amendment.

4.   Progress reports

4.1   Research Ethics Committees are required to monitor research with a favourable opinion. The Chief Investigator should submit a progress report to the Committee 12 months after the date on which the favourable opinion was given. Annual progress reports should be submitted thereafter.

4.2 Progress reports should be in the format prescribed by COREC and published on the website (see www.corec.org.uk).

4.3 Progress reports should include an annual list of any Serious Adverse Events occurring to research subjects (see paragraph 8.3 below).

4.4 The Chief Investigator may be requested to attend a meeting of the Committee or Sub-Committee to discuss the progress of the research.

5. Amendments

5.1 If it is proposed to make a substantial amendment to the research, the Chief Investigator should submit a notice of amendment to the Committee.

5.2 A substantial amendment is any amendment to the terms of the application for ethical review, or to the protocol or other supporting documentation approved by the Committee, that is likely to affect to a significant degree:

(a) the safety or physical or mental integrity of the trial participants

(b) the scientific value of the trial

(c) the conduct or management of the trial.

5.3 Notices of amendment should be in the format prescribed by COREC and published on the website, and should be personally signed by the Chief Investigator.

5.4 A substantial amendment should not be implemented until a favourable ethical opinion has been given by the Committee, unless the changes to the trial are urgent safety measures (see section 7). The Committee is required to give an opinion within 35 days of the date of receiving a valid notice of amendment.

5.5 Amendments that are not substantial amendments ("minor amendments") may be made at any time and do not need to be notified to the Committee.

6. Changes to sites *(multi-site studies requiring site-specific assessment only)*

6.1 Where it is proposed to include a new site in the research, there is no requirement to submit a notice of amendment form to the Committee. Part C of the application form together with the Principal Investigator's CV should be submitted to the relevant LREC for site-specific assessment. If no objection is notified, the Committee will extend the favourable ethical opinion to the new site.

6.2 Similarly, where it is proposed to make important changes in the management of a site (in particular, the appointment of a new Principal Investigator), a notice of amendment form is not required. A revised Part C for the site (together with the CV for the new Principal Investigator if applicable) should be submitted to the relevant LREC for site-specific assessment.

7. *Urgent safety measures*

7.1 The sponsor or the Chief Investigator, or the local Principal Investigator at a trial site, may take appropriate urgent safety measures in order to protect research participants against any immediate hazard to their health or safety.

7.2   The Committee must be notified within three days that such measures have been taken, the reasons why and the plan for further action. Notifications should be in the format prescribed by COREC and published on the website.

8.   *Serious Adverse Events*

8.1   Any Serious Adverse Event (SAE) occurring to a research subject must be promptly notified to the Committee where it is considered possible that the event resulted from their participation in the research. An SAE is an untoward occurrence that:

      (a)   results in death
      (b)   is life-threatening
      (c)   requires hospitalisation or prolongation of existing hospitalisation
      (d)   results in persistent or significant disability or incapacity
      (e)   consists of a congenital anomaly or birth defect
      (f)   is otherwise considered medically significant by the investigator.

8.2   Reports of SAEs should be provided to the Committee within 15 days of the Chief Investigator becoming aware of the event, in the format prescribed by COREC and published on the website.

8.3   An annual list of SAEs occurring in the research should be provided to the Committee with the annual progress report.

8.4   The Chief Investigator may be requested to attend a meeting of the Committee or Sub-Committee to discuss any concerns about the health or safety of research subjects.

8.5   There is no requirement to provide reports to other RECs in the case of multi-site studies.

9.   *Conclusion or early termination of the trial*

9.1   The Chief Investigator should notify the Committee in writing that the trial has ended, and provide a final report on the research, within 90 days of the conclusion of the research. The conclusion of the trial is defined as the final date or event specified in the protocol, not the completion of data analysis or publication of the results.

9.2 If the trial is terminated early, the Chief Investigator should notify the Committee within 30 days of the date of termination. An explanation of the reasons for early termination should be given.

9.3   Final reports, or reports of early termination, should be submitted in the form prescribed by COREC and published on the website.

10.   *Breach of approval conditions*

10.1 Failure to comply with these conditions may lead to suspension or termination of the favourable ethical opinion by the Committee.

Alder Hey Children's Hospital, Eaton Road, Liverpool L12 2AP
Telephone: 0151 228 4811

NHS

RESEARCH CONSENT FORM

To be completed by parent/person with parental responsibility

Title of study:...

....

Name of Investigator: ....

Permission is given for .... to be included in the above titled study.

(please print)

- I confirm that the above study has been fully explained to me ☐

- I was given opportunity for further explanation by the investigator ☐

- I have received a copy of the parent information document ☐

Participation in this study is entirely voluntary and there is a right to withdraw from the study without giving a reason and in the knowledge that treatment following withdrawal will not be affected.

Name of parent/person with parental responsibility ....

(please print)

Signature..

Signed in the presence of...
as witness to the above signature.   *(please print)*

Signature of witness ....   Date...

Job title and department if member of staff ....

Address if unconnected with the hospital ....

.... Post Code ....

Top Copy - To be retained in Medical Care Notes
Yellow Copy - To be retained by parent/person with parental responsibility
Green Copy - To be retained by the investigator

INVESTOR IN PEOPLE

Appendix 6
Interview schedule

Interview structure [for both single and joint interviews]
General Information:

1. Introduction of researcher, setting the scene [quiet room, seating, reiterating what the study is about, are they still happy to take part? Use of recording equipment, make sure it is audible, time limits, boundaries and issues of confidentiality, signing consent form].
2. Family data collection (simple demographics)- family history-parents mother / father / step parent / interviewed alone jointly if prefer to single interviews, how long after interview with one parent is this interview with second parent, age of child, attendance at school (yes/no), siblings and place in the family.
3. Start interview:

Interview Guide [for both single and joint interviews]
- Story telling or back ground questions

[e.g. From the beginning can you talk me through your experience after having your child? There are no right or wrong answers. I am interested in listening to what you have to say. At any point in the interview or when we have finished the interview we can briefly go over the story to make sure that you are happy with it. If you get worried or anxious or need a break then we can stop, take a break and if you then want to we can resume. I'll not interrupt your story very much but I'll probably ask you some questions to help me be clear about things.]

- To elicit description of experience, behaviour, actions and activities

[e.g. What are the most memorable experiences you have of your child as they have grown?]

- Opinion or value questions to gain information about the parents goals, intentions, desires and values

[e.g. Why did you make that choice?]

- Questions about feelings in order to obtain an understanding of emotional responses

[e.g. How did you feel when you were told about the diagnosis?]

- Questions about knowledge and factual information

[How many others have you shared your child's diagnosis with?]
[How have you learnt about your child's condition?]
General probing / follow up questions can be used as appropriate:
Can you tell me more about.........................?
Is there anything else................................?
I am curious about ....?
I am interested to hear more about ....?
What do you mean by ....?
How are you dealing with   ?
Can we go back to....................................?
You were telling me about.............................?
Aim is to: Encourage parents(s) to tell important moments in their lives. Asking questions that open up topics and allow parent(s) to construct answers, in collaboration with the listener, in the ways they find meaningful (Mishler, 1986). Avoidance of leading questions, biasing questions, double negatives, two questions in one, poor listening.

Transcription Agreement

Qualitative Study exploring the narratives parents tell about their child's genital surgery.

I ………………………………………… am aware of the need for confidentiality and anonymity in relation to my work in transcribing the tapes for the above study.

I am aware that I should not retain copies of the tapes, files or paper transcripts. I am aware that I should ensure than any of the materials related to the project should be password protected and should be kept in a locked 'cupboard'.

I am aware that any material related to the project should be secure during transit and that any sensitive materials will be given special consideration.

Signed …………………………………………(Name of transcriber)

Name…………………………………………(printed)

Date………………………………

(Please retain one copy of the agreement and return the signed copy for project records)

General story group	Fragments that add to story groups
Adaptation	Having to do something new for baby / child New skills Worst experiences
Adjustment to Hospital environment	Appointments and systems New faces Scary place
Alone	Wanting to be valued as an individual not jut as a parent Burden
Anxiety linked to anger	Destroying something Healthcare failing family Blaming parents Future anxiety Worry about surgery
Antenatal	Plan for how this would be, normality of it for everyone
Baby	Being a new parent
Baby health stories	Discovery Learning new skills Colostomies
Blame and Guilt	-transient
Bonding	Breastfeeding
Breaking news to family	Visitors Witness at routine clinics
Catastrophising	Death
Child and parent relationship	Emotional bond, caring for, possessive
Community	Taunting Victimisation Proving child's gender to community Teasing Persecution
Comparison	Others pregnant Sisters baby Glasses – the triviality of other peoples worries Media Education Siblings
Concordance	Medication
Coping	Endure Worry about what people think Anxiety Insular Avoidance Surgical aftercare Being special Hospital environments
Determination	Resourcefulness, adaptation
Disbelief	Why me thoughts
Discredited	Parents knowledge

	Not believed
Endings	Discharge from hospital services
	Loss of familiarity
	Healthcare as an extension of family
Expectation	And planning
Extended family relationships	Support
	Anger with them
	Let down by them
Failure	-transient
Fear	Something's gone wrong
	Of child's appearance
	Worry
	Lonely
Forgiveness	Hospital
	Reactions of staff
Freak show	
Friendships	Child
	Parental peer support in hospital environments
Future	Child relationships in the future
	Surgery
Gender	Issues and identity of child
	Treatments
	Genitals
	Gender uncertainty
Gratitude	Hospital staff
Grief	Funeral
	Bereavement
	Death
Hearing the news	Initial moment
	Shock
Hospital horror stories	Blame - transient
	Food
How you know (learn) about gender	Bloods
	Chromosomes
	Waiting
	Making own choices
Impact on others	Siblings, relationships
Joy	Birth / baby
Life Changing	No more children
	Being a carrier
	Not being able to have children
Limbo	Gender loss - uncertainty
Loss of maternal feelings	Bonding
Maternal health story	Postnatal
Mental health	Survival
	Counselling
Normality	Loss of opportunity to have normality
	Baby growth and sleep

	Having other children already Returning to work Surgery makes normal
Ongoing child stories	Worried for the future Physiology Health Medication
Only child stories	Father anxiety
Paranoid	No one else can care as well as I do
Parent long term health	Illness and disease
Parent versus professional	Becoming and expert Decisions for gender Battle
Parental loss of identity	How do I manage this, what story can I tell others
Past knowledge	Informed parent
Pessimistic stories	Always anticipating the worst
Photographs	Insensitivity Reinforcement Ill prepared
Pity for others	Sorry about child's future family
Pity from others	Sorrow from others unhelpful
Planning	Expectations?
Possessive	No one else can care as well as I do Trust
Post delivery	Shock? First moments of life
Pre-news story	Bad
Preparation	Surgery – going to theatre Future
Privacy	Parent Child Hearing news
Protection	Over protective Extended family Surgery Community Themselves and child
Proud parent	Ability Resilience Adaptability Relationships Memories
Puberty	Worries
Rationalising	Why something has happened
Realisation	Something is different
Rebirth	Once surgery complete
Recovery	Physically and emotionally
Re-evaluation	Life

	Giving something back
Regret	No more children Tough life experience Challenge to beliefs
Sad	Upset / Crying stories
Scared	Future and present
Secrets	Private information Lies
Seeking knowledge	Who tells me this Why cant they see I don't understand So many people's advice
Separation	New baby – going to theatre is separation
Sexuality	Childs Homosexuality
Sharing and regret	News – later regret
Shocking	Everything about the experience, understanding challenged
Siblings	Confusion
Support	Support groups Child getting support form extended family Father
Surgery	Waiting Planning and sharing information Choice Informed consent issues Decisions
Surgery - Expectations	Making you feminine Vaginal dilation Cosmetic Anal dilatation Management of catheters
Survival	Persistence
Teamwork	Relationships Listening to HCP's
Telling the child	Surgery Birth
Transfer	Peripheral to tertiary