A BOY OR A GIRL?
Parental, Family and Whanau Information Needs When a Child is Born with an Intersex/DSD Condition

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ABSTRACT

Is it a girl or a boy? This is a question that new parents assume will be answered at the birth, or even in the months leading up to the birth of their baby. To have a baby born where gender is not certain or to discover your child has a condition where their chromosomes and/or genital anatomy are not what are considered 'normal' for males and females shatters the expectations parents have for their child. While dealing with their shock and accompanying grief parents are caught up in major clinical decisions which impact on their child's future. Over the last fifty years there has been a medical paradigm of care that has recently been questioned in terms of the ethics and clinical treatment these families and children have received. This debate is ongoing and new parents are still being caught up in trying to make decisions with major implications for their child's life while negotiating their own, their family and friends, and the medical community's interpretations of major questions around what makes us male or female.

This study used a qualitative exploratory methodology to interview twenty one representatives from four key stakeholder groups in order to explore the information needs parents, families and whanau have when their child is diagnosed with an intersex/DSD (disorder of sexual development) condition. The groups were adults who self identified as having an intersex condition, parents of children with intersex/DSD conditions, representatives from advocacy and support groups and medical professionals these families would come in contact with.

Data was analysed looking at themes which arose from the narratives of those who spoke to the researcher and excerpts from the transcripts are used throughout the data chapters to illustrate the themes. 'Finding Out and Finding Information' examines the experiences of parents and families as they discover their child has an intersex/DSD condition and how this is impacted by the way health professionals talk with them and the information they receive. It reviews the internet as a source of information and the challenges the internet provides for parents. 'The Hard Issues' examines some of the dilemmas, ethical, legal, social and personal, brought up when making decisions for a child and the issues which parents confront in their day to day parenting. It also looks at the experience of growing up with an intersex/DSD condition. The Chapter on 'What is Helpful Information? Conclusions and Recommendations' reviews the advice and information parents would have liked to

receive when their child was born, and that they would offer to others. Finally recommendations are made for health professionals in terms of education and the information and help they can offer to parents, families and whanau.

This thesis explores the experience of those adults, families and children who have been treated under an existing medical paradigm, the work of those who want to effect change for these families and the information needs expressed by those who were willing to share their stories. It makes recommendations for improving the future experience families have within the healthcare system as they face the challenges and joys involved in having a child with an intersex/DSD condition.

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GLOSSARY

Ambiguous Genitalia Term used to describe outer genitalia which do not have the typical appearance of either a boy or a girl.

Complete Androgen Insensitivity/CAIS A genetic condition that makes XY fetuses insensitive (unresponsive) to androgens (male hormones). Instead, they are born looking externally like typical girls. Internally, there is a short blind-pouch vagina and no uterus, fallopian tubes or ovaries. There are testes in the abdomen or the inguinal canal

Congenital Adrenal Hyperplasia/ CAH Congenital adrenal hyperplasia refers to a group of inherited adrenal gland disorders. People with this condition do not produce enough of the hormones cortisol and aldosterone, and produce too much of androgen. Some forms of congenital adrenal hyperplasia are more severe and cause adrenal crisis in the newborn due to salt wasting. In this salt-losing form of congenital adrenal hyperplasia, newborns develop severe symptoms shortly after birth, including vomiting, dehydration, electrolyte changes, and cardiac arrhythmias.

Disorders of Sexual Development/DSD Term introduced in 2006 following the International Consensus Conference on Intersex as an umbrella term for conditions in which a person is born with a reproductive or sexual anatomy that does not seem to fit the typical definitions of female or male.

Gender While "sex" usually refers to a person's physical anatomy, the term "gender" usually refers to mental, social, and cultural characteristics, regardless of anatomy, related to being a boy, girl, man, or woman in our society

Gender Identity A person's innermost sense of himself or herself as boy or man, girl or woman. How one thinks of one's own gender: whether one thinks of oneself as a man (masculine) or as a woman (feminine.)

Gender Role Arbitrary rules, assigned by society, that define what clothing, behaviours, thoughts, feelings, relationships, etc. are considered appropriate and inappropriate for members of each sex. Some clothing, behaviours, etc. are considered appropriate for members of both sexes. Which things are considered masculine, feminine, or neutral varies according to location, class, occasion, and numerous other factors

Gonadal Dysgenesis/Swyer Syndrome Gonadal dysgenesis also known as Swyer syndrome is characterized by "streak gonads" in a phenotypic female with a 46XY karyotype. This condition is due to a mutation which inhibits the function of the Y-borne determinant that would normally cause the indifferent embryonic gonad to differentiate into a testis

Hermaphrodite and Pseudo hermaphrodite Terms introduced by medical authors in the Victorian period. The term "true hermaphrodite" was used for an individual who had both ovarian and testicular gonadal histology, verified under a microscope, "male

pseudo-hermaphrodite" for a person with testicular tissue, but either female or ambiguous sexual anatomy, and "female pseudo-hermaphrodite" for a person with ovarian tissue, but either male or ambiguous sexual anatomy. The terminology is now considered problematic with ISNA recommending that all terms based on the root "hermaphrodite" be abandoned because they are scientifically specious and clinically problematic. The terms fail to reflect modern scientific understandings of intersex conditions, confuse clinicians, harm patients, and panic parents.

Intersex Intersex is an umbrella term used for a variety of conditions in which a person is born with a reproductive or sexual anatomy that does not seem to fit the typical definitions of female or male. Intersex is a socially constructed category that reflects real biological variation

Klinefelter Syndrome 47, XXY or XXY syndrome is a condition caused by a chromosome aneuploidy. Affected males have an extra X sex chromosome. The principal effects are the development of small testes and reduced fertility

Partial Androgen Insensitivity/PAIS Partial or mild androgen insensitivity syndrome results when tissues are partially sensitive to the effects of androgens. XY individuals may have a variety of phenotypic expression

Prader Scale Staging system for the degree of virilisation of the external genitalia.

Sex One's anatomical designation as either male or female based on the presence or absence of primary sexual characteristics.

Sexual Orientation How one thinks of oneself in terms of whom one is sexually and romantically attracted to, specifically whether one is attracted to members of the same gender as one's own or the other gender than one's own. The inclination or capacity to develop intimate emotional and sexual relationships with people of the same sex (lesbian, gay), a different sex (heterosexual), or either sex (bisexual).

Starship Starship Children's Health, located in central Auckland, is part of Auckland District Health Board

Sources used for developing these terms; http://feminism.eserver.org/, http://www.senecac.on.ca/hr/redc/psp/glossary.html, http://www.endosociety.org/, http://en.wikipedia.org, http://www.dsdguidelines.org/, http://isna.org/.

Chapter 1

INTRODUCTION TO THE RESEARCH

I first came to this research after attending a Grand Round given by one of the registrars in our unit in the late nineties. She was presenting case studies from five babies with ambiguous genitalia, and followed up with a review of what I have since come to know as the medical paradigm of treatment for these infants. These babies were presented as medical emergencies requiring a gender assignment which would be enforced by early surgery. The whole presentation did not 'gel' for me and I came away thinking two things. Firstly I should be more interested in what happened to these babies once they left our service and secondly I questioned the ethics of doing early genital surgery to reinforce a gender assignment. At the same time stories of Intersex people and Intersex advocacy were appearing in the queer press, and thus began my exploration of Intersex issues.

My reading over the last 8 years has covered articles in medical and nursing journals, the work of Intersex advocacy groups, stories and narratives of those born intersex, historical writings, and explorations of gender issues. There has been a shift in the content of literature appearing in medical and nursing journals since the late nineties. The paternalistic "we know best and can fix this" attitude which had predominated in the medical community has been openly challenged and there is now some acknowledgement of a different paradigm of care. This acknowledgement however, has not necessarily equated to a change of practice among clinicians who treat these children and decisions involving care and treatment continue to raise ethical concerns. The primary issues addressed in the literature are initial gender assignment for these infants, the question of early versus later (when the child can consent themselves) genital surgery and whose right (the parents or the individuals) is it to consent to any surgical treatment. The journey that activists and more recently members of the medical community have embarked on in an effort to change the paradigm of care for those born with a range of intersex conditions has been mirrored by changes in nomenclature. Over the last 40 years there have been a number of changes in the language used to provide an umbrella term for, and debate over the terminology to use when there is an atypical sex anatomy, or discordance between any of the sexual characteristics including chromosomes, internal and external genitalia and

gonadal histology. This debate, which forms the background for this study, necessitates a review of the issues surrounding nomenclature and a discussion of the decision about what terminology to use throughout the thesis.

Debates Over Nomenclature

Literature from the 60's through to the early 90's uses terms based on the root 'hermaphrodite' (Dreger et al, 2005) or refers to infants with ambiguous genitalia. In the early to mid 90's, following the 1993 founding of the Intersex Society of North America (ISNA), those who had been treated under the existing paradigm began to speak out and the term Intersex came into use. This was in place of hermaphrodite which was considered demeaning and harmful (ISNA newsletter, May 2001). ISNA (2006) comments that they did not invent the word intersex; it had been used in medicine since 1923. However since the term has come into more common usage there has existed some confusion amongst both the medical and lay communities over the meaning of the term intersex and controversy over what it actually refers to or includes as an umbrella term. ISNA (2006) notes that meanings attributed to the term include infants in which there is a question about what sex to assign or an ambiguity about the "true" sex, any child in whom there is discordance between sexual characteristics including chromosomes, or as a synonym for the older terms based on hermaphrodite. With the advent of intersex activism meanings also came to include an experience of gender identity and a political identity. As such, the term intersex has been embraced by some, while others do not feel it is a good fit for them, or refuse to have their condition identified as such. Sherri Groveman (2006) comments on the large and diverse group of adults who have spearheaded the movement for improved care and the empowerment the word has afforded them. It allows a way of classifying their conditions, and speaking about their bodies and experiences while providing a sense of community. However there are also many parents and medical professionals who do not want to give a child a label with a politicized meaning (ISNA, 2006).

Sue Elford (2006), founder of the Congenital Adrenal Hyperplasia (CAH) support group in the UK comments that the majority of the CAH community do not consider themselves intersexed, or the condition to be an intersexed one. Emi Koyama (2006) notes that parents of intersexed children tended to refuse the label intersex only

accepting condition names such as CAH or AIS (Androgen Insensitivity Syndrome). She goes on to comment that an interpretation of the term by parents, that their baby was neither male or female, might make them more likely to pursue 'drastic measures to eliminate that factor, be it surgery or complete secrecy' (p.4). Dreger (2007) notes that doctors cannot agree on what people are talking about when they use the term intersex and parents can find the term shaming and stigmatising, the very things the intersex movement aims to remove from a diagnosis of these conditions. In 2006, following the International Consensus Conference on Intersex organized by the Lawson Wilkins Pediatric Endocrine Society and the European Society for Paediatric Endocrinology the following statement was issued.

> Advances in identification of molecular genetic causes of abnormal sex with heightened awareness of ethical issues and patient advocacy concerns necessitate a re-examination of nomenclature. Terms such as "intersex," "pseudohermaphroditism," "hermaphroditism," "sex reversal," and gender-based diagnostic labels are particularly controversial. These terms are perceived as potentially pejorative by patients and can be confusing to practitioners and parents alike. We propose the term "disorders of sex development" (DSD), as defined by congenital conditions in which development of chromosomal, gonadal, or anatomic sex is atypical. (p.e488)

ISNA have also recommended that the term DSD be used in venues where the medical care of children or infants is considered and have handbooks for parents and health professionals incorporating this term. ISNA (2006) states that they have found that with the term DSD being less 'charged' their message of patient-centred care has become more accessible to parents and doctors. The term DSD is defined as involving conditions which have the following elements.

1. congenital development of ambiguous genitalia (e.g., 46,XX virilising congenital adrenal hyperplasia; clitoromegaly; micropenis)
2. congenital disjunction of internal and external sex anatomy (e.g., Complete Androgen Insensitivity Syndrome; 5-alpha reductase deficiency)
3. incomplete development of sex anatomy (e.g., vaginal agenesis; gonadal agenesis)
4. sex chromosome anomalies (e.g., Turner Syndrome; Klinefelters Syndrome; sex chromosome mosaicism)
5. disorders of gonadal development (e.g., ovotestes)

Clinical Guidelines for the Management of Disorders of Sex Development in Childhood (pg.2)

It would seem though that rather than stemming the debate on nomenclature this statement has fuelled it. Letters to the British Medical Journal (BMJ) following the publication of the consensus statement provide an overview of the reaction to the term DSD. Of particular note is that of Milton Diamond whose work in the mid nineties challenged the existing medical paradigm based on the work of John Money (see Chapter 2 for a further discussion of the works of John Money and Milton Diamond). Diamond (2006) comments that the consensus statement still remains 'wedded to the notion that variations in sex development constitute "disorders" or "something wrong" that should be medically or surgically managed" (p.4). Diamond goes on to caution that "those who influence how medicine classifies individuals must be sensitive to the potential transformative power of the labels they assign" (p.4). He suggests instead the term variations in sex development or VSD be used, as the medical community does not know the biological purpose of such variations and controversy remains about when to intervene. He calls terms such as error or disorder an unwelcome arrogance on the part of medicine.

A number of letters also appeared about the term DSD from those who identify as intersex. David Cameron (2006) writes that he does not like to be defined by negative medical terms and would rather be seen in a positive way with humanistic attributes. Echoing these sentiments is Curtis E. Hinkle (2006) who states that "being labelled disordered dehumanizes us and reinforces the stigma and shame many of us have felt throughout our lives" (p.6). He voices a concern that these guidelines seem more about managing gender, not real health issues that intersexed people face. On the Intersexualite website he also observes that the choice of DSD comes from the same medical experts who have managed the lives of intersexed people over the past decades. He goes onto comment, that once a parent is told that their child has a disorder, they most likely will assume there must be some treatment to either cure or manage this disorder.

Margaret Simmonds (2006) of the AIS support group in the UK notes that DSD provides a terminology clinicians and funding bodies will feel comfortable with due to the lack of political/activism associations but feels the choice of the term disorder is 'bad news' (p.8). She reflects on the term being based in pathology as does Sherri G. Morris, of the US AIS support group who feels the term should not be used

for anything other than an interim period in the clinical setting until a more appropriate term is adopted. Comment is also made on the lack of international peer support group consultation. As Mani Mitchell (2006) of the Intersex Trust Aotearoa New Zealand (ITANZ) says, any change to medical terminology impacts variously on lives and "consulting us would perhaps of been a neat first step" (p.5). Advocates for Informed Choice (AIC), a group formed in the United States in 2006 through an Equal Justice Works Fellowship, with the aim of undertaking a coordinated strategy of legal advocacy for the rights of children with differences in sexual development has made a decision to use both terms, intersex and DSDs. They choose to refer to the term DSD as 'differences in sexual development' (AIC website).

Following the introduction of the term DSD (disorders of sexual development) in 2006 a decision needed to be made in respect to the language used in this thesis. As the author, I acknowledge the debate and criticisms over both of the terms intersex and DSD. Throughout this thesis I have used the nomenclature which was being used in the literature at the time that I am commenting on. This means that the terms hermaphrodite, ambiguous genitalia, intersex and DSD are all used as a reflection of the terminology being used in the literature being cited in the thesis. It is difficult to decide on a term which feels inclusive of all of those with these conditions so in places I have used 'intersex/DSD conditions' in an effort to respect the views of all of those who work to promote the voices of those affected by these conditions. The choice to continue using the term intersex respects those for whom the term has positive meaning or identity. Like the AIC group mentioned above, I feel the terminology 'differences in sexual development' would be better suited than the term 'disorders' as the former has a more positive meaning. The researcher also chose to take a middle ground position in regard to many of the issues that are raised in this thesis - for example the ethical debates that are raised concerning nomenclature (discussed above and in Chapter 5) surgery (Chapters 2 and 5), informed consent (Chapter 2) - so as not to alienate the individuals interviewed for this study, namely health professionals, parents of and adults with intersex/DSD conditions and advocacy and support groups.

Research Aims, Objectives and Contribution to the Field

While debate continues over nomenclature and 'best practice', there remain parents, families and whanau who are making decisions on a daily basis about their children, which will have long term ramifications for their children's well being and sense of self and identity. There appears to be a gap in the knowledge base, constituting the experiences of parents and families as they make critical decisions about their child's future. It seems important to consider what information needs parents have when their child is born with one of these conditions and they are then faced with major decisions. The aim of the research was to explore the information needs parents, families and whanau have when a child was born with an intersex/DSD condition by looking at the experiences these people had within the healthcare system. The objective was to use the information gained to produce a resource for parents to meet their information needs and to provide education for health professionals.

Thematic analysis of the narratives of 21 participants provides a further contribution to the existing literature on intersex/DSD conditions. The experience of finding out a child has an intersex/DSD condition at birth or during childhood, or the experience of discovering as a teenager or adult you have an intersex/DSD condition is explored. Insight is gained into the impact of the language that health professionals use, their treatment of individuals, and the information they provide on the ongoing experience of living with intersex/DSD conditions for both parents and individuals. The role of the internet in the discovery of information about intersex/DSD conditions by participants and the consequent influence this has on the information provided, and terminology used, to speak to those outside the immediate family is a new finding within this research. The thesis identifies that the vast amount of information from the practical to the political, and the ease with which information on genital variances and surgical options is able to be accessed, is a means of empowerment and also a potential source of confusion and stress for the participants. Parents interviewed for this research indicated, for example, that they worry that their child might access information before the parents themselves are able to discuss these issues with their child.

The medical and social literature reviewed in Chapter 2 derives predominantly from an American and European basis with three papers examining the Australian perspective. The issues of nomenclature, surgery, gender identity and genetics are given

a New Zealand perspective by this research, which in turn leads to the formulation of recommendations for information resources unique to the New Zealand experience for parents, families and health professionals. The debate over surgery raises the issue of early genital surgery versus waiting until an individual can provide consent themselves. The review of the literature in Chapter 2 highlights the fact that, due to the historical paradigm of treatment, there is a lack of data on the experience of those who have not had early genital surgery. This research contributes the narrative of a woman with CAH who did not have early genital surgery to the body of literature on individual experiences of growing up with an intersex/DSD condition. The answers supplied by participants to questions concerning the information they would have liked to have received about intersex/DSD conditions and the information they in turn offered to assist other parents also provides a unique resource for health professionals and future parents of children with these conditions.

Thesis Overview

This thesis presents a review of the literature concerning the paradigms of care those born with intersex/DSD conditions have received over the last fifty years, and continue to receive. The issues and ethical debates arising out of this care are presented. The aim of answering the question 'what are parental, family and whanau information needs when a child is born with an intersex/DSD condition' is approached using a qualitative exploratory methodology. The resulting narratives from the four key stakeholder groups are presented using a thematic analysis. Recommendations for meeting the information needs of parents, families and whanau and for the education of the health professionals they come in contact with in both the hospital and community settings are presented at the conclusion of the thesis.

Chapter 2 provides a review of the literature pertaining to intersex/DSD conditions. Intersex/DSD conditions are defined in terms of anatomy and physiology. A wide range of literature is reviewed, and critically analysed including: nursing and medical journals, academic books, the voices of intersex advocacy and support groups published in newsletters and online, personal narratives published in books and online, and articles published in the media and social literature. Major changes and debates

around treatment options and protocols form the background for this study. The literature is reviewed in terms of the history behind the medical paradigm of treatment, with an examination of western clinical management of intersex/DSD conditions from the 1960s to the present (2007). This includes the work of John Money and criticisms of his work by Milton Diamond. Clinical studies, and in particular the lack of long-term follow up studies prior to the mid 1990s are reviewed in terms of the impact on clinical management. Ethical debates over nomenclature, surgery and informed consent are presented and critical analysis of the differing viewpoints is offered. The rise of intersex advocacy groups and their impact on the ethical and clinical debate over the issues of surgery and informed consent are presented. Surgical issues include the timing of surgery (in infancy versus later when an individual can give informed consent), and the risks to the function and sensitivity of genitals by surgically altering them to conform to a pre existing, but subjective notion of 'normal' are considered. The rights of parents to consent to genital surgery for their infant, as opposed to the child's right to provide their own consent is examined in regard to ethical and legal debate.

Chapter 3 provides a detailed account of the methodology used to conduct the research upon which the findings within this thesis are based. A qualitative exploratory methodology was employed to canvas the views of multiple stakeholders who would inform the research from their personal perspectives. The four key groups were identified as parents of children with an intersex/DSD condition, adults who self identified as having an intersex/DSD condition, advocacy and support group representatives and the health professionals that these children and families may come into contact with. Interviews were conducted over 2005/2006 with 21 individuals. The resulting narratives were than examined using thematic analysis which enabled the substantive themes presented in subsequent chapters to be identified and the recommendations for future educational and informational resources to be developed.

Chapter 4, Finding Out and Finding Information examines the experience of discovering a child or an individual has an intersex/DSD condition, it explores the language used to convey information to parents and how this impacts upon their experience. It looks at the amount of, and the format in which, participants received information from health professionals both in hospital and community settings. Issues

specific to the condition Congenital Adrenal Hyperplasia (CAH) are presented. The internet is analysed both as a source of information and a source of stress for participants. Within this chapter it is identified that the internet was initially used to source information by individuals who felt poorly informed by health professionals. However, the ease with which this information could be accessed, in particular information on genital variances and surgical options, led participants to censor what they told those outside their immediate family in the hope of protecting themselves and/or their child against unwanted questions or suppositions about their gender assignment and any surgery.

Chapter 5, The Hard Issues presents some of the experiences unique to having an intersex/DSD condition. The ethical, legal, social and personal issues raised for participants are explored and related to the literature. These include nomenclature, surgery, gender identity and genetics. Nomenclature examines participants reactions to the term intersex, their interpretations of the meaning of intersex and choices made around nomenclature for them individually. Surgery examines the debate for and against early genital surgery in both the medical and social literature and relates this to the experience of, and choices around surgery for participants. This encompasses individual experiences of being informed or given sufficient information to provide informed consent by the New Zealand and Australian health systems (genital surgery for New Zealand children has been carried out in Melbourne since the late 1990s). The issue of personal interpretation and understanding of gender identity, gender assignment, gender roles and sexual orientation and the impact this might have on parental and health professional decisions is examined. The genetic basis for, and the increasing ability to determine genetic makeup antenatally, or in cases of IVF preimplantation of embryos, is related to participants experiences and debate over choices around 'what is normal'. The experience of growing up with an intersex/DSD condition is explored looking at participant's narratives as children, teenagers or parents.

Chapter 6, What is Helpful Information? Initial Conclusions and Recommendations presents participants views of the information they found helpful or alternatively the information they would have liked to receive at the birth of a child with, or at the diagnosis of, an intersex/DSD condition. Participant's answers to the

questions 'what information would you of liked to have had at the time your child was born'? or 'as you were growing up'? 'What information have you found helpful'? And 'what advice would you offer to other parents'? are presented. Participants views on the ways they were talked to and the oral and written information they received along with practical advice offered to other parents are compiled. This practical advice includes the important role of support groups and the necessity participants stressed, of being put in touch with support soon after the birth and/or diagnosis. The recommendations arising from the research study are presented. These encompass the areas of health professional education, written information for parents and the proposal of a team approach to the care of children, parents, families and whanau when a child is born with an intersex/DSD condition.

The research for this thesis was gathered at a particularly historical moment in which debates in the social and political arena were facilitating changes in treatment protocols for infants born with intersex/DSD conditions. The thesis concludes with a review of the conservative paradigm of treatment and ethical debates around early surgery and informed consent identified in the literature. Results from the thematic analysis of participants' narratives are discussed in regard to this existing literature and in view of the new findings identified by this research. Recommendations for further study are also considered. The following chapter provides a review of the literature.

Chapter 2

LITERATURE REVIEW

Is it a girl or a boy? This is usually the first question most new parents ask after a baby is born, and in these days of antenatal ultrasound and prenatal testing, often before birth. Concepts such as "we're unsure", that external genitalia may not be consistent with chromosomal genotype, or even that gender differentiation could occur along a continuum, rather than strictly male or female, do not occur to the majority of new parents. However it is perhaps not surprising that these concepts are not within the realm of thinking for most expectant parents. It is not only that they are not mentioned in baby books or discussed as possibilities by obstetricians or midwives, but more that it is not generally acknowledged in western society that such things may and do occur. Western society and in particular western medicine's answer over the last 40-50 years has been a paradigm of treatment which greeted the birth of an intersex child as a medical emergency (McGillivray, 1992; Myers-Seifer & Charest, 1992). Gender was determined as quickly as possible and reinforced with hormones and surgery. In the last ten years the growing voice of those treated under this paradigm and more recently, published papers in the medical and social literature have challenged this existing paradigm as unethical and non-respectful of the basic rights of intersex children, teens and adults (Dreger, 1998, 2001; Howe, 1998; Chase, 1999; Fausto- Sterling, 2000). This has led to an ongoing ethical and clinical debate about the best course of treatment for these infants.

There now exists a variance in results and recommendations from follow-up studies completed in different countries, with recommendations also differing dependent on the condition the child has. The most controversial issue continues to be whether or not surgery should be performed on these infants (Creighton, 2004). At the core of this ongoing debate is the fact that the birth of an intersex child challenges ideas around what and who determines gender and sexual identity, thinking about gender roles and the seeming importance of external genitalia in determining gender identity. It also brings up issues surrounding the rights of children to have accurate information

about their bodies, and to decide what happens to their bodies, versus the rights of parents and physicians to determine what is best for a child. The issue of informed consent involves parents' and physicians' making decisions that are embedded in sets of understandings and prejudices within society concerning gender identity and what is considered "normal" for males and females. Caught in the centre of this debate among clinicians, ethicists, advocacy groups and those themselves born with intersex/DSD conditions, are the estimated 1:2000 children (Kipnis & Diamond, 1998), who are born with variances in the development of chromosomal, gonadal and anatomical sex. Their parents and families are left trying to make decisions for their child which will have long-term implications for their future physical and psychological well being. This literature review presents the medical paradigm which has existed, under which thousands of children have received treatment, the ethical and clinical debate and criticisms around this paradigm, and the results of more recent studies and debates from the literature which aim to determine the care these infants and their families should now receive. It serves as a background to the research question "what are parental, family and whanau information needs when a child is born Intersex?"

Anatomy and Physiology

Between fertilisation and the determination of genetic sex, and the formation of the internal and external ducts and genitalia which a child is born with, there can be a number of alterations to what is defined in medical terms as the "normal" path to a male or female child. At fertilisation a combination of chromosomal material from both parents most often results in 46XX (female) or 46XY (male). However variations include 45XO (Turners Syndrome), 47XXY (Klinefelters Syndrome) and 45XO/46XY mosaicism. Until 6-7 weeks every fetus has the potential to develop typically male or female genitalia, possessing external genitalia that appear female. Gender differentiation then depends on a sequence of exact events which in turn are dependent on chromosomal, hormonal and receptor function (Frimberger & Gearhart, 2005). It is the presence of SRY or the Sex Determining Region of the Y chromosome which determines testis formation in a 46XY fetus and the absence which determines ovarian formation in a 46XX fetus. If there is absent or deficient SYR in a 46XY fetus this leads to partially formed testes. The differentiation of internal ducts and external genitalia is influenced by the androgens N testosterone and dihydrotestosterone and Mullerian Inhibiting Substance (MIS). It is the fetus's ability to secrete or respond to

these which leads to a variety of forms of internal and external genital development. The resulting conditions have than been broadly categorised into infants who are virilised females, undervirilised males, true hermaphrodites or those with gonadal dysgenesis (Lean, 2004). Lean (2004) goes on to describe these classifications. Virilised females have 46 XX chromosomes and internal female organ development but masculinised external genitalia. The most common of these conditions is Congenital Adrenal Hyperplasia (CAH). Undervirilised males are described as those with 46 XY chromosomes and incomplete masculinisation. This includes Partial Androgen and Complete Androgen Insufficiency (PAIS and CAIS), 5 alpha-reductase deficiency and Persistent Mullerian Duct Syndrome (PMDS). In CAIS individuals appear to have female external genitalia and without prenatal chromosome testing are often not diagnosed until puberty. True hermaphrodites are described as those with both ovarian and testicular tissues in the same or opposite gonads with XX, XY or XX/XY chromosomal makeup. Gonadal Dysgenesis refers to those individuals with 46 XX or 46 XY karyotype, female appearing external genitalia, streak gonads and persistent mullerian structures. Table 1 (Nabhan & Lee, 2007) classifies these same conditions using the Disorders of Sexual Development (DSD) nomenclature.

A Brief History of Clinical Management.

The birth of a child with genital variances had traditionally elicited an immediate response within a medical paradigm, which sought to "normalise" the situation as quickly as possible for all those involved. Two articles appearing in Seminars of Perinatology in 1992 (McGillivray, 1992; Myers-Seifer & Charest, 1992) summarise this response very well. Their findings are indicative of all the medical literature published on the treatment of these infants up until the late 1990's.

McGillivray (1992, p.365) described the birth of a child with ambiguous genitals as a "neonatal psychosocial emergency" where the most crucial feature of the infant (the genitals) is confused. Myers-Seifer and Charest (1992) suggested that a rapid and organised evaluation was required to assign appropriate gender, identify a possible life threatening medical condition and begin necessary medical, surgical and psychological intervention. A team approach to investigation was recommended including a neonatologist, endocrinologist, cytogenetisist, urologist, surgeon and psychologist. An urgent measurement should be 17-hydroxyprogesterone as an indication of Congenital

Adrenal Hyperplasia (CAH), this being the most common reason for ambiguous genitalia. Infants with this condition require fluid and electrolyte monitoring and may require mineral corticoids. A family and pregnancy history should be taken, chromosomes sent and a comprehensive physical examination done. The physical examination includes a measure of the phallus which should be "equal to or more than 2.5cm in length in term males" McGillivray (1992, p.366), and a measure of clitoral length which should "not exceed 1cm" Mcgillivray (1992, p.366). McGillivray goes on to state that any deviations from those standards may indicate incomplete or aberrant sexual development.

The three most important considerations in assigning gender are listed as the potential for an unambiguous appearance of genitalia, adequate sexual functioning and fertility. These factors for determining gender occur throughout the literature. The American Academy of Pediatrics, (2000) in a review of the Newborn with Developmental Anomalies of the External Genitalia, stated that the size of the phallus and its potential to develop at puberty into a sexually functioning penis are of paramount importance. They suggested that infants raised as girls would usually require clitoral reduction to result in a "normal looking vulva" (p.141). A French study looking at a diagnosis of true hermaphroditism stated that generally a female gender was chosen as it was easier to reconstruct functionally satisfactory female genital anatomy (Hadjiathanasiou et al, 1994).

Meyers-Seifer and Charest (1992) identified three historical categories for infants with ambiguous genitalia, which included the term hermaphrodite. The terms were female psuedohermaphroditism with 46XX genotype, ovaries and indeterminate or masculinised genitals, male pseudohermaphroditism with 46XY, testes and ambiguous or female genitalia, and gonadal dysgenesis which includes true hermaproditism. Standard clinical management for female pseudo hermaphrodites, even if severely virilised, was to raise them as female due to the presence of ovaries. Surgical intervention, including perineal reconstruction, clitoral reduction, labioscrotal reduction and vaginoplasty, would ensure that they had female appearing external anatomy. For male pseudo hermaphrodites, gender assignment was based on phallus size. If the penis remains < 2.5cm despite a trial of androgen therapy, then it was recommended that a female gender assignment should be given. Surgical treatment included clitoral reduction, vaginoplasty, gonadectomy and removal of wolfarain duct

tissues. Similarly with gonadal dysgenesis, and XY chromosomal material, a female gender assignment was given due to small phallus size.

Under the old paradigm of treatment psychological help was advised for the parents to help them understand their child's condition, accept the gender assignment and reinforce this for the child, (Meyers-Seifer & Charest, 1992). It was suggested that the anatomy and physiology of gender differentiation be explained and the genitals examined in the presence of the parents. The parents must then completely accept the gender assignment in order to feel secure about their ability to raise their child within the assigned gender. The parents may need help with planning communication with family and friends, and require information about future surgery and hormone treatment. Furthermore it was recommended that genital reconstruction occur as soon as possible to ensure a consistent response from parents and caregivers.

Under the interventionist paradigm of treatment Rossiter and Diehl (1998) described the goal of the medical team as allowing the child optimal opportunity to function and enjoy life in a gender of rearing which is compatible with hormonal, anatomic and functional capabilities. Surgical intervention is required to avoid gender confusion and to prevent undue psychological trauma. They suggested that raising a child in a male gender role for which they are non-functional sets the groundwork for future psychological challenges and psychological harm as an inadequate male.

The guidelines discussed above for the psychosexual management of intersex infants were described by Bradley (1998) as a sex assignment to the gender that carries the best prognosis for good reproductive and sexual function and normal appearing external genitalia. However in the case of XY individuals given a female gender assignment reproductive functioning was not preserved which is contradictory to the stated aims of the treatment guidelines. A decision made as early as possible with minimal uncertainty and ambiguity would, according to Bradley, lead to a stable gender identity.

The exploration of a paradigm of treatment as described above, and an examination of the critiques of this paradigm requires an understanding of its origins and the history behind it. Clinical treatment of infants with ambiguous genitalia was based in the work of John Money, a New Zealand psychologist who worked at John Hopkins University Hospital in Baltimore. It arose from Money's theories about gender identity development, and the subsequent recommendations for clinical treatment

which developed out of a single case study of twin boys. This case study went on to be presented in numerous publications since 1972 (Kipnis & Diamond, 1998; Colapinto, 1997) justifying the clinical paradigm of treatment for infants with intersex conditions. Money's research in the 1950s and 60s with intersexed children and adults led him to believe that all infants are born sexually neutral, and are malleable during a window period of up to 18-24 months when gender becomes fixed. The sexually neutral infant notices the presence or absence of a penis, observes the social distinctions between genders and then conforms to the standards of gender. Therefore if physical gender were unambiguous, normal behaviour would follow perceived anatomy (Kipnis & Diamond, 1998). In a paper on the Psychological Evaluation of the Child with Intersex Problems, Money (1965) stated: "It is not obligatory that assigned sex should agree with chromosomal or gonadal sex, but it should agree with external morphology, surgically corrected, and with hormonal sex correctly regulated at puberty" (p.55). The opportunity to test this theory presented itself with the case of twin boys born in the 1960s (Kipnis & Diamond, 1998). At eight months the boys were circumcised for phimosis but an electrocautery knife used on one of the boys severely burned and destroyed his penis. When he was 17months old, his understandably distraught parents consulted Money, having seen a report of his work on television. After five months they agreed to Money's recommendation to have the child surgically reassigned and raised as a girl. At 22 months his testis were removed and his scrotum reshaped to become a vulva. His parents were advised not to tell him the whole truth and that "she shouldn't know she wasn't a girl." (Colapinto, 1997, p. 35). Yearly evaluations were done by Money and reports were published describing a successful surgical reassignment and rearing as a girl. Money's work was seen as the epistemic foundation for the paediatric standard of practice in regard to intersex infants (Kipnis & Diamond, 1998). The inference was that genetic makeup and prenatal endocrinology could be largely ignored and that the penis had to be clearly absent or present. Given the surgical difficulty in creating a functional penis, most male infants with micropenis were reassigned a female gender, with repeated surgeries to "normalise" their genitals.

Despite, as Kipnis and Diamond (1998) point out, the fact that this was a single case study, Money's publications became the base standard for paediatric practice. Reiner (cited in Colapinto, 1997) estimates that as many as 15,000 sex reassignments have been performed since this case was first published. Suzanne Kessler (cited in

Colapinto, 1997) notes that almost all published literature on intersexed infants was written or co-written by John Money.

John Money did however have one longstanding critic, Milton Diamond, who at the time was a graduate student. As early as 1965 Diamond published a critique of Money's papers on sexual development proposing that intersex children may have an inborn capability to go both ways, but were not gender neutral at birth (Calopinto, 2000). Money retaliated in his book *'Woman & Man, Boy & Girl'* by referring to Diamond and his colleagues' work as "instrumental in wrecking the lives of unknown numbers of hermaphroditic youngsters" (Colapinto, 1997, p.16). Diamond continued to have an interest in, and concerns for the case. Finally in 1994, he located and interviewed the family involved and published in 1997, along with Keith Sigmundson, 'the truth' behind the "John/Joan" case.

On talking to John (pseudonym) himself, his mother and wife it became apparent that this was not the success story Money had claimed. Girls toys, clothes and activities were repeatedly offered to Joan and were most often rejected. She was teased daily at school about her appearance and mannerisms. By age seven she was rebelling against going to the John Hopkins consultations, due to embarrassment at the forced exposure of her genitals, and constant attempts to make her behave more like a girl. Between the ages of 9-11 Joan realised she was not a girl and expressed frustration that her feelings that she was not a girl, even in the absence of a penis, were ignored. At age 12 she rebelled against estrogens and refused to have a vaginal reconstruction, finally refusing to see Money anymore. By 14 the decision had been made to live life as a male, even before being made aware of his true story. With the support of another medical team John then began to receive male hormones and received a mastectomy, with surgery for phallus reconstruction occurring at ages 15 and 16yrs. At 25yrs John married and adopted his wife's children (Colapinto, 1997). He had in fact never developed a sense of a female identity, resented the treatment he received at John Hopkins and at the age of 14 reasserted himself as male. Kipnis and Diamond (1998) comment that medical intervention had added infertility, emotional trauma and ego loss to his initial injury of an accidental penectomy. Since these publications (Colapinto, 1997 and 2000; Diamond & Sigmundson, 1997; Kipnis & Diamond, 1998) more tragedy was to occur in the lives of David Reimer (John) and his family. His twin brother died of an overdose in 2002 and in 2004 David took his own life. In commenting on his suicide Colapinto (2004) said:

> In the end, of course, it was what David was inclined to brood about that killed him. David's blighted childhood was never far from his mind. Just before he died, he talked to his wife about his sexual "inadequacy," his inability to be a true husband. Jane tried to reassure him. But David was already heading for the door.

As well as publishing the follow-up story of David Reimer, Diamond and Sigmundson (1997) commented that there was no support for the postulates that individuals are either psychosexually neutral at birth or that psychosexual development is dependant on the appearance of genitals. They went on to recommend that any child born 46XY be raised as a male.

One of the most obvious questions would seem to be where, in over four decades of this paradigm of medical treatment, are the other follow-up studies on the approximately 15,000 intersex children given a surgical and gender reassignment? Had this been as Dreger (cited in Lewis, 2000, p.4) asks a "40 year plus, poorly run, unethical experiment"? There are in fact very few studies available in the literature prior to 2002. While there may have been some challenges to Money's theories, it appears they had little or no effect on practice. Kipnis and Diamond (1998) commented that there had been no systematic large-scale studies done. The literature reveals some small studies and individual case reports only. Reiner (cited in Lewis, 2000) followed a cohort of 14 46XY individuals with an intersex appearance of no penis but normal testicles. Twelve were reassigned female yet the parents reported that all displayed typically male behaviours during childhood. Six of the twelve switched themselves to the male gender between the ages of 5-12 years. A second study of twelve children has eight of the twelve reassigning themselves to a gender dictated by their XY chromosomes. Phornphutkul, Fausto Sterling and Gruppuso (2000) refer to three further case studies in which 46XY individuals were raised unambiguously as females, and later reassigned as males. Reilly and Woodhouse (cited in Dreger, 1998) interviewed and examined twenty patients with a diagnosis of micropenis in infancy who were raised as boys. All had erections and orgasms. The authors concluded that a small penis did not preclude a normal male role. Fausto Sterling and Laurent (2000) surveyed existing articles on vaginoplasty and found that in an evaluation of data on 314 patients from 80 different small studies, patients undergoing Vaginoplasties had poor outcomes. There were high frequencies of postoperative complications, requiring additional surgery in 30-80% of children, which led to additional and significant scarring. Vaginoplasties done in infancy had frequencies of stenosis as high as 80-85%. Thomas (1997) reviewed the cases of

twelve girls with CAH who had undergone surgery to create a cosmetically satisfactory clitoris and external genitalia. Six of the girls had a urogenital sinus despite vaginoplasties. Two of these girls also had hematocolpos (menstrual blood accumulating in the vagina). Clitoroplasty was deemed unsatisfactory in six girls with atrophy in five. Several of the clitoral reconstructions were withered and non-functional. Thomas referred to the results as "indifferent and, frankly, disappointing" (p.2). Kipnis and Diamond (1998) reported that their reviews of the literature had failed to turn up a single article on the hazards, psychosocial or otherwise, of a large clitoris.

Other Voices and Ethical Debate

Hand in hand with the lack of clinical studies on which this paradigm of medical and surgical treatment for intersex infants was based was a lack of ethical debate. Dreger (1998) comments that this silence, and the lack of ethical analysis have stemmed from the underlying assumption that the "normalising" surgeries have been viewed as necessary. However in the last fourteen years a growing body of literature has developed, initially from the intersex community, and more recently within medical literature, which raises the ethical issues which might have been expected to warrant consideration over the last 30 years.

Cheryl Chase, herself born intersex, founded the Intersex Society of North America (ISNA) in 1993. Its aims were to "end shame, secrecy and unwanted genital surgeries for people born with atypical sex anatomy" (ISNA website). The establishment of ISNA provided the opportunity and a forum for Intersex people themselves to begin to speak out about their experiences. They expressed feelings of betrayal by their parents and doctors, and shared their experiences of feeling shame and isolation. They pooled their knowledge that the genital surgeries had damaged their sexual function and that they had often developed a gender identity different from their sex of rearing. In 1995 ISNA applied to the National Organisation for Rare Disorders (NORD) asking that intersexed adults, and parents of intersexed children be informed of ISNA's existence as peer support. The application was rejected on the basis that they were promoting "experimental treatment" (Chase, 1999, p. 452). Initial letters written to physicians practising on intersex children urging them to reconsider their practice were universally ignored. An offer in 1996 to provide a patient's panel to surgeons conducting a symposium on paediatric genital surgery was also refused. This led to a

protest outside the American Academy of Paediatrics (AAP) meeting in 1996 which resulted in the AAP releasing a statement saying: "The Academy is deeply concerned about the emotional, cognitive, and body image development of intersexuals and believes that early genital surgery minimises these issues" (Chase, 1999, p. 452). However the views of ISNA were finally heard by some physicians and by the end of 1996 they were beginning to contact ISNA, stating that they were frustrated with some of their peers' insistence on tradition. By 1997 ISNA had a large group of adult intersexuals who would speak publicly, and the support of a number of sexologists, sociologists, psychotherapists, historians, some surgeons, physicians and ethicists (Chase, 1999).

It was at this time that Diamond and Sigmundson (1997) published their review of the 'John/Joan' case suggesting that parents and children be referred to appropriate and long-term counselling rather than immediate surgery and sex reassignment, because it seemed like a simpler immediate solution to a complicated issue. This was followed by the publication of their guidelines for dealing with individuals with ambiguous genitalia (Diamond & Sigmundson, 1997). These suggestions for treatment were based on their own experiences, input from colleagues and the intersexed community and their interpretation of the literature. They advocated a change in the language used, avoiding terms such as abnormal, maldeveloped or defective in terms of the genitals, so parents and children would accept the genitals as normal but atypical. A full history and physical exam including genetic and endocrine evaluation was recommended in conjunction with fully and honestly disclosing to the parents why there was a delay in assigning gender, respecting theirs and the child's confidentiality, and offering counselling. Gender assignment was not to be based purely on phallus size and function. They also developed recommendations for a number of individual conditions including Androgen Insensitivity Syndrome, Congenital Adrenal Hypoplasia and Hypospadias. The concern was primarily with how an individual with one of these conditions would develop and prefer to live post puberty. No surgery should be performed for cosmetic reasons and the use of steroids should require informed consent. Diamond and Sigmundson (1997) suggested a watch and wait approach, rather than the removal of gonads as a prophylactic measure. They acknowledged the difficulties that would arise for parents and children and recommended counselling by those skilled in the areas of sexual, gender and intersex matters. They further recommended that this counselling should be ongoing and available for the family and

the child individually. It should include frank and honest information and discussions around sex, sexuality, puberty, fertility, gender identity, and the possibility that this might change for an individual. They also recommended that, as early as possible, the family should be put in touch with a support group. Diamond's keynote speech to the AAP in October of 1998 called for a moratorium on early genital surgery (ISNA, 1999).

In 1998 the entire winter issue of the Journal of Clinical Ethics was devoted to the issue of Intersexuality. While it included articles by Diamond, Kipnis, other physicians and surgeons, of note is the inclusion of work by Cheryl Chase, founder of ISNA. All contributors called for a drastic change in medical practice. Chase (1999) notes comments by the editor that none of the surgeons who routinely perform intersex surgery were willing to contribute, nor was she able to find anyone to defend the ethics of the traditional model. This was followed by a range of publications and position papers by representatives of paediatric societies, clinical treatment working parties and patient support groups both questioning and condemning the traditional model of treatment for those born intersexed. In 1998 surgeon Justine Schober in the text 'Pediatric Surgery and Urology: Long Term Outcomes in Feminizing Genitoplasty for Intersex', recommended a change in practice (ISNA, 1999). In 2000 the Lawson Wilkins Pediatric Endocrine Society devoted a third of its conference to the controversy over medical management of intersex with Cheryl Chase giving the final address (ISNA, 2001). In 2000 the North American Taskforce on Intersex was also formed. It comprised of American and Canadian physicians in the fields of paediatric endocrinology, urology, psychiatry and genetics, along with representatives of support groups. It aimed to conduct long-term follow-up studies with the goal of providing future management guidelines. The task force was also available to provide a medical perspective to the news media. In July 2001 the British Association of Paediatric Surgeons Working Party on the Surgical Management of Children with Ambiguous Genitalia produced a statement taking into account the lack of outcome data on genital surgery and the views of patient support groups. They stated that although there is likely to be continuing pressure from parents for early corrective surgery, fully informed consent would require them to be aware of the possibility of non-operative management. Therefore they stated, there is a strong case for no clitoral surgery in lesser degrees of clitoromegaly. They proposed that vaginoplasty should be left until puberty, as should gonadectomy, and they argued against female gender assignment and surgery for undervirilised males.

In 1999 the High Court of Columbia, South America, issued two decisions regarding the ability of doctors and parents to surgically treat intersex children. The aim was to force parents to put the child's best interest ahead of their own fears and concerns about sexual ambiguity. The court went on to state that "surgery may actually be a violation of autonomy and bodily integrity, motivated by parents' intolerance of their children's sexual difference" (p.3). For children less than five years consent could only be given after accurate information about risks and alternative treatment paradigms rejecting early surgery were considered. It needed to be written and given more than once over an extended period of time. Children over five years could not be consented for by parents, as they were deemed to have their own autonomy. These decisions were based in part on a 10,000-word document from ISNA, and it has been their work which has played a fundamental part in challenging and beginning to change thinking and practice over the last decade.

The ISNA developed its own recommendations for the treatment of intersexed infants and children. The model was based on the avoidance of harmful or unnecessary surgery, qualified professional mental healthcare and empowering the intersexual to understand his/her own status and to choose (or reject) any medical intervention (ISNA website). They stated there should be no unnecessary or harmful genital surgery, a designation which includes vaginoplasty, clitoral reduction or resection, clitorectomy or repair of first degree hypospadias. ISNA acknowledged the need for surgery in severe second or third degree hypospadias, chordee extensive enough to cause pain, bladder exstrophy and imperforate anus. Early surgical intervention should not occur for either micropenis or clitoromegaly. The option of surgical and hormonal intervention should be further considered at puberty with the full informed consent of the individual concerned. Counselling should be available for the whole family by a professional with extensive training and experience in the areas of psychotherapy and sex therapy. There should be complete disclosure and support for patient autonomy along with referral to peer support. While a gender is assigned based on all the information available, it is recognised by both the parents and physicians involved that the assignment is preliminary and that the child may decide to change it later.

Having made the recommendations outlined above and with the start of a shift towards a new paradigm of care for infants who were born intersex ISNA recognised the importance of acknowledging the issues which would arise from this. As Dreger

(2001) pointed out, "it can be dammed hard to be intersexed or to have an intersexed child" (104, p.5). An important issue which arose was that, without surgery, the child would not be recognised as the assigned gender. How would it be in reality for parents to raise a child without early surgery to "normalise" the genitals? How would it be for children to grow up, go to school, and enter puberty? There was, and in fact still appears to be, little information available as to what it will be like for children not to have surgery, as this has not been an option under the old paradigm of treatment. However there does exist a wealth of knowledge from the personal stories of those treated under the interventionist paradigm, about the damage that surgery and non disclosure about their bodies and diagnoses has inflicted. Parents may ask whether their child will be recognised as the assigned gender without the surgery. The question should perhaps be 'recognised by whom?' Does the way your genital anatomy looks determine who you are as a person or whether you accept your child? Dreger (2001) stated that when people ask if your child is a boy or a girl you don't have to show them the genitals to answer. Howe (1998) asked if parents could overcome any prejudices they have and provide care and support for their child. While this may be a very real issue these authors stated that surgery is not an answer. Surgery does not ensure "normal" looking genitals and risks function and sensitivity. Furthermore it conveys and reinforces a message that there was something wrong or unacceptable about the genitals and the child to start with, thereby increasing any feelings there may be of shame, guilt and confusion. Howe (1998) commented on the fact that surgery actually serves to undermine parents' ability to love and accept their child unconditionally. Groveman (1998), in her experience of working with AIS families, found the most critical variable in achieving a better outcome for intersex people was not surgical management, but resources which educate parents and work through any anxieties and guilt. When parents are able to communicate their understanding and comfort with intersexuality then the child's self esteem can develop.

For the children themselves, Howe (1998) noted that we had yet to develop maximal approaches to providing help. What is clear is that children will need age-appropriate, truthful information. Groveman (1998) acknowledged that knowing the truth about being intersex can be temporarily traumatic, but it is nothing compared to the impact of discovering later in life you were lied to and deceived by those claiming to care most about you. Children need to become a part of the decision-making process, and to enable this must have open and accurate information and communication about

their bodies. The decision to proceed without surgery allows the individual a choice. Who are parents and physicians to decide whether or not an individual would be happy with their genitals?

Tamara Alexander (1997) looked at the links between children treated for intersex conditions and their experience of trauma, as compared to children who had been sexually abused. There is direct contact with the genitals by a person in power with the co-operation of the parents. Procedures can be painful, confusing and repeated, with children silenced or misinformed. She observed the same sequalae in both groups including depression, body image disruption, intimacy issues and post traumatic stress disorders. Fausto Sterling (2000) commented on the trauma of measurements of penile growth necessitating doctors masturbating boys, and girls requiring a dildo to be inserted into their vaginas to keep them dilated following vaginoplasty. There are also links in the literature between surgeries carried out on intersex children and the practice of Female Genital Mutilation (FGM). Holmes (1995) commented that FGM is seen as an act of violence which decreases sexual function and pleasure for women. How different is that from removing sexual, erectile, genital tissue from intersexed children? Why does western society condemn FGM yet ignore similar surgeries on intersex infants? Dreger (1998) suggested we should consider the historical and cultural basis for genital conforming practices, noting that if it is necessary to protect the rights and well being of African girls, it is equally important to consider the well being and rights of intersex children. Kipnis and Diamond (1998) questioned whether physicians should ever sacrifice the organic functionality of any nonconsenting child on the altar of cultural expectation.

Issues which accompanied the initial suggestions for a change in practice can be divided into two broad categories. Firstly there were the assumptions that have been made surrounding gender identity, and who and what determines this for an individual. Along with this are definitions of male and female, and what constitutes "normal" genital appearance and function. Secondly there were issues surrounding deception and the lack of accurate information for individuals and their parents, and the effect this has had. These issues encompass the area of informed consent and the rights of intersex infants and children versus the rights of their parents and physicians to make decisions which have been deemed as being in their best interests.

In the Child's Best Interests?

The base assumptions of society are that there are two genders which everyone can be assigned to, and that there is a correct way to behave within these genders as male or female. If these beliefs are challenged in any way, especially by the birth of an intersex child, then children can be reconstructed to fit the beliefs. Coventry (2000), in Ms Magazine, observed that the strict division between male and female bodies and behaviours is our most cherished and comforting "truth". Within these assumptions is an expectation of what male and female genital anatomy should look like and be capable of. Dreger (1998) noted that while an adequate penis must be of a certain size, have the capacity to become erect and flaccid and pass urine and semen at an appropriate time via an appropriately placed opening, for a reconstructed vagina to be a surgical success it needed only to be big enough to accommodate the aforementioned adequately-sized penis. Lubrication and sensation are not noted as important. Likewise a clitoris needs to meet a size stipulation, with sensation seen as a secondary consideration. This also assumes that sexual intercourse is the most important thing a human does (Lewis, 2000). Holmes (1994) noted that this thinking also presumes that a heterosexual mode of penetrative sex by the male is the appropriate one. A female could not possibly have a clitoris that may be capable of penetrative sex. Holmes goes on to comment on a patriarchal desire to protect the rightful place of the phallus and place value on its size. Medically and culturally male bodies must have penises and female bodies must not. There is no allowance for an individual to possess a combination of external or internal genitalia usual to both sexes. Genitalia must be strictly male or strictly female, reinforced with surgery and hormones where necessary. Fausto Sterling (2000) viewed these beliefs as Aristotelian categories which are not found in nature. Nature creates a whole lot of different forms. In addition, female fertility is valued and preserved whereas male fertility has often been sacrificed with a reassignment to female gender and the removal of testes. As Chase (1999) noted, with new infertility treatments there is the possibility of fathering a child in the absence of a penis.

The medical literature clearly voiced the concept that the birth of an intersex infant constituted a medical emergency (McGillivray, 1992; Myers-Seifer & Charest, 1992). While Congenital Adrenal Hyperplasia needed to be considered and electrolyte

imbalances treated promptly, the extension of the term medical emergency to the genitals and the implication that they too must be treated promptly lacked support. Dreger (1998) stated that ambiguous genitalia do not constitute a disease but instead a 'failure to fit a particular (and at present a particularly demanding) definition of normalcy' (p.8). Similarly Kessler (cited in Dreger, 1998) proposed that intersexuality does not threaten the patient's life: it threatens the patient's culture.

Another question this debate raised is what determines gender identity? Is it genetic makeup, prenatal hormonal environment, dependant on physical and genital appearance, a social construct or determined by an individual as a child, teen or adult? Also do perceptions about the relevance of all these factors within society play a part? The instances of intersex infants, who were raised as one gender and migrate back to a gender that would seem to be determined by their genes and prenatal hormonal environment, would appear to disprove the theory that gender identity is determined by the presence of unambiguous genitals and the social reinforcement of the assigned gender. The number of both genetic males and females, with genitalia that would be construed as normal under this paradigm, who as children, teens or adults feel uncomfortable in their gender and choose to change gender either temporarily or permanently with surgery and hormones also raises questions around the determinants of gender identity. Dreger (cited in Lewis, 2000) observes gender identity as being complicated, with various components interacting and mattering in different ways for different individuals. Fausto Sterling (2000) looks at male and female as standing at extreme ends of a biological continuum, with bodies containing a mix of anatomical components appearing between these ends. She contests that if nature offers more than two sexes then binary notions of male/masculine and female/feminine are merely cultural conceits. Mason (2001) comments that this uncertainty as to what determines gender identity increases the inherent dangers of operating on an intersex child. Not only may it destroy future fertility and sexuality but it may also compromise a chosen gender identity.

A further assumption behind the surgical treatment of an intersex child is that having "abnormal" genitals would psychologically damage such a child. It is only the combination of early surgery and parental conviction of the chosen gender that is meant to ensure the child will be happy and grow into a well-adjusted heterosexual adult (Chase, 1999). Dreger (1998) commented on the belief that talking truthfully with intersexuals and their families would undo the positive effects of the surgery. However

the stories of adult intersexed who have undergone surgical reassignment indicate that rather than resulting in feelings of being "normal", the surgeries and repeated medical attention emphasised how different they were. Howe (1998) suggested that the insistence on early surgery only reinforced to parents that the condition is a source of shame. The pressure placed on parents to raise their children unambiguously has led to a web of silence and deception, between the medical establishment, parents and children, and between parents and children themselves. Complete medical histories and the true reasons for surgeries have been hidden. It is the combination of this deception, repeated medical attention to their genitals, and the resultant surgery which appear to have caused, rather than prevented, psychological harm to a large number of intersex individuals. Dreger (1998) asked the question; why would you treat a psychosocial problem with surgery?

Preves (1998) who conducted in-depth life history interviews with intersex adults found that medical attempts to destigmatise intersexuality were in fact experienced as alienating and shaming. While Preve found there was a rich diversity of sexual identities and opinions, all those interviewed conveyed the conviction that being poorly informed, and encouraged to keep silent, about their differences and surgical attentions only enforced feelings of isolation, stigma and shame. Malin (cited in Dreger 1998), on talking with adult intersexuals, found that they were certain they would rather have kept the "abnormal" genitals they were born with than the "mutilated" genitals they were given. Sherri Groveman (1998), the US representative of the Androgen Insensitivity Syndrome (AIS) Support Group, says of her childhood:

> I spent my adolescence filled with shame, though I was never told the true details of my diagnosis. My trauma was needlessly compounded by my doctor's stony silence while examining me, and his asking me to lie naked on the examining table so that teams of interns and residents could inspect my genitals. Such experiences themselves, far more than the true facts I later learned about the nature of AIS, instilled a sense of freakishness that I have only recently shaken. (p.358)

Preves goes on to comment on the toxic mixture of silence and surgery which has destroyed relationships between parents and children. This deception and partial disclosure of the facts also raises the issue of informed consent. Can a parent give informed consent for their child and can older children receiving surgeries be informed if they are not made aware of their medical histories? Is it informed consent if they are not made aware the surgery is for cosmetic rather than medical reasons? Is it informed

consent if parents are not made aware of the lack of research or the controversial results surrounding the surgical and psychological outcomes of early surgical intervention? Mani Mitchell (2000) of the Intersex Trust Aotearoa New Zealand asks what the child's rights are and who speaks/ advocates for the child? When is it okay to treat parental distress by treating the children? Chase (1998) noted the underlying attitude that intersexuality is so shameful it must be erased before the child can have any say in what is done to their body. Holmes (1995) claimed that decisions are often based on the assumption that children are their parents' property and Dreger (1998) comments that parents are often receptive to offers to "normalise" their child's genitals.

A combination of inadequate or misleading information and parents' own fears and prejudices, often reinforced by the medical attention their child receives, means they see themselves acting in the best interests of their child. Holmes (1995) commented that while parents of intersex children may deserve some sympathy for being misled, these parents are also responsible for educating themselves about proposed medical treatments. She goes on to question whether "it is ethical to permanently damage the sexual organs of some people just so that those who (don't) care for them can be made more comfortable"? (p. 8). Beh and Diamond (2000) questioned whether any parent would consent to such extensive treatments if the lack of scientific evidence they were based on was revealed, or if they were told that the child might ultimately reject the treatment and be worse off for it. Kon (2006) in reviewing the book 'Surgically Shaping Children: Technology, Ethics, and the Pursuit of Normality' describes this debate in terms of the tension between the wish for children to look "normal" to gain acceptance in society and the desire to alter society to become more accepting of those who are "different.".

The issue of informed consent in regard to genital surgery on infants is currently under debate in the ethical, legal and medical literature. The traditional paradigm of care has presumed that physicians, and under their advice parents, have been acting in the best interests of the child by consenting to surgery. The ethical and legal debate focuses first on whether the information given to the parents was adequate and comprehensive enough to enable them to make that choice. It then goes on to question whether that is in fact a choice for parents to make. Should parents be able to consent to surgery on their child or should the decision be the child's when they reach an age to be deemed able to give informed consent. Greenberg (2003) in a review of

the Legal Aspects of Gender Assignment states that if parents consent based on incomplete information then that consent may not be valid. She notes that because of the desire of physicians to have the child identify with the chosen gender assignment, physicians may downplay or omit information to make parents more comfortable with their decision to approve surgical intervention. The San Francisco Human Rights Commission investigation into the medical 'normalisation' of intersex people (2005) noted that many parents have chosen 'normalising' interventions based on misinformation and/or coercion from doctors.

As an infant is unable to make a decision on surgery, and the push from the medical profession has been for early intervention the decision has thus been conferred on the parents. Lareau (2003) questions whether in fact by asking parents to give consent physicians are diminishing their incentive to consider whether 'normalising' genital surgery is medically necessary. Greenberg (2003) comments that parental decisions are generally accorded a significant degree of deference because legal institutions generally presume that parents will make decisions that will be in the best interests of their children. Hughes, Houk, Ahmed and Lee (2006) in the Consensus Statement on Management of Intersex comment on medical law and practice in the United States and note that courts assume parents know what is best for their child with parental decisions being deferred to unless a lifesaving treatment is withheld. In looking at best interests Carnevale (cited in Woods, 2007) notes that it is 'problematic for a parent to distinguish the child's interests from the parents' own interests' (p.241). Is choosing surgery actually managing the parents discomfort with their child's genitals? Greenberg (2003) states that ' although parents believe they are considering the best interests of their children when they make their treatment decision, it is difficult for parents to rationally assess whether they are focusing on their need to have a "normal" infant over the long-term interests of their child.'(p.283). The San Francisco Human Rights Commission (2005) notes that the Columbian Court, in making their 1999 decision to litigate around parents rights in providing informed consent for surgery on intersex children (see p.22), found that parents are more likely to make decisions based upon their own fears and concerns rather than what is ultimately best for the child.

Frader et al. (2004) reporting on a multidisciplinary group convened by The Hastings Centre, New York considering medical, psychosocial, and ethical issues surrounding the care of children born with atypical genitalia or later found to have intersex conditions stated that normalising surgery without the patients' consent lacks

ethical justification. Lareau (2003) states that parents should not have 'unfettered discretion' to consent to genital surgery as it causes categorical conflicts. These are conflicts that arise when a parent has an impaired ability to consider the best interests of the child. Impaired ability may result from such things as a fragile emotional state or fears around having an 'abnormal' child. This can then lead to the child's rights, such as the rights to erotic and reproductive potential as an adult, being possibly impaired by a medically unnecessary surgery in childhood. In argument to this Eugster (2004) proposes that withholding genital surgery overlooks the fundamental role of parents in ensuring a child's physical and emotional well-being. If a family can not accept their child with unaltered genitals then that would be detrimental to the child therefore consent to surgery is justified. She concludes that if parents maintain a prejudice for surgery despite a complete education about the risks then denying the surgery could be more disastrous than consenting. Therefore 'affording parents this authority does not present an ethical dilemma, since in our society all major decisions regarding minor children are traditionally made by parents' (p.429).

In New Zealand the legal age of consent is 16 years (Woods, 2007). For children under 16 years the issue of consent becomes more complicated. Woods (2007) notes that since 1993 the law relating to treatment decisions in children became the same as the law for adults, children are seen as individuals. The Code of Health and Disabilities Services Consumer Rights allows an assessment of competency which applies to young children. Woods (2007) states that 'if the consumer is not competent to make an informed choice, then it is expected that another person, who is entitled to consent on behalf of the consumer, will be allowed to consent on their behalf' (p. 58). This process can then be regarded as the norm in consenting to medical treatment for those less than 16 years. Parents are then left being asked to consent to treatments on behalf of their child while the child has rights under the above code to consent to the procedures themselves as far as competency allows. In regards to consenting to genital surgery the rights of parents to consent in New Zealand has not been tested in law. The fact that these children, in general, are funded to travel to Australia for surgery means that the parental consent to surgery is being sought out of New Zealand. However, it could be argued that at an institutional or state level parental decisions to go to Melbourne for surgery are supported, as the New Zealand state pays for this surgery and New Zealand clinicians, as gatekeepers in terms of access to funding, would need to support parents in this decision. In commenting on the Australian perspective in

regard to genital surgeries at Royal Melbourne Hospital, in the Cares Foundation Newsletter 2005, Elizabeth Kennedy, Corporate Counsel notes that parents of girls with CAH need a thorough understanding of the condition, the treatment options and proposals offered by the health care professionals to give proper consent and cautions that taking legal advice might be appropriate.

This then leaves two options, that parents do have the right to give consent or that they do not. If they do have the right to consent, then the focus needs to become making them fully aware of the controversies that exist in relation to the surgery. These include the outcomes and risks associated with surgery such as alterations to sensation and function, the questions around psychological well being in terms of either an individual undergoing a surgery which they did not consent to or having surgically unaltered genitals which are not considered 'normal' or 'conforming' for their assigned gender of rearing. Informed consent should also include information on the ethical debates surrounding whether parents can and should give informed consent. Lareau (2003) notes that there is a belief by some, that if parents were adequately informed they would decide against surgery anyway. Ford (cited in Lareau, 2003) states that 'the fact is that there is just not enough accurate information available on the benefits or consequences of genital-normalizing surgery for even the most well-meaning and contemplative parents to make truly informed decisions for their infants'(p.11). If parents do not have a right to give informed consent this then must become a matter of law within specific countries. For a moratorium against all genital surgeries to exist the issue would require debate as a legal matter. Greenberg (2003) notes that in the United States as yet parental consent to intersex surgery cases has not been litigated. This is because no one has a stake in bringing a lawsuit if the parents and physician are in agreement. However in view of the current controversies and the laws in Columbia this may be a future possibility.

Clinical Management- Contemporary Issues

Despite the lack of agreement among health and legal professionals relating to appropriate clinical management outlined above, there is now general agreement around being open and honest with children and their parents. There is an increased awareness of the need for support and education for the parents, and the importance of offering contact with support groups. Of interest is a follow-up study done by John

Hopkins hospital with John Money as one of the co-authors (Migeon et al. 2002). They followed up 96 intersex patients and determined that almost half of them were neither well informed about their medical or surgical history nor satisfied with their knowledge. In a report of their experience over 25 years with 250 patients, the Gender assessment team from the Children's hospital and Regional Medical Centre in Seattle (Parisi et al. 2007) comment that psychosocial support for families is a priority with their practice including a large educational component and resources from web based sites. The Seattle based Gender Assessment team give parents age appropriate recommendations for talking to their child in an honest, non stigmatizing way. They look to a future which includes partnership between clinicians, advocacy groups and parents. The consensus statement on Management of Intersex Disorders (2006), notes the importance of a multidisciplinary team, the important role of support groups and also open communication with the family.

The view that undervirilised XY infants should be reassigned female with feminising surgery no longer appears to have support. Parisi et al. (2007) note that a theme of their management is to match sex-of-rearing with chromosomal and gonadal sex wherever possible. Reiner (2005) in a review of 84 patients looking at declared gender identity as opposed to sex of rearing, concluded that genetic males with male typical prenatal androgen effects should be raised male. Genital surgery, however still remains a controversial issue. Recommendations vary from the avoidance of all surgery until the child can provide informed consent, through a suggestion that some procedures still be carried out on certain conditions through to a continuation of early surgery.

There are a number of recent studies which continue to report poor outcomes for vaginal and clitoral surgery. Gollu et al. (2007) looking at the results of 85 children managed surgically for ambiguous genitalia at the Ankara University School of Medicine in Turkey between 1988 and 2005 found that 29% of patients with feminising procedures and 34% of those with masculinisation procedures experienced complications requiring repeat operations and recommended delaying vaginoplasty until adolescence due to vaginal stenosis. Alizai, Thomas, Lilford, Batchelor and Johnson (1999) looked at the postpubertal outcome of feminizing genitoplasty in girls with CAH. They found very poor results with 13/14 girls requiring additional vaginal surgery and unsatisfactory clitoral surgery in 42% of

patients including atrophy. These authors also recommended delaying vaginoplasty until after puberty. Poor outcomes for vaginoplasty requiring repeat surgery were also reported by Stikelbroeck et al 2003. Krege, Walz, Hauffa, Körner, and Rübben (2000) looking at follow up for patients with Congenital Adrenal Hyperplasia also stated vaginoplasty should be undertaken at the beginning of puberty due to high levels of stenosis and further commented on the importance of psychological support in the treatment of children with CAH. London based gynaecologists Sarah Creighton and Catherine Minto have published, in collaboration with others, a body of work that has taken a comprehensive look at the outcome data and issues surrounding genital surgery in Britain. Creighton, Minto and Steele (2001) in their retrospective study of 44 adolescents showed results which concluded that most vaginal surgery could be delayed till after puberty and that genital ambiguity could not be corrected in infancy by a single procedure. A 2003 study by Minto, Liao, Woodhouse, Ransley and Creighton found when comparing adult women with DSDs that the group who had undergone surgery as opposed to those that had not, had a significantly higher rate of orgasmic difficulties and lack of sensation.

In 2004 Creighton reported on the London experience of long term outcome in feminizing surgery. She comments on the proponents of genital surgery citing stable gender identity, better psychosexual and psychosocial outcomes, and relief of parental anxiety as reasons for early genital surgery. She notes there is no evidence that surgery promotes a stable gender identity and reports on research at University College London Hospital where they have a multidisciplinary adolescent and adult intersex clinic which has conducted a number of follow-up studies. Results showed repeat procedures were common and could be avoided by delaying vaginal surgery to puberty, cosmetic clitoral surgery did not improve adult sexual function and might indeed cause damage and that genital sensation in women with CAH who had undergone surgery was markedly impaired. She comments that while cosmetic results on infants may relieve parental and clinician concerns there is scant evidence of "satisfactory postpubertal cosmetic or anatomical outcome" (p.45). Therefore the option of no infant genital surgery must be discussed with parents.

The aim of feminising surgery seems to be to provide a 'normal appearance' to female genitalia including clitoral size. In a paper in 2005 Lloyd, Crouch, Minto, Liao, and Creighton comment that there are surprisingly few descriptions of normal female

genitalia in the literature therefore when considering genital surgery the decisions regarding how much reconstruction is required and what constitutes a normal genital appearance must be completely subjective. They suggest educating parents on the wide variations in both appearance and dimensions of female genitalia. In a 2007 review of papers on long-term functional outcomes of female genital reconstruction in childhood Crouch and Creighton still conclude that vaginoplasty should be delayed until puberty and that all clitoral surgery will risk some damage to sensation and state that: "Parents and clinicians should be clear about this when planning and agreeing to genital surgery in childhood" (p.406). They also comment on the subjectivity of the words 'successful outcome' when publishing studies on genital surgery. Issues include a lack of information on pleasure or sensitivity, and the surgeon or author of the study rather than the patient or parent determining a pleasing cosmetic appearance or satisfactory anatomical function. They go on to question studies which report good functional outcomes in children, arguing how do you determine this if children are not sexually active? Reiner (2004) also questions the validity of research report outcomes stating that medical and surgical studies do not provide clear definitions of cosmetic appearance. In looking at reports in the literature of good surgical outcomes it then becomes important to carefully consider the definitions of 'good'. Sircili et al. (2006) report good results in 68% of patients with CAH undergoing feminising surgery but good is defined as "vaginal introitus stenosis adequately treated by vaginal dilations with acrylic molds" (p.211) and regular results as also requiring vaginal dilation but accompanied by pain and bleeding during intercourse.

Further recommendations are offered by the Consensus Statement on Management of Intersex Disorders (2006) which resulted from an evidenced based review of a large number of studies. Their comments on surgery following this review were;

> Some studies suggest satisfactory outcomes from early surgery. Nevertheless, outcomes from clitoroplasty identify problems related to decreased sexual sensitivity, loss of clitoral tissue and cosmetic issues. Techniques for vaginoplasty carry the potential for scarring at the introitus necessitating repeated modification before sexual function can be reliable. Surgery to construct a neo-vagina carries a risk of neoplasia. The risks from vaginoplasty are different for high and low confluence of the urethra and vagina. Analysis of long-term outcomes is complicated by a mixture of surgical techniques and diagnostic categories. (p.493)

The authors of the Consensus Statement go on to identify that there are no controlled clinical trials of the efficacy of early (less than 12 months of age) versus late (in adolescence and adulthood) surgery or of the efficacy of different techniques. Their recommendation is that the surgeon must outline the surgical sequence and subsequent consequences. Surgery is recommended only for severe virilisation with emphasis on functional outcome rather than strictly cosmetic appearance. For hypospadias, chordee correction and urethral construction are recommended. They also offer recommendations for the removal of testes in those with CAIS and PAIS. Surgical management should also consider facilitating the chance of fertility.

ISNA published in 2006 its' Clinical Guidelines for the Management of Disorders of Sex Development in Childhood and a Handbook for parents. While acknowledging the ongoing debates about 'best care' it aims to present a model which minimizes potential harm. They continue to recommend no surgery for cosmetic purposes until the child can make an informed decision. They state that parental distress should be addressed with competent mental health care and peer support.

Other studies published since 2000 (Farhat, 2005; Lean et al., 2005; Warne et al., 2005) however recommend early genital surgery continue as previously. Farhat (2005) publishing on the Toronto experience with surgical management of CAH states "we perform a perineal reconstruction as early as possible to minimize the period of gender uncertainty" (p.66). He goes on to comment that only a prospective study comparing early and delayed surgical interventions would change their current policy. Their feeling is the parents and child should not have to anticipate surgery in adolescence. This would however appear to ignore the data that reports these girls do in fact need repeated surgical management in adolescence anyway.

It is important to note that the majority of published reports describe the experience in Britain, Europe, and the United States and are not necessarily reflective of a single institution which has specialised services for intersex surgeries. Of interest is the 2005 follow-up report from Royal Children's Hospital (RCH), Melbourne as this is where the majority of children from New Zealand now travel for genital surgery. Lean, Deshpande, Hutson and Grover (2005) follow up on 32 patients, 47% with CAH who underwent genital surgery at RCH. They employed a standardised approach, also used by Creighton to enable the possibility of comparing outcomes between institutions and countries. They report an overall good (88%) cosmetic and anatomic outcome with patients who received 1-stage genital surgery. When

comparing their results to Creighton's poorer outcome results the suggestion was that the centralised service and specialised surgeon and team contributed to the difference. Their poorer results were in those patients who did not receive initial surgery at RCH. They note that the age of reconstruction did not have a significant impact on outcome but felt the results did not support abandoning genital reconstruction in childhood.

A further study from RCH, Warne et al, and (2005) which looked at long-term outcome for intersex conditions in terms of psychological, sexual and social outcomes found that most patients had positive psychosocial and psychosexual outcomes, although some problems were reported with sexual activity. They concluded that their results supported a model of care which included early genital surgery carried out at a centre of excellence with a multidisciplinary team. In a statement published in the Cares Foundation Newsletter 2005, John Hutson, Pediatric Urologist and Professor of Pediatric Surgery in Melbourne discusses his approach to parents regarding surgery on virilised girls with CAH:

> My plan thereafter is to see the families for discussion in the clinic about the various surgical options and the surgery required with regard to the amount of surgery and timing of surgery, etc. I put to the parents the alternative views of having surgery in early infancy or delaying this until later. I also ensure that they understand my own personal bias which is that early surgery has many significant advantages and very few disadvantages if done well. (p.5)

Despite these further follow-up studies and recommendations there still remains a spectrum of care which an infant with a variance in chromosomal, gonadal or anatomical sex may receive. This is dependant on the views and recommendations of the health care facility they attend. While some major US, Canadian and European hospitals have established protocols and a dedicated team approach others rely on the recommendations of individual physicians. Columbia remains the only country, which at present, has a law governing surgery on infants with DSDs. Therefore there is now a challenge for health professionals working with those born with these conditions to consider which paradigm of care they will work within. There is a choice to work within the existing paradigm while awaiting the further results of long term follow up studies, with an acknowledgement that informed consent requires parents have all the information available when making decisions. There is also a choice to work within a new paradigm of care which realises the rights of those born intersex to make their

own choices. These include choices around gender identity, the right to truthful information and to give informed consent to any medical or surgical intervention. What appears to be fundamental is the need to facilitate support and counselling for children and their parents so that children can grow up feeling empowered and with a healthy sense of self and body image.

Information Needs

In order to be able to work with the families of these children there is a need for practitioners to know what information parents/ families require to make decisions and provide support for their child. There has been little dedicated research into the information needs of parents when their child is born with an intersex/DSD condition. Reiner (2004) notes that what health professionals need "to know for appropriate interventions for these children and their parents is precisely what is lacking" (p.51). Ellen Feder (2002) comments in a book 'Feminist Perspectives on Dependency', on the lack of information that addresses the experience of parents making decisions about surgery. She states that parent's isolation and the striking failure to take into account their experiences were essential components of the medical paradigm of care around intersex. She interviewed three families, one with two children born with CAH in the early 60s, one with a child born with AIS which was discovered at age 12 years in 1986, and a family with a child born in 1990 with extra X chromosomes. She interprets their transcripts in terms of the concept of dependency. She refers to parents as 'dependency workers' - those who care for the inevitably dependant (children). As dependency workers parents then have to rely on others to provide resources, which they need to do their work as caregivers. In relation to medical issues everyday 'social knowledge' does not provide a parent with professional or technical information and they must depend on health professionals for 'expert knowledge'. It is the content or nature of this expert knowledge that than affects the decisions made and the ensuing relationship between parent and child. Of the two families with now adult children, Feder reported that all three children had felt violated by the surgery performed, constant examinations, and the secrecy surrounding their conditions. This had led to ongoing issues between the children and the parents who had made those decisions. Feder comments that the parents were not given the chance to imagine their children's lives in any way except as in need of immediate correction. She suggests that as dependency workers parents need to claim their rights to secure the future autonomy of their

children and recognise the child's right to choose surgery. Siminoff, Mercer and Sandberg (2007) looked at how families of children with CAH were making healthcare decisions. They found parents reported incomplete and inadequate information was received from healthcare providers for their needs as parent care givers and decision makers. The parents also felt they received incomplete or contradictory information on surgical issues.

As most parents with a child born with an intersex/DSD condition will either be in the NICU (Neonatal Intensive Care Unit) or have contact with NICU or paediatric staff it is useful to review the literature on parental needs surrounding information and how parents view information given by health professionals in these settings. In this setting there is a body of literature that examines the concept of information as support and as caring. Saunders et al (2003) comment that information sharing and collaboration are the cornerstones of family centred care. In a study of the nature and organisation of maternal needs in the NICU Bialoskurski, Cox and Wiggins (2002) found that 93% of mothers stated that the need for accurate infant related information was a priority. Consistent advice and information was needed in various formats to suit the needs of parents. This needed to be accurate, reliable and topical with questions answered honestly. Conner and Nelson (1999) reviewed literature on parent satisfaction and identified 11 dimensions of care which were important to parents. These included communication, consistent information and education. They found that communication including the sharing of factual information was the most reported domain of satisfaction for parents. They found that parents who were told of their child's diagnosis at birth or early in their child's life were more satisfied than those told later. In reviewing the range of support nurses can give Miles (2003) defined four overlapping components of support which included communication and information. This included helping parents to understand information they had been given and knowing their rights and responsibilities. The quality of this information/support affected parent's abilities to cope with an immediate crisis, the development of their parental roles and their competence in providing care post discharge. Hurst (2002) comments that based on reports from hundreds of families she has worked with a persistent issue for families is their difficulty in obtaining "empowering information" (p.41). She defines empowering information as that which is readily accessible, pertinent and understandable. It includes information about health care issues and

needs both within the NICU and beyond. Obtaining empowering information is requisite for kind, compassionate care.

The range of DSD conditions, uncertainty of paradigms of care, a lack of consensus regarding evidence based care, political debates with different communities of people with intersexed conditions, and the lack of research on information needs for parents outlined above suggest the need for a qualitative interview based project. This research would need to focus on the experience of individuals with and parents of those with intersex/DSD conditions with particular regard to their information needs.

Summary

Over the last fifty years the clinical treatment of children with an intersex disorder has been based on a medical paradigm of care arising from John Money's work in the 1960s. This paradigm focused on the birth of a child with an intersex condition as a 'medical emergency' with the genitals needing immediate surgical attention to reinforce a chosen gender of rearing. In the 1990s adults, who had been treated under this paradigm which involved secrecy and non disclosure of the true nature of individuals conditions, the surgery performed and the reasoning behind the assignment of a gender of rearing, began to speak out. They expressed feelings of shame, betrayal and isolation resulting from their treatment. At this time the ISNA proposed a new paradigm of care with genital surgery being withheld until an individual could choose and consent themselves. Medical and social literature also began to question the ethics of a paradigm which continued with few long term follow up studies and questionable clinical outcomes. Issues arising in terms of ethics and legality included whether genital surgery should be continued on infants and whose right it is to consent to surgery for an individual, and choices around an assignment of a gender of rearing based on genital appearance or size. Mirroring this ethical debate has been the issue of nomenclature (discussed in the introduction) and the use of the umbrella term intersex to reference a number of conditions. The term DSD which was proposed in 2006 also raises debate and controversy. There also exist a number of conditions which fall under these umbrella terms for which genital surgery is not an issue, but which

under the medical paradigm of care, have still been treated within a framework of non disclosure and secrecy.

It appears that information is a priority for parents when their child has a condition requiring contact with healthcare professionals. The quality and depth of this information, then impacts on their decision making ability and on their ability to give informed consent. In the case of children with intersex/DSD conditions the decisions made in the neonatal period will impact the rest of their lives. It seems important therefore to do further research into the information needs of parents and families making these decisions. This is especially relevant in view of the continued controversy and debate about treatment options.

Chapter 3

NOTES ON METHODOLOGY

This chapter presents the qualitative methodology employed to look at the research question of parental/whanau information needs when a child is born with an intersex/DSD condition. It reviews exploratory research as a methodology and presents the ethical issues which arose prior to commencing the research. Recruitment of participants and the interview process, data collection and management of the data are presented. The method of thematic analysis used to explore the narratives gained via the interviews is presented along with the plans for implementing and disseminating the research findings. At the time the research was proposed, ethics approval sought and participants recruited, (2003-2006) the term DSD had not been proposed therefore the term intersex will be used in this chapter in reference to the research.

Methodology

When trying to ascertain which information would be useful for parents as they received the news of their child's condition and began to make decisions, it became apparent that there was little dedicated research into the needs of parents when their child is born Intersex. There was sparse literature which looked specifically at parents of intersexed children. Ellen Feder (2002) interviewed three families and comments on the lack of information that addresses the experience of parents making decisions about surgery. There was also no research found on the experience of parents who were not having to make a decision about surgery but still had a child with a condition considered intersex. This lack of existing research pointed towards new research of an exploratory nature.

Nieswiadomy (1998) states exploratory studies are conducted when little is known about the phenomenon of interest. Brink (1998) describes the goals of exploratory research as being problem discovery and problem definition. A problem is identified and described, then explored in depth in a free-ranging way to form a

description of the experience. The assumptions underlying the design are that the topic has not been previously studied from the point of view of the informants and that the sample has personal experience or knowledge of the topic. Carnwell (1997) describes exploratory research as an approach in which the researcher explores a particular aspect of health or social care in order to seek the opinions and feelings of the participants. It is concerned with people's experiences and needs. Once the needs of particular clients or patients become apparent future services can be planned to meet their needs. The aim of this research then became focused around exploring the information needs of parents/families/whanau when their baby is born Intersex. It was hoped that by asking people about their experiences of having a child born with an intersex condition the information they provided could eventually be used in a resource for other parents.

A Qualitative Exploratory Methodology was employed with a plan to interview representatives from four key stakeholder groups. An exploratory design using small samples and unstructured interviews provides the researcher with flexible techniques with which to gain insight into the phenomenon (Brink, 1998). Within the exploratory methodology a responsive approach was used. Carnwell (1997) describes this approach as seeking to respond to issues from multiple perspectives and listen to multiple viewpoints by seeking the views of all the stakeholders. The stakeholders identified with parental information needs were, the parents themselves, those born intersex, intersex advocacy groups and medical professionals who become involved with these children and their families. It must be acknowledged that one of the main stakeholders could be identified as the children themselves, however the scope of this research did not allow for the interviewing of children with intersex conditions. It was hoped that by speaking to parents and adults with the conditions that some of the needs and experiences of the children themselves might be canvassed.

Carnwell (1997) describes the benefits of an exploratory approach to research as sensitivity to multiple viewpoints and an ability to deal with poorly focused concerns and conflicting views. The richness of the data assists in understanding a variety of issues. It was envisaged that the four groups of stakeholders might present very different viewpoints in terms of their experiences of intersex and their opinions of the information which parents/families may need to make an informed decision about their infant. In coming to this conclusion I drew on the different positions and experiences portrayed in written texts and examined some of the assumptions I myself

brought to the research from having undertaken a thorough review of the literature. Demi and Warren (1995) state, that in order for qualitative data to be deemed trustworthy the investigators must evaluate themselves as data collection instruments. Demi and Warren suggest that to be cognizant of ones own assumptions and ones tendencies helps minimize biases in what is eventually reported. Morgan and Drury (2003) also note the need for a researcher to document any insights or biases that may permeate and influence the interpretation of the data. Based on this the following issues and presumptions were documented and discussed with my supervisor.

Parents

From a parental, familial and societal aspect there is an expectation during pregnancy that parents will have a healthy, full term female or male baby. When a baby is born intersex and differs from what the parents expect they experience a grief reaction which may include emotions such as shock, anger, denial, and guilt (Groveman, 1998). In the midst of this grieving, parents are asked to make important decisions regarding their baby. Whether an informed decision is made is largely dependant on the nature and degree of information they are given. Parental decisions might be affected by a number of things including their desire to have a baby which looks 'normal' to them and others, and fears over how family/friends and others will react to their baby (Howe, 1998). Often there can be confusion between gender identity and sexual orientation as well as their own issues/fears around gender, gender roles, sexuality and sexual orientation.

It is presumed that parents want what is best for their child and that they have a right and an obligation to find out all the information they can/need to make an informed decision. However they may also see the advice of a medical practitioner as the 'right thing to do', and be presented with the opinion that the condition is one that can be 'fixed' with surgery. Dependant on where their infant is born they may only receive the opinion of one, or a limited number of practitioners. Alternatively they may be presented with, or have access to information which includes numerous viewpoints including those for and against early surgery. If parents want to decide against early surgery they may face opposition from health professionals. Another issue which has arisen out of the literature is that it may not even be the right of the parent to make a decision about early genital surgery on an infant (Holmes, 1995). I was aware that I

might be talking to parents who had made a decision which was questioned or even criticised in the literature. There was also the possibility that the focus on surgery apparent in the literature was not a major focus of concern for the parents.

Adults with Intersex conditions

It is probable that adult intersex people were treated under a medical paradigm that involved secrecy and lies surrounding their condition and treatment. They may not have been aware of the reasons behind their genital surgery. It was envisaged that there may be emotional and psychological issues related to this that would come out in the interviews. Their views on the information parents would need would be coloured/influenced by their own personal experiences and stories.

Health Professionals

It was considered that health professionals might be embedded in the medical paradigm of treatment and therefore see the exploratory research as questioning their practice. Health professionals were also being sought from the largest paediatric service these families come in contact with, so it was acknowledged that their views might not necessarily reflect the opinions of those around the country.

Advocacy Groups

Intersex Advocacy groups, by their own admission, have a political agenda to change the paradigm of care intersex infants receive (ISNA website). This would be reflected in the answers they provided to the research.

It was hoped that by examining extracts from conversations with health professionals, advocacy groups and the narratives of parents and people who identified themselves as intersexed or whose conditions were encompassed by the umbrella term intersex, I would be able to identify themes at work in this area.

Ethical Issues

Before it had begun the research raised a number of ethical issues. These included informed consent, and the confidentiality and involvement of vulnerable families and individuals. Demi and Warren (1995) describe vulnerable families as those that are susceptible to harm because of their socioeconomic status, their minority status or other stigmatizing status. In terms of this research having an intersex child or being intersex themselves may make the respondents vulnerable. Demi and Warren also state that sensitive research with vulnerable families is necessary to gain a better understanding of the needs and resources of these families and that the results may form a basis for change that empowers these families. It was hoped that by being given the opportunity to share their experiences and express their information needs, and the issues which surround these, respondents would feel they had been listened to and their opinions considered when producing a resource for those in a similar situation to themselves. However it was anticipated that there might be emotional and psychological issues related to their experiences that would come out in the interviews. I went to the interviews with the names and numbers of support groups as well as that of an ADHB Maori professional should the respondents require them. Another issue was that of internal confidentiality. Tolich (2002) describes this as the potential harm that might come from one insider recognising another insider in the final report. This is due to the interconnectedness of the participants within a small group or community. It could be considered that due to the small number of people with intersexed conditions, especially those with the same condition and the sharing of stories that has happened via advocacy and/or support groups that participants might recognise the narrative of another participant. While their identity is easily disguised from the general reader it may be obvious to another participant. It could also be an issue that something said in confidence to the researcher about a particular health professional could be recognised by that health professional should they read the research. As such this again classifies the participants as vulnerable and when considering quotes from the narratives it was important to reflect on how they might impact on the internal confidentiality of the data. To counteract this small community phenomenon short extracts have been provided from participants, which have been incorporated into paragraphs, rather than large extracts from interviews.

Ethical approval was sought from three committees. The first approval was sought and given by the Massey University Human Ethics Committee. Secondly the Maori Research Review Committee was approached and a meeting held with Mata Forbes, Maori Health Services Co-ordinator at the Auckland District Health Board (ADHB) to discuss the research. The researcher was committed to treating Maori participants with respect, both for themselves as individuals and as Maori. In order to address this I had included in my research proposal a commitment to:

- Explaining the terms and commitments of the study in clear understandable language.
- On completion of the study, offering to provide each participant with the study results.
- Providing an Opportunity for the participant's Hapu to discuss the implications of the project on family members, other members of the Hapu and all Maori.

Following this meeting the information sheet was amended to include an opportunity for participants to speak in Te Reo with an interpreter provided and the support of an ADHB Maori professional if they wished.

Thirdly ethical approval was sought from the Northern X Ethics Committee. This was a drawn out process with numerous amendments being required. The main issue was with the conditions I had listed as coming under the umbrella term Intersex. It was suggested that I would not find people with these individual conditions therefore a rewording was required. I was informed that I would not find any parents with children who had the conditions PAIS, CAIS or 5- alpha Reductase, and therefore I was required to remove this wording from the information sheets and advertisement texts. I was however, able to leave them on the sheets which would go to adults with these conditions. My argument to the ethics committee was that while Intersex is generally becoming a better known term there are still people who do not associate their, or their child's condition with the umbrella term Intersex so I felt it was important to list some of the more common conditions. To gain approval though, the conditions had to be left off, with the more general term Gonadal Dysgenesis being used instead. I feel this could have negatively impacted on my ability to recruit a wider range of participants.

Recruitment and Interviews

A qualitative research approach was chosen as it enabled the researcher to access participants who had experience in the research topic. Participants were sought intentionally for their ability to inform the research from their personal perspectives (Roberts & Taylor, 1998). Therefore the 21 people who participated in this research project may be regarded as comprising a purposive sample (Blaikie, 2000). Liamputtong and Ezzy (2005) note that in-depth interviews are most often used for research on sensitive topics or with vulnerable populations. This method of collecting data was also chosen to enable myself as the researcher to develop a dialogue with the respondents during the data collection stage.

The research design sought to recruit between fifteen and twenty four participants dependent on response. The aim was to talk to between six and ten parents/families and six-ten adults. It was envisioned 3-4 health professionals would be recruited for the study. Parents of intersex children, adult intersexed persons and health professionals were all sought within New Zealand. However as there is only one intersex advocacy group in New Zealand opinions from the largest Intersex advocacy group in the United States of America were also sought with the researcher travelling to Seattle to conduct interviews and gather information. The Congenital Adrenal Hyperplasia support group in New Zealand (CAHNZ) was also approached.

Parents/family/whanau were defined as any individual/s that provides primary care and support for a child. This could include biological, adoptive, or foster parents, same sex or re constituted families, grandparents or other family members who provide primary care for the child. Adult intersex persons were defined as those persons who self identify as intersex or with an intersex condition. Health professionals sought included representatives from the endocrinology, surgical and genetic disciplines. Intersex Advocacy groups approached were those self identifying as such.

Participants were approached via an open letter of invitation to participate in the research with all participation being voluntary. Parents/family/whanau were approached via an open letter in the Little Treasures magazine and the New Zealand Parents magazine. It was envisioned that this would only recruit parents but in fact these magazine advertisements yielded a number of adults with intersex conditions as

well. It was originally planned that further parents might be approached via open letters extended by private paediatricians, GPs and Plunket Nurses but this proved unnecessary as 9 families were recruited from the magazine advertisements and the support groups. Adult intersex persons were also approached via an open letter in Express Newspaper and via websites dealing with intersex issues and conditions, asking for New Zealand participants. Letters of invitation were also extended via the Intersex Trust Aotearoa New Zealand, (ITANZ) and the Congenital Adrenal support group (CAHNZ). These approaches yielded four adult participants with a further two resulting from the advertisements in parenting magazines. Intersex advocacy groups who were approached were ITANZ and ISNA (Intersex Society of North America). This provided 3 adults who were willing to talk about their experience of having an intersex condition as well as their role in representing an Intersex advocacy group. Health professionals were approached via Starship Children's Health, Auckland and Auckland City Hospital.

Research participants have the right to full disclosure concerning the nature and purpose of the research. All those who indicated a willingness to participate received a detailed explanation verbally and/or in writing, of what the research involved, including the aims and process of the research, and the participants' involvement. All respondents to the advertisements replied by email or phone and were then forwarded the information sheet by email or standard mail. They were offered the right to withdraw at any time and when further contacted about an interview two declined to participate any further. An additional opportunity to ask questions, make comments and voice any concerns they may have had about the project was offered at the commencement of the interviews.

Participants responded to my advertisements in the Little Treasures and New Zealand Parents magazines as well as the text of my advertisement being sent to people on the CAHNZ mailing list. I was able to interview 9 mothers, two of which had two children each with the same condition. The conditions were CAH, Klinefelters, and CAIS. I also received two abusive emails following my advertisement placements, one from a mother of a child with Klinefelters and one from the partner of a man with Klinefelters telling me they did not consider Klinefelters to be an intersex condition. Two adults responded to my advertisements in the Little Treasures magazine, one a mother and one a grandmother asking if I would like to interview them about their

own, rather than their child's condition. Other adult participants were recruited via support and advocacy groups. The adults identified as having, CAH, Klinefelters, CAIS, Gonadal Dysgenesis and one as Intersex without naming a specific condition. In total I spoke to six adults. Representatives from two advocacy and one support group were interviewed, ITANZ, ISNA and CAHNZ. Three health professionals were interviewed, an endocrinologist, a metabolic Nurse Specialist at Auckland District Health Board and a Genetic Associate for the Northern Regional Genetic Service. In total twenty one participants were interviewed.

Research participants have the right not to be harmed by a study. Due to the personal nature of the disclosures it was envisioned there might be occasions in which the participants experienced some emotional discomfort related to their experiences of having a child born intersex or being born intersex themselves. I came to the interviews prepared to provide immediate and appropriate emotional support if required, and with a list of support groups, and individuals experienced and qualified as counsellors if participants required further emotional support. I found that a number of participants wanted information on the support groups and one wanted information about ITANZ.

Data Collection and Management

I conducted the interviews with parents and adult intersex persons in Auckland, Taraunga, Wellington, Christchurch, Dunedin and Invercargill and travelled to meet participants. As it was important to establish a rapport with the participants interviews were held in a place nominated by the respondents. This was people's homes for the majority of the interviews but also included a workplace and my motel room at the respondent's request. I travelled to Seattle, Washington to interview two people from the Intersex Society of North America, to Wellington to interview one person from the Intersex Trust Aotearoa New Zealand and to Christchurch to Interview one person from CAHNZ. Professionals were interviewed in their workplaces in Auckland.

Data was collected via audio taped interviews with all the participants except the Metabolic Nurse Specialist who asked not to be audio taped. Written notes were taken during the interview and also directly following the interview, as an overview of what was discussed. A plan was formulated to use open in-depth interviews with key question prompts (the interview guidelines are reproduced in the Research Appendix).

Patton (1987) describes this form of interviewing as asking open ended questions, listening to and recording the answers, and then following up with additional relevant questions. Roberts and Taylor (1998), state that interviews allow an opportunity for people to express through language, their experiences, which may be different to those that could be represented in a prepared survey or questionnaire.

After a general introduction and explanation of why I was doing the interviews I asked parents and adults with intersexed condition to simply 'tell me your story'. The narratives that unfolded often brought up all the issues that I had hoped to cover with the more formal interview questions I had prepared. As the interviews progressed I seldom had to refer to these questions, only formally asking at the end 'what information would you like to see included in a resource for parents'? During the interviews my understanding of what the participants were saying was interrogated, as I checked with the respondents that my interpretation was correct. Verification is important to ensure that the researchers' life experiences do not unduly influence the understanding of the information presented by the respondent.

Interviews with the advocacy and support groups were more along the line of a mutual discussion on the many issues which arise for parents and adults with these conditions. Representatives of these groups were able to provide overviews of the common issues for parents and individuals who identified as intersexed. They were also able to provide me with a lot of written material to take away as a contribution to my research. At the time of the Seattle interview I received a draft copy of a North American based parent's handbook which has since become available online as a resource.

Interviews with health professionals were completed after talking to all other respondents. I was able to discuss in general some of the information from the interviews and again issues which arise for parents and adults were discussed. They were also able to offer practical information in terms of testing and investigations and outline treatment guidelines from Starship Children's Health (Auckland Healthcare Services) on the treatment of children with intersex conditions. The three health professionals with whom I spoke also offered to review any information which might be developed into a written resource for parents.

Methodological triangulation, for example mixed method, was originally developed in quantitative research in order minimise bias and to improve the validity of findings (Blaikie, 2000; Silverman, 2006). In qualitative research data triangulation involves the use of a variety of sources, such as consulting a range of literatures and conducting interviews with different categories of respondents, as they are unlikely to share the same biases (Blaikie, 2000; Liamputtong & Ezzy, 2005). In this research the credibility of the information obtained from parents of children and adults with intersexed conditions, health professionals and representatives of intersex support groups was also established through considering consistency in themes across interviews, with the available literature and with the researcher's experience as a nurse who has worked in the area for a number of years.

As the principle researcher I managed the data. Interview tapes and transcripts were kept in a locked cabinet at Auckland City Hospital while being analyzed and will be stored in a locked filing cabinet at Massey University following completion of analysis. Data base records and computer work are password protected. The database will be kept for ten years and safe guarded by myself. Access to the data was limited to myself and two research supervisors.

The audio taped conversations were transcribed ver batim and transferred to a CD rom copy. Interview notes were entered onto a word processor as soon as possible after the interviews. Audiotapes were the back up for written notes taken during the interviews. Procedures to maintain confidentiality were implemented including removal of identifying details.

Thematic Analysis

A manual method of thematic analysis was used to look at the data. Braun and Clarke (2006) describe thematic analysis as a method of analysing and reporting themes within data. It serves to organise and describe a data set rich in detail. It looks to find repeated patterns of meaning. Brink (1998) describes the analysis of exploratory research data as a fluid, flexible intuitive interaction between the researcher and the data. As the data are analysed within context, descriptions arise which make sense to both those who have lived the experience and those who have no prior knowledge of the experience. A theme is defined as something important about the data in relation to

the research question and representing a level of meaning within the data set (Braun & Clarke, 2006). The six phases of thematic analysis described by Braun and Clarke (2006) were used as a basis to approach a thematic analysis of the data from the 21 participant interviews. The researcher familiarised herself with the data by transcribing the interviews, reading the narratives contained in the transcripts and noting down initial issues the participants talked about. This served as a method of immersion in the narratives over the period of data analysis. For each narrative a note was made of important issues and the narratives quoted to illustrate these issues. The issues were then collated and used as a means of selecting and organising the data into potential themes (Braun & Clarke, 2006). The narratives as a whole were then compared looking for these themes and checking to see if the themes worked in relation to the extracts and quotes from the narratives. Final themes were than refined with definitions and names and these were grouped together when constructing the final analysis of the data.

Major themes within the interview material, which eventually became substantive chapters within the thesis, were identified as 'finding out', 'finding information', 'hard issues' and 'information needs'. In deciding on the substantive chapters for the thesis a number of themes emerged that seemed to fit logically together, for example participant's experience of being told about their condition/their child's condition, what kind of information that was given to them, what information was withheld and their experience of looking for and responding to information from various sources including the internet seemed to fit logically together under the heading 'Finding out and Finding Information' and this eventually became Chapter Four in the thesis. The themes were then contextualised into paragraph form (Carnwell & Daly, 2003) and interpreted in regard to previously examined literature. Chapter Six for example links the information needs identified by respondents with some of the literature that was previously considered in Chapter Two and situates this discussion in relation to documents containing practical advice that have recently become available via the web to people who are interested in finding out about and/or understanding a range of intersex conditions. The data from the four stakeholder groups was also compared in terms of the themes which had emerged. This comparison contributed to the reliability of the study in terms of one data source confirming that of another (Carnwell & Daly, 2003). An inductive analysis was used with the themes linking strongly to the data rather than the researchers' theoretical interest in the topic (Braun

& Clarke, 2006). A combination of analyst narrative and illustrative data extracts was used to present the themes. The aim was to present a story about the data and the topic (Braun & Clarke, 2006).

Plan for Dissemination and Implementation of Findings

At the time of the interview participants were offered a copy of the final report and given the opportunity to fill in a form requesting this be sent to them. The research findings will be disseminated via health professional journals and conferences. The findings will also be presented at conferences pertaining to Intersex Issues. I have been asked by the two magazines I recruited through, and the CAHNZ to write something for them on my findings. The findings will also be implemented in the design and content of an information resource or booklet for parents/familes/whanau and health professionals.

Chapter 4

FINDING OUT AND FINDING INFORMATION

'never underestimate how profoundly shocking it is for families to have any kind of diagnosis'
Co-ordinator of CAHNZ support group.

Introduction

This chapter explores the experience of discovering an intersex/DSD condition, for parents in their child, and for individuals realising themselves as a teen or adult. It looks at the language health professionals use to convey information to parents and individuals, and how this impacts on their perceptions of the condition. The effect of unsympathetic treatment by health professionals and the feeling an individual was a 'curiosity' or a clinical teaching opportunity is examined in relation to compounding parents' grief and shock when a child is born with an intersex/DSD condition. The amount of information participants received, how it was delivered and the ongoing information and support received in the community is examined in regard to how families coped once they were discharged from the hospital. The chapter looks at some issues unique to those with Congenital Adrenal Hyperplasia in terms of their ongoing experiences dealing with a lifelong condition with a medical component. Lastly the chapter examines the role of the internet in participant's experience of gaining knowledge about individual conditions, as well as the possibility of being exposed to some of the ethical debate around treatments which have been discussed in previous chapters. It brings up the issue of choices which were made around telling people outside of immediate family about individuals' conditions in light of the accessibility of information on the internet. This was a new finding to this research.

When pregnant there is an expectation that you will deliver a healthy, full term, male or female infant. Western media does a lot to reinforce this expectation, as pregnancy and parenting magazines, television advertisements and shops selling baby clothing and equipment all immerse prospective parents in a pink and blue world of boy or girl babies. With advances in prenatal testing and scanning parents often find out months before delivery whether they are having a 'boy or a girl', thereby further immersing themselves in a culture which expects them to know the gender of their

child. Parents also rely on this same testing to inform them of any medical condition their child might have, often leading to them feeling reassured their baby will be healthy when in fact the testing only identifies a few more common conditions such as heart defects or Down Syndrome. While there can be some mention of preterm delivery or conditions which may affect a baby these are usually one off stories which do not necessarily lead parents to believe the same thing may happen to them. Few stories of babies born with intersex/DSD conditions, or about intersex/DSD conditions, appear in the media. Stories that do appear of babies born with intersex/DSD conditions are usually dramatic in nature with titles such as 'my daughter was born a boy' (Adrenal Hyperplasia website). In recent times the media have been critical of the work of sexologist John Money ('Dr Money's horrible sex experiment', *The Dominion Post*, 2005; 'Dr Money and the Boy With No Penis' *TV3,* 8.30pm 23/06/05) while stories of adults living with 'intersex' conditions are often sensationalist and tend to focus upon the person's negative experiences of treatment ('the secret of my sex', Graham, 2005; 'mistaken identity' Hubbard, 2000). People often rely on the media to provide them with information about things with which they are not familiar (Lupton, 2003). Articles, like the ones outlined above, shape and are shaped by public understandings of intersex/DSD conditions giving little useful information as to how common these conditions are or how parents might cope. It is therefore not surprising that the news that their baby has an unexpected and often unimagined condition has a traumatic impact on new parents.

The overwhelming sense of shock and grief parents felt was the first thing parents talked about when I asked them to tell me their stories, and it dominated their recollections of the time after the birth of their baby. It also came through strongly in the narratives of adults in their recounting of how their own parents had felt about their births, or of their own experiences of finding out about their conditions.

Reading the words participants used to convey their sense of this time provides an insight into how affected they were by the news they were given.

> *'a terrible shock', 'pretty distressing,' 'enormous shock', 'big, really horrible thing', 'devastated', 'shocked in hospital, horrible', 'such a bad dream I've blocked most of it out', 'I was so messed up in my head I lost sense of direction'.*

These initial feelings of shock and devastation at the news of an intersex/DSD condition are consistent with the literature (Kessler, 1995; Groveman, 1998) and continued to affect participants' experience of health professionals and the healthcare system.

How We Are Talked To

People interviewed for this research project suggested that feelings of shock and grief were both intensified and compounded by the treatment and information received from the health professionals that they came in contact with. Themes common to a number of participants were the language and expression used by health professionals, the lack of information they perceived health professionals gave them and the treatment they and/or their children received when they were examined or treated by health professionals.

Mani Bruce Mitchell, head of the ITANZ tells of the words her mother heard at her birth fifty years ago, *'oh my god it's an hermaphrodite'*. Other adult participants also recalled the language used to talk about them and the feelings it evoked for their parents and themselves. As a teenager being seen at the medical school one participant was told *'you are going to end up in a textbook one day'*. Another describes her mother thinking the doctors thought she *was 'an interesting little specimen'* and the doctors asking her about whether she had taken drugs or hormones, *'almost a blame thing...what have you done'*.

Frank (1999), comments on medical staff seeing a patient's life as an open book or a chart. These participants felt their condition had been reduced to an interesting text book case. These participants also expressed dismay and anger about how health professionals treated them when they were in hospital with no regard to their privacy, feelings, or their own knowledge of what was happening with their bodies. When she was admitted for tests at the age of 23 years one woman, now in her fifties, with CAH described her experiences:

> *'when I went in it was absolutely shocking because I was young and I had these doctors coming in and out in and out in and out, no one ever asked me anything about me they just wanted to have a look'*

She recalls they never told her what they thought was wrong with her or explained about her condition *'they just want to look at your private parts, never explained why'….. 'I got really upset because in those days you didn't ask questions '.* In fact she never received an adequate explanation of CAH, even after leaving the hospital and returning to the clinic. *'I said what's that? He said 'well what we are going to do is put you on steroids'….. 'I said what is it? he said you are just producing a little more testosterone'.* She never received more information and went on to stop taking the steroids because of how they made her feel with the consequence of years of health problems for which other doctors saw her without ever requesting her hospital files with the diagnosis of CAH. She blamed the way she felt on childhood sexual abuse and was told it was her own fault, *'you're a hypochondriac, we are never going to get anywhere because a lot of it is in your head'.* It took 12 more years before a GP finally requested her medical records and was able to explain to her what CAH was and get her the treatment and help she needed. The result of her early treatment and lack of an explanation that she could understand was far reaching and extreme in its impact on her life. She now has major health problems from being left untreated for so many years including diabetes, osteoarthritis, and thyroid tumours. She describes the effect of the major mood swings, anger and depression on her family, *'it destroyed my marriage',… 'I had to explain to my children years later it had been an illness which made me act the way I did'.* It also negatively impacted on all her future encounters with the medical profession, *'no confidence in male doctors because I'd had that bad experience at the hospital and that had stayed with me and I thought no male doctor is ever going to touch me again'.* A lack of trust in the medical profession resulting from negative experiences of treatment has also been identified by intersex/DSD advocacy groups (ISNA website) and discussed in the literature (Feder, 2002). Howe (1998) and Groveman (1998) comment on medical secrecy and deception and the psychological harm repeated genital examinations can have.

Another adult woman with CAH, who had clitoral surgery as a baby, talks about her experiences of returning to the hospital as an adolescent knowing something was not right. *'I actually had to convince them that there was something a wee bit abnormal'.* She had a urogenital sinus, which she had to look up in a textbook, self diagnose, and then convince doctors she was right:

> *'clearly from original surgery it would have been obvious but if it wasn't for me saying this isn't alright and I guess my mother saying look she's bright, she's been looking it up and if she says she has a urogenital sinus then she probably has, nothing would have been done'.*

Yet another woman who was seen by doctors at the age of 14 because she wasn't entering puberty stated:

> *'I didn't get much information. I recall being told there was something abnormal with the chromosomes, there was something gone wrong and they didn't really know what they were going to find but they wanted to do some surgery to have a look and see what they could find out, and they needed to do some examinations and stuff'.....'I found the rectal examination awful, to this day I gag at the thought of it'*

Again she felt she wasn't treated with respect or sensitivity, *'I remember as a sweet nice young thing at 14/15 yrs old being asked if I minded a student in the room, I wasn't going to say no but I really did mind'.* Later when she was admitted for exploratory surgery she describes nursing staff as having:

> *'complete insensitivity to a 16yr old girl in a ward full of 45yr old women having hysterectomies'...'nurse asking when was your last period, I've never had one why do you think I am in here'.....'pre surgery coming to shave me' 'there is nothing to shave'.....'not being treated like a person'*

As these older participants were diagnosed at a time when the only paradigm of treatment was a medical one which operated under the guise of do not ask, do not tell, these stories are perhaps not unexpected. I had some hopes that in talking to parents with children born in the last ten years and particularly in the last five years, when the controversies around the treatment of children with DSDs have been explored and debated in much of the medical literature, I would hear different stories, unfortunately this was often not the case.

Some parents talked about their initial shock and distress on hearing of their child's condition and how hard it was to talk to friends and family members. For some parents it was obvious at birth that something was different for their child because of the child genitals. The confusion the parents felt about whether they had a boy or a girl was made all the more difficult because of the time taken to get test results back, but also intensified by the way they were talked to by staff in the hospitals. A mother of a child with PAIS tells *'she was born then the atmosphere changed'....'I asked whether we had a boy or a girl, husband went to say boy and midwife said girl'...'a lot of panic and confusion'....'it changed from half an hour, it just changed, could be a girl, could be a boy'.*

Another mother of a daughter born with CAH recalls:

'we took home a baby boy and 5 days later got a call to say chromosomes showed a girl.....please come back'....'up until than we were told that the options were you choose whether you want a boy or a girl'

One parent interviewed for this research described how hard it was for her not only to take the news in herself but also to talk to family and friends:

'the first initial thing was lets not tell anybody but how can you not, it's the first thing people ask'.... 'and we told anyone who got in touch we have a boy, didn't mention surgery would be required,'... 'then we didn't have a boy but a girl so than we really had to explain things so a lot of people neighbours and friends had it all explained to them'

The language which staff used had a major impact on how parents felt. One mother described her experience of being transferred to a bigger hospital after her baby was born,

'it was a rude awakening, they took her off me and a doctor made the comment 'oh my god I've never seen anything like it'...'it got worse and worse from there on in...no one could acknowledge what you had even the nurses didn't know how to deal with her, made me and my family embarrassed and I got depressed'

After another five days she was taken into a room with a couple of endocrinologists and a surgeon who told her:

'yes you probably have a boy but go home give this boy a girl's name and we'll see you in 5 months- this is what's going to happen, you are going to have surgery to make her a little girl because everything's not quite right, get on with it she's not dying'

The doctor minimised or dismissed the parents concerns by making a downwards comparison between their child's condition which is described as 'not life threatening' and children who have terminal medical conditions. This effectively silenced the parents, closing down any further opportunities for discussion. These parents felt bullied by the medical professionals they saw and felt they were guilt tripped into making decisions they now regret *'you're not a good parent, you're not good parents if you leave her like this what if she has to go to day care, remember you can't tell anyone'.* The comments by the medical professionals served to reinforce the sense of shame and secrecy these parents already felt. As members of the medical profession these doctors were already in a position to wield considerable cultural authority. The advise to *'don't tell people that you don't know well, don't tell relatives, keep it a secret which just about sent me round the twist'* further sought to dismiss this family and their concerns while cutting them off from a source of support, their extended whanau.

For those parents who found out about their child's condition via a blood test the sense of shock and grief was no less strong, and was again compounded by their treatment once they returned to the hospital for assessment and medical management. They also talked about how difficult it was to comprehend what they were being told about their child and how hard they found it to understand or ask questions of the staff. A mother of a child diagnosed on a Guthrie test with CAH talks about the GP coming to tell her the news *'I actually couldn't hear her talking, I could see her talking but I couldn't hear her'*. When she then arrived at the hospital with a barely conscious, critically ill daughter she says *'tests needed to be done quickly but the manner in which the house surgeon spoke was traumatic.. I needed an explanation and to be given the option to stay with my baby'*. Over the 2 weeks in hospital she talked of becoming incredibly isolated because *'I was consumed with anxiety'* and *'we were 'objects of curiosity'* which only added to the feelings of isolation and anxiety, which for her contributed *'a new level of trauma'.... 'I don't remember anyone coming and sitting with me at all or even offering a cup of tea'*. The initial trauma for this mother was learning her child had a condition that had made her critically ill and once recovered from this initial event she would need lifetime medication. The second level of trauma was added by health professionals who saw her baby as an object requiring testing without acknowledging her need to continue mothering her daughter, and those who saw her baby's genitalia as a curiosity to be examined rather than taking the time to talk with her about how she was feeling.

One participant who was a mother of a son admitted to hospital for failure to thrive and a subsequent diagnosis of CAH remembers *'the doctors didn't have a clue'*, one told her *'she had a spoilt child who just wanted his mother's milk and wouldn't feed'*. Another talks about how shocked she was at the *'onslaught of medicalisation'* and that *'it wasn't until day four in hospital a nurse realised how distressed we were and acknowledged the grief.* A mother of a daughter with CAH talked about her time at home with her midwife visiting *'I was exhausted- it was just unbearable- I thought I was going mad'* Because of stress she lost her milk at 4 weeks and was given a hard time by midwife about not breastfeeding, she considered that the midwife needed to be more helpful *'not make you feel guilty and like a crap mother'*.

When I talked to the co-ordinator of the CAH support group in New Zealand who has talked with many more families, one of the things she stressed was how important the way health professionals talked to families was and how vital it was to look at the way doctors, nurses and social workers 'hold' families. She suggested they needed to *'slow down and recognise this person is in a very slow way of thinking, and use a gentle and reassuring tone'*. You need to make sure the first thing you say is positive as it will be *'imprinted on their minds forever'*, which in turn will *'have huge ramifications for the future relationship between mother and child'*. Concerns about how information is given and families are treated by health professionals were echoed by parents interviewed for this research project. This suggests that health professionals who work with parents and families of children with ambiguous or unclear sex designation at birth may need to be more sensitive and develop better communication skills. The need for, and opportunity to educate health professionals both in the hospital and community settings, evolved as a clear issue during the research. These concerns are also reflected in the academic literature.

Kessler (1995) in a presentation on *Genitals, Identity, and Gender*, questioned what parents or children hear when medical professionals use language to negatively describe genitals. For example "Your genitals are inferior (less functional, ugly). We pity you and suggest you have corrective/cosmetic surgery" or "Your genitals signify something about your parents. They have misbehaved or are genetically unsuitable. They are embarrassed by you and your genitals" (Kessler, 1995, Table.2). She suggests a change in language might mean the message that is heard is more along the lines of "Your genitals are just another body-part that varies from person to person, like noses and ears, and it doesn't matter what they look like as long as they function well. We don't think that much about your genitals or our own" (Kessler, 1995, Table.2)

Alice Dreger (2002) comments on the issue of well-intentioned medical professionals dealing with intersex, who in hoping to provide a "quick fix," do not manage to adequately address parents' persistent confusion and distress. In a study looking at the psychological impact of genital anomalies on the parents of affected children, Duguid et al (2007) also found that parents felt poorly informed with the information they received being described as inadequate, incorrect or too emotive. Their study indicated a need for more adequate education and information for parents in respect to many aspects of care. This was also reflected in interviews with ten families of CAH affected children (Siminoff, Mercer, & Sandberg, 2007) who found

parents reported they received inadequate and incomplete information from healthcare providers for their needs as parent caregivers and decision makers.

What We Are Told.....Or Not Told

One of the strongest themes that arose in this research project was the total lack of information parents felt they received at the time of their child's birth and in the community once they went home. The information parents wanted ranged from a simple explanation of what their child had, through an adequate explanation of how and what drugs to give if they were needed for CAH, to desperately wanting to know what the future would hold. They talked of information being given grudgingly or in language they did not understand and of feeling it was not alright to ask for a better explanation or question any of the information they were given:

> *'I was distraught at the lack of information, no explanation of drugs, timing, what to record, no information on what would happen over the course of the hospital stay'.....'I had to drag information out of the doctors piecemeal'*

> *'always believed the medical fraternity knew everything you didn't second guess or ask them something, if we wanted to ask we felt dumb, so what's our baby got partial androgen insensitivity syndrome....words meant nothing'.....'I can't say I don't understand or they will think I'm completely mental'*

> *'sort of explained'....(but)... 'because of stress and trauma of realising your child has a condition which could kill her you don't take it in'*

This need for information was also highlighted by Duguid et al (2007). They found an unfulfilled need for accurate and adequate education and information for parents with respect to all aspects of the child's care, with parents wanting both condition specific and generic information. They wanted this information to be written and to be presented in a gradual and steady way following the initial presentation. Duguid et al (2007) found this need was highlighted throughout the study on the child's journey through the care pathway. ISNA offers a basic list of questions and information that parents should seek from health professionals (see Table 2). It would appear that very few of these questions were answered for the parents I interviewed and that information they were given was often not easily understood in the period immediately after they found out about their child's diagnosis.

Congenital Adrenal Hyperplasia

There were some issues which were specific to conditions, the main one being CAH. Of importance to those parents of children with salt wasting CAH was the issue of how to give drugs, what to do about children who vomited them back up and what to do if their children got sick. The first year was described as being consumed with the constant dosing and monitoring of the drugs, and worry about their child getting sick and going into an adrenal crisis. This often led to them isolating their child from others and in effect isolating themselves. Parents also described leaving hospital with no real idea of how to give the drugs or what to do about timing. Through a stressful process of trial and error, some asking for suggestions on the internet or finally being put in touch with the support group who could offer solutions, they were able to determine how to make it work for their child. They expressed a desire to be given some simple practical instructions on leaving the hospital.

'absolute nightmare to give drugs'
'wasn't told it could be given in milk'

'salt replacement is distressing with gagging and vomiting, how should I give it'

'up for an hour trying to give sodium, she vomited it back, how much more to give'
'when should you give, strictly eight hourly, should you wake a sleeping child?'

Another mother described the impact on her family:

'the stresses involved impact on everyone and everything. For the first six months it takes over your head...'who will look after him when he is sick if I am working?'

She went on to talk about wondering if...

'eventually all the medications and having a child with CAH will become normal and there may even be times you will forget that he has CAH or you don't clock watch, it will not be all consuming'.

A large part of the first year was also taken up with trying to protect their children from catching illnesses from others with the frightening consequence of a possible adrenal crisis if their child got sick. Parents described feeling alone with their child's condition and unable to seek support and help as others could not manage the day to day medical aspects of CAH. One mother described it as being:

'very very hard initially, constantly keeping him in a bubble, constantly watching him', ... 'I was very depressed after coming home from hospital, no-one else could care for him, I couldn't use childcare, friends and family couldn't visit as I was worried about germs, it effects your whole life'

Again the lack of information and support from the health professionals they saw continued to compound the stress for families in the community. This was especially true for families out of the Auckland region. Those in contact with Starship Children's Health in Auckland were, in general, happy with ongoing management but those in other cities continued to struggle. They described having to balance the knowledge they had acquired from their own research and information from the CAH support group with the attitude of the health professional managing their child. One mother stated: *'I worry he is not getting the most up to date treatments and have to balance that worry with a good relationship with medical staff'* . Another recalled finding out about heel prick testing from the support group but her child's paediatrician *'told me off for going behind his back, he does not like the support group. I have to approach him without upsetting him as he is the only person I can see'.* Travel was yet another issue which provided challenges for parents, from requiring letters explaining their child's condition and medication to having to demand their child be treated correctly when they became sick away from home. One mother described her experience of a small town hospital:

'she had an adrenal crisis and nearly died just because the doctor wouldn't phone Starship because he was the doctor and I was the mother and what the hell am I doing telling him what to do, ended up in stand up argument and had to threaten to hop on a plane and go to Auckland because he didn't believe me she was in adrenal crisis'

Siminoff, Mercer and Sandberg (2007) talked specifically to CAH parents to look at the issues related to CAH from a parental perspective. In interviewing 10 families they found the day-to-day medical management was paramount to parents. The parents also expressed a need to be taken seriously by healthcare providers as they felt they knew their children the best. Issues with consistent care and travel outside of Auckland were examples of health professionals having power and control over parents experience and ability to manage their child's condition. The variation in diagnostic and treatment options throughout New Zealand reinforced the need for a team approach and protocols which would be consistent throughout New Zealand for children with CAH. Collaboration with fellow health professionals to provide the most up to date treatment options for children irrelevant of where they lived in New Zealand would be a step towards improving the information and support health professionals could offer families. Upon discharge a letter from a Starship hospital paediatrician explaining a

child's medical history, condition and the treatment regime for emergencies and adrenal crisis may be another option for parents to provide them with a resource to manage situations where they feel their child is not receiving optimal treatment. Parents in this section talked about using the internet passively to find out information, or actively by requesting help though blog sites, this suggests that the internet may be a significant resource for some parents. Drawing on the narratives of the people interviewed for this research project the following section considers how the internet may be both empowering for parents of children and adults with intersex/DSD conditions and a source of stress.

The Internet

> *"I don't want to freak them out when they look something up on the internet and think – oh god mummy's a man"*
> Mother with Gonadal Dysgenesis

The internet provides a vast source of information which stretches from fact through fiction to fantasy. All of those I interviewed mentioned it in some capacity. While it was useful in terms of providing information about a condition, especially when they felt the medical profession had poorly informed them, many found it led to a sense of confusion and even fear after trying to navigate the myriad of information encountered when they entered a term or condition into a search engine. This sense of fear extended to worries over what might happen if family, friends, acquaintances and even their own children searched for information on a certain condition and came away misinformed. The very ease of availability of information was than influencing people's decisions about how much detail they revealed to others about their own or their child's condition.

As the internet becomes an integral part of our daily lives it is logical to assume that parents and families will turn to it as a source of information when given a diagnosis. Starke and Moller (2002), comment that seeking information and gaining knowledge about a diagnosis is a way for parents to "restore order in a chaotic existence" (p. 245). In a study looking at how useful parents find information on the internet Sim et al. (2007) found that 89% of parents surveyed looked for more information about conditions and 61% looked for information on long-term outcome. While overall (94%) parents found the information useful they also found the

information to often be too technical and too distressing. Of interest, no parent had a website suggested to them by a health professional.

If health consumers access the internet the question arises; how reliable is the information that they access? It could be presumed that anyone publishing on the internet has their own agenda, including the medical establishment when aiming to present factual information. Any information can be considered biased to the opinions and beliefs of the person writing it or the clinical norm for the institution providing it. When searching for information on the conditions of those participants I interviewed, a mixture of medical sites, support group sites and personal web pages and blogs appeared in a search engine. Semere et al. (2003), in looking at parental usage of the internet for medical information, comment that web sites which tend to be least accurate are those written by and for lay people. They suggest medical professionals provide their own information or links to a hospital or medical site that they know to be reputable (assuming that the medical professionals do not have their own agendas).

Despite the concerns outline above, the web sites of lay people allow for the telling of personal stories something which other parents may find invaluable. Skinner and Schaffer (2006) in looking at how genetic information and the internet affect parents search for, and understanding of a genetic diagnosis comment that the intersection of knowledge and media have led to a powerful social context which is changing families experiences and interactions. They note that as parents publish their children's case histories via personal web pages and share advice in chat rooms they take part in "increasingly influential productions of genetic knowledge" (p.16). They identified that parents searched for resources in layman's terms and the case descriptions, prognoses and treatment options offered by other families. Of interest was the fact that even those who said they benefited from their research talked about the anxieties online resources caused.

Parents and adults I interviewed who turned to the web as a source of information felt they had been given little or poor information by health professionals. They turned to the internet for information about the condition, how to manage it at home and for a long term prognosis. They talked about leaving hospital feeling confused and unsure of what the diagnosis meant, this uncertainty was often

compounded by a lack of knowledge by the community health professionals that they relied on, midwives, GPs and Plunket nurses. This led them to search the internet for answers and support. A mother of a child with CAH commented that she had to resort to a website for good information as her information on CAH was *"hand scribbled on paper"* by a paediatrician whom she described as having no bedside manner or empathy. Another mother commented that she *'craved information but didn't get it till I got home and started looking on the web'*. Another mother who received a diagnosis of Klinefelters for her 4yr old son said her *"best information came from the kindy who printed stuff off the internet"*.

For one woman the internet became the source from which she found out the name of condition she had. She didn't hear the term gonadal dysgenesis until she had her medical files transferred as an adult.

> *"I was never given an exact diagnosis, it was always left vague"....."I got on the internet recently and started nailing this gonadal dysgenesis thing, wanting more information"*

She figured out the exact diagnosis and condition from the internet and armed with a name contacted her physician to confirm the facts.

Participants turned to the internet for empowering information, however, they also commented on the negative and 'scary' information they encountered. *"So much on the internet that can scare the be Jesus out of you"*. This ranged from reading about side effects of medication not being managed correctly to personal stories about the devastating effect treatment by the medical profession had had on individual lives and the poor outcomes of genital surgery. Parents were also confronted by the controversies that exist in care and treatment, often for the first time. One mother, when trying to decide about surgery for her daughter found *'most stories were horror stories, anti surgery were the worst with the scenario of 30 year old surgeries'*. A mother of a child with CAH commented that she was *'freaked out'* by stuff she read on *'body hair and a girl looking like a little boy'* which served to reinforce her decision to have genital surgery performed on her daughter. Another mother noted that the internet has lots of information *'but it might not necessarily be in the right direction'*. Yet another parent of children with CAH stated that she did not advise getting on the net and hunting for sites, while some were good, some were *'highly militant, aggressive, anti establishment political machines powered by people with their own agendas'*. While she acknowledged that they are a legitimate expression of rage for those who

have been treated appallingly she stressed that she felt they are not necessarily what a teen or parent needs to read.

Discussions with participants about information they had found on the internet led onto many participants expressing concern over what others had or might do with information they found on the internet. These fears led them to censor what they told others about their own, or their child's condition. They stressed a need to protect their child from what others might say, and a concern about what their child might read if they accessed the internet themselves. They felt if they told people including child care providers, schools and the parents of their children's friends, the actual name of the condition it would only lead to teasing or worse still assumptions about gender and sexuality and questions about any genital surgery which might have occurred. These worries extended to how much and what they should tell their children before others mentioned something to them or they accessed the internet themselves and were frightened or confused by the information they encountered there. One mother stated *'I am happy to say they have a medical condition with adrenal insufficiency, don't want to say CAH in case people look on the internet and ask about genitalia and surgery'.* Another worried about her daughters' classmates saying *'kids may find out the name of her condition, look up on the internet and tease.'* Using the term Gonadal Dysgenesis worried one participant as she felt that if people knew her chromosomes were typically male they would say something to her partner which would make him uncomfortable *'it can be very strange for a man to have someone say your wife has the same genetic makeup as you- that's a bit weird isn't it'?*

One participant related the story of someone she knew socially, who was a nurse, asking about her child's condition and when she told her it was CAH, the same person then went and researched and proceeded to ask in front of her 4 year old *'has she needed surgery'* then said *'oh she looks like a normal girl'.* She felt that she had nurtured and protected her child, built up her self esteem and realised someone could come along and ruin all of that. She now feels it is parents place to protect their child and would go so far as to say *'a parent is within their rights to lie to do so'.* She advises people to be vague about what a medic alert bracelet is for or the condition a child has.

Many of the participants expressed concern over what to tell children as they got older (see chapter 5), but part of this was their fears over what their children would discover on their own.

> *'the information they will have access to is just so different from the information I had access to when I was younger and I don't want them to find out something or have someone tell them something that I wasn't ready to answer'* Mother with Gonadal Dysgenesis

Another parent explained that you *'don't want someone pre-empting what you would tell your children or them looking it up themselves'.*

Health professionals interviewed also expressed concern about the lack of regulation of information on the internet. The comment from the metabolic nurse specialist was that parents need to be referred to good websites as there is a lot of misinformation. Health professionals were also willing to share the websites they thought were appropriate. The feeling was that if parents were given a written, regularly updated list of 'good' websites they would feel less overwhelmed. Of note none of the participants I spoke to said that health professionals had provided them with good information about websites. This points to a need for a regularly updated list of websites with helpful information for parents. Contributions to this list could come from health professionals, support groups and other parents.

It seems interesting that having come from a paradigm of medical treatment that encouraged people to be quiet about their condition the very availability of a wealth of different and accessible information is still encouraging people to keep quiet about any variance in what is assumed to be the norm for males and females. Participant's experiences of the internet had provided them with more knowledge of the various intersex/DSD conditions but the large amount, and sometimes political content of the information on the internet had also caused them to become confused and scared at times. The issue of others looking up an individual's condition on the internet and the implications of this in terms of teasing at school or inappropriate comments to the individual or their family appeared to be new to this research and was not an issue which had arisen in the literature review.

Summary

The experience of finding out a child or yourself as an individual has an intersex/DSD condition was dominated by feelings of shock and grief for participants contributing to this research. This initial shock and grief was compounded by the language health professionals used to talk about individuals or their children, referring to them in the abstract as specimens or text book oddities. The language health professionals then went on to use to give or explain a diagnosis or condition left participants feeling uncared for as people, or stupid for not fully understanding. At the time of diagnosis participants described feeling confused about what the diagnosis actually meant. They asked that information be truthful, simple and practical and that it be delivered in a caring and empathetic way.

For mothers the way their baby was treated at birth, the uncertainty of the gender assignment before test results became available and the confusion they felt over the information they received all intensified their initial shock over the birth of a child with a condition they had not expected and in the majority of instances never heard of. For those with a diagnosis of CAH there were extra stressors associated with the medical aspects of the condition. Participants described being poorly informed about the medications, how to give them and how to manage them at home. The initial year at home was often dominated by fears around the possibility of an adrenal crisis and a growing sense of isolation as they felt the need to keep their baby away from others due to the risk from coughs and colds. Mothers described the experience of feeling they were the only ones who could provide care for their baby.

Managing in the community and negotiating relationships with a wide range of health professionals or a single health professional that appeared to be the only choice for ongoing care in their city, provided further challenges for parents. The lack of knowledge and understanding some health professionals had of the conditions or participants perception that health professionals were threatened by parents growing knowledge over time meant that their interactions were at times tense and a further source of worry about whether their child was getting the best or most up to date treatment. Consistency of care throughout New Zealand was also an issue raised by parents contributing to this research.

The internet, while a resource for information, some of which was helpful and for some the first detailed information they described gaining, also added fear and confusion to participant's knowledge base. The sometimes political content or the very real stories of how others with intersex/DSD conditions felt betrayed and damaged by the medical system intensified participants worries around the condition they themselves or their child had. The narratives around the experience of finding out about an intersex/DSD condition and seeking more information led on to participants talking about a range of difficult and complex issues which were often unique to having an intersex/DSD condition - these themes are taken up and developed in the next chapter of the thesis.

Chapter 5

THE HARD ISSUES

'the parenting journey involves lots of choices, parents who have a child with a rare condition have some different choices to make, they are tough choices at the hard end of the scale'
Parent of two children with CAH

Introduction

There were a number of issues that arose in my conversations with participants which could be deemed unique to the experience of having a child with an Intersex/DSD condition. This chapter looks at the issues of the nomenclature used to describe the conditions, issues around surgery and informed consent for surgery, gender identity questions and issues around future pregnancies and genetic testing. The chapter also examines the experience of growing up with an intersex/DSD condition and the issues which arise for children, teens and their parents.

Views On Nomenclature

'I found out she was classified intersex and I was upset by it because I thought she's not. She's a girl and she's not ambiguous anymore, I see she is because she was born an intersex baby'
Mother of a daughter with CAH

This section examines participants' reactions to the term intersex and their feelings about how the term related to their or their child's condition. It looks at the choices they made in terms of nomenclature when talking about themselves or their child. The ongoing debate surrounding nomenclature is reviewed in Chapter 1. The term intersex as an umbrella term for a number of conditions has been criticised as being confusing in terms of meaning and politicised in its usage (Dreger, 2006; ISNA, 2006). The term Disorders of Sexual Development (DSD) was proposed by the 2006 International Conference on Intersex and supported by ISNA, however this term has also been criticised for the use of the word disorder and the negative inferences this has to pathologise and manage those with the conditions if refers to (Cameron, 2006; Hinkle, 2006).

I had anticipated before I talked with participants that there would be issues over the use of the term intersex to describe different conditions. Several people had

indicated to me in their emails that while they did not consider their child intersex they did have one of the conditions I had written about, therefore they were willing to talk to me. At the start of the interviews there was generally some discussion as to why I had used the term and how participants felt about it. I explained I had used it as an umbrella term which described individuals born with anatomy or physiology that differs from what is usually expected for males or females. All of the parents of children with CAH interviewed did not consider their children intersex and preferred to just use the term CAH. They described feeling upset when they heard CAH children referred to as intersex because their interpretation of the term was, it meant their child had two sexes or there was confusion over their gender identity :

'enough to get your head around CAH without being put in the intersex basket first off found out via my own research and I was upset by it- I thought it wasn't fair'
'my husband doesn't like the term intersex, my son isn't like that'

These feelings also reflected the comments of Sue Elford (2006) of the UK CAH support group who notes that the majority of the CAH community do not consider themselves intersexed. The mother of a son with Klinefelters also stated:

'A's a boy- that's all there is to it, genes may say something different but he's got what he needs for being a boy'.

The narratives reproduced here reinforced the perception stated in the academic literature that for parents the term intersex is interpreted as meaning their child was neither a boy nor a girl.

There were other parents who could understand why the term would be used in reference to their child but they still did not wish to use it themselves. *'I can see why it's intersex because he is a male with a female chromosome'*

Three of the adults I spoke to self identified as intersex while another three understood the usage of the term but did not refer to themselves as intersexed. At the time of the interviews the term DSD was not in usage, I have since asked the head of CAHNZ if there has been feedback from any of their members on the change of nomenclature, her reply was:

I've had no feedback at all from CAH parents. I think most of them do not think in terms of "intersex" quite honestly. It's not a term we bandy about in conversation and if it ever has been used, it's met with some strong feelings. Not sure about the new term. I agree with Mani though

(last newsletter, but one) that it would have been nice if the intersex community had had their feelings canvassed about it. Such a big move and all the "experts" driving it. Same old same old.'

The participants I interviewed contributed two different views to the nomenclature debate. Firstly there were those for whom an umbrella term under which their condition was classified either angered or frustrated them. They did not wish to be lumped together with others who had a different condition and preferred to just identify with their own condition and others who had it. This view was most commonly expressed by those with CAH. The parents I spoke with did not consider their child with CAH to have an intersex condition. They expressed the view that by using the term intersex an inference was being made that their child was not the gender that corresponded to their chromosomes or their genital anatomy, this was true of parents who had chosen to have genital surgery on their daughters' virilised genitals. For these parents CAH was characterised by the medical component of the condition rather than any difference in genital appearance when their child was born. This could explain why they preferred the use of a term which explained the medical diagnosis, Congenital Adrenal Hyperplasia. The second group were the adults, and one parent who chose to identify with the term intersex, this included the conditions CAIS, PAIS, Klinefelters and one adult with CAH.

Surgery

'she will have reconstructive surgery and she will be a great girl'
Doctors words to the mother of a baby boy with PAIS prior to feminising surgery

Surgery has historically been performed for three broad groups of infants. Firstly for those with a female karyotype, predominantly those with CAH, to reverse the effects of virilisation in the form of a clitoralvaginoplasty. Secondly for those with a male karyotype, combined with a small phallus or ambiguous genitalia who were subjected to feminising surgery to reinforce their female gender assignment. Lastly for those with a male gender assignment with surgical treatment involving procedures such as orchiopexy, the 'repair' of chordee and hypospadias, the removal of mullerian structures, all with the aim of producing a more 'normal' male appearance and function. More recently, however, recommendations for, and views on, medical management have been changing as a result there is a lack of consensus in the literature regarding surgery on intersexed infants. This section reviews the ethical issues and debates surrounding surgery and informed consent. It examines older participants' experience

of a surgery they did not consent for and parental decisions around genital surgery, the information they received to enable them to make an informed decision and their experience of the surgery.

Surgery on intersexed infants is undoubtedly one of the most controversial issues in the care of these children. Emerging from a paradigm of care where surgery was performed swiftly to reinforce a gender assignment, the ethical issues surrounding when and indeed if surgery should be done continue to be debated in both the medical and social science literature (Blizzard, 2002; Daaboul & Frader, 2001). While the era prior to the mid nineties was notable for a sparsity of follow-up studies there are now a growing number of published papers which consider the outcomes of genital surgery (as discussed below). The recommendations arising from these studies, however, are not always in agreement on which way to proceed with some suggesting a continuation of early surgery (Farhat, 2005; Lean, Deshpande, Hutson & Grover, 2005; Warne et al, 2006), some recommending vaginoplasty be delayed until adolescence (Gollu et al, 2007; Alizai, Thomas, Lilford, Batchelor & Johnson, 1999; Stikelbroeck et al, 2003; Creighton, Minto & Steele, 2001; Crouch & Creighton, 2007) and others suggesting the results are poor enough to indicate delaying all surgery until a child can make an informed decision themselves (INSA,2006; San Francisco Human Rights Commission, 2005). There exists a variance in results and recommendations from different countries, with recommendations also differing dependent on the condition the child has (Consensus Statement on Management of Intersex disorders, 2006).

For those adults whose genitals were surgically altered to reinforce a gender assignment and create 'normal' looking appearance there are a number of issues. These surgeries were often done without fully informing the parents of the diagnosis and risks of surgery, the children then grew up with their condition, and reason for the initial and subsequent surgeries being kept a secret from them (Diamond, 2004). Preves (1998), comments that this approach of being poorly informed, and encouraged to keep silent, led to an enforcement of feelings of isolation, stigma and shame. Added to this the surgeries were often unsuccessful in terms of outcome. In their 2000 review of the small studies published on outcomes for surgeries Fausto Sterling and Laurent found poor outcomes with vaginoplasties done in infancy having frequencies of stenosis as high as 80-85%. In 2006 the Lawson Wilkins Paediatric Endocrine Society (LWPES)

and the European Society for Paediatric Endocrinology (ESPE) reviewed the data on longer term outcome and had the following to say on surgical outcomes:

> outcomes from clitoroplasty identify problems related to decreased sexual sensitivity, loss of clitoral tissue and cosmetic issues. Techniques for vaginoplasty carry the potential for scarring at the introitus necessitating repeated modification before sexual function can be reliable. Surgery to construct a neo-vagina carries a risk of neoplasia. ...Analysis of long-term outcomes is complicated by a mixture of surgical techniques and diagnostic categories. (p.154)

In talking with adults who would have undergone surgery under this older paradigm of care I was able to speak to two women with CAH, one who had undergone surgery as an infant and one who had never had surgery due to her CAH being undiagnosed until she was in her twenties. One considered herself lucky that the surgery had not significantly damaged her genitals and the other was very pleased surgery had never been performed. In describing her surgery one of the women states *'I was lucky that I wasn't basically completely neutered for want of a better word like something you would do to a cat and I was at the whim of that surgeon on that day in terms of my future sex life and I am really grateful it was partial'*. She goes on to say she feels her surgery was satisfactory but feels she was just lucky and that if she had not been she *'would probably have been a pretty bitter person if it had of been the case because I didn't have a choice it wasn't an informed choice for me cos I was a baby'*.

For the woman who didn't receive surgery, on discussing what it was like for her to have virilised genitals she comments, *'I didn't know if I was normal or not because you don't go round saying hey can I look at your private parts'*. It wasn't until she was an adult and saw her sister at the pool that she noticed a difference, *'you know how I found out I went and got some porno books and that's how I found out, no one said this is what's normal and this is what's abnormal'*. She did remember that her husband said *'my you're big there'* but she didn't think anything of it. When I asked if she would have wanted to have surgery her reply was *'me personally I don't think I'd bother with the surgery now I'm quite happy I've got it...I had wonderful orgasms, never any problems, multiple orgasms'*. Both of these women indicate that they value sexual functioning over the appearance of their genitals. This is in contrast to the focus upon the appearance of the genitals in a 'pre sexual' infant where how the genitals look is more immediate then longer term concerns about sexual functioning. One of the participants indicated that having genitals that were different did not impact

upon her gender identity development as a child or on her ability to marry and have a 'normal' heterosexual relationship. This is in contrast to the argument that states genital appearance is linked directly to successful gender identity development (Eugster, 2004; Farhat, 2005).

I asked both women about parents choosing surgery. The woman who had undergone surgery stated *'when you are a baby you are at the whim of your parents or the surgeon or the paediatrician so it's almost like as parents you have to advocate for your baby and do the best with the information you have as a parent'.* If she herself had to make the decision now for a child, she would research and talk to surgeons *'have to be really strong in my conviction I'm making this decision for you my baby and I'm making it for these reasons or actually I'm leaving it to you so that when you're an adult and have the where withal to rationalise and understand about relationships'.* She thinks she would struggle with the decision, it would be really hard *'as a mother of a child do I determine their actual sexual journey or does nature do that and has nature already made a start on that'.* For the woman who hadn't had surgery her opinion was that she was *'pleased I didn't have surgery because I would rather be who I am I think mucking around changing genitalia I think is wrong, I think it should be your decision not your parents, I really do because no-one knows how you are going to turn out later in life.'* Both women refer to surgically unaltered genitals as 'natural' - surgical alteration is man-made or artificial. They suggest that decisions about genital surgery need to be made in light of the future implications for gender identity and sexuality. Complexity arises because this kind of 'normalising' genital surgery is not simply a medical decision; it has deeply social consequences.

Both women brought up the issue of whose right it was to decide about surgery. There have been a number of statements published suggesting that parents do not have the right to choose surgery for their child. In 2005 the San Francisco Human Rights Commission published its recommendations following their investigation into the Medical Normalisation of Intersex People. They consider that any 'normalising' interventions done without the patient's informed consent are inherent human rights abuses and violate the medical standards of informed consent. The Consortium on Disorders of Sex Development recommends delaying any surgery which is non life threatening until the patients themselves can participate in decision making. This however comes with an acknowledgement that parental distress may lead them to want surgery to 'normalise' the genitals and a multidisciplinary team will be required to

provide support and counseling for the family and later the child. They state parents and families need to be made aware of the controversies that exist in regards to surgery. The Lawson Wilkins Pediatric Endocrine Society (LWPES)/European Society for Paediatric Endocrinology (ESPE) Consensus Group statement on management of Intersex disorders published in 2006 recommends that only surgeons skilled in these surgeries should perform them and they have an obligation to outline the surgical sequence and subsequent consequences from infancy to adulthood. It goes on to state that evidence is lacking to support the theory that cosmetic surgery in the first year alleviates parental distress and promotes bonding. It also suggests leaving vaginoplasty until adolescence, offers recommendations for the timing of the removals of gonads in certain conditions and maintains an emphasis on preserving future fertility.

Parisi et al. (2007) in their review of 250 patients seen by the Gender Assessment Team at Children's Hospital, Seattle over the last 25 years comment on the change in approach to, and the question of the appropriateness of, genital surgery. They state it is now their policy to offer surgery as an option rather then the standard treatment with an explanation to parents about the existing data on the two approaches. They do however note that most parents still choose to proceed with genitoplasty in infancy with only a few parents choosing to delay surgery. In their interviews with parents Siminoff et al. (2007) found parents felt they received incomplete or contradictory information on the necessity of, and optimal timing for, surgery on external genitalia. This led some to delay a decision until they felt they were fully informed and could find an experienced surgeon. Duguid et al. (2007) found a substantial number of parents were concerned about the stigma or ridicule associated with their child's condition, but only a small number of parents felt that a delay in surgery would lead to additional ridicule at school.

In 2005 Lean, Deshpande, Hutson and Grover published their results for cosmetic and anatomic surgeries done at Royal Children's Hospital (RCH) in Melbourne. They cited good outcomes for those surgeries as long as initial surgery was done at RCH by a specialised surgeon. Those surgeries done by non specialist surgeons prior to referral to RCH showed poorer outcomes, leading them to recommend that only surgeons experienced in these surgeries should perform them. What constitutes experience was not specified however the inference is that volume of surgeries equals

expertise. The results include 15 surgeries on girls with CAH. Of interest they state that the age of reconstruction did not have a significant impact on results but they also felt the study did not support the abandonment of genital reconstruction in childhood. These results are of particular significance to the interview results presented in this thesis as RCH is the recommended centre for surgery for children from New Zealand with the government funding surgery at RCH.

In talking with an Endocrinologist from Starship Children's Health their policy of surgery is as follows *'We do not sex reverse ever'*, those who are born with XY chromosomes and a small phallus are assigned a male gender and do not undergo feminising surgery. However there is a recommendation for surgery for girls with CAH for two reasons. Firstly the positive outcome results from Melbourne where the surgery on New Zealand patients is currently performed and secondly the risk of multiple urinary tract infections and subsequent renal damage due to the prolonged urethra in significantly virilised infants. Prior to 1997 surgeries were performed at National Women's Hospital and the outcomes from these were *'awful'* with fistulas, stenosis and often the requirement for daily dilations. The recommendation for treatment in Melbourne is in light of the expertise of their specialist surgical team and the acknowledgement that the poor results identified in other overseas studies (where surgical expertise is lacking as surgery is perhaps performed only every 5-10yrs by an individual surgeon) might mean a different recommendation if Melbourne was not an option.

The co-ordinator of CAHNZ discussed the issue of surgery saying parents needed to hear both sides of the issue, with encouragement needed for health professionals to present both views to parents. Her suggestion was that parents are told there is information to help them make choices that are right for them and they are entitled to that information. They may choose not to have surgery because the virilisation is minor or they may want to wait until their child is older and can give their full consent. She suggested issues to consider are situations where the genitals may be exposed to others such as *'day care and nappy changing'* and *'issues with extended family'*. She also recommended the Melbourne team if surgery was the choice as *'Melbourne offers a team approach, nowhere in NZ offers seamless care for genital surgery'*. She also commented on

how agonised women in particular are about these decisions which was often made more difficult by their isolation.

Parents I spoke to who had had the experience of genital surgery on their infants fell into two groups, those who were happy with their experience and those who considered their experience to be appalling. The parents who expressed satisfaction were those of girls with CAH. For one mother surgery was suggested before they left the hospital and she described it as being *'not a hard decision'*. She felt surgery was *'a big decision but can't see why people wouldn't'* although she did feel it was *'scary to decide to have your child chopped up and stitched up'*. She felt it helped to talk to others who had done it and that they *'didn't want their daughter to grow up looking different and getting picked on'*. They did not send their daughter to preschool until after the having travelled to Melbourne for the surgery. In describing the surgery she commented that *'she looks pretty normal'*, the *'surgeon said anyone who wasn't medical wouldn't know the difference'* and *'she has no extra bits, surgery went really well'*. She was glad they did it but also expressed the sentiment that they were *'glad not to have to decide again'* when their 3rd child, a boy, was also born with CAH.

Another mother described getting conflicting information before leaving hospital *'one doctor very pro, one very anti...well he wasn't very anti but he said you don't have to do this you know'*. They were put in touch with the CAH support group and ITANZ and felt they were able to obtain information that let them *'make as an informed decision as we can'*. She spoke to other mothers of teenage girls with CAH and decided:

> *'we wanted the vaginal surgery done but not the clitorectomy as we had heard so many stories...why would you do this to me this is the most sensitive part and um we went to Melbourne to have surgery done and in Melbourne changed our minds and had the clitorectomy done as well just because no-one in NZ had actually explained the surgery to us properly'.....'good to have it over and done with in one hit and he could guarantee me success' .....'I feel I made the right decision...I mean you can screw up your child's life by sending them to the wrong school'*, (or) *'birthmark on the face could be just as damaging as choosing unnecessary surgery'*.

Later in the interview however she did question her decision a little:

> *'so I hope I made the right decision, we will find out in another 12-13 years I suppose...obviously we can't know till it gets put to use...you can only hope for the best can't you'.....'if it all goes to custard later on I would be stuck with the well I had all the information but you know 30 years ago as a mother you weren't given a choice but this is what has to be done, you can get out of the blame, but for me it is my fault'*.

This parent was interesting to talk with in that she had taken the time to gather a large amount of information on the arguments for and against surgery. She had spoken with an advocacy group, a support group, other parents, and medical professionals and come to a decision not to consent to a clitorectomy. The decision however was able to be reversed within one meeting with another medical professional when she was shown photos of other girls who had undergone surgery. Duguid et al, (2007) found in their study of parents of children with genital anomalies that one of the most common coping strategies used by parents was a reliance on clinical staff to make the right decisions and a reliance on the perceived 'treatability' of the condition. She seemed to have a good understanding that her daughter might not be happy with the decision later in life but could justify it in terms of it being no worse than the social impacts of defects such as a birth mark and other damaging social experiences that a child might be exposed to such as 'the wrong school'. She expressed awareness that decisions to consent or refuse surgery have consequences and envisages the potential for a fraught relationship between herself and her daughter if the decision turns out to be the wrong one for her daughter. She would consider herself to blame as she made the decision however at no point did this mother or any of the others who had chosen surgery express the sentiment that it was not their right to make the surgical decision.

The most negative surgery experience came from a mother of a child born with PAIS whose child had undergone surgery in Australia in the late 1990's to reinforce a gender reassignment to female despite male chromosomes. She describes the explanation she received from the surgeons *'this is the way it is, she can't be fixed with what she's got, or he can't be fixed, it had to do with small phallus size, so small, she had a testis and a half, basically it was easier to make a hole than a pole'.* She feels she was bullied into the surgery and feels a huge amount of pain and anguish about what happened.

> *'I have that guilt everyday and we live with that everyday that we signed those forms and we have ruined this kids' life because we were told to do it', 'knew it wasn't right but doctors knew best'.*

The consent forms were signed with no counselling and surgery occurred at 5 months *'we signed those forms with no counselling, we were grieving a loss he's no longer a male'.* Of the surgery she states *'surgery was really crude...now we know it honestly does not work, she started having problems at a year and a half'.* She feels that her daughter will be 'a *freak, she was a freak who was intact and she could have made that decision.'* Surgery in this instance has led to a child with reconstructed genitals which are now in opposition to the gender

identity the child has developed. This has had a huge negative impact on the child's psychological development and the family's wellbeing. Currently *'there are two psychologists working on what to tell her'... 'she can't get on with adults or other kids.'*

Discussing surgery with participants highlighted the fact that doctor's motivations for recommending surgery and parents reasons for seeking treatment can be variable. It also reinforced the need for parents to be fully informed about the issues. These issues include the lack of agreement in the literature as to whether the treatment should occur at all, international variability in treatment recommendations, and outcomes differences according to condition and the person's age and corporeality. Parents need to know there is a debate about whether surgery should be performed or not including the view that it might not be their decision to make. They need the time and support to look at all the issues surrounding surgery so they make a truly informed decision. It appeared that some of the parents spoken to for this research project were not fully informed or given enough time to make these decisions.

Gender Identity

'do you put it down to all the extra testosterone she was exposed to or is it just who she is'
Mother of a 4 year old with CAH

The birth of a child with female chromosomes but typically male appearing genitals, male chromosomes and typically female appearing genitals, undervirilised male genitals or chromosomes which are not XX or XY challenges definitions and perceptions about the relationship between biological sex and social gender. Parents, family members, health professionals and society in general all have preconceived ideas of what makes infants girls or boys. This section examines the issue of gender identity and what determines this for an individual. It looks at participants' experience of developing their own gender identity or for parents the experiences of watching your child develop their own gender identity. It also reviews studies in which girls with CAH are reported to prefer 'typically' male gendered play and the higher incidence of a homosexual sexual orientation in women with CAH.

One of the issues which arise for people when considering a child with an intersex/DSD condition is their own interpretations and world view of what it is to be male or female within their society and culture. This can then lead to an examination, or

confrontation of their definitions of the concepts of gender assignment, gender identity, gender roles, and sometimes the interchange ability (albeit wrongly) or confusion between these terms and the term sexual orientation. Gender assignment is the 'boy' or 'girl' question usually answered at the birth of a baby or in many cases during pregnancy by the observation of typically male or female genital anatomy or chromosomes. It is also referred to in the literature as assigning a sex to a baby with the literature further referring to gender of rearing and sex of rearing (Blizzard, 2002; Creighton, 2001). An intersex child is always given a gender assignment but often this is delayed or in some cases changed as test results become available. Gender identity is how a child or adult identifies themselves, male, and female or in some cases as both or even adopting the term intersex. An individual's gender identity may not necessarily correspond to the gender assignment that they were given as an infant. Gender identity is thought to be determined by complex interactions, endocrine influences pre and postnatally, genetics, and environmental and psychosocial experiences (Ahmed, Morrison & Hughes, 2004). Gender roles are culturally and socially defined 'norms' for the behaviour of males and females and include such stereotypes as girls playing with dolls and boys with trucks. Sexual orientation is a persons' preference for same gender, opposite gender or partners of both genders. Sexual orientation is also defined as heterosexual, homosexual or bisexual. How all of these come together varies from individual to individual, as Ahmed, Morrison , and Hughes (2004) comment 'It is one thing to have a vulva, vagina, clitoris, breasts, ovaries, 46XX karyotype, etc, but it may be quite another thing being female, feminine, or a woman' (p.847). Creighton, (2001) notes that there is no indication in a newborn of gender identity or sexual orientation whether a child has an intersex condition or not. Of importance, Hughes, Houk, Ahmed, and Lee (2006) from the Consensus Group on management of Intersex Disorders also point out that a homosexual sexual orientation as an adult does not correspond to an initial incorrect gender assignment. They also note that following their review of the literature there is still much to be clarified about the determinants of gender identity in individuals with DSD's.

The traditional paradigm of medical and surgical treatment of intersex children has sought to categorise them into male or female based on outward genital appearance and a predicted heterosexual orientation, i.e. surgeries to allow penetrative heterosexual sex. (Ahmed, Morrison & Hughes, 2004; Daaboul & Frader, 2001). The ethical issues

behind this are then, who and what determines what makes an individual male or female, and should those who generally decide (the medical profession) get to perform surgery to enforce their definitions when this might not be how the individual identifies. Conversely if surgery does not occur will the individual be confused and psychologically damaged by having genitals that do not conform to what has been deemed female or male. In agreeing to surgery are parents trying to deal with their own issues about gender identity and sexual orientation. Two issues arose in talking with participants on issues surrounding gender assignment and identity. The first was around children and adults who did not identify with the gender assignment they were given at birth. The second was around girls and women with CAH who exhibited typically male behaviours and the link between this and their antenatal exposure to androgens.

The potential for a difference between gender assignment and gender identity is one of the reasons that genital surgery is opposed. (ISNA website; Blizzard, 2002). Mazur (2005) in a large review of results for 46XY individuals with CAIS, PAIS or micropenis found 9% of those with PAIS changed gender. Migeon et al (2002) in a long term study of 46XY individuals found that 23% were dissatisfied with their assigned gender. The parent I spoke to with a daughter with PAIS who is chromosomally male feels surgery should not have been done to reinforce a female gender. In describing her daughter she says *'knows she is different but doesn't know why, knows she is different from her sisters'.... 'plays with typically male toys'.... 'more personality traits that are typically male'.... 'hated the pink frilly dresses and dolls, wants to cut her hair' ....'tries peeing standing up'.* In short her daughter *'wants to be a boy'* and is rebelling against the stereotype of *'nice little girl'.* She does not however know what she should tell her daughter and when or how she should let her know the truth. She is very angry that genital surgery was done on her child and felt bullied into consenting. She would like to be able to tell the surgeons *'you've ruined her life, she's got no genitals, she's nothing she's not here or there, why would you treat children like that'.* The experience of this family would appear to support the proposition that genital surgery should not be performed until the child is old enough to develop and express their gender identity and consent to the surgery themselves.

There have been a number of studies looking at gender role and masculine behaviour in girls with CAH and the relationship to their pre natal androgen exposure.

Nordenström, Servin, Bohlin, Larsson and Wedell (2002) looked at the degree of fetal androgen exposure, on toy play and toy preference in girls with CAH. They concluded that girls with CAH played more with masculine toys than controls and that there was a dose-response relationship between the degree of fetal androgen exposure and the degree of masculinisation of toy play and preference. Berenbaum, Duck and Bryk (2000) compared girls with CAH to their unaffected sisters and also found that they played more with 'boys toys', and were more likely to use aggression when provoked. It could also be argued that the experience of having CAH and having undergone numerous medical examinations and having been subject to different experiences due to the medical nature of their condition may cause aggression under stress that should not be solely attributed to androgen exposure. Hall et al (2004) also concluded that behavioural masculinisation in girls with CAH was a consequence of prenatal androgen exposure. Berenbaum and Bailey (2003) looked at this gender behaviour in relation to gender identity in girls with CAH. They concluded it was not possible to predict across behaviours, particularly from gender-role behaviour such as toy play to gender identity or sexual orientation.

Other studies also identify a majority of those girls and women with CAH given a gender assignment of female also having a gender identity of female. Gastaud et al (2007) looking at sexual and reproductive outcomes in women with CAH found that none of the women expressed doubts about their gender assignment. Dessens, Slijper and Drop (2005) in a study of over 250 women with CAH found that 94.8% raised female later developed a gender identity as girls and women. There were however 5.2% who did have serious problems with their gender identity and the authors point out that this is higher than the prevalence of female-to-male transsexuals in the general population of chromosomal females. They did go on to conclude that no matter how severely virilised, those who were chromosomally female with CAH should be given a female gender assignment. Blizzard (2002) however offers a challenge to dogmatically declaring all those with CAH and 46XX chromosomes should be raised female. Citing a lack of evidence based research he questions whether those who are markedly virilised with complete fusion of the urethrolabial and urethrolabial folds with a resultant penis-like phallus (Prader 5 sex staging) should be considered for a male gender of rearing. This would avoid any surgical procedures and he queries whether 'the loss of sexual pleasure and the acquisition of an inadequate vagina may be too high

a penalty to pay'. (p.620). The question in determining a gender assignment for 46 XX girls with CAH is are they going to identify as female as they get older. Even if a number do not would this be any higher than the number of girls without CAH who later identify as male, and should this affect gender assignment? In this study all of those with CAH who appeared virilised at birth had been given a female gender assignment, and while none expressed dissatisfaction with their gender identity all, either through self report or in the case of children parental observations, engaged in and strongly identified with behaviours that could be described as 'typically male'.

Sexual orientation is another issue which has been examined in females with CAH. Dittmann, Kappes and Kappes (1992) in comparing girls with CAH with their sisters found that twenty percent of the patients wished for and/or had had homosexual relationships as opposed to none of the sisters. Gastaud et al. (2007) also found twenty percent of their participants had homosexual inclinations. Of the adult women I talked with who had CAH both identified as heterosexual however both mentioned identifying more with a typical 'male gender role'. One explained that a doctor had told her mother that *'CAH females because they have been exposed inutero to testosterone often have more male type features- the wee tomboy or sporty kid or more aggressive of which I was all of the above'*. She felt however that it had been a good thing as she *'played a lot of sport, I was assertive as a young person'*. The other explained that *'all the testosterone gave you the aggression and the drive'* and for her this translated to an *'extremely high sex drive'*. A number of parents also commented on their daughters preferences for typically 'male toys'. One described her daughter as *'has no interest in dolls, likes dinosaurs, trucks and cars'* she had wondered whether this was due to the CAH or just *'who she is'*. Another described her daughter as *'a tomboy and very sporty'*. She also saw these as positives, especially the sports as the fitness level would help with *'weight issues'*. Of the four girls with CAH, whose parents talked to me, none were expressing a gender identity that was not female but at ages 15, 9, 5, and 4 years there is a possibility that this is not resolved for these children yet.

Genetics

'Guilt over both of you having a recessive gene and giving your child this condition which is so totally devastating and life threatening'.
Mother of a child with CAH

For many intersex conditions there is a genetic component and this raises issues for parents in terms of a diagnosis, carrier testing of children and the possibility of using genetic screening in future pregnancies. This section looks at the implications of intersex/DSD conditions being genetically determined. It reviews the genetic determination of individual conditions and the ethical issues of carrier testing of children, pre implantation genetic testing and the termination of pregnancies affected by an intersex/DSD condition. It examines the effect a genetic basis has had for participants in terms of future pregnancies, genetic testing and decisions around when and how to tell their children about the genetic component of their condition.

As there is a genetic basis to a number of intersex conditions this raises issues for parents in terms of a diagnosis, carrier testing and future pregnancies. It can also be associated with feelings of guilt or blame for the child's condition. One woman with CAH described her mothers experience as fearing *'through the whole next pregnancy, whole lot of grief that went with parenting babies a long time before much was known about condition'.* Another mother described her feelings that CAH wasn't just *'this baby but the rest of family and subsequent pregnancies'.* It was not simply a matter of dealing with a condition in one child but the implications and impact it had on any further reproductive decisions, not only for them as parents but for all their present and future children. Genetic testing also meant that parents or some family members felt they could assign an explanation or blame for the condition. A mother of a son with Klinefelter Syndrome recounted how when she found out her son's diagnosis and told family members her mother said to her *'oh it's your fault he's got it'.* Skirton (2006) in exploring the experience of twenty sets of parents with a paediatric genetic referral found all the mothers interviewed expressed guilt over their perception of being the cause of their child's condition. Duguid et al (2007) found that 26% of parents in their study expressed concerns about the condition recurring in their future children or their affected child or the child's sibling's offspring.

CAH is an autosomal recessive condition, with 90% of cases being caused by 21-hydroxylase deficiency (White & Speiser, 2000). If both parents are carriers then

there is a 25% chance a child will have CAH and a 50% chance a child will be a carrier. Once a child is born with CAH this raises the question of carrier testing of siblings and extended family members and decisions about testing in future pregnancies and taking steroids. Mothers who have had a previous child with CAH have the option of taking dexamethasone in the early weeks of pregnancy to prevent or reduce virilisation in an affected XX fetus. After a chorionic villus biopsy (CVS) is performed between 6-11 weeks then the treatment can be stopped for unaffected fetus's or XY fetus's with CAH (Bianchi, 2002; Woodhouse, 2004). However there remain questions about the long term effects of dexamethasone and it has unpleasant side effects for the mother, excessive weight gain, severe striae, hypertension, and hyperglycaemia (White & Speiser, 2000). As Bianchi, (2002) comments "a major concern is that seven of eight fetuses will have unnecessary exposure to dexamethasone during an early, and presumably vulnerable, period of gestation" (p.689)

The parents I spoke with who had a child with CAH had made different choices about future pregnancies. For two women an active decision had been made not to have further children because of what was involved with taking care of their child with CAH. Both had wanted more than one child but for one *'my biggest concern was virilisation rather than the condition itself, I was told about dexamethasone, wasn't willing to take it', 'couldn't cope with two with CAH'*. For the other woman *'I heard about the tests no way, I never wanted an only child but wouldn't go through it all again'*. For another woman her child with CAH was her last but she felt if she had a CAH child to start with she wouldn't have gone on to have five children, she described it as *'like playing Russian roulette'*. She stated she would worry more about *'the effects of dexamethasone than virilisation'* if there was a further pregnancy.

For those who did go on to have further pregnancies the experience of getting dexamethasone prescribed and taking it was not an easy one. For one woman it was *'hard to get good info on dex, told by some not to do it'*. She experienced doctors who did not warn her about future pregnancies and another who refused to prescribe the steroids. Once on them she found them *'horrible, absolutely disgusting'*. She also felt that having to make a decision to commence them early in the pregnancy meant it *'takes the spontaneity out of conceiving'*. A second woman also found it hard to get information about dexamethasone, her child's paediatrician told her it was no longer used and she had to rely on the support group for information. She too found it *'left me a blithering*

mess'. The pregnancy was described as a 'surprise' but her decision prior to that, to have no more children was not influenced by CAH. While she described *'disappointment'* that this third child also had CAH she was *'pleased he was a boy so we didn't have to go through that side of things' (referring to virilisation and surgery which had been issues for her daughter).* The head of CAHNZ felt that it was important to talk with parents about future pregnancies once they are more in *'maintenance mode'*. She felt there needed to be an information sheet about dexamethasone framed in a neutral manner with pros and cons laid out, and stressing that it is a parental choice.

The second issue these families faced was carrier testing. Once a child has a diagnosis genetic testing can identify the specific gene mutation leading to CAH. This information can then be used to look for CAH in future pregnancies or for preimplantation genetic diagnosis, or to look at siblings or other family members for carrier status. Each of these choices also presents some ethical questions. Carrier testing of children is considered controversial due to concerns over effects on a child's self esteem, self concept or anxiety if told early versus their ability to adjust to the knowledge over time if they are a carrier (Borry, Stultiens, Nys, Cassiman, & Dierickx, 2006). The issue of informed consent arises, i.e. is it the parents' right to give consent to carrier testing or should the decision be the child's as they reach an age where they may be sexually active and be making reproductive decisions, as carrier testing impacts only on their reproductive future? In a review of published normative ethical and clinical guidelines concerning the genetic carrier testing of minors, Borry, Fryns, Schotsmans and Dierickx (2006) found that carrier testing of children could be reasonably deferred until the child had the intellectual capacity to decide when and if the testing should be done. They concluded that carrier testing should not happen until a child could give informed consent. They did however note that these were the views of medical and ethical professionals not parents. Campbell and Ross (2005), when looking at Parental Attitudes and Beliefs regarding the Genetic Testing of Children, found that most parents believed they should be the final decision makers.

Preimplantation genetic testing allows embryos that are to be implanted in an IVF procedure, to be tested for genetic conditions, then allowing for the implantation of unaffected embryos only. In a study of 250 cycles of preimplantation genetic diagnosis (PGD) including screening for CAH, Fiorentino et al (2006), surmised that 'PGD will play an increasing role as a specialized clinical procedure, becoming a

useful option for many more couples with a high risk of transmitting a genetic disease, to prevent the birth of children affected by monogenic defects.' (P.683). Ethical concerns around PGD are the subject of medical and social debate (Ettorre, 2002; Kanavakis & Traeger-Synodinos, 2002; Robertson, 2003) and include the selection and determination of what is 'healthy' or 'normal' in society and the destruction of embryos not considered 'perfect'. It is however an option that may become more readily available to parents with a known genetic condition. Genetic Services, Auckland stated that they did not perform carrier testing of siblings till they were of reproductive age although adult family members could be tested for carrier status. While none of the parents I spoke with had the option of PGD as they were not IVF pregnancies a number did know about the option of finding out the carrier status of their children or already knew due to prenatal testing in order to determine whether they could stop taking dexamethasone. This then left them making decisions about what to tell their children regarding their carrier status.

Klinefelter Syndrome results from sex-chromosome aneuploidy, with 80% of cases being 47XXY, and half of the cases being paternally derived (Lanfranco, Kamischke, Zitzmann, & Nieschlag, 2004). In pregnancies with a prenatal diagnosis of Klinefelter's there is a high incidence of termination. Holmes-Siedle, Ryynanen and Lindenbaum (1987), found in a study of forty cases of sex chromosome aneuploidy 62% of the pregnancies were terminated. Marteau et al (2002) also found in a study of 111 prenatal diagnoses of Klinefelter's 44% of pregnancies were terminated. The determining factor influencing termination appeared to be the professional background of the health professional providing post diagnostic counselling. Lanfranco et al (2004) describe this as a 'high proportion for a syndrome of variable phenotype that can present with a very benign clinical picture' (p.280). Men with Klinefelter's almost always have azoospermia yet with ICSI (Intra Cytoplasmic Sperm Injection) where a single sperm is injected into an egg, fertility is now possible. However with this comes a higher than average risk of a child with 47 XXY or 47 XXX (Lanfranco et al, 2004). This in turn leads to further ethical issues of PGD and selection or termination of an XXY pregnancy. On speaking with genetic services they stated they had the feeling '*that you offer parents different information antenatally as opposed to after birth, information is different when you have no choice abut terminating vs. continuing.*'

The woman I spoke to with a son with Klinefelter Syndrome felt that knowing it was genetic would effect a conscious decision to have more children. When she became 'accidentally' pregnant with another child she says she was *'pleased she wasn't a boy'* but was shocked that pregnancies were terminated for Klinefelter's saying *"but still healthy babies'....'oh nasty- that's never been a consideration'*. Later in the interview however she mentioned that her husband had wanted to terminate the pregnancy in case of another child with Klinefelter's and threatened to leave if *'there were big problems'*. In regards to her son's future infertility she says *'rather upsetting time once it was explained he would never have children'*. She also explained that it was good to have that knowledge so she could pass it on to him and any future partner he may have but was uncertain of how to have that conversation with him.

AIS is an x linked recessive condition (New et al. 2001) affecting the human androgen receptor gene. Therefore a carrier can transmit the condition to one in two of their male offspring and in heterozygous form to one in two of their daughters (Hughes & Deeb, 2006). Individuals with AIS can therefore be traced through generations of families, although in 30% of cases the mutation arises spontaneously. Conditions of androgen insensitivity are varied and present different issues in terms of fertility and reproduction. Individuals are 46XY with differing phenotypes. In CAIS (complete androgen insensitivity) individuals appear phenotypically female although they do not have ovaries or a uterus. The gender of rearing is almost always female. Except in cases (and this is likely to become more common) where prenatal testing has revealed a male karyotype this condition is often not diagnosed until puberty with primary amenorrhea as the most common presentation or if diagnosed pre-puberty with the presentation of inguinal hernia. In PAIS there are varying degrees of under virilisation and infertility and gender of rearing is varied with a large number of assignments to a female gender with feminising genital surgery (Melo et al., 2003; Viner, Teoh, Williams, Patterson & Hughes, 1997). For the child with PAIS whose mother I spoke with this was the case, feminising surgery had been done to reinforce a female gender of rearing. Her mother was told *'she will have reconstructive surgery and be a great girl'*. The child however has grown up with a male gender identity and her mother at the time of the interview was struggling with finding a way to tell the truth about her surgery and gender assignment but stated *'If H wants to become male it's okay'*.

Those with gonadal dysgenesis or Swyer syndrome have female appearing external genitalia and a uterus but no ovaries so pregnancy is possible with the use of donor eggs in those raised with a female gender assignment. Migeon and Wisniewski (2003) comment that in their experience few women with Swyers syndrome become pregnant and query whether this is due to a lack of desire to pursue this fertility option or a lack of knowledge as to its' possibility. The woman I interviewed with this condition felt that parents worry a lot about the fertility of their children, and that parents of a child with this condition needed to know that it is not the genetic component that is important *'It wasn't knowing I had a Y chromosome, that wasn't the important thing, not being able to have children was the important thing.'* She had been able to go on and have children via donor egg IVF and comments on how important is was to her to know this was an option: *'what a difference an egg donor has made to our lives'.* She further commented that there was *'actually nothing wrong with me I just don't have ovaries'* and considered this a healthy place to start IVF from, as opposed to years of infertility.

A genetic basis to a number of intersex/DSD conditions means parents face further hard decisions once their child is born. They need to consider further pregnancies both in terms of having another baby and the possibility of preimplantation genetic testing and the possibility of termination. In the case of CAH there are decisions regarding dexamethasone to lessen virilisation and the side effects for both mother and infant. There is also the issue of the carrier status of the child with the condition and their siblings, whether and when to test for this and how to, and when to tell their child/children about their carrier status. Responses from participants indicated a need for information on genetic testing and carrier testing. Parents also wanted to know how to go about telling their children about the future implications of their conditions in terms of genetics and reproduction.

Growing Up

> *'I don't like to be a different girl because I just want to be normal cos if I break a bone and they don't give me an injection I might die and I don't want that to happen"*
> 9 year old girl with CAH

This section looks at what is it like to grow up with an intersex/DSD condition, to go through puberty and on into adulthood. It examines how the decisions made for an individual as an infant impact on their life and the issues which arise for children and their parents and families. Participants views of whether life becomes better or harder are presented and the question of when and how to talk to children about their condition is examined.

Case studies and narratives from those treated under the traditional paradigm highlight a number of issues which have arisen. The theoretical framework underpinning the paradigm meant it was based on secrets and non disclosure. Physicians did not necessarily fully inform parents and they in turn were advised not to reveal anything to their children in the hope this would reinforce the gender assignment given. As these children grew many were unaware they had an intersex condition and never fully understood why they had undergone surgery or why they had repeat surgeries. When they did discover the truth it often reinforced the feelings of shame and stigma they had over being different and led to feelings of anger and resentment over their treatment. They were also dissatisfied with the surgical outcomes and the damage done to their genitals. (Diamond 2004; Frader et al, 2004; Lev 2006; Liao, 2003).

Diamond (2004), describes this sequence of events thus;

> typically, patients discover their condition from an inadvertent family slip, community gossip, personal investigation into puzzling aspects of their lives, or mix-ups at the doctor's office. Without openness, the patient discovers that his or her condition is shameful in the minds of parents and doctors. They wonder why they were not accepted and loved as they were and on what grounds it was decided that they could not manage the information. Also, the patient learns that s/he has been deceived since childhood by the people who should have been the most trustworthy—parents and physicians. All of this is damaging. To the extent that these children are misled, as they mature to adulthood they cannot act rationally from a realistic appraisal of their medical condition. (p.598)

Follow up studies have often focused on surgical results rather than the experience of growing up with an intersex/DSD condition. Ogilvie et al (2006),

comments that little authoritative information exists on the social and emotional well-being of women and men with CAH. Hughes, Houk, Ahmed, and Lee (2006) note that clinical practice focuses on gender and genital appearance as key outcomes, but the salient issues for those affected are the stigma and experiences associated with having a DSD.

The last ten years has seen some long term follow-up studies looking at the psychosocial effects of these surgeries being published. Midgeon et al (2002), reviewed 39 XY individuals, 21 assigned a male and 18 a female gender of rearing. They found that 60-70 percent of the participants had sought counselling for issues such as concerns about romance and dating, sexual function, problems with family members, depression, substance abuse, and teasing. Liao (2003) in discussing psychological aspects of conditions associated with atypical development of the reproductive-genital system reported that clients were distressed by medical practitioner's previous non disclosure of aspects of their condition and its implications. Their current concerns often related to having to discuss atypical genitalia with sexual partners. Johannsen, Ripa, Mortensen and Main (2006) looked at the quality of life and psychosocial well being in seventy women with disorders of sex development. They found that there was an impaired quality of life and more affective distress especially in CAH patients and virilised 46, XX and 46, XY females. This included fewer women in relationships and with children, more suicidal thoughts and a higher rate of psychiatric counselling than in the control group these women were matched with. The authors surmised this resulted from the trauma caused by distressing diagnostic procedures, the chronic illness per se and psychosocial consequences of the disorders.

There are recommendations in most of the academic literature pertaining to intersex/DSD conditions for families and individuals to receive a comprehensive approach which includes psychological counselling, social work support and referral to support groups. Frader et al (2004) comment that parents should tell children in age appropriate fashion about their condition. They note that the differences should provide opportunities to explore the value of individuality and diversity rather then being occasions for humiliation and shaming. Slijper (2003) suggests that sexual counselling should begin pre adolescence because of hormone replacement therapy and the need for further vaginal surgery. This was based on a study by Minto (2003) who found that the 28 sexually active participants in her study had sexual problems.

Diamond (2004) also states that health professionals should be open to what 'young patients tell you and help them probe their questions and doubts about identities and preferred behaviors'. These authors do not however offer practical written advice for what exactly parents or health professionals should say to these children. Duguid et al (2007) looked at the psychological impact of genital anomalies on the parents of affected children. They found that over 60% of parents expressed concerns about the condition being associated with ridicule or stigma and also had difficulties in discussing the condition with relatives and friends.

All parents I talked with expressed their feelings of how hard the first year or two were, and how their child's condition affected the family as a whole. Statements such as *'it changes the rest of your life'* and *'affects your whole life'* were common. A mother of a child with CAH describes how it affected her expectations of parenthood *'as a parent you know you will give up a portion of your life, with a child with a dependant medical condition you give up a huge portion of your life'*. Another stated the *'stresses involved in caring for him impact on everyone and everything'*. For some it had a significant impact on their relationships, one woman described the *'huge strain on marriage'* while another stated the birth of her child with PAIS *'affected family and marriage, marriage imploded'*. One mother of a child with CAH described her life as being *'constantly watching and on guard'* and how she than went on to *'become isolated'*.

As babies grew into preschoolers the issues became centred on childcare and the possibility of going to preschool. The expectations that they could go back to work, have their child in day care or preschool and have their child have experiences such as play dates were changed for these parents. Parents stated that it was *'hard to get childcare someone to baby-sit'* and *'I worry about working, who will look after him if he gets sick'*. Another mother stated *'he has a caregiver I wouldn't consider a crèche'*. For another mother the decision was for her daughter *'not to go to preschool till after surgery because of her genitalia'*. Social occasions also raised issues of how much and what to tell others about their child's condition. In talking about her daughter's CAH one mother described why she tells people they found out via a blood test rather than her genitalia at birth *'not because I'm ashamed or anything like that but I think she doesn't need the bother, it's within family and friends'*. Mothers of preschoolers with CAH expressed their worries over leaving their child with others *'birthday parties, can't leave on her own, other parents scared about emergency*

meds' and *'can't just leave with anyone, or go home from kindy for a play date without explanations and dealing with peoples questions'*. Parents of children with CAH still had ongoing worries about their medical condition *'when she is sick, can't just let her sleep it off, worried about adrenal crisis, do we need to go to Starship'*. For one family it impacted their decision about where their family should live *'want to live out of Auckland but having heard stories of other hospital services wouldn't feel comfortable being too far from Starship'*.

This was also the time when parents first began to wonder what to tell their children about their condition. Most parents of children with CAH had begun offering simple explanations about their medical condition but had not talked about any other issues. One mother stated her daughter *'doesn't know anything about surgery knows she has to take medicine to make her healthy and knows when she has gastro bug she has to go to hospital'*. Another mother stated her daughter *'knows about her medical condition and her medic alert bracelet'*. The school years also brought up issues for parents in terms of what to tell their children. One mother talked about the issues surrounding the viewing and touching of her child's genitals at yearly medical checkups *'she needs to be checked yearly for virilisation she was embarrassed at last clinic'..... 'I need to keep her safe only doctor can look and only if mummy is there really hard to explain without scaring her'*. She went on to talk about how hard it was to teach her child about privacy but also that *'she needed to let the doctor look'*. Addressing these issues also had a positive effect on their relationship she described the effect of these talks as *'makes you closer can talk about the body and how it works why it happens'*. Another mother described her daughter as knowing *'she is different anatomically, why do my bits hang out, why don't I have a hole'* but she then went on to state she didn't *'have the tools to tell her what happened'*. A mother of a son with Klinefelter syndrome also stated she wanted to know *'how to tell my son, he has no understanding'*. He has some developmental issues with coordination and memory and *'has no idea about Klinefelter's'*.

None of the parents I spoke to felt they had adequate information resources to have these personal and what they perceived to be difficult discussions with their children. The parents of younger children all expressed concern over what to tell them when they reached adolescence. One mother stated *'how to talk to your child in age appropriate ways would be good, to be sent out up to date info as they get older'*. Another talked about how her daughter would *'need to know about surgery and see a gynaecologist who could explain and make sure things are normal.* This would *'need to start at 9 or 10 years and build a relationship from there'*. She also stated *'I need more information about puberty'*. A mother of a

son with Klinefelter's talked about her fears for her son because all she had been able to read about puberty was about *'violent young men without testosterone'* and *'the high number in prison'*.

I was able to talk to two parents about their adolescent children. One mother told the story of her daughter telling her she *'halved her medication because of weight concerns'* although she thought she was 'an *intelligent aware child*, and they had had many conversations about why the medication is important. The head of the CAH support group talked about the different issues that arise for adolescents. She described these issues as being *'sensitive and even traumatic'*. In terms of medications *'when they take over control of medications they need another lot of info, this is why it is important, this is what will happen if you stop/ half/ change doses, these are the risks'*. There is also *'extra complexity around sexuality'* and *'issues around tampons, dilation, sex'*. She described how it may be hard to talk about sexuality for parents, and the need to find someone your child can talk to, and to start a relationship with a health professional earlier than puberty. She suggested adolescents could go into appointments by themselves and that they should *'build trust and a relationship before any vaginal /genital exam is undertaken'*

One of the few groups to address these issues with practical information is ISNA. When I travelled to talk with Jane Goto and David Cameron from ISNA in Seattle they gave me a draft of a booklet which has become available since 2006 in a downloadable format from the ISNA website.
(http://www.dsdguidelines.org/files/parents.pdf). This is an excellent resource which talks about developmental stages for children from birth through adolescence, addresses the specific issues which might arise for a child with a DSD and gives practical examples of how to talk to children of different ages.
An example from the section on talking with pre schoolers (p.23)

Child to parent	Why does my penis look different than the other boys'?
Parent to child	Your penis looks different because you have something called [specific name of DSD]. Before you were born, when you were growing in Mom's womb, your penis formed in a different way because of this.

For talking to children 6-11yrs (p.29)

Parent	Would you like to know more about your surgery? Maybe you'd like

to child to know about why we chose it, and what it seemed to be like for you as you healed after it? Would you like to know how I was feeling about you at the time of your surgery?

For talking with adolescents (p.34)

Parent to teen I'm not sure whether or not you feel comfortable talking to me about issues of sexuality right now. I just want you to know that if you do have any questions to ask, I will always be open to talking with you. We can talk about absolutely any issue or question that you would like to bring up. I might blush, but I do want to talk! (Pause. If necessary, add :) If you would rather talk to other people about sex, I can help you talk with other teens or with a professional therapist.

After completing my interviews I took the opportunity to post a paper copy or the web link of the 'Consortium on the Management of Disorders of Sex Development Handbook for Parents' to those who expressed an interest in having this resource. This resource was not available at the time this research was proposed.

Of the adults I talked to there seemed to be a general acceptance of their conditions despite often having had traumatic experiences as children or adolescents. One woman with CAH stated *'I'm still a human being, this is the body I've got, accept it and if someone else wants to look at my body and criticise me than they don't understand life as far as I'm concerned because we are all different'*. Another woman in talking about how CAH had affected her life and how she managed it said *'I was infertile by virtue of poor control and a tiny little bit of non compliance that lives within me in that I've always thought so what my 17 OHP is high, I was born with it high and it's the way my adrenals are and I'm not that hell bent on having it absolutely normal'*. Another with gonadal dysgenesis stated *'I don't see that my life is any different from any other woman despite the fact that I have a Y chromosome instead of a second X'*.

Summary

The diagnosis of a child with an intersex/DSD condition brings up complex issues for parents, family and whanau. For those parents whose child is born with genitalia that mean tests need to be done to determine an initial gender assignment, right from birth they are challenged and need to think about issues which most new parents do not consider. The outward appearance of typically male or female genitalia usually answer the 'is it a boy or a girl' question and thoughts about future gender identity or possible sexual orientation are not often the subject of discussion following the birth of a baby. For parents of a child with an intersex/DSD condition choices

about future pregnancies involve thinking about more than the ideal age gap for an individual family. The genetic determinates of a number of intersex/DSD conditions make reproductive decisions more considered. A lack of accurate information about these choices can further add to the complexity of decision making for parents. For those where genital surgery is a consideration or in some cases a recommendation the amount of information available concerning the controversies surrounding surgery, and whether a balanced view of the ethical issues, specifically informed consent and whose right it is to give that consent, also add to the complexity of parental decision making.

While parents are considering these hard decisions they also need to negotiate a way of, and the degree to which they tell family, extended family, friends, and even strangers about their child's condition. As a child grows and preschool or day care and then school enter their lives these issues must be considered again. This time also brings the issues of what and how to tell a child about their condition. Parents in this study did not perceive they received age appropriate information about child development and how to talk to and answer their children's questions. Issues such as having to explain to a child why she needed to let a doctor examine her genitals were in opposition to the messages parents considered they should be giving their children to keep them safe. Adolescence, when sex and sexuality become topics of necessity between parents and children, was a time participants with younger children expressed having worries about. These 'hard issues' which arose in participants narratives emphasised the lack of information available to parents and the need for this to become available to help parents make informed decisions and to be able to involve their children in these decisions as they matured.

Chapter 6

WHAT IS HELPFUL INFORMATION? CONCLUSIONS AND RECOMMENDATIONS

'parents are so strongly influenced by advice that they are given especially if they are terrified or feeling guilty or they are thinking oh my god this child's not normal or I don't like this child or how will this child ever survive'
Woman with CAH

Introduction

The question which this research sought to answer was 'What information do parents need when their child is born with an intersex/DSD condition'? Chapter 4 on 'finding out' highlights the fact that parents interviewed for this research project felt they were poorly informed at the time of their child's birth and that many continued to feel poorly informed until they were put in touch with a support group. At the time of the interviews participants still felt they did not have enough knowledge and support to be able to talk to their child or anticipate what issues might arise as their child grew older. A number of adults within intersex/DSD conditions that I spoke to also felt they had grown up being unaware of what their diagnosis meant and the implications for their future health and well being. My final questions to participants included 'what information would you of liked to have had at the time your child was born'? or 'as you were growing up'? 'What information have you found helpful'? And 'what advice would you offer to other parents'? Drawing upon the information provided by people who participated in this study, this chapter considers the immediate, intermediate and long term information needs for parents and adults living with intersex/DSD conditions. It concludes by providing recommendations for the education of health professionals, written information for parents and identifies the need to strengthen the team approach to the treatment of children with intersex/DSD conditions.

Participants Answers

At the time of first hearing of their child's or their own condition participants stressed the importance of the way health professionals talked to them. They wanted the news to be delivered in a gentle but straightforward way, and advised health professionals to;

'be gentle with the way you say it'
'don't launch into long winded words'
'need medical information straight'
'be very aware of peoples feelings'
'if possible give a name and label to the condition'

These answers highlighted the need for health professionals to have good inter-personal skills as well as some education themselves around intersex/DSD conditions so they could focus on the needs of parents and families rather than a pathologising diagnosis. Health professionals need to be able to give parents accurate information about conditions or if an immediate diagnosis is not available, which is often the case when a newborn presents with genitalia which differ from the norm, and be able to talk with parents about which tests will need to be done. Health professionals need to be aware that parents or individuals hearing about an intersex/DSD condition for the first time may be shocked and go through a grieving process. The way parents and families are talked to and the language used will potentially have a long term impact on how they understand and accept their child's condition. Information needs to be accurate but delivered in an empathic and caring way. If a health professional is unsure about a diagnosis or a condition they need to be honest with parents or an individual. Over-reactions or panic by staff in a delivery room will only serve to increase a parents' sense of fear or distress. This education needs to be available not only to health professionals in a hospital setting but also to those in community settings. Participants stressed that;

"It should be more widely known about during pregnancy'
'GPs and plunket nurses need good information'
'need to challenge doctors for information'

One participant with CAH who was also a health professional advised other health professionals to:

'Set up realistic expectations about what they might be having to deal with, think about or live with, if you have a baby who is intersexed you should be having that discussion'. 'you don't want to terrify them but don't want to shield them from something they will have to confront in real life'

Two participants suggested the words they would have liked to have heard, one when her child was born, and the other as a teenager when she discovered her diagnosis.

'say we think your child has an intersex condition we are not sure whether you have a boy or a girl'

'need someone in hospital who is really tuned up on this stuff, who can sit down on the end of your bed and say you're probably feeling really crap now, here's what I know about this'

The first participant indicated that it is acceptable for health professionals to communicate their uncertainty about the gender of and/or medical diagnosis for their child. The second participant suggests that information and empathy are important. She identifies a preference for support by health professionals who are knowledgeable about the everyday realities faced by people living with intersex/DSD conditions.

Written Explanations

Participants expressed a desire for some simple written explanations for themselves or their families;

'a simple one page explanation you can give to people who need to know like your parents'
'basic handout about the condition, explanation of what it is'
'what to tell people in the first few weeks, want people to know you have had the baby but not ask if it is a boy or a girl'
'parents have poor access to information, don't get given written information in a language they can understand'
'need clear accurate information without medical jargon'

While there are some written resources available from overseas the only parents who had received these were those with previously-diagnosed CAH children, once they were put in touch with the support group. None of the participants had received any written information at the time of birth or at the time their condition was diagnosed. Parents expressed a desire for simple explanations of the conditions and/or the tests their infant would have in a language which they could also use to explain the situation to their family or friends. Participants also expressed a need for explanations that they could give to others if their child's gender was uncertain at birth and a period of time would be needed before a gender could be assigned. This suggested a need for the development of written information specific to the New Zealand experience which could be given to parents initially at the time of the birth of a child where an initial gender assignment was in question. Once a specific condition was diagnosed or parent had had time to digest and question the initial information further written information could be given including the parent's handbook from ISNA or condition specific information from groups such as CAHNZ.

In Depth Discussions

Once a gender or diagnosis had been established participants identified the need for an in-depth discussion which covered some of the more ongoing issues they would have to face. This included a desire to know what would be hard for them as well as the positive aspects of their child's condition.

'in-depth talk to answer the many questions'
'parents and children need to know they are on a learning curve'
'It is the ongoing management of the disease which is challenging'
'assurance there is a good prognosis'

Once they knew the diagnosis parents expressed a desire to sit down and talk in depth about going home and issues that would arise in the future. These answers again emphasised the lack of adequate information being received both prior to and after leaving hospital. This research has identified that a lack of a team approach in some New Zealand hospitals has led to poorly informed parents, with participants who lived outside of Auckland being more likely to suggest that their concerns and questions were not adequately addressed. In ISNA's (2006) handbook for clinicians the emphasis is on the need for a team approach

> Although many children born with DSDs are healthy and require little medical management, having families connected with multidisciplinary teams as early as possible may ensure that familiar, expert care givers will be available when psychological, surgical, or medical needs do arise. Additionally, the challenges brought on by the environment of a developing child and family will require ongoing assessment and possible changes to established treatment goals. (pg.9)

Honesty

Honesty was a topic that came up repeatedly in participant's answers. This theme was consistent with the international literature in this area (Howe, 1998; Preves, 1998; Groveman, 1998). Participants wanted honesty from health professionals and stressed the importance of being honest with your child.

'be honest'
'secrets wreck lives'
'impacts whole family, need to tell truth',
'be honest with your kids – it effects your whole body'
'talk openly with your child'
'makes you closer, can talk about the body and how it works, why it happens'
'times when it is tough'
'starting dishonestly sets you up for your whole life'

'what's wrong with the truth?'

The potential destructiveness of anger, shame and secrecy to relationships between parents and their child and/or individuals with intersex/DSD conditions and the medical profession was considered in the literature associated with intersex/DSD conditions discussed in Chapter Two (Groveman, 1998; Diamond, 2004). For some of the New Zealand participants there was also anger at the way they felt they had been lied to or misinformed or the way they had been advised to keep secrets from their child or family.

'was told don't tell people she has CAH, just say an adrenal insufficiency but I think 'don't be ashamed, don't be embarrassed or your child will be'
'we were told don't tell people that you don't know well, don't tell relatives, keep it a secret which just about sent me round the twist'
'don't tell it's for her best interests'

How honest to be with people outside participants direct family was more controversial with some participants expressing the view that parents should be told it is justified to be vague with people outside their immediate family.

'initially told people about CAH because she was only slightly virilised'
'I found people were uncomfortable now if someone comes up in the street and asks about medic alert I feel it is okay to fudge or even lie to protect my child's privacy'

In interacting with non-family members both parents prioritised privacy over honesty. These extracts suggest that they were initially open to acquaintances about their child's condition, but through trial and error had learnt to be vague in order to protect their children from adverse reactions. The statements reproduced in this section highlight a need for information for parents which looks at ways to talk to their children, their family and to people outside their immediate family, about their child's condition.

Surgery

When asked what advice or information parents should receive about surgery participants stressed that parents should know both sides of the argument. This was consistent both for those who had decided to proceed with surgery and those who felt they had not received adequate information or had been bullied into surgery.

'parents should not be bullied'
'need both sides of the story for informed consent'
'important to know it is your choice'
'isn't just because the doctor says do it you should do it'
'never withhold prednisone but surgery is a whole different question'

Participants identified the need for impartial information and respect for the choices that individuals may make regarding treatment as key issues. Those who advocated surgery expressed a desire for others to have practical information about Melbourne and the Royal Children's Hospital, including photographs and the information about government funding for surgery in Melbourne.

'surgical before and after pictures are good not particularly what you would want to carry round in your pocket and show off at the pub but if you have a daughter who is going through surgery it is interesting and nice to know they will come out looking so normal'.
'go to someone who is doing it 2-3 times a week'
'Melbourne is a state funded option'.

These interview extracts suggested that, when making a decision about going to Melbourne for treatment, parents felt that specific information about outcomes, the expertise of the medical team and cost was lacking. This desire for practical information also arose when discussing future pregnancies and genetic testing.

'need to know about future pregnancies, steroids, how long results take to come back after CVS and female chromosomes'
'when the parents are in more maintenance mode talk about future pregnancy taking dex, sheet with info, framed in neutral manner, for and against, stress it is parents choice'
'gene testing- isolate gene in family'
'genetic testing can be difficult and take a long time, different assays, different countries, no clear protocols need a protocol developed for parents'

Detailed practical information, genetic counselling, and the need for the development of clear protocols arose as concerns for parents once they were considering a future pregnancy.

Practical Information

For families of a child with CAH participants also talked about having practical information on giving salt and medications. All parents of CAH children I spoke to talked of how much easier the early time at home would have been if someone had talked to them about the medications, how to give them, how important timing was, what happened if their child vomited.

'practical advice on giving meds how to get salt down, the biggest hurdle initially'
'do you stick to the clock, wake them to give medication'

They also wanted to know about adrenal crisis and have a clear plan about what to do if their child was sick. The Co-ordinator of CAHNZ felt that *'most families have an incomplete understanding of how to deal with vomiting and diarrhoea'.*

'worry about an adrenal crisis would have been good to know you will have an action plan for an adrenal crisis'
'guidelines on big issues such as gastro, vomiting, diarrhoea, adrenal crisis, broken bones, fever concussion, major injury'

Parents of children with CAH said that they went home feeling ill-prepared and appeared to lack basic information about how to give medications, to manage the condition and cope with potential health crises. Parents understanding about the lack of basic information given to new parents of children with CAH is evident in the tips that participants were willing to share about administering medications and salt.

'wasn't told it could be given in milk'
"how to get salt in- squirt it in'
'squirt straight to the back of the mouth huge difference'
'put meds in while baby is sucking on empty breast'
'give before a feed'
'now know you can slip them the meds when they are asleep'
'don't miss tablets'

These parents of children with CAH were also able to offer some specific advice for other parents in terms of going to school and managing in the community. At school suggestions included

'have their own kit'
'photo of her in staff room with information on, same for class teacher/ reliever'
'make sure office staff knows'
'have an information sheet explaining your child's condition print out and attach to car seat straps in case of an accident'
'Hydrocortisone injections are not carried on ambulances'
'have an action plan for an adrenal crisis'

Participants expressed a willingness and a desire to share practical information on drugs and other issues specific to CAH with future parents of CAH children. This was information they wished someone had shared with them before they took their child home rather then having to be on a steep learning curve of self discovery. It was only once they were put in touch with the support group they began to receive information and often this was months and sometimes years after their child's initial diagnosis.

Sources of Support

The important role of support groups and having others to talk to was stressed by all of the parents and adult participants. It is interesting to note that in their 1997 guidelines Diamond and Sigmundson recommended that families be put in touch with support groups as soon as possible yet only one parent had been given the CAHNZ information before leaving hospital, all the others had found it via the internet or from the endocrinology department or the metabolic nurse from starship, sometimes quite some time after their baby was born. Participants all stressed new parents with a baby with CAH should immediately be given the CAHNZ information. For those with other conditions, no information was given about support groups or contact information for other adults with, or parents of children with, the condition *'we were promised we could talk to other families...never happened'.*

In talking about the importance and value of the CAHNZ support group participants stated;

'you should be put in touch with support group, paediatricians and NICUs should put people in touch'
'to know about the support group from the start'
'would be awful without the support group'
'nice to have someone who had it to talk to'
'give contact numbers for the support group plus medical people'
'give support group number'
'list of parents willing to chat with others'
'support group important to point you in the right direction'
'nice to know you are not the only one'

Parents suggested that information about the CAH support group needed to be provided in the beginning. This suggests that perhaps the value attached to the CAH support group by parents underscores the importance of support groups for parents of children with other intersex/DSD conditions. Lastly participants expressed a desire to have heard stories about life getting better and people living ordinary lives with these conditions. They also felt it was important that new parents heard these stories.

'you need to see pictures of healthy CAH kids, and CAH kids taking their medications'
'life is normal, life isn't different'
'hearing people talk about their experiences and need NZ stories'
'it's not the end of the world, it's a condition you can treat'
they grow up and can do the same things other kids do- sport, travel it is manageable'
'aware of the good stuff...you can have kids, IVF is an option'
'hear stories about people who have a normal life, have children, look feminine'

'I do not let her medical condition stop her doing anything- don't wrap her in cotton wool – as long as she has her emergency kit'
'helped to think that horrible things that have to happen in the first few weeks are such a small proportion of your day'
'1/2 hour is taken up with blood tests and medicine, 23.5hrs you can mother just the same'
'at some stage life will become normal, medication won't be an issue, they will take in their sleep'

Chapter 4 discussed the prospect of parents going through a process of grieving about not having a perfect child or their child not having the kind of future that they had envisioned (Dreger, 2002; Woods, 2007). Having access to stories about people with intersex/DSD conditions living ordinary lives may help the parents re-imagine a positive future for their child. Participants suggested that having this information available for parents and families at the time of diagnosis would offer both support and reassurance at a time when parents feel their world has been turned upside down.

The resources available from ISNA in the form of their Clinical Guidelines and Handbook for Parents address many of the concerns and suggestions raised by participants in this study. The table of contents from the Parents Handbook (reproduced below) provides an overview of many of the issues which arise for parents. This handbook and the CAHNZ newsletter both present stories of ordinary families and people living with these conditions.

Welcome to Parents
You Are Not Alone!, What Are DSDs?, What Causes DSDs?, Acceptance Takes Time, Speaking About Shame, to Try to Get Beyond It, Helping Yourself to Help Your Child
Your Child's Development, and How to Talk with Your Child
Ages 12-36 Months Ages 3-5 Years (Pre-Schoolers) Ages 6-11 Years. Puberty Adolescence (11-18) Your Life Together
How to Talk with Others
What to Tell Your Friends and Family in General If Your Newborn's Gender Assignment is Delayed, What to Tell People Who May Think DSDs are Sinful, Tips on Interacting with Teachers and Day care Providers, Talking with Your Child's Medical Care Providers
Answers to Common Questions
Helpful Handouts About Disorders of Sex Development (DSDs), How Genital Development Happens, Things to Do and Things to Avoid, Preparing for a Medical Appointment, Record-Keeping and Journaling, Thoughts from Fellow Parents and from Adults with DSDs
Other Resources (Where to Learn More)
Websites, Support Groups, Diagnosis-Specific Information, Finding a Child Psychologist Books Devoted to DSD Issues, Videos and Television Broadcasts

,Journal and Magazine Articles, General Parenting , Sex Education for Your Child (ISNA 2006)

These handbooks were released in March 2006, a date following the completion of the participant interviews. The CAHNZ newsletter provides information in a variety of formats. It reviews research papers on CAH, reports on trends in the management of CAH, shares personal stories of those living with CAH, and offers tips on practical issues such as giving medications and travel.

Given the lack of information participants interviewed received it could be surmised that it is unlikely that parents with a child born since this time will have received these booklets from the health professionals they have come in contact with, but they may have accessed the booklet themselves while searching the internet. It would seem appropriate that health professionals provided parents with this booklet at the time of diagnosis. The booklet, while a potential resource for parents, also flags the need for further information that is relevant to the New Zealand context.

The desires and suggestions participants expressed to the researcher in terms of parent's information needs allowed the formation of a number of recommendations. This chapter goes on to look at practical initiatives which can be taken to meet the need for truthful straightforward information delivered in a gentle and caring way. It takes into account practical advice for the initial time after diagnosis, the experience of going home and ongoing management and coping skills in the community.

Recommendations

Recommendations are presented as an end result of this research. They encompass the information offered by participants and the literature reviewed throughout the thesis. They focus firstly on the need for health professionals to become more educated about intersex/DSD conditions. They offer practical examples of how written information could be provided for parents and look at the formation of a team approach within New Zealand to the clinical care of children and their families.

Education for health professionals

The narratives of participants and their views on helpful information highlighted a need for health professionals to have more information about intersex/DSD conditions. This pertained not only to health professionals in a hospital setting but also those in the community such as midwives, GPs and Plunket nurses. The information needs to be twofold; an understanding of intersex/DSD conditions and the current controversies over treatment and an understanding of how to talk to parents empathetically and honestly about these conditions.

Opportunities to educate health professionals could include the following;

- Lectures for midwifery, nursing and medical students
- Presentations at nursing and medical conferences
- Teaching sessions in neonatal and obstetric units in Auckland and around New Zealand

ISNA has produced a booklet to complement its parent handbook entitled Clinical Guidelines for the Management of Disorders of Sex Development in Childhood. This handbook is a collaboration between clinicians, parents of children with DSDs, adults with DSDs and advocacy groups. At the time this research was conducted this was in the production stage and I received a preliminary copy when visiting ISNA in Seattle. The booklet includes scripts for talking with parents. The handbook is now available in PDF and online format at http://www.dsdguidelines.org/. This booklet could be made available to health professionals in paper or electronic form. It is noted that DHBs throughout New Zealand have variable policies regarding the use of internet resources as an information source and teaching resource, and it would be necessary to comply with these within individual DHBs.

The researcher intends to present the findings of this research project in a number of formats including a grand round presentation at Auckland City Hospital, international and national paediatric and nursing conferences and in a journal publication.

Requests were also made from Little Treasures and Kiwi Parent at the time the recruitment advertisements were placed, for a follow up article. This would also meet the expressed need of participants to have intersex/DSD conditions more widely recognised and known about.

Written information for parents

Written information was requested by all participants. At the time the research was proposed the researcher had envisioned a single information booklet as an end product of the research. ISNA, however have since produced an invaluable resource for parents in the form of their Handbook for Parents. This covers all areas of information for parents including information on what DSDs are, how to talk to your children through developmental stages, how to talk to others, stories from other parents and adults with DSDs and information on other resources and support groups. All of these were issues that arose in this research. While an excellent resource, and one all parents should receive, the entire booklet could be a little overwhelming for parents at the birth of their baby. Many participants expressed the need for information to come in stages. Most intersex/DSD infants in New Zealand present to a neonatal service because there is a question about their genitalia and gender assignment at birth. Some premature infants may present with CAH while in the NICU while other infants with non virilising CAH will present to a Paediatric hospital when the results of their Guthrie test become available or because they have become increasingly ill at home in the initial week. For infants presenting to the neonatal service with a variation in genital anatomy I propose a small leaflet, which will be passed around clinicians, specialist nurses and advocacy groups for consultation following the completion of the thesis, with the following information;

Introduction

Congratulations on becoming new parents and welcome to your new baby. We know the first question many new parents ask is "Is my baby a girl or a boy?". Someone from the paediatric service has come to see you and look at your baby because this is a question we haven't been able to answer just by looking at your baby's genitals. Parents are often very shocked when told this kind of news, but we'd like to reassure you that we see a number of babies each year whose genitals are different from what you might expect to see in a female or male baby. Although your baby has a condition you have probably never heard of, these

conditions are not that uncommon. We will do a number of tests over the next few days to find out more about your baby's condition so we can help answer the question "Is my baby a girl or a boy?".

What condition might my baby have?

When developing in the uterus, all babies start out with the same sexual organs. Boys tend to develop a penis, scrotum and testes, whereas girls tend to develop a uterus, ovaries, vagina and clitoris. There can however, be a number of things that happen in the early weeks of development which cause variation in a baby's reproductive organs, either inside or outside. These are a biological variation and very seldom indicate anything life threatening. There are medical terms which cover a large group of these conditions and two you might hear being talked about are "intersex" and "DSDs" (Disorders of Sexual Development). Some people don't like these terms, "intersex" because it has become a political term, and "DSD" because of the word 'disorder', but at present these are the terms used. Individual conditions have their own names and the tests we do over the next few days will help to give a specific name to your baby's condition.

What tests will my baby have?

The condition that most commonly causes genitals to look different is Congenital Adrenal Hyperplasia or CAH. We will do a blood test to check for this, as this condition does have a medical complication, as babies with CAH lose too much salt from their bodies, which can be life-threatening. We will also do blood tests to look at your baby's chromosomes and the level of certain hormones in the blood. We may need to give some medication to your baby, then do blood tests to look at the response to these medications. Your baby will have an ultrasound to check the internal organs. We will also need to examine your baby. You can be present for all these tests and hold your baby throughout them when possible, or immediately afterwards. You can ask as many questions as you like and we will explain what we are doing at the time. Some tests give a result immediately while others may take hours or a few days. There are a number of health professionals we may need to speak to or ask to come and see you and your baby. We will try our best to have as few people as possible examine your baby. Some of these other people may be endocrinologists (looking at your baby's hormones), genetics (to do with your baby's chromosomes) and urology (looking at your baby's ability to produce urine). We will inform you when we get results, and as soon as we have them all we will sit down with you and discuss the findings. We realise waiting is hard and as soon as we have these test results we will be able to talk to you about the gender to raise your child in.

The test results

The results of the tests will help us find out which condition your baby has and then there will be some decisions to make. The biggest of these will be which gender to assign to your baby. Sometimes this is easy to decide and sometimes a little harder. We will discuss everything with you and the final decision will be yours. We suggest you choose the gender your baby is most likely to identify with as they grow up but it's important to keep in mind that all people – whether they have a genital variation or not make this decision as they mature, and may choose not to identify with the gender they've been given as a baby. This does not have to be an instant decision. We will give you as much help, information and support as you need. You can talk to your midwife or nurse, the paediatric staff from the neonatal unit or the paediatric service, a social worker or a patient advocate. We will work with you to get any support or help you need, including putting you in contact with other parents whose children have this condition. If your baby has CAH we will be able to discuss the medical issues which come with this condition. In most cases your baby will be able to remain in the postnatal ward with you while we wait for these results.

What should I tell other people?

We understand that waiting for test results can be very hard and that you will have family, whanau members and friends waiting to hear news about your baby. You might feel confused and scared, or that you don't want to tell anyone what is going on. Our experience with parents over the years however, says it is good for parents to have the support of family and friends at this time. A helpful thing to say to them might be;

Our baby was born with a kind of variation that happens more often than you hear about. Our doctors are doing some tests to figure out whether our baby is probably going to feel more like a boy or a girl. We expect to have more information soon, and then we'll send out a birth announcement with the gender and the name we've chosen. We appreciate your love and support and we're looking forward to introducing you to our little one in person soon. (based on the (ISNA handbook, 2006)

Other things you can do are share a picture of your baby with people or have people come and meet your baby.

Where else can I find information?

There is a lot of information about these conditions on the internet but other parents have expressed that they have often found it very overwhelming or sometimes frightening and confusing. As soon as we have a name for your baby's condition, we can give you a lot of specific information about the condition both in our talks with you and as written material. We will also give you information about support groups and the addresses of good

websites. We also have some good resources which answer a lot of the questions which parents have such as how to talk to their child, what will it be like to grow up, how do I talk to other people about my baby's condition and how can I meet other parents of children with the same condition?

The pamphlet could be supplemented with the list found in Table 2 questions for your medical provider (see the Research Appendix page 15).

Specific Information

After initial tests have been completed more sources of information could be made available to parents. Once a diagnosis has been made I would recommend having specific information packs for parents. The largest number of infants with an intersex/DSD condition will have CAH. The information pack could contain;

- An introduction written by CAHNZ and Starship endocrinology
- Contact details for CAHNZ and a copy of their latest newsletter
- A booklet on CAH (these are available from http://www.rch.org.au/cah_book/index.cfm?doc_id=1375&)
- Practical information about administering medications and salt
- Practical information about managing normal childhood health crises such as vomiting and diarrhoea,
- Practical information about adrenal crisis
- Practical information about what to do in the event of an accident (such as a broken bone).
- The addresses of websites recommended by health professionals and support groups
- Contact information for the metabolic nurse specialist and genetics and specific information on blood tests

- Contact information for other support groups such as ITANZ and ISNA and the names of other parents who are willing to be contacted.

- Information about genital surgery and the debate over surgery

- Detailed information about future pregnancies and taking dexamethasone

In order to put this information pack together I would return to CAHNZ, Starship endocrinology and the metabolic nurse specialist and the geneticist I spoke to for this research, for their input and then work to distribute it to neonatal units and paediatric medical wards throughout New Zealand. This could be combined with some in-service teaching for staff.

There is less information available for other conditions but I have obtained booklets on Klinefelter Syndrome from the U.S Department of Health and Human Services and PAIS and CAIS written by Gary Warne of Royal Children's Hospital in Melbourne and available in downloadable format from the internet.

The information packs should also contain a copy of the ISNA Handbook for Parents. There is so much good information in this booklet that addresses that information needs expressed by participants in this study.

A Team Approach

Lastly the research highlighted the need for a team approach for parents and children. This needs to happen in Auckland and especially throughout New Zealand so children are receiving consistent care and families are receiving consistent information. A team approach would allow parents and children access to support as children grew especially as they approach the teenage years. This approach would allow the development of a list of health professionals that individuals could see who had an understanding of intersex/DSD conditions. A number of participants talked about the importance of finding a good gynaecologist whom their daughter could establish a

rapport with before they became teenagers. ISNA recommends a team approach because

> The issues surrounding DSDs are multidimensional requiring cooperation from a number of disciplines in order to provide effective diagnosis, treatment, and support. A multidisciplinary DSD team typically consists of members from each of the following disciplines (in alphabetical order):
>
> Child Psychology/Psychiatry
> Genetics and Genetic Counseling
> Gynecology
> Nursing
> Pediatric Endocrinology
> Pediatric Urology
> Social Work (ISNA, 2006 p.10)

To organise a multi disciplinary team should be a goal for those providing services to parents of, and individuals with intersex/DSD conditions in New Zealand. There are models overseas on which this team approach could be based.

Conclusions To The Thesis

Each year a number of infants are born with conditions which fall within the umbrella terms intersex and DSD. With advances in pre natal testing a diagnosis is sometimes available prior to delivery, and for some infants it is apparent at birth due to variances in genital anatomy which make it difficult to assign a gender based purely on outward genital appearance. Others are diagnosed as the result of a newborn screening blood test, and yet others receive their diagnosis during childhood or adolescence. A review of the medical and social literature from the last forty years identified a medical paradigm of surgical intervention to 'normalise' genitals (Myers-Seifer and Charest,1992) and a process of gender assignment based predominantly on phallus size (McGillivray,1992). This paradigm was based in the 1960s work of John Money. In the 1990s follow up occurred of the case study on which Dr Money's theories, and the subsequent interventionist model of treatment, were based on, along with reviews of the few clinical outcomes studies which had been published on the resulting genital surgeries. Combined with the growing voices of intersex individuals treated under this paradigm and the emergence of intersex advocacy groups it became obvious that the existing paradigm of medical and surgical intervention had adversely affected the lives of thousands of children and adults born intersex. While some were satisfied with their treatment for others surgery has left them with non-functioning, mutilated genitals.

This, and the deceit and silence from physicians and their families had resulted in psychological scarring. The questionable foundation for the traditional paradigm, and the lack of follow-up studies, combined with a complete disregard for the issues of informed consent and autonomy then created a basis for a major ethical debate.

The ethical debate centred on issues of the legitimacy of early genital surgery for 'cosmetic' reasons and parental rights in terms of giving informed consent for surgery, the detrimental effects of giving a female gender assignment to XY individuals based on phallus size, and the atmosphere of secrecy and deceit which had occurred between physicians and parents, and in turn between parents and children surrounding the true diagnosis for individuals and the reasons for surgeries. This debate, questioning, and criticism of the existing paradigm has led to the publication of a number of larger follow-up studies since 2000. There have been some changes to practice in terms of promoting a more open and honest approach to discussing intersex/DSD conditions with parents and individuals and a move away from assigning under virilised XY infants to a female gender of rearing (Parisi et al, 2007; Reiner, 2005), however surgery still remains a controversial issue. A review of contemporary follow up studies (Gollu et al, 2007; Alizai et al, 1999; Farhat, 2005; Warne et al, 2005; Crouch & Creighton, 2007; Houk et al, 2006) identifies recommendations ranging from a complete moratorium on all infant genital surgery, through a selective approach to surgery based on degree of virilisation and parental wishes to a continuation of early surgery until long term follow up studies on infants who do not receive surgery can be compared to current surgical results. Accompanying this debate has also been the issue of nomenclature in regards to an umbrella term which encompasses a number of conditions where there is atypical sex anatomy or discordance between any of the sexual characteristics including chromosomes, internal and external genitalia and gonadal histology. While the term DSD now appears consistently in the medical literature debate continues around the appropriateness of using the term 'disorder' to label individuals (Cameron, 2006; Hinkle, 2006; Diamond & Beh, 2006).

What appears to be lacking in the follow up studies is research looking at the experience of parents when it is apparent at birth or shortly after that a child has an intersex/DSD condition or a diagnosis is made during childhood or adolescence (Reiner, 2004). Issues other then genital surgery such as genetics, gender identity, the medical component of certain conditions, and the experience of growing up with an intersex/DSD condition are also underrepresented in the contemporary literature. This

research sought to address the issue of parental, family and whanau information needs when a child is born with an intersex/DSD condition. A qualitative exploratory approach was undertaken and the narratives of twenty one participants were reviewed using thematic analysis.

The experience of discovering a child or an individual themselves had an intersex/DSD condition was characterised by feelings of shock and grief. These feelings were negatively intensified by the language health professionals used and the treatment of infants and individuals as objects of 'curiosity' or teaching opportunities. Concerns expressed about having a child that was different either because of genital anatomy or another aspect of an intersex condition such as the medical component of CAH were further escalated by a lack of information. For those individuals with CAH, or parents of a CAH child a lack of practical information on administering drugs and adrenal crisis led to increased feelings of isolation. Lack of information, both oral and written was a recurring theme at the time a child or adult was first seen, around diagnosis and once they returned home to the community. Parents expressed feeling inadequately equipped to talk with their child about issues that would arise as their child grew older. The role of the internet in providing information to participants and consequently influencing decisions they made about revealing information concerning their child's or their own diagnosis was a new finding to this research.

The issues of nomenclature, surgery, informed consent, gender identity and future pregnancies and genetic testing were reviewed in relation to the medical and social literature and the information from participants' narratives. While some participants could relate to the term intersex and three self identified as intersex, others, in particular parents of children with CAH did not consider the term intersex applied to their children as they interpreted it to mean there was confusion over their child's gender identity. As the term DSD was introduced following the completion of the interviews only the co-ordinator of CAHNZ and ISNA were able to offer opinions on the use of this term.

Participant's experience of surgery and the issues surrounding early genital surgery also reflected the differing opinions offered by the literature. A number of parents of CAH children reported positive experiences of their daughters' surgery in Melbourne while a parent of a child with PAIS had felt bullied into surgery by a number of doctors. She felt her child had been irreversibly harmed by the genital surgery and an inappropriate female gender assignment. Of the parents spoken to,

varying degrees of information was reported as having been given by health professionals to allow an informed decision, however all parents expressed the opinion that it was their decision to make.

The genetic basis of a number of intersex/DSD conditions had raised issues for participants in terms of future pregnancies, testing during pregnancy and carrier testing of siblings. Three participants had made an active decision not to pursue another pregnancy due to the risk of another child with the same intersex/DSD condition. For those with a child with CAH already the experience of obtaining and taking dexamethasone to decrease virilisation in another affected fetus, was a difficult one.

The narratives of participants provided an insight into the experience of growing up with an intersex/DSD condition, and of issues which arise for parents. The birth of a baby with variances of genital anatomy forced parents into considering issues about gender identity most new parents do not have to think about. The impact of the first year on relationships with partners and on the family as a whole was stressed by all participants. Issues unique to the preschool years included others caring for a child, the scheduling of any genital surgery and how to answer questions from those outside the immediate family. None of the parents spoken to felt they had adequate information to talk to their children about their conditions, especially as their children approached adolescence.

These 'hard issues' and the lack of information which participants' expressed receiving led on to the formation of recommendations for the education of health professionals and the formulation of written information and information packs for parents. Incorporated into these resources were the answers and suggestions of participants when asked about their information needs and advice to others. The desire for a team approach to the care of children with these conditions which was consistent throughout New Zealand was also a key finding of this research. While relating participant's narratives to the literature from America, Europe and Australia this research also offers a resource unique to the New Zealand experience of having an intersex/DSD condition.

Suggestions for Future Research

The issues identified in this thesis suggest some areas for future research. Of the 21 participants interviewed 2 were Maori, while the remaining participants identified as Europeans. To form a more comprehensive picture of the information needs of New Zealand families there is a need to canvas the views of a wider range of ethnic groups. It would have been beneficial to speak with further Maori, and also Pacific, people in regard to issues raised in the thesis.

As identified in the thesis, follow up research on those with intersex/DSD conditions has predominantly focused on surgical outcomes. The experiences of people, and their families growing up with, and living with intersex/DSD conditions is an area where there is generally a need for further research. Specific research is also needed on the challenges of, and information needs for, adolescents with intersex/DSD conditions, this is particularly relevant as those born in the 1990's when a challenge to the paradigm of care began to come about, are now approaching their teenage years.

In terms of the recommendations resulting from the thesis there needs to be a review of current DHB protocols regarding the birth of a child with an intersex/DSD condition. While the researcher is able to do this within her own DHB setting, more consultation would be required before making changes to clinical treatment protocols throughout other DHBs. The DHB protocols and the views of health professionals and patients around the use of the internet as an information source and teaching resource, also need to be investigated in regard to the resources which can be utilised and provided for health professionals and families.

The recommendation that a team approach which provides a consistent approach to care throughout New Zealand also requires research into strategies that would be effective for developing a team approach to care within and between institutions.

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RESEARCH APPENDIX

Table of Contents

A BOY OR A GIRL?

Parental Information Needs When A Child is Born Intersex

PARTICIPANT CONSENT FORM

This consent from will be held for a period of five (5) years

Circle one

English	I wish to have an interpreter.	Yes	No
Maori	E hiahia ana ahau ki tetahi kaiwhakamaori/kaiwhaka pakeha korero.	Ae	Kao
Samoan	Ou te mana'o ia i ai se fa'amatala upu.	Ioe	Leai
Tongan	Oku ou fiema'u ha fakatonulea.	Io	Ikai
Cook Island	Ka inangaro au i tetai tangata uri reo.	Ae	Kare
Niuean	Fia manako au ke fakaaoga e taha tagata fakahokohoko kupu.	E	Naka i

I have read the Information Sheet and have had the details of the study explained to me. My questions have been answered to my satisfaction, and I understand that I may ask further questions at any time. I have had the opportunity to use whanau support or a friend to help me ask questions and understand the study. I understand that taking part in this study is voluntary. I understand I may decline to answer any particular question. I understand that my name will not be used unless I give permission to the researcher.

I agree/do not agree to the interview being audio taped.

I wish/do not wish to have my tapes returned to me.

I agree to participate in this study under the conditions set out in the Information Sheet.

Project explained to me by.....

Signature: ..... **Date:** .....

Full Name - printed .....

Please send me a summary of the findings ☐
(There will be a delay between data collection and publication of results)

Address to send findings to

Researcher

G.L McCarthy
Masters Student, Massey University
[redacted]

A BOY OR A GIRL?
Parents Information Needs When Their Child is Born Intersex

INFORMATION SHEET

Researcher(s) Introduction

My name is Leigh McCarthy. I am a Masters student at Massey University. I also work as an Advanced Practitioner in the NICU (Neonatal Intensive Care Unit) at National Women's Hospital. Each year we see a number of babies born with conditions which fall under the umbrella term Intersex. These include conditions such as Congenital Adrenal Hyperplasia (CAH), Partial and Complete Androgen Insensitivity (PAIS & CAIS), Gonadal Dysgenesis and Klinefelters Syndrome. At present we have no written resource available to give parents and families which might answer the many questions they have when their child is born with one of these conditions. As part of my Masters work at Massey University I would like to explore the information needs parents/ families/ whanau have when their child is born Intersex. I would like to extend an invitation for you to speak with me about your experiences. My goal is to be able to use this information in producing a written resource for parents/ families and health professionals which addresses the many questions and issues which arise when a child is born Intersex.

Participant Recruitment

- I have sought participants for this research by open letter of invitation.

- I acknowledge that due to the personal nature of the disclosures participants may make during the interviews there may be occasions in which the participants experience some emotional discomfort related to their experiences of having a child born intersex or being born intersex themselves. This information sheet provides the names and contact details of people who may act as support persons for you in regard to any issues which arise during the course of the interviews.

Participant involvement

- Participation in this research will involve an interview at a place of your choosing. I anticipate the interviews taking between 1-1.5 hrs. I will travel to interview you. If English is not your first language I will be happy to work with you to provide a mutually agreeable interpreter.

Project Procedures

- Data collected in the interviews via an audiotape, and transcribed by myself. Data will be used to contribute to a resource for parents/families and health professionals. There will be no identifying information in the data and material which could personally identify you will not appear in any report on this study.

- The principle researcher will manage the data. Interview tapes and transcripts will be stored in a locked filing cabinet at Massey University. Data base records and computer work will be password protected. The database will be kept for ten years and safe guarded by the researcher. Access to the data will be limited to the researcher and the research supervisors.

- You can indicate on the consent form if you wish the results of the research to be forwarded to you.
- There will be a delay between the collection of data and publication of the report.

Participant's Rights

You are under no obligation to accept this invitation. If you decide to participate, you have the right to:

- decline to answer any particular question;
- withdraw from the study at any time up until the completion of data analysis, October 2005
- ask any questions about the study at any time during participation;
- provide information on the understanding that your name will not be used unless you give permission to the researcher;
- be given access to a summary of the project findings when it is concluded.
- you also have the right to ask for the audio tape to be turned off at any time during the interview.

Support Processes

For Maori health support, or to discuss any concerns or issues regarding this study, please contact Mata Forbes RGON, Maori Health Services Co-coordinator / Advisor, 5th Level, GM Suite, Auckland City Hospital. Tel 307 4949 extn. 23939 or [redacted]

If you have any queries or concerns regarding your rights as a participant in this study, you may wish to contact a Health & Disability Advocate, telephone no. 0800 555 050 Northland to Franklin.

Project Contacts

If you have any questions about the research please contact the researcher Leigh McCarthy on
021 777 896 or email leighmccarthy2004@yahoo.co.nz.

The research supervisor Associate Professor Denise Dignam can be contacted at
School of Health Sciences Albany Campus, Private Bag 102 904, North Shore MSC Auckland
Ph 64 9 4140800 ext 9167 Cell 64 (0)21 414299 D.M.Dignam@massey.ac.nz

Committee Approval Statement

This project has been reviewed and approved by the Massey University Human Ethics Committee, ALB Application 04/067If you have any concerns about the conduct of this research, please contact Associate Professor Kerry Chamberlain, Chair, Massey University Campus Human Ethics Committee: Albany, telephone 09 414 0800 x9078, email humanethicsalb@massey.ac.nz.

This study has received ethical approval from the Auckland ethics Committee.

ADVERTISEMENT/LETTER FOR EXPRESS NEWSPAPER

Intersex?

I would like to talk with adults born Intersex. I am an Advanced Practice Nurse in a NICU (Neonatal Intensive Care Unit). At present we have no written resource available to give parents and families which might answer the many questions they have when their child is born with one of these conditions. I feel it is vital that the voices of intersex people are heard and reflected in such a resource. I would like to extend an invitation for you to speak with me about your experiences. If you are willing to talk with me please contact me via Email: [redacted]z

ADVERTISEMENT/LETTER FOR PARENTING MAGAZINES

Children born with Intersex Conditions

My name is Leigh McCarthy. I am a Masters student at Massey University and I also work in an Advanced Practice role in the NICU (Neonatal Intensive Care Unit) at National Women's Hospital. Each year we see a number of babies born with conditions which fall under the umbrella term Intersex. These include conditions such as Congenital Adrenal Hyperplasia (CAH), Gonadal Dysgenesis and Klinefelters Syndrome. At present we have no written resource available to give parents and families which might answer the many questions they have when their child is born with one of these conditions. I would like to explore the information needs parents/ families/ whanau have when their child is born Intersex. I would like to extend an invitation for you to speak with me about your experiences. My goal is to be able to use this information in producing a written resource for parents/ families and health professionals which addresses the many questions and issues which arise when a child is born Intersex. If you would be willing to be interviewed about your experiences as a parent or as a family member of a child born with one of these conditions please contact me.

Email: [redacted]

Phone: [redacted] I will be happy to return your call at my expense.

Interview Guidelines for Adult Intersex People.

Tell me what it was like growing up?

Did you understand the tests/ and or surgeries that were being done and why?

Were you involved in decisions about your medical treatment?

What things were said to you growing up by medical professionals that were helpful and what things were unhelpful?

What did you feel you needed more information about as a child/ teen or adult?

Where did you look, who did you ask for information?

What information do you think it would be helpful for parents of a child born Intersex to have?

If there was a written resource for parents what would you like to see included?

Interview Guidelines for Health Professionals and Intersex Advocacy Groups

What information do you think it would be helpful for parents of an Intersex child to have?

If there was a written resource for parents what would you like to see included?

Interview Guidelines for Parents /Families of an Intersex child.

Tell me what it was like when your baby was born?

Did you understand the tests that were being done and why?

What things were said to you in the first few weeks and months that were helpful and what things were unhelpful?

What did you feel you needed more information about, at the time of your baby's birth?

Where did you look, who did you ask for information?

What decisions regarding your child's care and treatment have you been asked to make?

What information was helpful in making these decisions?

What information do you think it would be helpful for other parents to have?

If there was a written resource for parents what would you like to see included?

AUCKLAND
DISTRICT HEALTH BOARD
Te Toka Tumai

Auckland District Health Board
Greenlane Clinical Centre, Green Lane West Auckland 3, New Zealand
Telephone: 09 638 9909
Website: www.adhb.govt.nz

Service: **ADHB** Research Office Office. Level 2, Bldg, 14. GCC Postal' PB 92189 Auckland
Phone: 630-9943
Ext: 4085.4077 and 3122
Fax: 630 -9796 or 4996
Email: [redacted]

25 January 2005

Gabrielle McCarthy
[redacted]

Tena koe Ms
McCarthy

RE: Research project 2004/291 (A+3096) -A Qualitative Descriptive, Exploratory Study Examining the Information Needs of Four Key Stakeholder Groups in Regard to Parental Information Needs When a Child is Born Intersex

Thank you for sending us a copy of the revised participant information sheet. The study has been approved.

Please send a copy of the final report to the Maori Research Review Committee (c/o Jenny Ma, Research Office, Level, Bldg 14, Greenlane Clinical Centre) at the conclusion of the study.

We wish you the very best in your research.

Noho ora mai,

SECRETARIAT
MAORI RESEARCH REVIEW COMMITTEE AUCKLAND
DISTRICT HEALTH BOARD

Massey University
AUCKLAND

OFFICE OF THE
OEPUTY VICE-CHANCELLOR -AU
Bag 102904
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T Deputy Vice-Chancellor -Auckl
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9516
F 6494140814

20 September 2004

Gabrielle McCarthy
C/o Associate-Professor Denise Dignam College of Humanities and Social Sciences Massey University
Albany

Dear Gabrie!le

HUMAN ETHICS APPROVAL APPLICATION -MUAHEC 04/067
"A **Boy or** a **Girl-** Parental Information **Needs When** a **Child is Born Intersex"**

Thank you for your application. It has been fully considered, and approved by the Massey University, Albany Campus, Human Ethics Committee to proceed to the Health and Disability Ethics Committee, Auckland.

Could you please forward to us a copy of the letter of response from HDEC, once that committee has considered your application?

If you make any significant departure from the Application as approved then you should return this project to the Human Ethics Committee, Albany Campus, for further consideration and approval-

Yours **sincerely**

Professor Brian Murphy **Deputy Chairperson, Human Ethics Committee Albany Campus**

cc Associate-Professor Denise Dignam College of Humanities and Social Sciences

Northern X Regional Ethics Committee
Ministry of Health
3rd Floor, Unisys Building
650 Great South Road, Penrose
Private Bag 92 522
Wellesley Street,
Auckland

e-mail: [redacted]
website: http:/www.newhealth.govt.nz/ethicscommittees

17 March 2005.

Ms Gabrielle L. McCarthy
[redacted]

Dear Gabrielle,

AKX/04110121 **A qualitative descriptive, exploratory study examining the information needs of four key stakeholder groups in regard to_parental information needs when a child is born Intersex: PIS/Cons V#2, 15/03/05**

Thank you for your amendments, received today

The above study has been given ethical approval by Northern X Ethics Committee for the Northern Region. A copy of that Committee's members is attached.

It should be noted that Ethics Committee approval does not imply any resource commitment or administrative facilitation by any healthcare provider, within whose facility the research is to be carried out. Where applicable, authority for this must be obtained separately from the appropriate manager within the organisation.

Certification
It is certified as not being conducted principally for the benefit of the manufacturer or distributor and may be considered for coverage under ACC.

Accreditation
This Committee is accredited by the Health Research Council and is constituted and operates in accordance with the Operational Standard for Ethics Committees, March 2002.

Documents Approved:

.Information Sheet/Consent Form V#

Progress Reports
The study is approved until 17 March 2006. Should you wish to extend the time, a progress report is required for this study by that date

Table 1

Summary of the various types of Disorders of Sex Development with the revised nomenclature

Revised Nomenclature	*Types*
Sex Chromosome DSD	45,X Turner syndrome (mosaic, isodicentric Xq, ring chromosome, etc.) 47,XXY Klinefelter syndrome (and variants)
46,XY DSD	Defects in testicular development Complete gonadal dysgenesis(Swyer syndrome) Partial gonadal dysgenesis (WT1, SOX9,SF-1 mutations) Gonadal regression Ovotesticular DSD Disorders in androgen synthesis or action Androgen biosythesis defects (e.g. 17-HSD,5a-RD, StAR, POR, 3b-HSD, 17,20 lyase,Leydig cell hypoplasia or aplasia) Defects in androgen action (CAIS, PAIS) Others Hypospadias, micropenis, cloacal extrophy,congenital anomaly syndromes Persistent Mullerian duct syndrome
46,XX DSD	Defects in ovarian development Ovotesticular DSD Gonadal transformation (e.g. SRY translocation) Gonadal dysgenesis Disorders of androgen excess Fetal: CAH (11 hydroxylase deficiency,21 hydroxylase deficiency, 3b-HSD, POR) Fetoplacental: aromatase deficiency, POR Maternal: luteoma of pregnancy, exogenous androgen Others Congenital anomaly syndromes,vaginal atresia, cloacal extrophy, MURCS
Ovotesticular DSD	46,XX/46,XY (chimeric) 45,X/46,XY (mixed gonadal dysgenesis)
46,XX Testicular DSD	XX sex reversal (SRY translocation)
46,XY Complete gonadal dysgenesis	XY sex reveral (SF1, WT1, SOX9) Swyer syndrome

CAIS, Complete androgen insensitivity; DSD, disorder of sex development; 17-HSD, 17-hydroxysteroid dehydrogenase; 3b-HSD, 3bhydroxysteroid dehydrogenase; MURCS, Mullerian, renal, cervicothoracic somite abnormalities; PAIS, partial androgen insensitivity; POR, P450 oxidoreductase; 5a-RD, 5a-reductase deficiency; SF1, splicing factor 1; SOX9, sex-determining region box 9; StAR, steroidogenic acute regulatory protein; WT1, Wilms tumor gene.

Table 2

Questions for your medical care provider

1. Do you know my child's exact diagnosis? Ask the doctor to write down the condition's name and where you can learn more. Keep in mind, many DSDs can look similar, so sometimes it takes weeks or even months to figure out the correct diagnosis. In this case, ask your child's doctors to write down for you the diagnoses they are considering. You need to know as much as possible because you are your child's primary caregiver.

2. How can I get complete copies of my child's medical chart and lab results? Also be sure to get copies of the medical charts and lab results from the hospital where your child was born. Being persistent about receiving copies at the time of care will save you hours of frustration later. Having copies of the records will allow you to consult others easily and make it possible for you to give your child a written record of your child's medical care when your child matures. You have a legal right to copies of all your minor child's medical records.

3. If a lot of medical people are coming to look at your child, ask: "Who really needs to examine my child personally?" If your child is in a teaching hospital, your child will likely be used as a teaching tool for medical students, nursing students, residents (doctors in training), etc. We have heard from many adults with DSDs that repeated medical displays of their genitals seriously harmed them (and sometimes their parents). You should therefore limit exams to those truly necessary for your child's care. If your child's doctor is a resident (a doctor in training) you should also permit the supervising attending physician to examine your child. Remain with your child during examinations to comfort and advocate. Resist having the medical team take pictures of your child's genitals; parents of children with DSDs who are also medical professionals tell us those pictures are almost never necessary for a child's medical care.

4. Does this hospital's team have a parent liaison or a working relationship with support groups for families with DSDs? How can I contact them? If your child's doctor isn't sure, ask your medical team's social worker, psychologist, or nurse. Peer support is probably the MOST important thing for parents. Meeting another parent who has lived for years with a child with a DSD will help you realize you are not alone, and that your roller-coaster of emotions and experiences is normal. Peers will also help you work your way through the medical and school systems and will explain jargon and resources.

5. Would you please give me a referral to a psychologist, psychiatrist, and/or social worker who has experience dealing with gender issues and birth anomalies, so I can get someone to help me with my mixed emotions (fear, confusion, guilt, joy, curiosity, etc.)? Note that caring medical doctors—including endocrinologists, urologists, and surgeons—may try to provide counselling to you and your child, but most have neither the time nor training to do it well. Push for professional psychological support for yourself and your child. Getting that does not mean you're crazy or weird; it means you have found yourself in an unusual situation and know enough to get expert help.

6. Is my child having any immediate medical problems? If so, what are they, and what are the treatment options? What is the danger of doing nothing right now? Most children born with DSDs are healthy; they have no immediate medical problems. Most can be taken home safely and joyfully as soon as test results show there are no immediate medical problems. (Examples of immediate medical concerns are failure of urine to drain and salt-losing in congenital adrenal hyperplasia.) Let your care providers know you want to take your child home as soon as possible, so you can get on with the business of getting to know your precious new family member. Sometimes well-meaning doctors feel that they have to offer you a procedure now, even when it can really wait. Ask your doctor or nurse if there are home-based resources available to you that might speed up getting discharged and back home. If your child requires some monitoring, sometimes this can be done at home with the help of a specially trained home-visit nurse.

7. Which gender assignment (boy or girl) do you think my child should be given? Which gender do you think my child is most likely to feel as my child grows up? What are your reasons? Doctors can help you figure out if your child is likely to feel like a girl or boy in the long run. Evidence suggests that children exposed to high levels of androgens before birth are more likely to grow up to feel masculine.

But no one can predict with certainty what gender a child will ultimately grow up to feel. Your opinion about your child's gender assignment matters a lot because you are the one who will raise the child. Keep in mind your child doesn't need any surgery to be labelled a boy or a girl. We know of many men and women who were raised with bodies labelled "ambiguous" who did well. (Do not let anyone tell you that delaying this kind of surgery is equal to "raising your child in a third gender." It is not.) Children with DSDs do not change their original gender assignments very often. If your child grows to act gender "atypical," that is not because you or your child have done anything wrong; it just means your child is different from the statistical average, and the best thing you can do is to provide love and support for your child's individuality.

8. Here's a useful general question: Can we wait until my child can make the decisions about optional medicines (like hormones) and procedures (like surgeries)? Waiting until a child can decide about optional medicines and procedures is supported by the American Academy of Pediatrics. The AAP also says that you and your child have the right to know everything you can about the procedure being offered. We recommend you download a copy of the AAP policy and go over it with your child's doctors (see www.cirp.org/library/ethics/AAP). If the doctor says "your child will need this surgery when he or she becomes sexually active," ask why the surgeries can't wait until then. As Sherri Groveman (a lawyer with a DSD) points out, your child is also going to need a computer when the time comes for college, but that doesn't mean you need to buy one now! Letting your child decide will let your child know he or she in charge of his or her own body.

9. If the doctors are offering genital surgeries to change the way your child's genitals look, ask: Why do you think my child needs this genital surgery? What evidence do you have that this will help my child in the long run? If your child needs a surgery to save her life, obviously it is a good idea! If your surgeon wants to do a surgery to change how your child looks, pause and consider waiting. What we know about people who grew up with "ambiguous genitalia" tells us on average they do well! You may understandably worry that your child will be emotionally hurt by having something other than average-looking genitals, but the evidence suggests your child won't be, especially if you're open, honest, accepting, and supportive. Surgeries may leave your child with diminished health, diminished sexual sensation, scarring, a poor cosmetic outcome, and an unintended message that your child needed to be "fixed" to be accepted by you. So consider waiting and letting your child decide whether to take the risks. You may discover your child is fine with the way your child is, especially if you let your child know you are.

10. You might want to ask the surgeon: How many of these particular surgeries have you done? What have been all the outcomes, both in terms of physical well-being and psychological well-being? No surgery works every time; find someone who is honest and realistic. If you decide to go with a procedure, choose the one that has been shown to be of proven benefit for people with your child's condition. If there's no evidence about what works, think about waiting until your child can decide whether to risk an experimental procedure. Make sure to ask what improves the quality of life for patients, since that will be a central concern for you and your child.

11. If surgeons are offering removal of your child's gonads (testes, ovaries, etc.), ask: Is this medically necessary right now? If your child is facing a significant immediate threat of gonadal cancer, then there's good reason to take the gonads out. But sometimes surgeons remove gonads from infants with DSDs even though they pose no significant immediate medical danger because they think this will spare the child from being aware of the gonad removal later. Yet adults who have been through this tell us they were not ultimately spared the psychological trauma of gonadectomy—they just had the choice taken away from them. There are often good reasons to leave healthy gonads in, including the significant benefits of natural hormones and increasing your child's chances of becoming a biological parent. (Reproductive technologies are increasingly allowing previously-infertile people to be biological parents.)

12. If the doctors are offering hormone treatments, ask: Is this medically necessary right now? What is the evidence, and what is the danger of doing nothing right now? Some hormone treatments are necessary to keeping children physically healthy and to preserving their fertility. But some are optional you can wait for your child to decide what's right for him or her. Many hormone treatments come with effects that are not reversible.

13. Can you introduce me to someone with a similar condition who has been treated the way you recommend, and someone with a similar condition who was treated with an alternative? This won't give you a scientific sample, but it will let you meet some adults with DSDs who can help you think about what your child might want from you in the long run. They may also know of good people for you to talk to and may have some important information about your options.

14. If you are feeling overwhelmed and stressed, ask: Can you help me get professional mental health support? I'm feeling overwhelmed and I think I need help. You can also ask your own personal doctor (your family practice doctor, your intern, your gynaecologist, etc.) for this kind of referral. Make sure you tell him or her how you are feeling. Sometimes people will think you are coping okay when you are really feeling overwhelmed.

15. Finally, if you are feeling like you are emotionally strong and have become well educated about your child's DSD, ask the doctor: Would you please give my name to other parents in your practice who might need someone to talk to? It doesn't matter if their children have exactly what mine does, I just want to be supportive of parents in similar situations. Also consider letting your doctor know about good resources you have found that might help other families.

(http://www.isna.org/articles/tips_for_parents)