Parents need to protect: influences, risks and tensions for parents of prepubertal children born with ambiguous genitalia

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Abstract
Aim and objectives. The aim of the study was to explore parents’ experiences of and the influences, risks and tensions associated with their child’s genital ambiguity and the place reconstructive surgeries had in their lives.

Background. Research into the clinical management of disorders of sex development is a complex and growing field of study. A small number of studies have indicated that the decisions parents make with regard to their child’s care during infancy are confounded by moral, social and emotional factors which can influence their choices.

Design. Narrative methods.

Methods. In-depth interviews elicited stories from 10 mothers and five fathers of 11 prepubertal children.

Results. A key theme from the data focused on the parents’ motivations to protect their child from the real or perceived threats arising from other people’s attitudes and responses to their ambiguous genitalia. Several rationales for their protective behaviours were described, which encouraged the parents to actively manage their child’s care.

Conclusion. The dilemmas that parents face in the complex and challenging situation following the birth of a child with ambiguous genitalia require early support from insightful professionals such as nurses. This support could better prepare the parents to recognise and discuss their protective behaviours and the impact these may have on their child’s future.

Relevance to clinical practice. These findings draw attention to the parents’ need for nurses to be knowledgeable about the impact that disorders of sex development can have on family’s emotional, psychosocial well-being and decision-making. Nurses’ awareness of the debate surrounding genital surgeries is important if they are to understand the motivations that underpin parents’ decision-making and how to support them sensitively. Nurses are the health professionals who have the most consistent direct contact with these families throughout the child’s life and, therefore, can provide support in the ongoing decision-making process.

Introduction
The actual number of people worldwide who are born with ambiguous genitalia secondary to
having a disorder of sex development (DSD) (also referred to as intersex) is unknown (Leidolf et al. 2008). Surgical decision-making and the management of infants, children and young people with DSD are under major debate. A current dilemma faced by parents and professionals focuses on the complex ethical and social problems associated with either performing or withholding genital surgeries in infancy (Spriggs & Savulescu 2006). Parent’s motivation to consent to early genital surgeries (which within the context of this study refers to irreversible genital surgery undertaken in childhood without the child’s inclusion in the decision-making process) for their child can be influenced by discussions with clinicians (Douglas et al. 2010). The perceived psychological needs of the parents when faced with a child of uncertain sex have been cited as a motivational factor in surgical decisions (Murray 2009). Against a background of controversy and uncertainty, the questions arise as to who should decide and who has the authority to consent to childhood genital surgeries and what role nurses have in supporting or challenging parents’ decision-making.

In this study, we address, from the parents’ perspective, what they see as the impact and place of early genital surgeries on their children and the influence this has on their decision-making.

Background
For some babies born with ambiguous genitalia, the child’s sex is unclear despite expert examination (Ahmed et al. 2011). Surgeons have in the past been criticised for labelling DSD and genital ambiguity as an emergency situation (Murray 2009). Views, opinion and clinical practice surrounding early genital surgery have changed considerably over the last two decades in the Western world, yet variation between cultures continues to exist (Gillam et al. 2010). It has been suggested that surgical approaches to remove, diminish or alter genital ambiguity in non-life-threatening circumstances are neither necessary nor a sufficient measure by which to guarantee well-being (Tamar-Mattis 2006). Parents’ understanding of the long-term outcome and possible risk of genital surgeries in childhood is unknown (Roen 2004, Karkazis et al. 2010). Poor decision-making by adults during a child’s childhood can lead to distress in that individual with DSD later in their life (Gillam et al. 2010). Whether parents appreciate the potential consequences of early genital surgery that could include failed surgery, a change to future reproductive opportunity, impact on later sexual intimacy or a misalignment between the surgical outcome and later gender identity is uncertain. Further, MacKenzie et al. (2009) qualitative study suggested that adults with DSD may query their bodily differences and their gender identity during adult life.

Several authors describe parents’ interpretations of the need for genital surgeries in childhood as ‘erasing’ ambiguity or banishing disruption (Tamar-Mattis 2006) or as part of the quest to find the child’s true gender (Gough et al. 2008). Parents are considered to be at the frontline in understanding their child’s best interests and they are responsible for decision-making (Wiesemann et al. 2010). Their interpretation and decisions about a synonymous sex and gender for their child are influenced by individual knowledge, cultural and social experiences and beliefs (Parens 2006).

The aim of the study was to explore parents’ experiences of their child’s genital ambiguity and the place reconstructive surgeries had in their lives. This enquiry facilitated in-depth exploration of the influences, risks and tensions that parents of children with ambiguous genitalia experienced. Data presented here focus specifically on the parents’ preoccupation with wanting to protect their child as it was found to be a major influencing factor in their perception of responsible decision-making.
**Design and methods**
This study employed narrative enquiry, to collect, analyse and interpret parents’ stories to better understand parents’ experiences of their child’s genital ambiguity and contribute to knowledge of the condition.

**Methods**
The topic area of ambiguous genitalia in childhood was observed in practice by the first author to be complex. To develop understanding of the parents’ experiences and build a picture of the place their child’s DSD had in their lives, a qualitative approach was adopted. The research used narrative enquiry as a methodology that enabled us to collect parents’ stories, explore their tacit knowledge and clarify issues when we were not sure of our understanding. Narrative analysis was used to examine, interpret and synthesise parents’ stories about their experiences. In using a narrative approach, we were able to learn about parents’ experiences and examine their understanding not solely based on ‘causal determinism or relationships’ but by appreciating how parents related to their experiences (Snape & Spencer 2003). We were able to explore the social structures, systems and behaviours that influenced the parents’ thinking.

Qualitative in-depth interviews were undertaken twice, 10–12 months apart, with seven parents, while eight parents were interviewed once; this was because of several children undergoing surgery during the research study time period. In using a narrative approach, we were able to explore the impact of having a child with DSD, capture parents’ experiences of their child’s genital surgeries and look at how this changed over time.

The parents were asked to share detailed stories of experiences, behaviour, actions and activities. Opinion and value questions were used when necessary to gain information about parents’ goals, intentions and desires along with questions about feelings that aimed to draw out emotional responses and explore beliefs. All interviews were undertaken by the first author at a venue chosen by the parents, often the family home. Data collection took place over a 16-month period.

**Participants and narrative interviews**
Fifteen parents were included in the study from October 2004–February 2006. Ten participants were mothers and five fathers, all lived in United Kingdom. All of the children had had a range of genital surgeries within the last 3–10 years; some children had had early surgery, whilst others had multiple surgeries over a period of time. The surgeries included genitoplasty, vaginoplasty, clitoroplasty, severe hypospadias and chordee, removal of uterine remnants and gonadectomy.

**Ethical considerations**
Approval from the Local Research and Ethics Committee, University and Trust Research and Development Committee was sought and gained. The parents received written and, when requested, further oral information about the study. Prior to each interview, they were contacted to explain the purpose of the interview in more detail. Written informed consent was collected both prior to starting and after completion of the interview. This was in response to the ethics committee’s concern that parents might reveal sensitive information during the interview that they wished they had not shared. The parents were also aware that they had the right and opportunity to withdraw their consent at any time, while psychological support was available if they experienced any emotional challenges brought about by sharing
their stories. The parents were recruited by the first author and were neither known to her nor part of her clinical caseload. Issues of possible tensions in being both a researcher and a clinician were discussed with the ethics committee and explored throughout data collection within the researcher’s academic and clinical supervisory teams. Confidentiality was maintained and assurance given that pseudonyms would be used in reports, papers and presentations.

**Data analysis**

Interviews were audiotaped and transcribed verbatim and analysed according to Labov (1982) and Riessman (2008). Each interview was read and listened to several times to build an overall impression of the experience, and findings were discussed amongst the authors. Analysis of the interviews resulted in the uncovering of many stories that were, through an interpretative process, brought together as three themes. These themes focused on stories about their child being different as a result of ambiguous genitalia; emotional, social and life stories which revolved around parenting a child born with ambiguous genitalia and stories that explored both the early and ongoing interactions with healthcare professionals. Embedded within all the themes were three recurring elements of shock, protection and anxiety. Specifically, the element of protection crossed all three themes with detailed stories in relation to privacy, social interactions with others, genital surgeries and the child’s future relationships. This study focuses on protection because this was found to be something parents were particularly sensitive about (and at times felt criticised for) as it influenced their decision-making and was integral to their movement towards finding a sense of harmony about their child’s initial genital ambiguity.

**Findings**

The following sections are organised around the element of protection that comprises of parents’ stories about seeking to maintain privacy, social experiences and interactions, the role of genital surgery and parents’ concerns about their child’s future relationships. Illustrative quotations using pseudonyms are used throughout. The term child is used generically to include all stages of childhood development.

**Parents’ stories of seeking to maintain privacy**

The news of a baby’s birth is often a celebrated event shared with an extended social network. Questions about the health of the baby and mother are common place along with enquiries about the sex and name of the child. Often, shortly after the birth, the parents in this study were confronted with the need to manage, withdraw or change information about their baby. The realisation that they could not contain all the information and conversations about their child was a threat to their ability to safeguard their child. Parents described the postnatal ward, baby clinic and changing their child in public places as settings in which their child’s genital ambiguity could be discovered.

Postnatal wards were often difficult environments for mothers as they described feelings of being alone, afraid and angry with little privacy when medical professionals came to talk about their baby, as Chloe recalled: ‘I remember a mother in the next bed to me said, ‘what’s that test’? ‘Do we all have that’? And I was thinking ‘no’! And I thought ‘oh my God, I’m not going to be saying to you, we don’t know whether my baby is a girl or a boy, I’m not saying that’. In the early period of waiting for results, the parents were faced with not knowing how to respond to other people; sharing information in the early days carried the risk of regretting it later.
For those parents who shared information, the act of disclosure was based on the belief that it could limit inquisitive questions, as Maria explained, ‘we knew that whoever was told would know for his full life’. She continued, ‘I didn’t have a problem with speaking about it or discussing it, but people did say to me, “well we’ll know him when he’s 16 so, you know, so don’t say too much” and I suppose that’s right in a way because of the children that he was born with, he’s gonna go all the way through school with them children and them parents will always know’ (Maria). When her friends questioned her approach to sharing information, Maria did begin to doubt her openness. She talked about how ‘everyone in that community will remember that child, the one that was born that we didn’t quite know whether he was a boy or a girl, so I do think of that now and I wonder whether I would have done it differently’. Her worry was that in trying to ensure that genital ambiguity was not something to be ashamed of, her disclosure could influence how others would interact with her son in the future.

The parents’ concern about how others would react when their child was undressed was informed by their own initial reactions and those of the midwives and other healthcare professionals. Maria reflected on her worries about ‘going to the clinic and having him weighed. It was like everyone’s gonna see him undressed, and this is like horrendous’. Even though Maria had shared the story of Neil’s birth condition with friends, the risk of them seeing him undressed was a significant worry. While some mothers attended baby clinics and had babysitters, others chose to avoid situations they sensed may result in questions ‘people [couldn’t] change my baby, because I hadn’t told people’ (Chloe). In attempting to avoid the perceived threat of others knowing, some of the parents let opportunities pass that would allow them to participate in parent–child activities such as play group and nursery. These instances perhaps reflect how parents negotiated protection and weighed up the precautionary measures such as avoiding activities they believed could have the capacity to harm their child’s future well-being.

Parents’ stories, social experiences and interactions
The parents’ motivation to protect their child from possible threats was grounded in their own beliefs of binary sex (i.e. children are either born male or female) and their desire to avoid potentially negative social and emotional outcomes for their child. The parents had the task of sharing the news of their child’s condition with family and friends, and they needed time and support to think about how they could do this. Some of the parents recalled being fearful about sharing information about their child’s initial uncertainty as it could result in their child being vulnerable to unwarranted nosiness from other people. Three parents had direct experience of negative encounters as a result of their child’s ambiguous genitalia. The desire to protect their child was amplified by the insensitive and intrusive nature of enquiry. Both Imogene and Harry were made aware that their community knew of their daughter’s ambiguous genitalia when they experienced direct questions: ‘some of the things that people have said have just been horrendous... we were out weren’t we in a club and the barmaid said to me, “have you got that baby, they didn’t know whether it was a boy or a girl”’. So it was just like a knife all the time, you know, because everybody knew (about) us having the baby’. They described how they were worried that people would gossip about their daughter and exclude her or tease her for being different in the future.

Faye’s local community expressed a cruel interest in her baby, Gabby, ‘I had one woman ask me, “what it was like to have a child that was born with a gaping hole between its legs?”’ Even though she had previous family experience of a relative with ‘mixed sex’, she did not recall that they had encountered the level of curiosity she had, so she had not anticipated the
reactions of strangers in her community towards Gabby. In reappraising the threat, Faye wondered how she could shield Gabby from the ‘amazing’ and untrue stories. For example, she was accused of having ‘paid private to give my child a sex change cos I didn’t want (all) boys’, she was astonished that people had considered she would so this. Being unable to participate in daily activities without fear of reprisal was initially frightening for Faye. However, she believed in her own resilience and self-efficacy, she could move on and ‘cope’ by going out less and ignoring the ‘people spitting at me in the street’. Yet she remained fearful that the level of curiosity would manifest itself as cruelty towards Gabby in the future. Faye’s desire to avoid this potential negative outcome resulted in her moving house, losing those friends who had supported her and changing her elder son’s school.

Although the majority of parents did not share Faye’s or Imogene’s and Harry’s experiences, some remained fearful of a perceived threat or possible hostility as Oliver described ‘[if] the community knew, it would possibly have a detrimental effect to Rose’s wellbeing’. Having control over sharing information about their child helped parents to navigate and understand ways in which to protect their child from the perceived (and for some real) risks of disclosure. Parents described this as keeping information ‘private’ (not secret). It also meant they had to guard against having ‘weak moments’ (Chloe) where they would share information with people that did not really need to know.

Based on their experiences and beliefs, the parents believed that surgery could possibly reduce the risk of their child’s ambiguity by making it less obvious to others.

**Parents’ stories about the role of genital surgery**

The majority of parents were thankful their child ‘didn’t remember’ early surgical experiences, but they did not use this as a reason to ‘not talk about’ previous surgery with their child. Like several other parents, Chloe considered ‘it was not good [for Edward] to look different’ and she needed ‘some clever surgeon to fix it’. Although a small number of parents also used the word ‘fix’ to describe their initial interpretation of the role of surgery, their reflections during the interviews framed it as a protective action taken on behalf of their child and that they understood that surgery was not curative.

Over time, the decision to opt for early surgery was revisited by some parents in the light of experiences, new knowledge, observational learning or discussion with some professionals, including nurses. One such change was described by Andrea, her motivation to protect Bobby became focused on halting additional surgeries as she had learnt more about his condition and he had begun to voice his own opinion. This resulted in Andrea and Brian having opposing views on both the timing and multiplicity of Bobbie’s surgeries. Andrea viewed multiple surgeries as ‘cosmetic’, and therefore, these could wait while Brian considered that surgery was ‘corrective’ and needed to be completed. Other parents re-examined their own decisions and their confidence, knowledge and authority to question surgery, Christine described ‘It’s hard either way you look at it, isn’t it. You just want what’s best’. She spoke about Daniel’s multiple surgeries having what she believed were a short-term negative outcomes on his emotional well-being.

Social and cosmetic tensions motivated many parents. They believed their child was in danger of ridicule ‘I just felt it needed to be repaired he was going to the toilet and everything so I suppose it was more cosmetic. I knew it [ambiguous genitalia] wasn’t having an effect on his health, apart from the reflux he had. But cosmetically he wasn’t weeing from the right place, and I suppose in the long term I was thinking well when he starts going to the loo, not
in his nappy you know. And going to nursery and things like that, that is when it would be noticeable.... I wouldn’t want him to be singled out as being different, just because his willy doesn’t look right. That can start the bullying thing, so I suppose in a way, you just want them to be the same as everyone else and you’re protecting them, really’ (Anne).

Conversely, Kerran questioned ‘If she was protecting her daughter by not agreeing to a clitoral reduction’. Initially, she had believed ‘it is a parental choice’, and she wanted surgery for her daughter but following discussion with a professional she began to question ‘What’s right...what are the long term implications...I’d never do it without my daughter’s consent’. At times, some parents worried about later gender identity issues. For Daniel having elements of both male and female were described by Christine as a worry ‘but as long as it’s not causing him any problems then, you know that’s a relief’. For Christine, the perceived threat was to Daniel’s later gender identity that possibly made him vulnerable in the future to attack from other people who could be intolerant towards his sexual orientation.

**Parents’ concerns about their child’s future relationships**

Parents felt the need to protect their child’s future intimate relationships by saving their child from possible ‘embarrassment’. The dilemma of sharing too much information with the young person too soon was a concern. By not sharing information early enough, some parents believed that their child would not recognise that their genitalia were different from others as David described his fears as ‘I’m just trying to protect him, when he’s sixteen and all the hormones start going he’s only got to try and go with a girl. Next thing you know it’s round the school, I can see it coming’. David was anxious about sharing too much information with Edward too quickly as he felt it could impact on his self-esteem and confidence in developing future relationships. He movingly summed up the struggle he faced in relation to his son by saying ‘I’m just trying to protect him’ and not knowing ‘how do I protect him, really’.

When their child decided to share information about their own body with other children, the parents’ protective motivations and desire to keep their child safe were challenged. Sian considered Tara’s decision to share information as a positive move and not something ‘she’s got to hide’. All of the parents thought that their child should have an understanding and knowledge about their condition in order to be able to share information about themselves. In learning how and when to share information with their child (about genital surgeries, DSD and genital ambiguity), the parents believed they were protecting their child from future uncertainty while encouraging the early development of positive self-esteem.

Conflict occasionally arose between a parent and child when the child was resistant to the parent wanting to talk, as recalled by Maria ‘I didn’t think he [Neil] had a problem with his genitalia at all and then....they started swimming....he wouldn’t change. Won’t talk about it, silence. Just gets very very upset and very agitated and won’t talk about it’ (Maria). She wanted him to have an understanding about his body and believed his behaviour at the swimming baths would result in his peers ‘taking the mick out of him’. Maria made a decision to wait until he was ready to talk about it, and she encouraged her husband to lead the discussion with Neil about sex and relationships.

**Discussion**

In line with other studies, we report that having a child with ambiguous genitalia has an immediate and ongoing impact on parents (Duguid et al. 2007, Fedele et al. 2010). For the parents in our study, assessment and re-evaluation of their response to the news of their
child’s ambiguous genitalia brought about a ‘response efficacy’, the belief that taking protective action on behalf of their child will work (Floyd et al. 2000, p. 411). The parents believed in their own ability (self-efficacy) to do what they thought was right in order to protect their child from the risks and dangers they believed may arise as a consequence of genital ambiguity. Gillam et al. (2010) argue that parents have a principal ethical responsibility to protect and promote their child’s interests. However, being a parent does not necessarily mean that they will always make good decisions for their child; their wishes may be in conflict with what their child as a young person or adult might have chosen for themselves. Parents’ expressed an innate and overwhelming desire to protect their child, and this informed their decision-making to opt for early genital surgeries. Adults’ instincts to protect children from distressing situations can result in children not engaging in later decision-making (Coyne & Harder 2011). As parents’ made decision’s for early surgery, the chance that the child could take absolute ownership of their DSD is lost; major decisions have already been taken on the child’s behalf, and it is too late to reverse what has been done. We know from studies of adults with DSD that there can be an alternative view, but this was missing from many of the parent’s narratives about decision-making. The desire to protect needs to be explored and balanced with the right of the child to make future decisions.

Spencer (2011) notes that nurses are involved in supporting parents through difficult decisions concerning their child’s treatments and their role often includes advocacy. Nurses involved in caring for this group should be cognisant of the complex ethical and social concerns surrounding early surgery so that they are in a good position to listen to parents’ concerns and discuss these in an informed way. By participating in these discussions with the parents, the nurse can then bring a unique perspective to other heath professional involved in the care of the child with DSD and their family.

Being a parent of a child with ambiguous genitalia brought a heightened sense of responsibility for all the parents in this study. Although genital ambiguity is not overtly visible, there were times when the child’s ambiguity could potentially be observed by others (parents’ family, friendship and community groups) which reflect the findings by Karkazis (2008). The parents’ desire to protect their child motivated them to build a sense of social congruence in their lives as also seen in the work of Carmichael and Alderson (2004). Developing this congruence took time. Occasionally, protection manifested itself as a form of voluntary exclusion from the usual social engagements, associated with parenting such as deciding not to attend baby clinic. Through seeking to shield their child, the parents assumed the role of ‘information controllers’ (Goffman 1963), and this in itself created tensions for parents. The parents had to weigh up the cost of taking precautionary measures, such as not sharing news with extended family and friends about their child’s condition, against the possible benefit of receiving additional personal and emotional support from a wider social network. Controlling or concealing information from family and friends was not a natural act for parents, but the tension created by this was outweighed by the possible risks of their child being singled out as different.

In particular, and aligned to the findings of Gupta et al. (2010) and Duguid et al. (2007), the parents in our study were concerned about reducing the risk of their child being teased or bullied. Similar to Kirk et al.’s (2010) findings, the parents in our study believed that their child with DSD was vulnerable. It was clear that the parents were constantly alert to threats arising from other people’s attitudes and responses to their child’s genital ambiguity. When other people were unable to easily match the child to either being a boy or girl, the parents responded by evaluating the risk to the child and their family. Other writers considering
threats to health have noted similar internal and social responses (see Protection Motivation Theory, Rogers 1983, Boer & Seydel 1996).

Prior to surgery, the parents worried about the visual information that their children could inadvertently ‘convey through their bodies’ (Goffman 1963). Genital surgeries held the potential for reducing the risk of their child’s body giving them away which has also been reported by Dayner et al. (2004). Parents had to balance the risks associated with the surgery against the risks they perceived to their child’s social identity, future self-esteem and safety. It was unclear whether the majority of the parents had been presented with the option not to operate on their child’s genitalia. Similar to other studies (Chertin et al. 2005, Crawford et al. 2009), the parents in this study appeared to use the aesthetic outcome as a measure of the success of childhood genital surgeries because genital surgeries offered a degree of unity between sex, gender and physical function that was considered to be impossible without surgery (see also Dayner et al. 2004, Jürgensen et al. 2006, Rebelo et al. 2008).

Limitations
The small sample represented a range of parents whose children had different underlying diagnosis of DSD (a full summary can be found in Sanders et al. 2011a,b). The study only followed up participants over a 16-month period and included those parents whose children had had reconstructive surgeries from a single centre.

Conclusion
The findings reflect the strategies and actions taken by the parents to protect their child in the short and longer term. Nurses and other professionals need to understand the impact that parents’ need to protect has on their decision-making and balance this against the perspectives of young adults with DSD who have expressed alternative views about early surgery. Because early surgical intervention is not a guarantee of future well-being, further longitudinal research with both parents and young adults could provide valuable insight into longer-term outcomes. Furthermore, future research could include an exploratory study of young people with DSD, their experiences of genital surgeries and growing up with DSD and families interactions with/influences from health professionals.

Exploring the parents’ ongoing need to protect their child is an area that nurses should consider exploring so as to better support them in a shared decision-making process that includes the child’s perspective. The desire to protect, whilst positive in many respects, has both foreseeable and potentially unforeseen implications. Opportunities to support children and families can arise during other professional encounters such as reviews of medications and outpatient clinics. Regardless of whether a nurse has particular expertise within endocrine, urology, genetics, gynaecology or other areas of care, limited evidence exists to guide practice and to provide a platform for debate, discussion, support and sharing of current practice (Sanders et al. 2011a,b). It is important that nurses have an understanding of the role they play in explaining the options available to parents and supporting them when they face difficult decisions. The nurse needs to be aware that decisions shaped by parents’ desire to protect their child through early surgical intervention will have ongoing consequence throughout the child’s childhood and adult life. Nurses need to have knowledge of the challenges parents may continue to experience and they also need to be skilled communicators.

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Conflict of interest
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