

The psychological impact of genital anomalies on the parents of affected children

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Abstract

Background: There is scarce information on how parents cope with children with genital anomalies. **Participants & methods:** Twenty-six parents of 25 children with a median age of 0.5 years (r, 5 days–10.8 years) were recruited through the Scottish Genital Anomaly Network and had a quantitative assessment of parenting stress and coping patterns; a qualitative assessment by a semi-structured interview was also performed in 19 parents. **Results:** In five parents, the total stress score was above the 85th centile, denoting clinical levels of stress. Three parents showed reduced coping pattern scores for social support, self-esteem and psychological stability and three showed a reduction in utilization of communication with medical professionals. The scores did not correlate with each other or the extent of genital anomaly in the child. Semi-structured interview analysis revealed parents' need for more knowledge about the imminent surgery, post-operative care and their desire for written information that could complement the time-restricted contact with the clinical team.

Conclusion: In the majority of cases, parents did not display abnormal levels of stress or coping on quantitative assessment. The semi-structured interview provided further information about the parents' level of coping and potential for stress and highlighted the need for more effective exchange of clinical information at a critical period of the parent–child relationship.

INTRODUCTION

Genital anomalies are encountered at birth in approximately 1 in 300 births (1). They range from relatively simple structural abnormalities such as glanular hypospadias to other more complex genital anomalies. As adults with previously corrected genital anomalies recount their experiences, clinical management of these conditions and, especially, those that are more complex, is undergoing a reconsideration with a greater emphasis being placed on improved functional, rather than, cosmetic outcome (2,3). Clinicians are also considering delaying surgery until a later stage when the patient may be in a position to provide informed consent (4). The move towards multi-disciplinary decision-making models has led to the parents being seen as central to the decision making process (5,6).

Child behaviour, life events and parental personality characteristics can raise parenting stress which, in turn, can adversely affect the quality of caregiving, parent–child interactions and child behaviour (7). Coping is used to mediate stress and a family can utilize specific cognitive and behavioural coping strategies to reduce this stress. It is likely that parenting stress and coping patterns may influence long-term psychological outcome of the child (8,9). Whilst there are many published reports of the role of chronic disease as parental stressors, at present, there is little published evidence on the needs of parents of children with genital

anomalies and how best to support them. Therefore, it is important to understand the stresses, if any, that they may face and what methods they use to cope with their child's condition, in order, to ensure positive psychosocial outcomes for the child. The purpose of this study was to examine stress and coping patterns amongst parents of children with genital anomalies by using previously validated questionnaire-based quantitative psychological assessment tools, as well as a semi-structured qualitative interview.

METHODS

Subjects

Children and their families were recruited through the Scottish Audit of Genital Anomalies, a prospective survey of children presenting to the Scottish Genital Anomaly Network (SGAN). All children presenting for the first time with a genital anomaly (other than isolated glanular hypospadias or isolated unilateral undescended testis) were eligible for entry. The parents were presented with information about the study at presentation and following their approval, the study coordinator (AD) was notified of their details and she subsequently contacted the parents for recruitment into the study. Between July 2003 and October 2004, informed consent for this study was obtained from the parents of 43 children (42 boys, 1 girl) out of 79 notified cases. Due to logistical constraints, the questionnaires

were completed by face-to-face interview and the total number of completed questionnaires for quantitative assessment were obtained from 26 parents of 25 out of the 43 children (22 mothers and two fathers and in one instance, with both parents) and qualitative assessment was performed in 19 of these 26 parents (12 mothers, two fathers and in five instances, with both parents) in their own home. The studies were performed at a median age of 0.5 years (r, 5 days–10.8 years). These 25 children consisted of boys in whom the karyotype was 46XY in 21, 45XO/46XY in 3 and 46XX in 1 case. The median External Masculinization Score (10) was 9.5 (range, 6.11); further clinical details of these 25 cases, as well as those in whom consent was not obtained are presented in Table S1. The study was approved by the Multi-Centre Research Ethics Committee and informed consent was obtained from all participants.

Parenting stress index/short form (PSI/SF)

The 36-item PSI short form is derived from the full-length PSI and is used to determine the total stress score which provides an indication of the overall level of stress experienced. Total stress on the PSI short form correlates at 0.94 with the full-length form. Test-retest and alpha reliabilities for the PSI short form total score are 0.84 and 0.91, respectively (9). The normative data were derived for each PSI scale from the raw data frequency distribution of scores from the mothers in the normative sample (9). Scores above the 85th centile for the normative sample were regarded as achieving clinical levels of stress (9).

Coping health inventory for parents (CHIP)

The CHIP is a 45 item self-report measure designed to assess parents' perceptions of the behaviours they are currently using to manage family life when they have a child who is seriously and/or chronically ill (8,11). Respondents are asked to record the helpfulness of each coping behaviour in their family on a scale of 0 to 3, with 0 indicating 'not helpful' and 3 indicating 'extremely helpful'. The scales break into three discernible patterns: Pattern I (family integration, co-operation and an optimistic definition of the situation), Pattern II (maintaining social support, self-esteem and psychological stability) and Pattern III (understanding the health care situation through communication with other parents and consultation with the health care team).

Semi-structured interviews

Families were interviewed by AD using a semi-structured protocol, examining: the disclosure of the child's condition, information received or accessed by the parents, sources of support, concerns about the forthcoming surgery, concerns about the child's future and suggestions for future service development. The interviews lasted between 30 and 90 min and were recorded, transcribed and analysed using a QSR N6 qualitative package (QSR International, Cardigan, UK). Initially, the text was separated into 'meaning units' (MUs) (12) identified as portions of text that expressed one concept relevant to the study. MUs relating to the same phenomenon were then collected into a category, which was given a de-

scriptive label. The category content and labels were refined in an iterative process as coding and analysis of subsequent transcripts built on the initial coding. Once the initial categories were completed, these were clustered together at a higher level of abstraction. Finally, the relationship between these higher-order categories was examined. Memos were used throughout analysis to aid this process (13).

Statistical analysis

The data were presented as medians and 10th and 90th centiles. Bivariate analysis was performed using Pearson rank correlations to compare the relationships between subject dependent variables. Data were analysed using XL STAT V7.0 (Addinsoft, France) and Microsoft Excel 2000 (Microsoft Corp, Redmond, WA, USA). The alpha value of statistical significance was 0.05.

RESULTS

Total PSI

In 21/26 (81%) cases, the total PSI score was within the normal range (0–85th centile) and in five cases the score was above the 85th centile denoting subclinical levels of stress. There was no relationship between the EMS and the PSI score.

CHIP

Three parents out of 24 showed reduced scores for social support, self-esteem and psychological stability and three showed a reduction in utilization of communication with medical professionals and parents as a method of coping. There was no relationship between the EMS and the CHIP scores.

Qualitative analysis

Seven principal themes emerged from the data. These were: general experience, handling the subject of genital anomalies, concomitant stressors, sources of social support, coping strategies used by the parents, the impact of the condition on the child and the family and suggestions on improving the clinical service. Issues raised and the percentage of parents who raised them are presented in Table S2.

General experience

Information that was inadequate (10 parents), incorrect (three parents) or too emotive (two parents) at birth was a very common subtheme raised by parents. The emotional vulnerability of the mother during the postnatal period was also raised by a number of parents.

Handling the subject of genital anomalies

Over 60% of parents interviewed reported difficulties in discussing the condition with relatives and friends. In addition, four out of 19 parents (20%) admitted that the condition was even difficult to discuss between the mother and father. Thirteen out of 19 parents (68%) had some concerns about the condition being associated with ridicule or stigma. Although a number of parents mentioned that their child

received an appropriate level of sensitive handling when admitted for surgery, three parents reported a greater need for professional sensitivity when teaching trainees. Four out of 19 parents also wished for greater support for explaining the condition to the affected child.

Concomitant stressors

Complications associated with pregnancy, delivery and prematurity were reported as stressors by one third of parents. Other stressors specific to the affected child included ongoing problems with the child's own cognitive development. As expected, problems with other offspring, as well as other forms of social upheaval were reported by some parents as stressors.

Sources of support

Almost 75% of parents reported that relatives and consultant surgeons were their greatest source of support. A quarter of the parents reported that they did not find it helpful to seek support from their own relatives. In general, hospital-based clinical staff were reported to be a greater source of support than primary care-based personnel.

Coping strategies

Parents used a number of coping strategies to deal with their child's condition. Relying on the clinical staff to make the right decisions and relying on the perceived 'treatability' of the condition were the commonest employed coping mechanisms. Placing their child's condition in perspective with other affected children was also commonly employed.

Impact of condition on child & family

The commonest concerns of the parents under this theme included general concerns of an anaesthetic and as well as surgery and future concerns of fertility and sexual function. Eleven out of 19 (58%) parents reported that they were unclear about the expected appearance of the genitalia following the operation. Most of the concerns under this theme were related to surgery (Table S2). Delay in surgery as leading to further ridicule at school was only raised as an issue by 2 (11%) parents. Five (26%) parents raised concerns about recurrence of the condition either in future offspring of the parents, the affected child or the affected child's siblings.

Suggestions on improving the clinical service

A substantial number of parents reported the need for written information on general issues such as the condition and specific issues such as cleaning affected genitalia before and after surgery. The need for gradual and steady information about the condition following the initial presentation was also reported, as was the need for some illustrative examples of the expected appearance post-operatively. In this category, eight out of 19 (42%) parents reported the desire to have a local network of families with children affected with similar conditions. Other sources of support such as a website and a link person who would be available at the initial presentation was also highlighted by some parents.

DISCUSSION

This study is among the first to investigate the level of stress and the potential sources of stress faced by parents of children with genital anomalies. Studies looking at parents of children with congenital anomalies, such as, heart disease, and other congenital developmental conditions, have generally found evidence of increased stress when compared to parents of typically developing children (11,14–16). In the current group of children, predominantly boys with XY undermasculinization, the parents did not generally display levels of stress that would require professional intervention. Whilst some of the offspring were quite severely undermasculinized at birth, the average EMS was relatively higher than that reported in other cohorts (17). Whilst it is possible that the generalizability of our findings may be limited to a cohort with this degree of undermasculinization, other investigators have shown that the parenting stress or level of coping is more likely to correlate to parental perception of risk than the clinical perception of severity of illness (11, 18,19). It is possible that the chronicity of the genital anomaly or the timing of the interview in relation to initial presentation or interventions such as surgery may alter parenting stress or level of coping but this will require a longitudinal study.

The qualitative assessment by semi-structured interview provided a valuable adjunct to the objective assessment and highlighted clear differences between these two types of assessment. The study indicated a great need for accurate and adequate education and information for parents with respect to many different aspects of the child's care. The need for this information was highlighted at many points of the child's journey on the care pathway starting from soon after birth to the point of surgery and beyond. Dayner et al. have previously reported the importance of transmitting accurate and understandable information and the parents' desire to understand the basis of the underlying problem, choices of treatment available and the realistic outcomes that might occur (6).

The concerns raised in the qualitative interviews highlighted the parents' needs for both condition specific as well as generic information. For instance, whilst they had concerns about sexual function and fertility, one of the major concerns raised related to the general concept of anaesthetics and surgery. This generalized, non-specific anxiety may not be just due to the perceived risks of these procedures but may also be related to their perception of culpability in deciding to proceed with surgery that may result in pain and distress for the child. A substantial number of parents reported being concerned about the stigma or ridicule associated with the condition, but only a small number of parents felt that a delay in surgery would lead to additional ridicule at school.

The interviews served as a first step in characterizing the potential factors that may lead to parental stress. It has provided a glimpse into the type of support and coping mechanisms that parents employ and, most importantly, it provides a benchmark that can be used to improve the provision of service for parents of children with genital

anomalies. Our current study also raises a question about the reliability of using standard quantitative questionnaires for the purpose of identifying parents with clinical levels of stress or problems with coping. It is possible that a questionnaire that is adapted from the qualitative data may provide more specific insight into the concerns of parents with genital anomalies. Quantitative assessment, such as PSI, can be subject to false negative results when the respondent is not being honest about their feelings, acting defensively or disengaged with the whole process and the PSI score is very low (9). However, only one respondent had a borderline low PSI score.

At about 50%, the recruitment rate into this study was low but not as much as another study of quality of life and psychological outcome of adult women with androgen insensitivity syndrome by our own group (20). However, these studies involved face-to-face interviews and given the geography of Scotland they were very resource intensive; our study sample was, therefore, further depleted because of logistical restraints on performing the interviews. We could not ascertain the reason for non-consent but our data suggests that the severity of the anomaly was similar in those who consented to those who did not. Although it is possible that some parents may wish not to discuss issues concerning genital anomalies, as highlighted in the qualitative interview, a recruitment rate of just over 50% was also encountered in a study of maternal stress and coping in the field of end-stage renal disease (21).

In summary, the parents of the children studied in this cohort, predominantly boys with XY undermasculinization, did not generally display abnormal levels of stress or coping on quantitative assessment. The semi-structured interview provided further information about the parents' level of coping and potential for stress and highlighted the need for more effective exchange of clinical information at a critical developmental period of the parent-child relationship.

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Supplementary material

The following supplementary material is available for this article:

Table S1 Range of clinical features in cases where consent was obtained as well as those where consent was not obtained for the study.

Table S2 Themes, subthemes and percentage of respondents (n, 19) who raised the theme.

This material is available as part of the online article from: <http://www.blackwell-synergy.com/doi/abs/10.1111/j.1651-2227.2006.00112.x>

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