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Predictors of Posttraumatic Stress in Parents of Children Diagnosed with a Disorder of Sex Development

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Abstract The aims of the current study were twofold: (1) to assess the prevalence/severity of posttraumatic stress symptoms (PTSS) as well as cognitive and emotional responses in parents whose children were diagnosed with a disorder of sex development (DSD); and (2) to assess factors which contributed to PTSS. We hypothesized that parents would show elevated levels of PTSS and that negative cognitive and/or emotional responses would be predictive. Participants were parents of children diagnosed with a DSD. Thirty-six mothers and 11 fathers completed a measure of posttraumatic stress and reported difficulties in the domains of cognition (e.g., confusion) and emotion (e.g., grief). Using multiple regression, we determined factors contributing to parental PTSS. Reported PTSS was high: 31 % of mothers and 18 % of fathers met the threshold for caseness for Posttraumatic Stress Disorder. Regression included: child sex, parent sex, child age at diagnosis, years since diagnosis, genital ambiguity, father occupation, cognitive confusion, and emotional distress. Only cognitive confusion contributed significantly to variance in PTSS. Parents of children with DSD may experience the diagnosis as traumatic, evidenced by high rates of PTSS in the current report. Assessment of reactions to their children's diagnoses revealed that cognitive confusion, and not emotional distress, predicted PTSS. In this case, direct cognitive interventions may be applicable. Though psychological support is widely recommended, no detailed intervention has been offered. Our findings suggest that we may directly apply models successful in other areas of pediatrics, such as pediatric oncology. Future studies may assess the usefulness of such an intervention.

Keywords Ambiguous genitalia · Intersexuality · Disorders of sex development · Posttraumatic stress

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Introduction

One in 4,000 infants is born with abnormalities of external genitalia sufficient to warrant formal investigations (Achermann & Hughes, 2011), often resulting in the diagnosis of a disorder of sex development (DSD) (Hughes, Houk, Ahmed, Lee, & LWPES1/ESPE2, 2006; Lee, Houk, Ahmed, & Hughes, 2006; Pasterski, Prentice, & Hughes, 2010a). Such a diagnosis, defined as a condition where chromosomal, gonadal, or anatomical sex is atypical, presents a challenging clinical emergency: Immediate and long term concerns include gender assignment, genital surgeries, gonadal malignancy, potential gender dysphoria, and disclosure to parents and patients (Hewitt & Warne, 2009; Sutton et al., 2006). Over the past two decades, treatment models have emerged with the common theme of a multidisciplinary approach to management, including as many as possible of the relevant medical subspecialties, including psychiatric/psychological support services (Lee et al., 2006; Liao, Tacconelli, Wood, Conway, & Creighton, 2010; Pasterski et al., 2010a). An audit of services in Europe (Pasterski, Prentice, & Hughes, 2010b) suggested that theoretical models have been put into practice with 95 % of 60 centers surveyed employing the majority of the recommended subspecialties. With these teams in

place, attention has turned to detailed protocols and logistics for delivering specialized services with patient and family needs at the center of consideration.

For parents, the experience of having a child diagnosed with a DSD is increasingly being understood as a traumatic event (e.g., Duguid et al., 2007). While not all cases are life-threatening, as with salt-wasting and dehydration in congenital adrenal hyperplasia (CAH), the event may nevertheless be experienced as a threat to the integrity of the child. An evidence based model for treating and supporting families with a child diagnosed with cancer, the Pediatric Psychosocial Preventative Health Model (Kazak, 2006; Kazak et al., 2006) identifies events such as diagnosis and emergent medical care as potentially traumatic events (Kazak et al., 2007). The model articulates that it is the interaction between the objective nature of the event and the subjective interpretation of the event which renders it as traumatic or not. This model allows for the identification of at risk families and sheds light on an opening for direct intervention. Likewise, accessing parents' subjective interpretation at the point of disclosure in the case of DSD may offer insight toward a more holistic health care protocol for patient and family.

With regard to potential sequelae from trauma experienced by parents at the point of disclosing a DSD diagnosis, we may consider the existing framework for posttraumatic stress symptoms (PTSS) outlined in the DSM-IV-TR (American Psychiatric Association, 2000): "[E]xperiencing, witnessing, or confronting events that involve actual or threatened death or serious injury, or a threat to the physical integrity of self or others." Symptoms are clustered in terms of intrusive thoughts about the event, avoidance of reminders of the event, and/or hypersensitivity with regard to that or similar events.

Though increased levels of distress have been reported in parents of children severely injured or diagnosed with grave illnesses (Kazak et al., 2004; Mastroyannopoulou, Stallard, & Lenton, 2006; Winston et al., 2002), there is only a single report specific to stress in parents of children born with a DSD (Duguid et al., 2007). In that study, though 5/26 (19.2 %) of parents scored above the clinical cut-off on the Parenting Stress Index (PSI), Duguid et al. concluded that stress was not primary feature for such parents. If one included parents whose scores were subthreshold in that study, the percentage would no doubt increase, warranting formal clinical attention. Furthermore, while models of health care delivery have been tailored to include the potentiality of PTSS in families of children diagnosed with cancer (Kazak et al., 2007), such a detailed conceptualization of patient and family experience has not yet been issued with respect to DSD.

The aims of the current study were twofold. First, we aimed to determine whether parents of children diagnosed with a DSD experience PTSS. Secondly, given our conceptualization of the potentially traumatic event, where subjective interpretation may lead to distress, we aimed to assess the relationship between the subjective response to learning about a diagnosis of DSD and posttraumatic stress.

Method

Participants

Tables 1 and 2 show sample characteristics. Thirty-six mothers and 11 fathers independently completed a measure of PTSS and rated their cognitive and emotional reactions to learning about their child's diagnosis. The 47 parental reports represented 31 female children, 16 male children, and included 36 families. Two mothers in our sample each had two children. Their data are presented as separate reports as they had different experiences. All but two (96 %) of the parent participants were Caucasian (two were of Indian/Pakistani/Bangladeshi origin). With respect to inclusion criteria and DSD, we included parents of patients with both 46,XX DSD and 46,XY DSD (see Table 2). Note that while CAH in males does not technically fall under the umbrella of a DSD, as they do not evince a chromosomal, gonadal, or anatomical anomaly, some have suggested that it should be due to the risk for testicular adrenal rest tumors (TART) and potential gonadal dysfunction/infertility (Claahsen-van der Grinten et al., 2007). To be optimally inclusive, we included these cases for comparison as children who experience the same pathophysiology, but without genital ambiguity.

Procedure

Parents were recruited from the endocrine clinic where they were receiving treatment ($N = 83$) or from a patient support network (UK CAH Support Group; $N = 140$). Those who indicated interest in the study were invited for an interview and were given a set of questionnaires pertaining to their own and their child's psychological and emotional well-being. Procedures were developed in conjunction with service users and according to national ethical standards and were approved by a local medical ethics board.

Measures

Impact of Events Scale Revised (IES-R) (Creamer, Bell, & Failla, 2003)

The IES-R is a revised version of a widely used self-report measure of posttraumatic stress. Twenty-two items comprise three subscales (see Table 3). The total scale has good internal

Table 1 Sample characteristics

	Males $N = 16$	Females $N = 31$	p
Child M age (SD) at diagnosis (in years)	0.69 (1.49)	1.42 (3.10)	ns
Child M age (SD) at participation (in years)	5.69 (3.93)	8.52 (5.48)	ns
Mean years (SD) since diagnosis at parent interview (in years)	5.00 (4.23)	7.10 (4.81)	ns

Table 2 Disease characteristics

Patient	Diagnosis	Karyotype	Assigned gender	Genital ambiguity	Gender assignment in question	Age at diagnosis	Parent reporting
01	Anorchia	46,XY	M	Yes	No	Newborn	M,F
02	GD	Mixed	M	Yes	Yes	Newborn	M
03	Undiag	46,XY	M	Yes	Yes	Newborn	M,F
04	CAH	46,XY	M	No	No	4y	M,F
05	CAH	46,XY	M	No	No	Newborn	M
06	CAH	46,XY	M	No	No	Newborn	M
07	CAH	46,XY	M	No	No	Newborn	M
08	CAH	46,XY	M	No	No	Newborn	M
09	CAH	46,XY	M	No	No	Newborn	M
10	CAH	46,XY	M	No	No	Newborn	M
11	CAH	46,XY	M	No	No	3 years	M
12	CAH	46,XY	M	No	No	Newborn	M
13	CAH	46,XX	M	Yes	Yes	Newborn	M
14	GD	Mixed	F	No	No	14 years	M
15	Undiag	46,XY	F	No	No	Newborn	M
16	Undiag	46,XX	F	Yes	No	Newborn	M
17	Undiag	46,XY	F	Yes	Yes	Newborn	M
18	AIS	46,XY	F	No	No	5 years	M,F
19	AIS	46,XY	F	No	No	Newborn	M
20	AIS	46,XY	F	No	No	2 years	M
21	CAH	46,XX	F	Yes	Yes	Newborn	M,F
22	CAH	46,XX	F	Yes	Yes	5 years	M,F
23	CAH	46,XX	F	Yes	Yes	Newborn	M,F
24	CAH	46,XX	F	Yes	Yes	Newborn	M,F
25	CAH	46,XX	F	Yes	Yes	Newborn	M,F
26	CAH	46,XX	F	Yes	No	Newborn	M,F
27	CAH	46,XX	F	Yes	Yes	Newborn	M,F
28	CAH	46,XX	F	Yes	Yes	Newborn	M
29	CAH	46,XX	F	Yes	No	Newborn	M
30	CAH	46,XX	F	Yes	Yes	Newborn	M
31	CAH	46,XX	F	Yes	No	Newborn	M
32	CAH	46,XX	F	Yes	No	Newborn	M
33	CAH	46,XX	F	Yes	Yes	Newborn	M
34	CAH	46,XX	F	Yes	Yes	Newborn	M
35	CAH	46,XX	F	No	No	1 year	M
36	CAH	46,XX	F	No	No	7 years	M

Patients 10, 32 and 11, 31 are two sets of siblings. Their data are presented as separate reports as the children and their families had different experiences

CAH congenital adrenal hyperplasia, *AIS* androgen insensitivity syndrome, *GD* gonadal dysgenesis, *Undiag* undiagnosed

consistency (Cronbach's $\alpha = 0.96$) as do the subscales (intrusion: 0.94; avoidance: 0.87; hyperarousal: 0.91). Sum scores were calculated for each subscale and subscales combined. While this measure was not designed as a clinical diagnostic instrument for Posttraumatic Stress Disorder (PTSD), cut-off scores have been commonly employed. We considered 33 as a theoretical cut-off for caseness (sensitivity = 0.91; specificity = 0.82). Parents were asked to specifically focus on the experience of disclosure when making their ratings. This method was based on a similar report where parents of children diagnosed with cancer were assessed for PTSD at a later point in time (Kazak et al., 2004). See Table 1 for mean age since diagnosis.

Cognitive and Emotional Responses

As a direct measure of response to disclosure, parents were asked to what extent the experienced the following: confusion and disbelief (cognitive response) and shock, shame, anger, guilt, grief, and relief (emotional response) using a 5-point Likert scale (1 = Not at all; 5 = Very much). The first five emotion items were chosen based on a previous report specific to androgen insensitivity syndrome (AIS) (Hughes et al., 2012; Slijper, Frets, Boehmer, & Drop, 2000). Relief (reverse coded) was added per request by service users at the study development/review stage. Mean scores across all items showed good reliability (Cronbach's

Table 3 Impact of Events Scale-Revised

Item		Subscale
01.	Any reminders brought back feelings about it	In
02.	I had trouble staying asleep	In
03.	Other things kept making me think about it	In
04.	I felt irritable and angry	Hy
05.	I avoided letting myself get upset….	Av
06.	I thought about when I didn't mean to	In
07.	I felt as if it hadn't happened or wasn't real	Av
08.	I stayed away from reminders about it	Av
09.	Pictures about it popped into my mind	In
10.	I was jumpy and easily startled	Hy
11.	I tried not to think about it	Av
12.	I was aware that I still had a lot of feelings….	Av
13.	My feelings about it were kind of numb	Av
14.	I found myself acting or feeling like I was back….	In
15.	I had trouble falling asleep	Hy
16.	I had waves of strong feelings about it	In
17.	I tried to remove it from my memory	Av
18.	I had trouble concentrating	Hy
19.	Reminders caused me to heave physical reactions….	Hy
20.	I had dreams about it	In
21.	I felt watchful and on guard	Hy
22.	I tried not to talk about it	Av

In intrusion subscale, *Hy* hyperarousal subscale, *Av* avoidance subscale

$\alpha = 0.61$). Higher emotional response scores were more negative and higher cognitive scores indicated greater difficulty in comprehension regarding the DSD diagnosis.

Statistical Analyses

Independent samples and one-sample *t* tests were conducted to evaluate posttraumatic stress in mothers and fathers compared to each other as well as to a published cohort of parents whose children have survived cancer (Kazak et al., 2004). Next, we employed two 2 × 2 repeated measures ANCOVAs, with parent occupation as a proxy for socioeconomic status as the covariate, to assess cognitive and emotional responses as a function of PTSS levels and parent sex. Finally, we performed a multiple linear regression to assess the impact of specific parental responses (cognitive and emotional) as well as other patient/familial characteristics on PTSS.

Results

Table 4 shows means for the IES-R total score and subscales. Mothers and fathers reported overall levels of posttraumatic stress similar to each other and similar to published means for mothers and fathers of children who have survived cancer (Kazak et al., 2004). The only differences in comparison to parents of children who have survived cancer were that mothers from our sample reported fewer symptoms on the intrusion subscale $t(31) = -1.97$, $p < .10$, and fathers from our sample reported fewer symptoms on the avoidance subscale, $t(9) = -3.66$, $p < .01$. In terms of caseness, 11 (31 %) mothers and 2 (18 %) fathers scored above the clinical cut-off of 33.

Cognitive and Emotion Responses

To further elucidate the relationship between cognitive and emotional responses with respect to PTSS, we conducted a 2 (Caseness) × 2 (Domain) repeated measures ANCOVA, with father occupation as a covariate. To establish PTSD caseness, we used the recommended cut-off score of 33+. There was a main effect of Domain, $F(1, 45) = 9.12$, $p < .01$, such that parents rated higher levels of cognitive confusion than negative emotional response. The Caseness × Domain interaction approached significance, $F(1, 45) = -1.70$, $p < .10$, suggesting that those who met the cut-off for PTSD caseness reported more cognitive confusion compared to those who did not meet the cut-off. This was not the case for emotional response. Simple effects analysis revealed a similar pattern. Though neither cognitive confusion

Table 4 Scores (and SDs) for the IES-R

Scales	Mother reports $N = 36$	Father reports $N = 11$	Diff. between parents	Mothers compared to mothers of cancer survivors $N = 146$	Fathers compared to fathers of cancer survivors $N = 106$
	M (SD)	M (SD)	*p*	*p**	*p**
IES-R total	25.32 (15.45)	20.33 (10.74)	ns	ns	ns
Intrusion	11.29 (6.59)	9.10 (6.14)	ns	ns	ns
Avoidance	7.80 (5.09)	5.00 (3.10)	ns	ns	.001**
Hyperarousal	6.23 (5.07)	4.20 (3.54)	ns	ns	ns

Parents in the comparison group were interviewed between 1 and 10 years post treatment

* Comparisons made using one-sample *t* tests

** Parents from our sample showed fewer symptoms

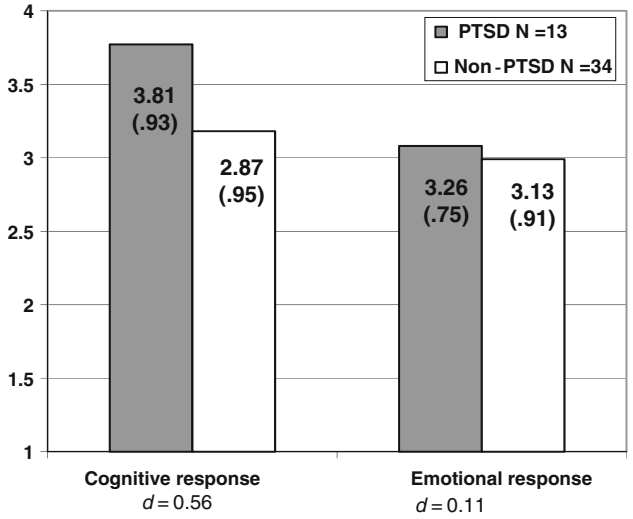

Fig. 1 Means (and SDs) for parents' cognitive and emotional responses as a function of posttraumatic stress. Note that participants scoring 33+ on the IES-R were considered threshold for PTSD

nor emotional response were significantly different between those who met the caseness cut-off and those who did not, effect sizes for the two comparisons suggest that the interaction might be significant with a larger sample, $d = 0.56$ for cognitive confusion and 0.11 for emotional response (Fig. 1).

We also conducted a 2 (Sex of parent) × 2 (Domain) repeated measures ANCOVA, with father occupation as a covariate, to see if mothers and fathers differed in their cognitive/emotional reactions to learning about their children's diagnoses. Again, there was a main effect of Domain such that parents reported more cognitive confusion that emotional distress, $F(1, 45) = 9.91, p < .01$.

Multiple Linear Regression

To investigate the relationship between parental cognitive/emotional responses and other patient/family characteristics with PTSS, we conducted a multiple linear regression, including eight predictors (see Table 5). Parents' post-traumatic stress scores (IES-R sum) served as the criterion variable. The overall model was significant, $R^2 = .35, F(8, 45) = 2.52, p < .05$. With regard to the eight predictors, only cognitive confusion explained a significant amount of the variance in parents' PTSS, $\beta = .56$, $p < .01$. Age at diagnosis approached significance, $p < .10$, such that those whose children were diagnosed at an earlier age had higher levels of PTSS.

Discussion

Findings from the current study suggest that not only do parents of children diagnosed with a DSD experience considerable levels of PTSS, but that this psychological distress may vary as a function of cognitive confusion about the event. Both mothers and fathers reported overall levels of PTSS that were comparable to those reported by parents of children diagnosed with other disorders, in this case cancer (Kazak et al., 2004). Thirty-one percent of mothers (11/36) and 18 % of fathers (2/11) in our study reported clinical levels of distress. Both the level of distress and the similarity between mothers and fathers were consistent with other reports (Kazak et al., 2004). Second, when we entered measures of cognitive confusion and emotional distress into a regression model that also included other specific patient/family characteristics, we found that cognitive, but not emotional, distress predicted PTSS. In fact, none of the other factors in the model accounted for significant amounts of variance. Finally, the difference in effect sizes in domain as a function of PTSD caseness suggested that those parents who met the clinical cut-off for PTSD showed higher levels of cognitive confusion compared to those who did not, while emotional responses were similar between the two groups. This finding was consistent with another report ranking uncertainty about diagnosis/management as particularly stressful for parents (Crissman et al., 2011).

Table 5 Regression coefficients and p values for predicting posttraumatic stress

Predictor	β	t	p
1. Sex of parent	0.18	1.25	ns
2. Sex of rearing of child	0.13	<1	ns
3. Genital ambiguity	−0.06	<1	ns
4. Child age at diagnosis	−0.28	−1.86	<.10
5. Years since diagnosis	−0.19	−1.39	ns
6. Father occupation	0.14	1.04	ns
7. Emotional index	−0.27	−1.36	ns
8. Cognitive index	0.53	3.06	.004

Model was significant, $F(8, 45) = 2.52, p < .05$

With respect to other predictors in the model, genital ambiguity did not contribute to the variance in PTSS as one might have expected. While gender is a highly salient feature and the condition of ambiguity at birth seems a likely candidate for parental distress, analysis suggests that it did not account for the high levels of PTSS in our sample. This was again consistent with the report mentioned above ranking uncertainty overall as more critical than concerns about genital ambiguity (Crissman et al., 2011). Furthermore, Duguid et al. (2007) concluded in their study specific to parents of children with genital anomalies that the parents did not report excessive levels of stress.

Because the time elapsed between receiving the child's diagnosis and participation in the current study may have played a factor in parents' recall of events, we included years since diagnosis in our regression. That this factor did not contribute significantly to the variance suggests that recall was not a limitation in the current report. Finally, as it was possible that parental cognitive functioning may have contributed to the variance in our measure of cognitive confusion, we asked parents about their

level of employment as a proxy of socioeconomic status/intellectual ability. Level of employment had no effect on PTSS.

Intuitively, it seems the high rate of PTSS in the current report may be attributed to the experience of life-threatening crises, such as salt-wasting in CAH (Woelfle, Hoepffner, & Sippell, 2002). While we did not have large enough samples within each sex/diagnostic category to make comparisons, e.g., a comparison between girls with AIS (Hughes et al., 2012) and boys with CAH (Woelfle et al., 2002), when we examined the mean scores for PTSS by child gender and diagnosis, the highest scores were for 2 mothers with daughters whose condition did not present with such a crisis (IES-R scores = 49 and 62), but rather whose children lacked a definitive diagnosis. In this case, not knowing the diagnosis seems to have contributed to PTSS.

In terms of limitations, we experienced relatively low participation, though this is consistent with similar studies including patients with a DSD (Crissman et al., 2011; Duguid et al., 2007; Slijper et al., 2000). This may in part reflect parents' wish to avoid such a difficult subject matter. In two cases of non-participation, the reason given was that the parents wished to avoid any reminders of the child's diagnosis. Such reasoning suggests that rates of PTSS in parents of children with DSD may well be higher than reported here. Avoidance behavior features prominently among PTSS. In addition, the largest cohort from which we invited participants, i.e., the CAH Support Group UK, had also been invited to participate in at least two other large scale studies being conducted in the UK at that time. In one study, many of the families were asked to return repeatedly over a 3 year period. It is very likely that many families were unable to make further efforts to participate in research. Sample size notwithstanding, our findings were consistent with reports in other domains, such as childhood cancer, and this lends credibility to the findings. Furthermore, reports of case management and outcome are meaningful whatever the sample size when the disease process in question is rare as is the case with DSD.

Implications

The data reported here are important for understanding factors which contribute to negative outcomes for parents, and potentially entire families, of children diagnosed with DSD. While specialists have suspected that the delivery of information to patients and families is important, until now there have only been general suggestions as to protocols. Though these protocols often call for the inclusion of psychological input, patient leaflets, and user-friendly websites, our findings suggest that additional education of all members of the multidisciplinary team (MDT) at the outset may be also beneficial in order to reduce the amount of confusing information being given to parents.

Even more specifically, however, these data suggest that we have the opportunity to employ and intervention which may alleviate distress in parents and perhaps change the course of development for the child and family. Evidence suggests that giving tailored information about diagnosis/prognosis has stress-reductive effects (Kitamura, 2005). By specifically assessing potentially traumatic events in the case of DSD, as in well-established protocols for managing childhood cancer (Kazak, 2006; Kazak et al., 2006, 2007), we may intervene at the juncture between objective events and perceived events for the parents. While we recognize that post-traumatic stress experienced by parents of children with a DSD is but one of the psychosocial challenges they face, we hope to clarify a blueprint for an incremental change. Indeed, the role of the psychological support system within the context of the MDT needs to be more clearly devised and to include a risk assessment (for trauma) in addition to the recommended information giving sessions (Cohen-Kettenis, 2010). Our findings suggest that the latter may avert further complications in the unfolding of events from disclosure, to treatment protocols, to follow-up and after care.

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