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Parent-Rated Severity of Illness and Anxiety Among Caregivers of Children Born with a DSD Including Ambiguous Genitalia

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Abstract

Background/Aims: Parents of children born with a disorder of sex development (DSD) often experience anxiety, but risk factors, including parental perception of the severity of their child's DSD, have not been examined. We hypothesized that severity of illness (SOI) ratings would relate to parental anxiety, and would be higher for parents of children with a potentially life-threatening DSD (e.g., 21-hydroxylase deficiency).

Methods: Eighty-nine parents (M_{age} =33.0, 56.2% mothers) of 51 children ($M_{age\ in\ months}$=8.7) with a DSD including ambiguous genitalia were recruited from 12 specialized DSD clinics.

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Parents completed questionnaires prior to genitoplasty, 6-months post-genitoplasty, and 12-months post-genitoplasty (if completed). Data were analyzed with linear mixed modeling.

Results: Parental anxiety decreased over time, χ^2 (1)=10.14, p<.01. A positive relationship between SOI and anxiety was found, with SOI being a strong predictor of anxiety (b = 0.53, p<.01; χ^2 (1) = 5.33, p < .05). An SOI by time interaction indicated SOI had an increasing effect on anxiety over time, b=0.06, p<.05; χ^2 (1) = 6.30, p < .05. There was no diagnosis by SOI interaction.

Conclusion: Parental anxiety decreased over time, but those with higher SOI ratings reported greater initial anxiety followed by slower resolution over time. Underlying etiology of DSD had no effect on the relationship between SOI and anxiety.

Keywords

Anxiety; Disorders of Sex Development; Psychological aspects of disorders; Severity of illness

Disorders of sex development (DSD) are a group of congenital conditions that result in an array of medical and psychosocial consequences.[1] Some of these conditions are characterized by ambiguous genitalia at birth, thus requiring caregivers to make important decisions regarding whether or not to proceed with genital surgery (i.e., genitoplasty), when to proceed with surgery (if at all) and how to assign a sex of rearing for their child.[2] Some, but not all, types of DSD are also potentially life-threatening if not treated medically, such as congenital adrenal hyperplasia (CAH) due to 21-hydroxylase deficiency. A significant subset of caregivers of children born with DSD including genital ambiguity report elevated symptoms of anxiety, depression, and posttraumatic stress, as well as reduced quality of life.[3] Few risk factors for parental distress among caregivers of children with DSD have been identified, but current evidence suggests that parenting factors, such as parental overprotection and increased parenting stress contribute to caregiver anxiety, whereas degree of genital ambiguity may be unrelated.[4,5] However, it is unclear if other condition-specific factors, such as the potentially life-threatening status of the underlying condition, contributes to distress for parents. The current study aimed to examine how parent perceptions of the severity of their child's condition, independent of degree of genital ambiguity, influences parents' anxiety over time. This factor was anticipated to be particularly relevant for parents of females with CAH due to 21-hydroxylase deficiency.

Importantly, models of stress and coping suggest that both objective medical condition characteristics, as well as parental perceptions of these same conditions impact adjustment outcomes.[6] Currently, there is limited understanding of how perceived illness severity relates to parental distress in families of children born with a DSD. However, the literature on other pediatric chronic illnesses shows that parent ratings of child health and vulnerability impact caregiver distress above and beyond objective physician ratings of illness severity.[7–9]

Thus, we examined parental anxiety as it related to parent-rated severity of illness (SOI) of their child over the course of the first 12 months following their child being recruited into a prospective study of DSD outcomes. Three specific hypotheses were tested: 1) parental anxiety will decrease over time, 2) parental ratings of SOI will predict parental anxiety over

time, such that higher baseline severity ratings will be associated with persisting anxiety, and 3) parents of children with CAH due to 21-hydroxylase deficiency will report higher ratings of SOI than parents of those with other types of DSD that are not life threatening.

Methods

Participants and Procedures

Eligible participants included families with children aged ≤ 2 years with moderate to severe atypical genitalia as defined by: a Prader[10] 3–5 rating in a 46,XX child, or a Quigley[11] 3–6 rating in a child with a 46,XY, or a 45,X/46,XY chromosomal complement. Patients with prior gentioplasty or non-urogenital congenital malformations were excluded. Parents were recruited from 12 DSD clinics from across the U.S. and were approached and consented during regularly scheduled clinic appointments. Parents completed a demographics form and a battery of questionnaires as part of a larger prospective study assessing medical and psychosocial outcomes in families impacted by DSD. As such, part of this dataset has been previously reported.[3,5,12] At the initial report, an 86.4% enrollment rate of all eligible participants across sites was obtained, and the current attrition rate is 19.00%.[3] For this study, parents returned questionnaires in-person or via mail at three time points: baseline (prior to genitoplasty, if genitoplasty was chosen), 6-month follow-up after genitoplasty if performed, or 6 months after baseline if no surgery was pursued, and 12-month follow-up. When 2 parents of a child completed the surveys, they did so independently of one another. Families were compensated for travel costs for study visits. The study was approved by the Institutional Review Boards at all participating sites.

Measures and Forms

Demographics Form and Chart Review.—The demographics form was completed by parents and included: child gender of rearing, child age and date of birth, diagnosis, parent relation to child, parental marital status, race/ethnicity, parent age, and household income. The medical chart review also yielded information regarding karyotype and diagnosis.

Severity of Illness Scale (SOIS).—The Severity of Illness Scale is a 6-item Likert scale assessing the severity of the child's condition, including items that measure impairment in functioning and required hospital visits.[13] Responses ranged from 1–7 with response options dependent on the item content (e.g., "independent functioning, requires no assistance" to "requires complete assistance"; "likely to improve" to "likely to worsen"). Items are summed to create a single total score, with higher scores indicating greater SOI. Of note, the measure was originally designed as an objective rating of severity to be completed by physicians. For the purpose of this study, the questionnaire was completed by caregivers to determine the parents' perception of the severity of their child's condition. Thus, after review of the developmental appropriateness and skewness of item responses, two items (e.g., "Describe the degree of impairment for this child" and "How much does this child participate in age appropriate activities") were dropped from the measure for all analyses. Due to this novel use of the SOIS, the current study is exploratory in nature and use of this version of the SOIS will require confirmation with additional investigations. The measure

demonstrated good internal consistency (α = .80) and excellent test-retest reliability (α = .92).

Beck Anxiety Index (BAI).—The Beck Anxiety Index is a 21-item, self-report measure of anxiety symptoms, including both physiological sensations (e.g., numbness, heart pounding) and cognitive symptoms (e.g., fear of dying, fear of losing control).[14] The BAI is widely used, with standardized cut-offs evaluating anxiety among adults in a variety of contexts.[14] The measure demonstrated high internal consistency (α = .92) and good test-retest reliability (α = .75).

Overview of Statistical Analyses

First, descriptive analyses and bivariate correlations were conducted. Following Thompson and Gustafson's[6] transactional model of stress and coping, illness and demographic variables, including child gender of rearing, child age, parent sex, parent age, parent race, and family income, were controlled for in all other analyses analyses.

Next, multi-level modeling (MLM) was used to test the hypotheses, since MLM accounts for similarities among ratings from the same parent, or from parents within the same family, who have shared experiences with their child.[15] The models were tested with Stata Version 11.2 using maximum likelihood estimation. Intraindividual differences were modeled at Level 1, between-parent differences were modeled at Level 2, and between-family differences were modeled at Level 3. Likelihood-ratio tests after estimation were used to determine if more complex models improved fit over simpler models. The statistical significance of the variance explained by the fixed and random effects of each additional predictor was also evaluated.

An unconditional model tested the variance of BAI scores without any predictor variables or controls. Next, a conditional model including the control variables was tested, and was subsequently used as the simpler comparison model. Time was then added as a predictor in a fixed effects model and random effects were tested. Baseline SOI rating was then added to the fixed effects model as a predictor, and random effects were tested. Finally, interactions between SOI rating and both time and DSD diagnosis (i.e., CAH or other), were tested.

Results

Demographics

Participants were 89 parents (M_{age} = 33.01, SD = 6.52; 56.2% mother) of 51 children with a DSD including ambiguous genitalia at birth (M_{age} = 8.70 months, SD = 6.34 months; 57.3% female). Parent participants were primarily Caucasian and affluent. The sample consisted of 24 children with CAH due to 21-hydroxylase deficiency, who were all reared female, and 27 children with another DSD condition not considered to be life-threatening, such as androgen insensitivity syndrome or 5-alpha reductase deficiency. Six children in the current sample did not undergo genitoplasty and reasons for this choice were not documented. Demographic information is presented in Table 1.

Preliminary Analyses

Parents reported an average SOI rating of 10.48 (SD = 3.46) and an average anxiety score of 7.43 (SD = 8.31) at baseline, with 14.60% of parents reporting moderate to severe anxiety symptoms above the clinical cut-off (Table 2.). At the 6-month follow-up 13.56% of parents were above the clinical cut-off for anxiety symptoms, and 10.42% of parents remained above the cut-off at 12-months. Mother's (M = 9.34, SD = 8.59) reported higher anxiety than fathers (M = 4.97, SD = 7.34) at baseline ($F(87) = 5.79$, $p < 0.05$), but there was no difference between mother (M = 10.72, SD = 3.42) and father (M = 10.18, SD = 3.55) ratings of SOI at baseline ($p > 0.10$), or between mother and father reports of anxiety at 6-months or 12-months ($ps > 0.10$). As only six families in the current sample elected not to have genitoplasty, the small sample size precluded statistical comparison. However, this subset of parents are characterized by an average anxiety score of 9.00 (SD = 14.57) and average SOI rating of 12.33 (SD = 3.98) at baseline, with one parent reporting moderate to severe anxiety symptoms.

Bivariate correlations were conducted, and significant relationships were found between BAI scores across time and baseline SOI rating, as well as between these two primary variables and several covariates. Parent relationship to the child (i.e., mother or father), parent age, child gender of rearing, child age, parent race, and family income, were identified as covariates to be included in subsequent models. The correlation matrix is included in Table 3.

The unconditional model provided the average BAI score across all participants (7.02). This unconditional model was used as a reference to test the variance accounted for by the addition of each predictor. Two Intraclass Correlation Coefficient (ICC) values were obtained, due to the three-level structure of our model. The first is the proportion of variance due to family differences (ICC = 0.26) and the second is the proportion of variance due to parent differences (ICC = 0.63). Since the proportion of variance due to parents differences was larger than that due to family differences, a three-level model was warranted.

Demographic and Illness Covariates

In the conditional model, when accounting for clustering (i.e., differences between mothers and fathers, differences between families), child gender of rearing ($p = 0.68$), child age ($p = 0.64$), parent race ($p = 0.12$), and family income ($p = 0.16$) were not significantly associated with BAI scores. However, parent age did have a significant effect ($b = -0.38$, $p < .05$), such that older parents had lower anxiety. The overall model including the controls predicted a significant proportion of variance in BAI scores, above the unconditional model, $\chi^2(1) = 18.55$, $p < .01$. Thus, the illness and demographic covariates were retained in all subsequent models.

Trajectories of BAI Across Time

A significant negative relationship was found between time and BAI scores ($b = -0.55$, SE = 0.17, $p < .01$), such that on average, BAI scores decreased over time. A likelihood-ratio test after estimation indicated that time accounted for a significant proportion of variance in BAI scores, above that accounted for by the controls only, $\chi^2(1) = 10.14$, $p < .01$. In this model,

parent age (b = −0.41, SE = 0.15) and child age (b = 0.24, SE = 0.10) were significant covariates. The random effects of time between parents ($\chi^2(1)$ = −0.00, p > 0.10) and families ($\chi^2(1)$ = −0.00, p > 0.10) were not significant. This model suggests that parent anxiety decreases over time.

Effect of SOI on BAI Across Time

A significant positive relationship between SOI scores and BAI scores was found, b = 0.53, SE = 0.23, p < .01. This suggests that at baseline, a parent with a one point higher SOI score will have a 0.53 point higher BAI rating, as compared to a parent with a one point lower SOI rating. Time (b = −0.55, SE = 0.16), parent age (b = −0.42, SE = 0.15), and child age (b =0.25, SE = 0.10) remained significant predictors in this model. A likelihood-ratio test after estimation indicated that SOI scores accounted for a significant proportion of variance above the model with only time and covariates as predictors, $\chi^2(1)$ = 5.33, p < .05. There was no random effect of SOI between parents ($\chi^2(1)$ = 0.15, p > 0.10) or between families ($\chi^2(1)$ = 0.18, p >0.10). This model suggests that parents who perceive greater severity report greater anxiety.

An interaction between time and SOI score suggested that the effect of SOI rating on anxiety is moderated by time. The interaction between time and SOI rating (b = 0.06, SE = 0.02) significantly improved the amount of variance explained in BAI ($\chi^2(1)$ = 6.30, p < .05). This indicates that a higher SOI rating at diagnosis relates to a slower decrease in anxiety over time, as compared to a lower baseline SOI rating. Finally, an interaction between SOI rating and diagnosis was tested, and did not significantly improve the amount of variance explained in BAI ($\chi^2(1)$ = L20, p > 0 .10). This indicates that the effect of SOI rating on anxiety symptoms did not differ between parents of children with CAH and parents of children with other DSD diagnoses.

Discussion

The present study examined parent ratings of the severity of their child's illness and their anxiety symptoms over a period of time following the child's recruitment into a prospective study of DSD outcomes. Importantly, the current study found that parental perception of illness severity at baseline significantly predicted their anxiety symptoms across time. The relationship between perceived severity and anxiety symptoms was also moderated by time, such that those parents with higher SOI ratings at baseline had a slower rate of improvement in anxiety symptoms. Overall, a significant subset of parents (approximately 14.6%) experienced moderate to severe levels of anxiety symptoms at baseline. On average, parent-report of anxiety significantly decreased from baseline to the 12-month follow-up for parents who opted for genital surgery for their child, but it decreased slower for those parents with higher SOI ratings. However, our hypothesis that this relationship would be stronger for parents of children with a life-threatening DSD such as CAH due to 21-hydroxylase deficiency, and that these parents would report greater SOI, was not supported.

Across conditions including asthma and inflammatory bowel disease, increasing illness severity, as rated by physicians, has been associated with greater parental depressive and anxious symptoms, but some reports demonstrate that objective illness severity is unrelated

to parenting stress.[16–18] Thus, the present study is important as it aligns with research in other medical conditions suggesting that parents' *subjective appraisal* of the severity of their child's illness may have a greater impact on parent adjustment than objective measures.[7–9] Additionally, the importance of examining parental perception of SOI is further supported by the well-established literature suggesting that overprotective parenting practices, a possible response to perceived severity of illness, are associated with negative child outcomes.[19]

These findings are also consistent with previous DSD research, such that a minority of parents continue to experience elevated anxiety over time, while most parents report a reduction in symptoms.[12] The results from our analyses reveal a more comprehensive understanding of the trajectory of anxiety symptoms and demonstrates the lasting impact that initial perception of illness severity can have on adjustment outcomes.

Interestingly, the current study diverged from past reports indicating that parental adjustment outcomes differ by the child's sex of rearing.[5] In the present study, parents rated children with CAH versus those with other DSD diagnoses as having equally severe conditions, despite only those with CAH having a life-threatening condition. Since all children in this sample with CAH were reared female, the lack of a significant interaction indicates that the relationship between perceived severity and parental anxiety does not differ by child sex of rearing or diagnosis. Thus, parent-rated severity, rather than diagnosis, accounts for risk for elevated anxiety symptoms across time.

This study also suggests the need for early intervention for the parents who experience emotional difficulties across the first year following their child's DSD diagnosis. The benefit of interventions aimed at providing parents with education about DSD and cognitive-behavioral techniques to help restructure negative views and to cope with the stressors associated with having a child with a complex medical condition could be tested. Additionally, our findings demonstrate that the Severity of Illness Scale is a useful screening tool for identifying those parents who are at-risk for challenges in adjustment. Further, moderating and mediating factors that impact the relationship between parental perception of illness severity and adjustment outcomes, such as illness uncertainty, should also be evaluated.[12,20] Perceptions of uncertainty may interact with perceived illness severity, potentially resulting in increased and lasting anxiety symptoms. Thus, the inclusion of additional mechanisms in future analyses may better explain the current findings and provide greater information for the design of interventions.

Although the current study allows for generalizability and temporal interpretations that were previously hindered by the mainly retrospective and cross-sectional literature, the findings should be interpreted in light of a few limitations. First, the study relied solely on self-report measures, which raises concerns regarding response bias and shared method variance. Multi-modal assessment of anxiety symptoms and appraisals should be used in future studies. For instance, due to imperfect measurement of SOI perceptions, qualitative interviews may be valuable. As our sample was collected from specialized DSD clinics, often necessitating that families have the resources to locate and travel to the clinic, most of the parents report relatively high family incomes and are Caucasian, suggesting there may be a potential

sampling bias. This could limit the generalizability of our findings to families of different racial and ethnic backgrounds or socioeconomic statuses. Further, due to the small proportion of our population that declined surgery, we were unable to compare differences among those families who did or did not choose surgery. Future studies could investigate these differences, particularly as they may relate to the effect of surgical or non-surgical cosmesis outcomes on parental anxiety. Finally, physician ratings of illness severity and other objective measures of the child's DSD were not collected, but should be controlled for in future analyses. Despite these limitations, the present study identifies parents' initial perception of the severity of their child's DSD as a critical risk factor for maladaptive outcomes.

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Table 1.

Demographics

	Child (n = 51)	Caregiver (n = 89)
Ethnicity		
Caucasian		69.7% (n = 62)
African American		6.7% (n = 6)
Asian/Pacific Islander		5.6% (n = 5)
Multi-Racial		4.5% (n = 4)
Other		6.7% (n = 6)
Not Reported		6.7% (n = 6)
Hispanic		21.4% (n= 19)
Age - Mean (SD)	8.7 (6.3) *	33.0(6.5)
Sex		
Female	56.9% (n = 29)	56.0%
Male	39.2% (n = 20)	43.8%
Unsure	3.9% (n = 2)	
Karyotype		
46,XY	33.3% (n= 17)	
46,XX	51.0% (n = 26)	
45,X/46,XY	5.9% (n = 3)	
Other	9.8% (n = 5)	
DSD Diagnosis		
CAH	47.1% (n = 24)	
Other (e.g., 5-alpha Reductase Deficiency, Androgen Insensitivty Syndrome)	52.9% (n = 27)	
Received Genitoplasty	88.2% (n = 45)	
Family Income		
0–14,999		6.7%
15,000–29,999		19.1%
30,000–49,999		14.6%
50,000–69,999		1.1%
70,000–89,999		9.0%
90,000–99,999		9.0%
100,000+		40.4%

* Note: Age for child is in months.

Table 2.

Descriptives for BAI and SOIS

		N	M	SD	% Above clinical cut-off*
BAI					
	Baseline	89	7.43	8.31	14.60%
	6-Month	59	6.66	9.81	13.56%
	12-Month	48	4.46	7.02	10.42%
SOIS					
	Baseline	89	10.48	3.46	

*Note: Clinical cut-off for the BAI refers to the Moderate to Severe range of symptomology.

Table 3.

Bivariate correlations

Variables	1	2
1. BAI	-	
2. SOIS	0.22**	-
3. Child Sex	-0.12	0.01
4. Child Age	-0.05	−0.02
5. Parent relation to child	−0.20**	−0.17*
6. Parent Race	−0.10	0.03
7. Parent Age	−0.32***	0.02
9. Family Income	−0.28***	−0.02

Note.

* $p < .05$,

** $p < .01$,

*** $p < .001$.

Note. Time point of report, clustering effects, and suppression effects are not accounted for in this matrix.

Note. Variable coding includes: Child Sex, Female = 0, Parent Relation to Child, Mother = 0, Parent Race, Caucasian = 0, African American = 1, Asian/Pacific Islander = 2, Multi-Racial = 3