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## Predictors of Psychosocial Distress in Parents of Young Children with Disorders of Sex Development

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## Abstract

**Purpose:** To evaluate demographic, financial, and support predictors of distress for parents of young children with Disorders of Sex Development (DSD) including atypical genital development, and to characterize early parental experiences. This extends our previous findings to identify those parents at risk for distress.

**Materials and Methods:** Participants included mothers (n = 76) and fathers (n = 63) of a child (n = 78) diagnosed with DSD characterized by moderate to severe genital atypia. Parents

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completed a demographic questionnaire, measures of anxious and depressive symptoms, quality of life, illness uncertainty, and posttraumatic stress symptoms, and rated their satisfaction with the appearance of their child's genitalia.

**Results:** Caregivers' depressive and posttraumatic stress symptoms were comparable to standardized norms while anxious symptoms were below norms. A subset of parents reported clinically elevated symptoms. Twenty-six percent of parents endorsed anxious symptoms, 24% endorsed depressive symptoms, and 17% reported posttraumatic stress symptoms. Levels of illness uncertainty were lower when compared to parents of children with other chronic illnesses. Differences by parent sex emerged, with mothers endorsing greater distress. Lower income, increased medical care and travel expenses, and having no other children were related to increased psychosocial distress.

**Conclusions:** Early psychosocial screening is recommended for parents of children with DSD. Clinicians should be aware that financial burden and lack of previous parenting experience are risk factors for distress.

## Keywords

Disorders of Sex Development; intersex; psychosocial; parents; psychological distress

Disorders of sex development (DSD) are a set of congenital medical conditions that are collectively rare, with combined prevalence rates ranging from 1:100 to 1:5,000,<sup>1–3</sup> while rates of individuals with secondary atypical genitalia are approximately 1:5000.<sup>4</sup> Compared to other rare conditions, the psychosocial aspects associated with DSD, including parental outcomes, remain understudied.<sup>1</sup> It is known that some parents are at risk for experiencing psychosocial distress after their child's DSD diagnosis, including symptoms of depression, anxiety, and posttraumatic stress.<sup>5–7</sup> Uncertainty about gender assignment, barriers to understanding complex medical information, and worries about stigma pose unique challenges for parents.<sup>8,9</sup> Psychosocial functioning is differentially affected by parent sex, with mothers experiencing greater anxious and depressive symptoms than fathers.<sup>7,10</sup> Additionally, parents report worse health-related quality of life for boys than girls on proxy measures of child functioning.<sup>8</sup>

Though data on parental and family adjustment to having a child with DSD are emerging, interpretation is limited due to retrospective study design, small sample sizes, and failure to include fathers. The aim of this paper is to characterize parent psychosocial distress and evaluate early predictors of distress for mothers and fathers of children with DSD including atypical genital development who are participating in a prospective multisite study. Such information is crucial as parents are expected to participate in shared decision-making with their child's healthcare team on how best to treat genital atypia.<sup>11</sup> This report expands our preliminary findings that a significant minority of parents (16% to 25%) reported anxious and depressive symptoms, lower quality of life, and symptoms of illness uncertainty and posttraumatic stress.<sup>12</sup> Given the study's exploratory nature, specific hypotheses are not set forth. However, because financial burden,<sup>12</sup> support,<sup>13</sup> and family variables (e.g., previous parenting experience<sup>13</sup>) have been linked to distress, we expect to identify relationships with these variables.

## Methods

#### **Participants**

Participants included mothers (n = 76), and fathers (n = 63) of a child (n = 78) diagnosed with DSD including moderate to severe genital atypia. A subset (8 mothers, 12 fathers) only completed demographic questionnaires and cosmetic ratings, electing not to complete measures of psychosocial functioning, leaving 68 mothers and 51 fathers. There was no significant difference between this subset of 20 parents and the rest of the sample. Parents were recruited at 12 DSD clinics across the US within two years of the child's birth ( $M_{Age} = 9.36$  months, SD = 6.94) and prior to genitoplasty (if the child underwent surgery) between September 2013 and November 2017. Families were ineligible if the child had other organ system complications. Families were invited to participate regardless of the medical and/or surgical treatments they opted for their child (non-surgery =7 children; 9%). A Prader rating of 3-5 in children with 46,XX DSD or a Quigley rating 3-6 in children with 46,XY DSD or 45,XO/46,XY sex chromosome DSD, was used to classify the degree of atypical genital development at enrollment. For further child information, see our previous report concerning diagnoses, phenotype, and sex of rearing.<sup>14</sup>

## Materials

A demographic questionnaire included child's age, sex of rearing, type of DSD, and parent age, sex, marital status, race/ethnicity, education, employment status, number of other children, and income.

The *Cosmetic Appearance Rating Scale* is a measure of satisfaction with the appearance of the child's genitals at study enrollment. It utilizes a single item 4-point Likert-scale from *"good"* to *"very dissatisfied"*. Parents and physicians completed this scale.

The *Beck Anxiety Inventory* (*BAI*)<sup>15</sup> is a 21-item, multiple-choice self-report measure of anxious symptoms. Item scores range from 0 to 3 (higher scores represent greater anxiety). Normed total scores of 0-7 represent minimal, 8-15 mild, 16-25 moderate, and 26-63 severe anxiety, with scores of 14 or greater deemed clinically significant.<sup>16</sup> Internal consistency in the current sample was excellent (a = .92).

The *Beck Depression Inventory-II (BDI-II)*<sup>17</sup> is a 21-item, multiple-choice self-report questionnaire assessing depressive symptoms. Item ratings range from 0 to 3 (higher scores represent greater depressive symptoms). Normed total scores of 0-13 indicate minimal, 14-19 mild, 20-28 moderate, and 29-63 severe depression. Scores of 14 or greater are clinically significant.<sup>18</sup> Cronbach alpha for the current sample was good (a = .89).

The *SF-36*<sup>19</sup> is a 36-item self-report measure of health-related quality of life (QoL). Item responses are provided on a variable Likert-scale (higher scores indicate better health-related QoL). Two composite scores can be calculated (i.e., physical and mental health). Mental health scores <42 are clinically significant. Cronbach alpha for the current sample was excellent (a = .92).

The *Parent Perception of Uncertainty Scale (PPUS)*<sup>20</sup> is a 31-item, parent self-report scale assessing uncertainty (e.g., ambiguity, unpredictability) concerning their child's DSD (e.g., "It is unclear how bad my child's physical discomfort will be"). Responses are provided on a 5-point Likert-scale (higher scores indicate greater uncertainty). Parents of children with other illnesses have reported mean scores of 70.3 or greater.<sup>21</sup> The Cronbach alpha for the current sample was excellent (a = .91).

The *Impact of Events Scale (IES-R)*<sup>22</sup> is a 22-item, self-report measure of posttraumatic stress symptoms (PTSS). Parents are instructed to consider their child's illness when responding to items. Responses are provided on a 5-point Likert-scale (higher scores indicate greater PTSS). A score 33 is considered clinically significant. Internal consistency for the current sample was excellent ( $\alpha = .94$ ).

#### Procedures

Approval was obtained from each site's institutional review board. Families were consented and enrolled at scheduled clinic visits. Participants were compensated \$50 for their participation.

## **Statistical Analysis**

Statistical analyses were conducted in SPSS 24. Mean scores for each measure were compared to standardized norms (BAI and BDI-II nonclinical norms, PPUS and IES-R cancer parent norms). Bivariate correlations were conducted between continuous demographics (parent age, child age, household income, distance to urologist) and all outcome variables. Dependent samples t-tests analyzed caregiver gender differences with each outcome. Analysis of variance evaluated whether child received a diagnosis, child sex of rearing, parent race/ethnicity, levels of support, financial burden, other children in the home, and increased expenses were related to caregiver distress levels. The above analyses included all participants; however, as the current sample included a small group of parents who opted for no surgery, exploratory descriptive analyses were conducted.

## Results

Parent demographics are presented in Table 1. A comparison of the current sample's outcomes to norms is in Table 2, and surgery versus no surgergy parent outcomes in Table 3. Parental distress symptoms were correlated (see Table 4). Outcome predictor analyses are presented in Table 5. Child sex of rearing was primarily concordant with karyotype (94%); therefore, analyses utilized child sex of rearing.

#### Parental Outcomes

The average parent rating of satisfaction with their child's genital cosmetic appearance was 2.68 (SD = .96; Table 2), with 20 (15%) rating appearance as good, 32 (23%) satisfied, 58 (42%) dissatisfied, 28 (20%) very dissatisfied. Physicians (n = 14) reported greater dissatisfaction than mothers (t(73) = -4.26, p <.001) and fathers (t(61) = -5.45, p <.001), with physicians rating 4 (5%) good, 2 (3%) satisfied, 38 (49%) dissatisfied, 32 (41%) very

dissatisfied. Physician satisfaction with cosmetic appearance did not differ for children based on sex of rearing.

Anxious symptoms in parents were significantly lower than population norms<sup>16</sup> (M= 8.19, SD= 8.88). Seventy-four (62%) parents presented with minimal, 20 (17%) with mild, 18 (15%) with moderate, 6 (5%) with severe anxious symptoms, and 1 (1%) parent had missing data. Thirty-one (26%) parents exceeded the cutoff for clinical symptoms.

The average reported depressive symptoms for parents was comparable to population norms (M = 8.93, SD = 9.50).<sup>23</sup> Eighty-four (75%) parents reported minimal, 10 (9%) reported mild, 13 (12%) moderate, 5 (4%) reported severe symptoms, and 7 (6%) had missing data. Twenty-eight (24%) parents exceeded clinical cutoff.

The average score for parental health-related QoL was 46.40 (SD = 10.97) for mental health and 55.54 (SD = 6.48) for physical health. Twenty-six (27%) parents scored at or below the clinical cutoff for diminished mental health QoL, and 23 (19%) parents had missing total scores.

Illness uncertainty was significantly lower (M = 65.21, SD = 15.90) than other childhood illnesses,<sup>21</sup> yet 45 (38%) caregivers endorsed symptoms equal to or greater than the average.

Parents reported PTSS with a mean of 17.31 (SD = 16.42), which is comparable to standardized norms.<sup>24</sup> Twenty (17%) scored at or above the clinically significant range, with 2 (2%) parents missing data.

#### **Exploratory Analyses of Non-Surgery Group**

Seven children did not receive early genital surgery (see Table 1 for demographics). While this small sample is underpowered to perform statistical comparisons (7 mothers and 4 fathers), the following are surgery and non-surgery means respectively (see Table 3 for means and standard deviations): cosmesis (2.75 and 1.91), anxious (8.19 and 8.10), depressive (8.88 and 9.44), uncertainty (65.22 and 65.10), and PTSS (17.45 and 15.80).

#### **Predictor Analyses**

**Family predictors.**—Forty-seven (60%) children were being reared as a girl, 27 (35%) as a boy, and two (3%) children had no assigned sex of rearing per parent report. Nineteen children (24%) had no identifiable etiology underlying their DSD. Forty-three (55%) mothers and 34 (44%) fathers had other children. Mothers rated greater dissatisfaction with cosmetic appearance of their child's genitals than fathers (t(60) = 2.12, p = .039). Mothers also reported more anxious symptoms (t(48) = 2.94, p = .005), depressive symptoms (t(46) = 3.85, p < .001), PTSS (t(48) = 4.33, p < .001), and worse mental health QoL (t(38) = -3.63, p = .001). No difference was identified between mothers' and fathers' physical health (t(38) = -.036, p = .971) or illness uncertainty (t(45) = .042, p = .967). Older fathers reported lower levels of anxious symptoms (p = .005). No significant difference was found based on child sex of rearing. Fathers reported greater PTSS with an unknown diagnosis (t(1,46) = 7.31, p = .010). Mothers reported more dissatisfaction with the appearance of their child's genitalia (t(1,73) = 6.86, p = .011), higher levels of anxious symptoms (t(1,65) = 4.18, p = .045), and

greater illness uncertainty (F(1,62) = 5.36, p = .024) if they had no other children. Driving distance to the DSD clinic was not related to distress or uncertainty.

**Financial predictors.**—Twenty-eight percent of parents reported household income below \$20,000 (19% of families). Due to their child's diagnosis, 34% of parents reported increased mental health expenses, 37% increased medical expenses, and 58% increased travel expenses. For fathers, higher household income was related to lower anxious symptoms (p = .002), lower depressive symptoms (p = .003), lower PTSS (p = .018), and better physical health-related QoL (p = .024). Household income was not related to mothers' distress. However, mothers reporting financial concerns endorsed significantly greater depressive symptoms (R(2,59) = 6.28, p = .003) and lower mental health-related QoL (R(2,50) = 6.30, p = .004) than those with no financial concerns. For mothers, greater medical expense were associated with greater dissatisfaction with their child's genital appearance (R(1,73) = 4.26, p = .042). Mothers with increased travel expenses reported greater depressive symptoms (R(1,62) = 6.97, p = .010) and worse mental health-related QoL (R(1,51) = 7.06, p = .011). Whereas, fathers with increased travel expenses endorsed higher PTSS (R(1,48) = 4.33, p = .043). Mental health expenses were not related to levels of distress.

**Support predictors.**—The most helpful support parents received since their child's diagnosis was emotional support (35%), help caring for their child (29%), and financial support (6%). Mothers who reported help with expenses reported better mental health-related QoL (F(1,51) = 5.27, p = .026). No other levels of support were related to parental outcomes.

## Discussion

The current study evaluated predictors of psychosocial distress in parents of young children with moderate to severe genital atypia due to DSD and includes both fathers and mothers, as well as parents who opted for early genitoplasty for their affected child and those who did not.

As a group, caregivers reported symptoms comparable to norms (i.e., depressive symptoms, PTSS) or below norms (i.e., anxious symptoms and illness uncertainty).<sup>16,21,23,24</sup> However, a subset reported anxious, depressive, and posttraumatic stress symptoms in the clinical range. Moreover, parents with high symptoms of distress in one domain were likely to experience distress in another (e.g., greater anxious and depressive symptoms). It is not known to what extent parents received mental health care, or if the lower distress scores are representative of psychosocial interventions throughout their child's care. However, parents experiencing greater distress received care for their child across sites.

Overall, mothers are at greater risk of distress than fathers, which is consistent with parents of children with cancer (another complex illness) shortly after diagnosis.<sup>13</sup> Mothers also endorsed greater dissatisfaction with their child's genital appearance than fathers, and physicians rated even greater dissatisfaction. It is unknown if physician opinion influenced parents' satisfaction, distress, or decision making. Although parent sex predicted distress,

child sex of rearing did not, which differs from previous studies with smaller sample sizes.<sup>10</sup> Mothers with no other children were at greater risk of distress. The extant literature on postpartum depression and anxiety has conflicting results as to whether distress is higher in first-time mothers or experienced mothers.<sup>25</sup> It is possible that some anxious symptoms are due to postpartum anxiety, which occurs in about 8.5% of mothers.<sup>26</sup> Further examination of how the child's DSD impacts new mothers mental health is certainly needed.

Financial burden predicted both maternal and paternal distress. Parent financial concerns should be evaluated, in addition to income, for families attending DSD clinics, as parents with higher income also experienced distress. Although referral to specialty DSD clinics is recommended by professional societies,<sup>27,28</sup> due to expert multipledisciplinary support, it is not known whether travel and financial constraints is assessed as a risk factor.

## Limitations and Strengths

The current study included an ethnically homogenous sample of caregivers with high incomes and high levels of formal education. Therefore, these results may not generalize to all parents, and future studies should evaluate whether distress differs in parents with other incomes and educational backgrounds. Furthermore, the extent of mental health services provided prior to the birth of their child or after, is not known for this sample of parents. Future studies should evaluate the benefits of DSD-specific psychosocial interventions, as well as risk or protective factors for parents already receiving mental health services. Lastly, the current study included a small sample of parents who opted for no surgery for their child. Although the current results indicate lower (better) cosmesis ratings in the non-surgery group than the surgery group, the small sample size limits speculation as to whether this is a clinically meaningful difference. Future studies of families electing no surgery are needed as biomedical policies evolve concerning early genital surgery in children with DSD.<sup>29</sup>

## Conclusion

Overall, parent psychosocial functioning is similar to the general population; however, a subset report clinical anxious and depressive symptoms, PTSS, and impaired QoL. Demographics are associated with distress in ways previously undescribed. Knowledge of parent mental health status, including predictors of parent distress, is needed to assist caregivers as they participate in decisions concerning complex treatment plans for their children. Therefore, interdisciplinary care including psychological screening<sup>1,30</sup> of parents, with consideration of financial resources and previous parenting experience, is recommended.

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## References

- Sandberg DE, Gardner M, Callens N, et al. Interdisciplinary care in disorders/differences of sex development (DSD): The psychosocial component of the DSD – Translational Research Network. Am J Med Genet Part C Semin Med Genet. 2017;175:279. [PubMed: 28574671]
- 2. Thyen U, Lanz K, Holterhus PM, et al. Epidemiology and initial management of ambiguous genitalia at birth in Germany. Horm Res. 2006;66:195. [PubMed: 16877870]
- Sandberg DE, Pasterski V, Callens N. Introduction to the special section: Disorders of Sex Development. J Pediatr Psychol. 2017;42:487. [PubMed: 28499017]
- 4. Sax L How common is intersex? A response to Anne Fausto-Sterling. J Sex Res. 2002;39:174. [PubMed: 12476264]
- Kirk KD, Fedele DA, Wolfe-Christensen C, et al. Parenting characteristics of female caregivers of children affected by chronic endocrine conditions: A comparison between disorders of sex development and type 1 diabetes mellitus. J Pediatr Nurs. 2011;26:e29. [PubMed: 21911316]
- Pasterski V, Mastroyannopoulou K, Wright D, et al. Predictors of posttraumatic stress in parents of children diagnosed with a disorder of sex development. Arch Sex Behav. 2014;43:369. [PubMed: 24085468]
- Wolfe-Christensen C, Fedele DA, Mullins LL, et al. Differences in anxiety and depression between male and female caregivers of children with a disorder of sex development. J Pediatr Endocrinol Metab. 2014;27:617. [PubMed: 24859514]
- Alpern AN, Gardner M, Kogan B, et al. Development of health-related quality of life instruments for young children with Disorders of Sex Development (DSD) and their parents. J Pediatr Psychol. 2017;42:544. [PubMed: 27026663]
- 9. Rolston AM, Gardner M, Vilain E, et al. Parental reports of stigma associated with child's disorder of sex development. Int J Endocrinol. 2015;2015:1.
- Wolfe-Christensen C, Wisniewski AB, Mullins AJ, et al. Changes in levels of parental distress after their child with atypical genitalia undergoes genitoplasty. J Pediatr Urol. 2017;13:32. [PubMed: 28041823]
- Speiser PW, Arlt W, Auchus RJ, et al. Congenital Adrenal Hyperplasia due to Steroid 21-Hydroxylase deficiency: An Endocrine Society clinical practice guideline. J Clin Endocrinol Metab. 2018;103:1. [PubMed: 29099927]
- Suorsa KI, Mullins AJ, Tackett AP, et al. Characterizing early psychosocial functioning of parents of children with moderate to severe genital ambiguity due to disorders of sex development. J Urol. 2015;194:1737. [PubMed: 26196734]
- Sultan S, Leclair T, Rondeau E, et al. A systematic review on factors and consequences of parental distress as related to childhood cancer. Eur J Cancer Care (Engl). 2016;25:616. [PubMed: 26354003]
- Finlayson C, Rosoklija I, Aston CE, et al. Baseline characteristics of infants with atypical genital development: Phenotypes, diagnoses, and sex of rearing. J Endocr Soc. 2019;3:264. [PubMed: 30623164]
- Beck AT, Epstein N, Brown G, et al. An inventory for measuring clinical anxiety: Psychometric properties. J Consult Clin Psychol. 1988;56:893. [PubMed: 3204199]
- 16. Bardhoshi G, Duncan K, Erford BT. Psychometric meta-analysis of the English version of the Beck Anxiety Inventory. J Couns Dev. 2016;94:356.
- 17. Beck AT, Steer RA, Brown GK. Manual for the Beck Depression Inventory-II. San Antonio, TX: Psychological Corporation; 1996.
- Viinamäki H, Tanskanen A, Honkalampi K, et al. Is the Beck Depression Inventory suitable for screening major depression in different phases of the disease? Nord J Psychiatry. 2004;58:49. [PubMed: 14985154]
- Bowling A, Bond M, Jenkinson C, et al. Short form 36 (SF-36) health survey questionnaire: Which normative data should be used? Comparisons between the norms provided by the Omnibus Survey in Britain, the Health Survey for England and the Oxford Healthy Life Survey. J Public Health Med. 1999;21:255. [PubMed: 10528952]

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- Mishel MH. Parents' perception of uncertainty concerning their hospitalized child. Nurs Res. 1983;32:324. [PubMed: 6567851]
- 21. Mishel MH. Uncertainty in Illness Scales Manual. Chapel Hill, North Carolina: University of North Carolina-Chapel Hill; 1997.
- 22. Weiss DS, Mamar CR. The impact of event scale-revised. In: Wilson J, Keane T, eds. Assessing Psychological Trauma and PTSD: A Practitioner's Handbook. New York: Guilford Press; 1997:399.
- 23. Erford BT, Johnson E, Bardoshi G. Meta-analysis of the English version of the Beck Depression Inventory-Second Edition. Meas Eval Couns Dev. 2016;49:3.
- Yonemoto T, Kamibeppu K, Ishii T, et al. Posttraumatic stress symptom (PTSS) and posttraumatic growth (PTG) in parents of childhood, adolescent and young adult patients with high-grade osteosarcoma. Int J Clin Oncol. 2012;17:272. [PubMed: 21748258]
- Yi-Han C, Jian-Pei H, Heng-Kien A, et al. High risk of depression, anxiety, and poor quality of life among experienced fathers, but not mothers: A prospective longitudinal study. J Affect Disord. 2019;242:39. [PubMed: 30170237]
- 26. Goodman JH, Watson GR, Stubbs B. Anxiety disorders in postpartum women: A systematic review and meta-analysis. J Affect Disord. 2016;203:292. [PubMed: 27317922]
- Hughes IA, Houk C, Ahmed SF, et al. Consensus statement on management of intersex disorders. J Pediatr Urol. 2006;2:148. [PubMed: 18947601]
- Bakula DM, Sharkey CM, Wolfe-Christensen C, et al. Recommendations for the establishment of disorders/differences of sex development interdisciplinary care clinics for youth. J Pediatr Nurs. 2017;37:79. [PubMed: 28888337]
- 29. Sundai M Revisiting the limits of professional autonomy: The intersex rights movement's path to de-medicalization. Harvard J Law Gend. 2018;41:1.
- Ernst MM, Gardner M, Mara CA, et al. Psychosocial screening in disorders/differences of sex development: Psychometric evaluation of the Psychosocial Assessment Tool. Horm Res Paediatr. 2019;175:279.

## Table 1.

## Parent Demographics

	Complete Sample		Surgery Sample		Non-Surgery Sample	
	Mothers	Fathers	Mothers	Fathers	Mothers	Fathers
N	76	63	69	59	7	4
Age M(SD)	31.3 (5.6)	34.4 (8.2)	31.7 (5.3)	34.7 (8.1)	27.6 (6.6)	29.3 (10.0)
Race/Ethnicity N (%)						
Hispanic	17 (22)	8 (13)	14 (20)	8 (14)	3 (43)	0 (0)
Black/African American	6 (8)	5 (8)	6 (9)	5 (8)	0 (0)	0 (0)
White/Caucasian	52 (68)	41 (65)	46 (65)	38 (64)	6 (86)	3 (75)
Asian/Pacific Islander	4 (5)	4 (6)	3 (4)	4 (7)	1 (14)	0 (0)
Multiracial	3 (4)	1 (2)	3 (4)	1 (2)	0 (0)	0 (0)
Other	5 (7)	6 (10)	5 (7)	6 (10)	0 (0)	0 (0)
Marital Status N (%)						
Single, Never Married	13 (17)	3 (5)	11 (16)	3 (5)	2 (29)	0 (0)
Divorced	2 (3)	1 (2)	2 (3)	1 (2)	0 (0)	0 (0)
Married to Parent of Child with DSD	59 (78)	51 (81)	55 (76)	49 (83)	4 (57)	2 (50)
Living with Parent of Child with DSD	8 (11)	10 (16)	7 (10)	9 (15)	1 (14)	1 (25)
Partner Parent of Child with DSD	1 (1)	0 (0)	1 (1)	0 (0)	0 (0)	0 (0)
Highest Education Attained N (%)						
Some High School or Less	3 (4)	2 (3)	2 (3)	2 (3)	1 (14)	0 (0)
Finished High School/GED	9 (12)	8 (13)	7 (10)	7 (12)	2 (29)	1 (25)
Some College or Associates Degree	23 (30)	21 (33)	20 (28)	13 (22)	3 (43)	2 (50)
Bachelor's Degree	24 (32)	18 (29)	23 (32)	18 (30)	1 (14)	0 (0)
Graduate Degree	16 (21)	11 (17)	16 (23)	11 (19)	0 (0)	0 (0)
Employment Status N (%)						
Part-Time	10 (13)	5 (8)	8 (12)	5 (8)	2 (29)	0 (0)
Full-Time	39 (51)	49 (78)	34 (49)	46 (78)	1 (14)	3 (75)
Part-Time Student	2 (3)	2 (3)	2 (3)	2 (3)	0 (0)	0 (0)
Full-Time Student	3 (4)	2 (3)	2 (3)	2 (3)	1 (14)	0 (0)
Stay at Home	26 (34)	3 (5)	23 (33)	3 (5)	3 (43)	0 (0)
Unemployed	6 (8)	1 (2)	5 (7)	1 (2)	1 (14)	0 (0)
Other	4 (5)	1 (2)	4 (6)	1 (2)	0 (0)	0 (0)
Household Income N (%)						
$0 - 19,999^{\dagger}$	14 (18)	6 (10)	9 (25)	5 (8)	5 (71)	1 (25)
20,000 - 39,999	13 (17)	10 (16)	13 (18)	10 (17)	0 (0)	0 (0)
40,000 - 59,999	7 (9)	6 (10)	7 (10)	6 (10)	0 (0)	0 (0)
60,000 - 79,999	8 (11)	7 (11)	8 (11)	7 (12)	0 (0)	0 (0)
80,000 - 99,999	8 (11)	7 (11)	7 (10)	6 (10)	1 (14)	1 (25)
100,00 +	25 (33)	23 (37)	24 (34)	22 (37)	1 (14)	1 (25)

Note. The sum for participant race is greater than the sample due to participants being able to select Hispanic as ethnicity as well as a race. The sum for marital status and education are additionally greater than the sample size due to the opportunity to select more than one option. The sample of

no surgery parents (n = 11) is underpowered for statistical analysis. The above demographics are a description of the surgery and no surgery parents.

 $^{\dot{7}}\text{Below the federal poverty line for a family of 3.}$ 

## Table 2.

Parental Outcome Measures with Comparison to Published Norms

Measure	<u>Sample</u> Mean ± SD	<u>Norms</u> Mean ± SD	<u>Mothers</u> Mean ± SD	<u>Fathers</u> Mean ± SD
Child's Cosmetic Appearance	$2.68\pm.96$	Not Applicable	$2.79\pm.93$	$2.59\pm.94$
BAI	$8.19\pm8.88\overset{*}{}$	$9.89 \pm 8.76^{16}$	$9.88 \pm 8.44$	$5.53\pm8.25$
BDI-II	$8.93 \pm 9.50$	$8.39 \pm 6.89^{23}$	$11.50\pm11.35$	$5.30\pm6.40$
SF-36 physical health	$55.54 \pm 6.48$ ***	$50.0\pm10.0$	$55.34\pm7.30$	$55.38 \pm 5.58$
SF-36 mental health	$46.40 \pm 10.97^{***}$	$50.0\pm10.0$	$43.36\pm11.52$	$51.27 \pm 8.42$
PPUS	$65.21 \pm 15.90^{***}$	$70.3 \pm 14.5^{21}{}^{\dagger}$	$65.46 \pm 15.44$	$65.37 \pm 16.47$
IES-R	$17.31 \pm 16.42$	$18.5 \pm 16.1^{24}$	$21.53 \pm 18.15$	$11.72\pm11.98$

*Note.* Standardized norms reflect a combination of males and females; therefore, parent means are combined for a total sample comparison. BAI = Beck Anxiety Inventory; BDI-II = Beck Depression Inventory-II; SF-36 = Health related quality of life; PPUS = Parent Perception of Uncertainty; IES-R = Impact of Event Scale-Revised.

\* p<.05,

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*p* < .01,

\*\*\* p<.001

<sup>†</sup>Norms based off parents of children with Leukemia.

#### Table 3.

Outcome Measures of Surgery and Non-Surgery Parents

Measure	<u>Surgery</u> Mean ± SD	<u>No Surgery</u> Mean ± SD
Parent Rating of Cosmetic Appearance	$2.75\pm.94$	$1.91\pm.83$
Physician Rating of Cosmetic Appearance	$3.41\pm.60$	$2.14 \pm 1.22$
BAI	$8.19 \pm 8.67$	$8.10 \pm 11.48$
BDI-II	$8.88 \pm 9.64$	$9.44 \pm 8.16$
SF-36 physical health	$55.48 \pm 6.59$	$56.14 \pm 5.53$
SF-36 mental health	$46.57 \pm 10.81$	$44.55\pm13.25$
PPUS	$65.22 \pm 15.99$	$65.10 \pm 15.75$
IES-R	$17.45\pm16.19$	$15.80 \pm 19.63$

*Note.* The sample of no surgery parents (n = 11) is underpowered for statistical analysis. The above means are an exploratory description of the surgery and no surgery parents. BAI = Beck Anxiety Inventory; BDI-II = Beck Depression Inventory-II; SF-36 = Health related quality of life; PPUS = Parent Perception of Uncertainty; IES-R = Impact of Event Scale-Revised.

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#### Table 4.

#### **Correlations of Parental Distress**

	Cosmesis	BAI	BDI-II	PCS	MCS	PPUS
Cosmesis						
Cosmesis	-					
BAI	.055	-				
BDI-II	.022	.676 ***	-			
PCS	.130	175	418 **	-		
MCS	.031	597 **	807 **	.305 ***	-	
PPUS	.009	.259 ***	.271 **	202 **	391 ***	-
IES-R	.226*	.565 **	.557 **	149	588 **	.347 **

*Note.* Negative numbers occur on physical health related quality of life (PCS) and mental health related quality of life (MCS). This demonstrates that quality of life decreases as the other distress variable increases. BAI = Beck Anxiety Inventory; BDI-II = Beck Depression Inventory-II; PCS = Physical health related quality of life; MCS = Mental health related quality of life; PPUS = Parent Perception of Uncertainty; IES-R = Impact of Event Scale-Revised.

.10 to .30 = small correlation; .30 - .50 = moderate correlation; .50 = large correlation

\*\* p<.01,

\* p<.05.

## Table 5.

Analyses for Predictors of Cognitive Appraisals and Distress

<i>Means</i> Mother (Father)	Cosmesis	BAI	BDI-II	SF-36 Physical Health	SF-36 Mental Health	PPUS	IES-R
Parent Gender							
Female	2.79*	<b>9.9</b> **	12.1***	55.3	43.4**	65.5	21.4 **
Male	2.59*	5.5**	5.4 <sup>***</sup>	55.4	51.3**	65.4	11.8 **
Child Gender of Rearing							
Girl	2.7 (2.4)	10.5 (6.2)	12.6 (5.3)	57.1 (55.2)	41.8 (50.0)	64.2 (63.3)	19.9 (9.7)
Boy	2.9 (2.9)	11.0 (4.1)	11.1 (5.7)	53.0 (55.1)	42.2 (54.8)	68.2 (67.8)	26.2 (14.4)
Unsure	1.5 (2.0)	1.5 (3.5)	.5 (2.0)	56.2 (57.3)	55.9 (52.5)	62.5 (72.0)	7.0 (14.0)
Diagnosis							
Known	2.8 (2.5)	10.6 (5.2)	12.8 (4.8)	57.0 (55.6)	41.3 (51.3)	64.0 (63.9)	21.9 ( <b>9.0</b> *)
Unknown	2.9 (2.9)	10.4 (6.2)	9.5 (7.4)	53.5 (54.0)	46.3 (51.1)	67.1 (68.3)	24.3 ( <b>18.8</b> *)
Other Children							
No	<b>3.1</b> *(2.7)	<b>12.8</b> *(4.5)	13.2 (4.6)	55.6 (55.2)	42.3 (51.9)	<b>70.4</b> *(66.4)	25.6 (13.0)
Yes	<b>2.5</b> *(2.5)	<b>8.4</b> <sup>*</sup> (6.0)	10.5 (5.9)	56.0 (55.3)	42.5 (50.7)	<b>61.5</b> *(63.8)	18.6 (10.4)
Financial Concerns							
A Lot of Concerns	2.5 (2.4)	13.8 (6.6)	<b>20.3</b> **(7.3)	53.9 (55.2)	<b>49.9</b> **(49.7)	64.1 (74.1)	23.8 (17.6)
No Concerns	2.9 (2.6)	6.8 (4.1)	<b>6.5</b> **(2.4)	57.4 (54.4)	<b>34.9</b> ** (56.7)	60.0 (54.9)	17.4 (8.6)
Increased Expense			()		(2007)		
Mental Health							
Yes	2.8 (2.6)	10.6 (2.7)	13.6 (4.4)	56.7 (54.8)	40.0 (50.8)	63.5 (64.4)	23.5 (10.8)
No	2.8 (2.6)	10.2 (6.7)	10.6 (5.8)	55.2 (55.5)	43.9 (51.6)	66.5 (65.3)	20.5 (12.0)
Medical							
Yes	<b>3.1</b> *(2.3)	8.7 (5.0)	11.0 (5.3)	56.4 (55.2)	44.2 (50.1)	66.4 (66.9)	24.2 (12.1)
No	<b>2.6</b> *(2.7)	11.3 (5.5)	12.1 (5.3)	55.4 (55.2)	41.4 (52.0)	64.8 (63.7)	20.1 (11.3)
Travel							
Yes	2.6 (2.6)	11.5 (6.7)	<b>14.1</b> *(5.3)	55.4 (54.7)	<b>40.0</b> *(50.9)	65.2 (65.9)	21.2 ( <b>15.0</b> *)
No	3.0 (2.5)	8.1 (3.7)	<b>7.1</b> <sup>*</sup> (5.3)	56.9 (55.7)	<b>48.7</b> *(51.6)	65.8 (64.1)	22.4 ( <b>8.6</b> *)
Support Received			(0.0)		(0.1.0)		,
Money for Expenses							
Yes	3.3 (2.3)	6.4 (1.3)	8.8 (1.3)	59.6 (56.8)	<b>54.5</b> *(56.4)	60.0 (52.0)	11.8 (8.3)
No	2.7 (2.6)	10.7 (5.6)	11.9 (5.6)	55.5 (55.1)	<b>41.5</b> *(50.9)	65.7 (65.5)	22.4 (11.8)
Emotional Support	× /		× /	· · /	(0000)		、 <i>,</i>
Yes	2.9 (2.5)	9.3 (5.9)	10.8 (5.7)	55.1 (54.0)	45.2 (52.3)	64.3 (67.9)	21.2 (10.4)
No	2.7 (2.6)	11.0 (5.1)	12.2 (4.4)	56.2 (55.8)	40.8 (50.9)	66.0 (63.9)	21.8 (12.1)
Help Caring for Child							
Yes	2.9 (2.5)	10.4 (5.7)	11.8 (4.9)	56.0 (54.4)	42.4 (52.1)	69.0 (70.4)	22.9 (12.2)
No	2.7 (2.6)	10.4 (5.2)	11.6 (5.4)	55.7 (55.6)	41.7 (50.9)	64.1 (63.0)	21.1 (11.3)

<i>Means</i> Mother (Father)	Cosmesis	BAI	BDI-II	SF-36 Physical Health	SF-36 Mental Health	PPUS	IES-R
Household Income (r)	.15 (.24)	18 (- <b>.42</b> **)	16 (- <b>.42</b> **)	.11 ( <b>.35</b> <sup>*</sup> )	.26 (.29)	.002 (04)	04 (- <b>.34</b> *)
Distance to Urologist ( <i>r</i> )	.004 (.08)	03 (.42)	07 (.01)	.02 (10)	.24 (26)	.16 (12)	11 (24)
Parent Age (r)	.06 (.11)	22 (- <b>.40</b> **)	10 (14)	24 (.04)	.23 (.19)	05 (.19)	09 (12)
Child Age ( <i>r</i> )	.14 (10)	.14 (.09)	01 (05)	.05 (19)	.02 (04)	12 (20)	02 (07)

*Note.* Mother and father mean scores presented unless the predictor is a continuous variable, in which case the Pearson correlation coefficient is reported (*r*). Significant relationships reported in bold.

\* p<.05,

\*\* p<.01.

\*\*\* p<.001