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Original Article

Impact of Disorders of Sex Development on Egyptian Parents and Factors Affecting Them

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Abstract

Background: Disorders of sex development (DSD) are a category of congenital diseases characterized by aberrant internal and external genital structure development. Parental adjustment and functioning have been highlighted as being at risk in this environment.
Objective(s): To compare the impact of children with XX, DSD and XY, DSD on their families, and factors influencing the burden on these parents.
Methods: A cross-sectional study was conducted on parents of all 72 children with DSD who were diagnosed and followed up regularly between January and May 2021 at Alexandria University Children's Hospital's Endocrinology Outpatient Clinic, Egypt. The Pediatric Quality of Life Inventory™ (PedsQL™) Family Impact Module (PedsQL™ FIM) (Arabic version) version 2 was used for assessing family impact and economic burden.
Results: The current study included 72 children with DSD, 57 diagnosed with 46,XX, and 15 with 46,XY. The mean age of studied children was 7.93 ± 4.03 years. Parents of children with XX, DSD had lower scores (reduced family function) on all categories (except for family relationships) of the PedsQL™ FIM version 2 questionnaire, with no statistically significant difference. The worry domain had the lowest mean scores, with 33.86 ± 21.59 in children with XX,DSD and 45.33 ± 29.79 in those with XY,DSD. Univariate and correlation analyses found that having a sibling with the same condition had a statistically significant (r=-0.359,p= 0.002) negative impact on the family.
Conclusion: XX,DSD had more negative impact on parents than XY,DSD, especially in the worry domain. Additionally, having siblings with similar conditions exhibited a strong correlation to creating a negative influence.
Keywords: Disorder of sex development, Family Impact, PedsQL™ family impact module

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INTRODUCTION

Disorders of sexual development (DSD) are a category of congenital diseases characterized by aberrant internal and external genital structure development. (1) Due to the ambiguity of the external genitalia, affected individuals may be identified from birth, others may show signs of neonatal virilization, delayed or missing puberty, or infertility later in life. (2)

Sex chromosomal DSD, 46,XY DSD (previously male pseudohermaphrodite (PH)), and 46,XX DSD (formerly female pseudohermaphrodite (PH)) are the two main diagnostic categories in the new DSD categorization. Congenital Adrenal Hyperplasia (CAH) is the most common cause of atypical genitalia. (3, 4) DSD is a hereditary autosomal recessive adrenal gland condition that affects both boys and girls, with a frequency of 1/10,000 and an annual incidence ranging from 1/5,000 to 1/15,000. (5)

The presence and severity of atypical genitalia, decisions about rearing gender (and also the possibility of gender reassignment), sex chromosome discordance, ongoing controversies about the risks and benefits of early genital surgery, anticipated stigma, accompanying shame, and economic burden are all examples of stressors faced by parents of children with DSD, as well as patients themselves as they grow older. (6)

Given the critical role of the family in a child's disease adaption and the influence of the disease on the family, functioning is a major concern in pediatric chronic health disorders.(7, 8) Parental adjustment and family functioning as a whole have been highlighted as being at risk in this environment.(8) Additionally, family economics, parenting abilities, and caregiver psychosocial functioning all have an impact on the health outcomes of children with chronic illnesses including DSD. (9)

Although there are a number of well-developed generic measures of family functioning, such as the Family Environment Scale,[10] there are few instruments that specifically measure the impact of pediatric chronic health conditions on parent and family functioning. The Pediatric Quality of Life Inventory™ (PedsQL™) Family Impact Module (PedsQL™ FIM) is a multidimensional tool that can be used on its own or in conjunction with the PedsQLTM Measurement Model. [11]

Limited studies have investigated the impact of Disorders of Sex Development of children on the lives of their parents. Therefore, this research aims to compare the effects of XX,DSD, and XY,DSD on their families. We also aim to look into the many elements that may influence the family impact of parents of children with DSD

METHODS

Study design, setting, and population

A cross-sectional study was carried out on the parents of all children with DSD who were diagnosed and followed up regularly at the Alexandria University Children's Hospital's Endocrinology Outpatient Clinic, Egypt between January and May 2021.

Inclusion and Exclusion criteria

All parents of children (2-18 years) diagnosed with DSD by an expert pediatric endocrinologist, who agreed to participate were included in the study. Children with a chronic medical condition, such as diabetes mellitus or hypertension, as well as neurodevelopmental and/or psychiatric disorders, such as Attention Deficit Hyperactivity Disorder (ADHD), Autism Spectrum Disorder (ASD), or anxiety disorders, were excluded from the study.

Data collection and tools

I: A pre-designed questionnaire was used to collect demographic and clinical data of the studied children; namely age, sex, family history (similar condition, consanguinity, structure, residency, educational status), antenatal and perinatal history (pregnancy course, gestational age, status of newborn, birth weight), age at diagnosis, duration of diagnosis, surgical history (age at the time of surgery, stages of surgical intervention), medications and for those with Congenital Adrenal Hyperplasia (CAH), whether controlled or not. We considered the child controlled when the 17- Hydroxyprogesterone level was within normal using the lowest dose of steroids. Data was taken from the patients' files, and missing data was obtained from the parents.

II: Assessment of the impact of children's illness on their parents was done using the Arabic version of the PedsQL TM 2.0 Family Impact Module (PedsQL™ FIM version 2). [12] The PedsQL™ FIM (Arabic version) showed good validity and reliability for the total PedsQL™ Family Impact Scale as well as its subscales in measuring functioning in families of children with chronic health issues, where Cronbach's alpha score was above 0.93 for the total scale. [12] Similarly, investigators from Canada and China showed its validity and reliability in measuring functioning in families of children with complex chronic health issues. [13, 14] This Arabic version of the PedsQL™ FIM version 2 is made up of 36 items in 8 dimensions including 6 items for physical function, 5 items for emotional function, 4 items for the social function, 5 items for cognitive function, 3 items for communication, 5 items for worry, 3 items for daily activities, and 5 items for family relationships. It is a 5-point rating Likert scale ranging from "never" to "almost always" as follows: 0 if it is never a problem and 4 if it is almost always a problem. Items were then reverse-scored and linearly transformed to a 0 – 100 scale (0 = 100, 1 = 75, 2 = 50, 3 = 25, 4 = 0) so that higher scores indicate better functioning (less negative impact). [13] Scale scores should not be computed if more than half of the items on the scale are missing. Scores of the scale included: The **total mean score** equals the sum of scores of all 36 items divided by the number of answered items; **mean score** of each subscale equals the sum of scores of the items over the number of answered items. The sum of scores of the items divided by the number of answered items in the physical, emotional, social, and cognitive functioning subscales yields the Parent Health-Related Quality of Life (**Parent HRQL)** summary score (20 items); the sum of scores of the items divided by the number of answered items in the daily activities and family relationships subscales yields the family functioning summary score (8 items). [15]

Each participant was given a printed copy of Arabic version of PedsQLTM FIM version 2 to complete while waiting for their follow-up appointment with the physician. To reduce biases, a member of the research team was there to supervise, answer any questions, help with the filling out procedure, and check that the quality goals were accomplished. The questionnaire was completed in the required amount of time (about 25 to 30 minutes).

Ethical considerations

The study protocol was approved by Alexandria Faculty of Medicine's Medical Ethics Committee. Written informed consents were obtained from children's parents after explaining the aim of the study and assuring about confidentiality of collected data.

Data management and statistical analysis

Data entry and analysis were done using the Statistical Package for Social Science (SPSS Inc., Chicago, IL,

USA) version 20. [16] The Kolmogorov-Smirnov test was employed to ensure normal distribution. Descriptive statistics were calculated as the means and SDs for continuous variables and as frequencies and percentages for categorical variables. Chi-squared ($\chi 2$) and Fisher's exact tests were used as the tests of significance for categorical variables. For normally distributed quantitative variables, the analysis of variance test (ANOVA) was used to compare more than two groups, while the Post Hoc test (Tukey) was used for pairwise comparisons. Mann Whitney test was used to compare two groups with improperly distributed quantitative variables. For abnormally distributed quantitative variables, the Kruskal Wallis test was used to compare more than two study groups, whereas the Post Hoc (Dunn's multiple comparisons test) was used for pairwise comparisons. The most independent/ influencing factors for Pediatric PedsQL^{TM} Family Impact Module were determined using univariate and multivariate analyses. The statistical significance level was considered when the P value was < 0.05 for all statistical tests.

RESULTS

Table 1 reveals that there were 72 children with DSD in the current study, 57 with the diagnosis of 46,XX, and 15 with 46,XY. The mean age of studied children is (7.93 ± 4.03) years. More than half of the parents is from a rural area (69%). In terms of education, 50% of parents had graduated from secondary school, and 8.3% had a college degree.

All cases of 46,XX DSD had congenital adrenal hyperplasia For children with 46,XY; 46.7 % of cases had partial androgen insensitivity and 53.3 % had 5 alpha-reductase deficiency. According to family history, 20 patients (35.1%) of the 46,XX group had a sibling with the same condition, and 11 patients (19.3%) of the 46,XX group had died sibling with the same condition, whereas the 46,XY group had no history of a sibling with the same condition or died sibling with the same condition. In comparison to the clinical characteristics of children with XY,DSD, children with XX,DSD had a statistically significant longer duration of disease (9.0 years ranging from 5.9 to 11.0), history of surgical operations (89.5%), and family history of a sibling of similar condition(35%). (Table 2)

Except for family relationships, parents of children with 46XX,DSD had lower scores (reduced family function) on all categories of the PedsQL™ FIM version 2 questionnaire, with no statistically significant difference. The worry domain had the lowest mean scores among family effect modules, with (33.86 ± 21.59) in children with 46XX,DSD and (45.33 ± 29.79) in those with 46XY,DSD. (Table 3)

Univariate and correlation analyses found that having a sibling with the same condition had a statistically significant (r=-0.359, p= 0.002) negative impact on the family among the 14 factors investigated. (Figure 1)

Table (1): Sociodemographic data of studied DSD children and their parents

Sociodemographic data	Cases (n = 72)	
	No.	%
Age of the children		
Min. – Max.	2.0 – 16.0	
Mean ± SD.	7.93 ± 4.03	
Median (IQR)	8.40 (3.8 – 10.4)	
Sex of the children		
Male	15	20.8
Female	57	79.2
Residency		
Urban	23	31.9
Rural	49	68.1
Education of the parent		
Illiterate	10	13.9
Read and write	14	19.4
Primary school	6	8.3
Secondary school	36	50.0
High education	6	8.3
Occupation of the parent		
Not working	8	11.1
Working	64	88.9
Number of family members		
Min. – Max.	3.0 – 7.0	
Mean ± SD.	5.0 ± 0.93	
Marital status of the parents		
Married	71	98.6
Divorced	1	1.4
Widow	0	0.0

Table (2): Clinical characteristics of studied DSD children

	46XX (n = 57)		46XY (n = 15)		Test of sig.	P
	No.	%	No.	%		
Age at diagnosis (days)	**(n = 57)**		**(n = 15)**			
Min. – Max.	1.0 – 1095.0		1.0 – 1460.0		U= 348.5	0.253
Median (IQR)	7.0(1.0 – 30.0)		32.0(1.0 – 272.5)			
Duration of illness(years)	**(n = 57)**		**(n = 15)**			
Min. – Max.	1.11 – 16.0		1.0 – 10.30		U= 160.0*	<0.001*
Median (IQR)	9.0(5.9 – 11.0)		3.10(2.0 – 5.4)			
Controlled	**(n = 57)**		**(n = 0)**			
Yes	42	73.7	–	–	–	–
No	15	26.3	–	–		
Operated	**(n = 57)**		**(n = 15)**			
No	6	10.5	10	66.7	$\chi2$= 21.654*	FEp <0.001*
Yes	51	89.5	5	33.3		
Stages of operation	**(n = 51)**		**(n = 5)**			
One stage	45	88.2	5	100.0	$\chi2$= 0.659	FEp= 1.000
Two stages	6	11.8	0	0.0		
Treatment	**(n = 57)**		**(n = 0)**			
Prednisone	47	82.5	–	–	–	–
Hydrocortisone	10	17.5	–	–		
Diagnosis	**(n = 57)**		**(n = 15)**			
Congenital adrenal	57	100.0	0	0.0		
Partial androgen insensitivity	0	0.0	7	46.7	$\chi2$= 65.504	MCp <0.001*
5 alpha reductase	0	0.0	8	53.3		
Sibling of the same condition	**(n = 57)**		**(n = 15)**			
No	37	64.9	15	100.0	$\chi2$=7.287*	FEp= 0.007*
Yes	20	35.1	0	0.0		
History of dead sibling	**(n = 57)**		**(n = 15)**			
No	46	80.7	15	100.0	$\chi2$=3.417	FEp= 0.105
Yes	11	19.3	0	0.0		

χ^2: Chi square test MC: Monte Carlo FE: Fisher Exact t: Student t-test
U: Mann Whitney test p: p value for comparing control and total cases *: Statistically significant at p <0.05

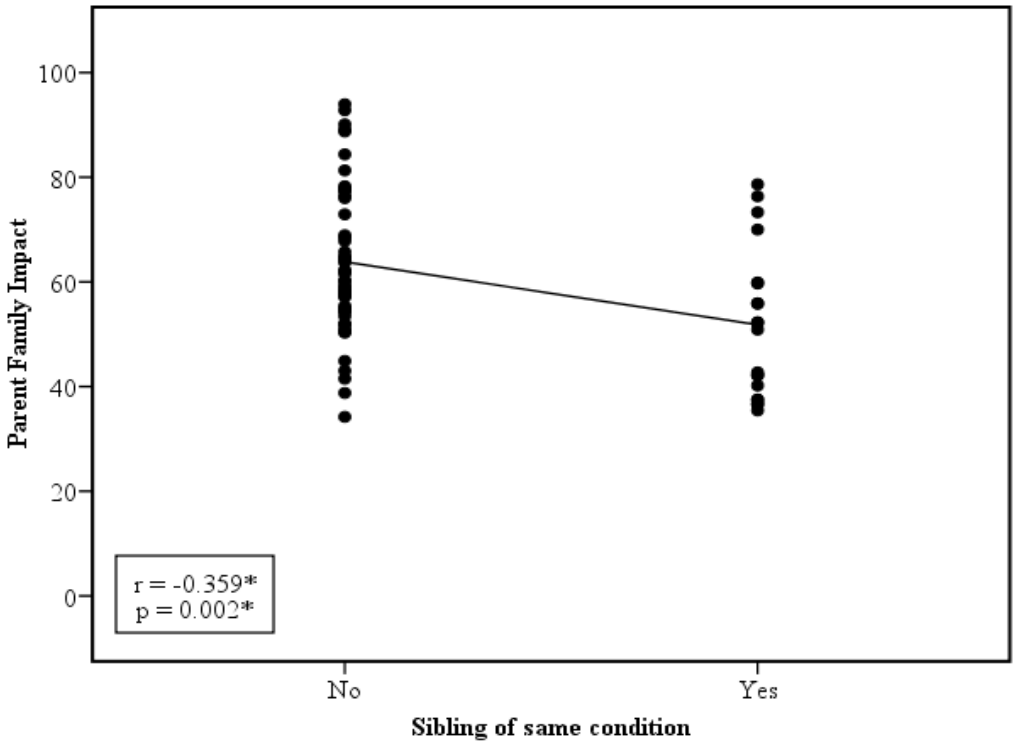

Figure (1): Correlation between parent Family Impact mean score and sibling of the same condition for total DSD cases

Table (3): Comparison between the 46XX and 46XY according to PedsQL™ FIM version 2 questionnaire

	46XX (n = 57)	46XY (n = 15)	t	P
Physical Function				
Min. – Max.	8.30 – 100.0	25.0 – 95.83		
Mean ± SD.	59.18 ± 22.12	65.55 ± 22.07	0.993	0.324
Median (IQR)	58.33 (41.7–70.8)	70.80 (50.0–81.2)		
Emotional function				
Min. – Max.	5.0 – 100.0	20.0 – 90.0		
Mean ± SD.	54.18 ± 22.32	62.33 ± 20.08	1.284	0.204
Median (IQR)	55.0 (40.0–65.0)	75.0 (47.5–75.0)		
Social function				
Min. – Max.	18.75 – 100.0	18.75 – 100.0		
Mean ± SD.	67.31 ± 20.86	72.50 ± 25.64	0.817	0.416
Median (IQR)	68.70 (50.0–75.0)	75.0 (56.3–96.9)		
Cognetive function				
Min. – Max.	25.0 – 100.0	25.0 – 100.0		
Mean ± SD.	61.66 ± 20.85	66.67 ± 25.47	0.790	0.432
Median (IQR)	62.50 (50.0–75.0)	75.0 (45.0–82.5)		
Communication				
Min. – Max.	0.0 – 100.0	25.0 – 100.0		
Mean ± SD.	44.57 ± 27.22	59.99 ± 27.14	1.953	0.055
Median (IQR)	41.66 (25.0–66.6)	50.0 (37.5–79.2)		
Worry				
Min. – Max.	0.0 – 80.0	0.0 – 100.0		
Mean ± SD.	33.86 ± 21.59	45.33 ± 29.79	1.685	0.096
Median (IQR)	30.0 (20.0–45.0)	45.0 (22.5–67.5)		
Daily activity				
Min. – Max.	25.0 – 100.0	25.0 – 100.0		
Mean ± SD.	74.66 ± 21.50	75.55 ± 27.00	0.135	0.893
Median (IQR)	83.30 (62.5–91.6)	83.30(50.0–100.0)		
Family relationship				
Min. – Max.	25.0 – 100.0	0.0 – 100.0		
Mean ± SD.	81.67 ± 18.38	79.0 ± 27.14	0.450	0.654
Median (IQR)	90.0 (75.0–95.0)	80.0 (72.5–100.0)		
Parent HRQL summary score				
Min. – Max.	20.0 – 100.0	22.10 – 96.25		
Mean ± SD.	60.59 ± 18.48	66.74 ± 19.70	1.131	0.262
Median (IQR)	61.75 (47.9–71.3)	62.81 (55.2–82.8)		
Family functioning summary score				
Min. – Max.	25.0 –100.0	12.50 – 100.0		
Mean ± SD.	78.15 ± 18.48	78.61 ± 23.52	0.080	0.936
Median (IQR)	84.15 (68.3–89.2)	82.50 (73.3–96.7)		
Total score of family impact				
Min. – Max.	35.45 ± 92.81	34.21 ± 93.95		
Mean ± SD.	59.33 ± 14.69	64.86 ± 16.36	1.267	0.209
Median (IQR)	57.97 (50.5–68.9)	59.22 (53.9–77.4)		

t: Student t test p: p value for comparing 46XX and 46XY *: Statistically significant at p <0.05

Table (4): Univariate and multivariate analyses for the sociodemographic and clinical characteristics affecting parents as reported by the PedsQL family impact module questionnaire

	Univariate		#Multivariate	
	B (95%C.I.)	p	B (95% C.I.)	p
Age (years)	-0.667 (-1.546 – 0.212)	0.135		
Sex (male =1 / female=2)	-6.343 (-14.623 – 1.937)	0.131		
Age at diagnosis (days)	-0.006 (-0.019 – 0.007)	0.389		
Duration of illness	-0.615 (-1.475 – 0.245)	0.158		
Controlled	-2.780 (-11.682 – 6.122)	0.534		
Operated	-4.351 (-12.886 – 4.185)	0.313		
Residency	1.806 (-5.848 – 9.461)	0.639		
Education	0.093 (-2.782 – 2.968)	0.949		
Occupation	1.066 (-10.306 – 12.438)	0.852		
Number of family members	-3.242 (-7.016 – 0.532)	0.091		
Marital status	3.629 (-26.904 – 34.162)	0.813		
Special diagnosis	1.076 (-4.309 – 6.462)	0.691		
Sibling of same condition (no = 0/ yes = 1)	-12.019(-19.468– -4.570)	0.002*	-12.019(-19.468–-4.570)	0.002*
History of died sibling	-8.032 (-17.782 – 1.718)	0.105		

Beta: Standardized Coefficients C.I: Confidence interval
#: All variables with p < 0.05 were included in the multivariate *: Statistically significant at p < 0.05

DISCUSSION

Disorders of sex development (DSD) are a type of aberrant chromosomal, gonadal, or anatomical sex development. If an infant is born with ambiguous genitalia, he or she will be subjected to a variety of diagnostic and therapeutic procedures, such as surgery, hormonal treatments, and long-term monitoring, all of which, in addition to the disease itself, can cause significant distress to the patient and his family. Parents frequently cite perceived or real child-focused stigma as a reason for not seeking aid and support from their community of relatives and friends. [17] To our knowledge, no studies have investigated the impact of DSD of children on their parents either in Egypt or other countries of the Arab world.

The present study found that, according to the PedsQL™ FIM version 2 questionnaire, XX,DSD had a more negative impact on the family than XY,DSD although the difference was not statistically significant. We believe that parents of children with XX,DSD may face more challenges than parents of children with XY,DSD including the need for surgical interventions to normalize external genitalia. Furthermore, having a female with DSD carries a greater burden, particularly in the face of certain social and cultural challenges in low-income nations, where there is a great deal of misunderstanding and stigma. In addition, the studied children with XX,DSD had a longer duration of illness (mean 9.0 years) than those with XY,DSD (mean 3.10 years) with statistical significant difference (U= 160.0 , P < 0.001) which might be explained by the fact that cases of CAH need long term medical treatment and follow up unlike those with XY,DSD.

The worry domain had the lowest mean scores among family impact modules, which means, the highest field of impairment on family function in children with XX,DSD and in those with XY,DSD . In agreement to these results, a study polled 51 parents of infants under the age of two who had been diagnosed with moderate to severe DSD but had not yet been assigned a gender. Out of the 51 included parents, 18% said they had clinically significant symptoms of depression, and 25.4% said they experienced moderate to severe anxiety. Post-traumatic stress symptoms (PTSS) were reported by 15% of the participants. [17] Similarly, few studies have revealed that a DSD diagnosis can be stressful for parents.[18-20]

In the current study, a high percentage of the participated parents (68%) live in rural areas, which we feel may have an extra impact on the families' quality of life challenges, due to either poverty, ignorance, illiteracy, lack of medical access, and psychiatric services. Zainuddin et al. also underlined the influence of underdeveloped countries on CAH patients. [21]

Social stigma in developing countries was also reported by Joseph AA et al. who conducted

interviews with 205 Indian children with DSD and their parents and discovered several issues including high maternal distress because many mothers believed the condition had been passed down through them. Also, late or misdiagnosis, significant discrimination from medical and paramedical staff, and inadequate pre-operative information provided to the parent was found. [22] Adult patients in Indonesia [23] and Nigeria [24] reported similar social difficulties and challenges to care in rural settings.

Regarding the investigated factors affecting the family impact in the current study, having siblings with the same condition is found to have a significant negative influence on the family. The autosomal recessive nature of DSD with high rates of consanguineous marriage increases the incidence of having a similar condition in the family. This is understandable because having more than one sick child puts a family's psychological and financial stress levels through the roof. Pasterski et al., [18] investigated different characteristics in the parents of 47 children with DSD (child sex, parent sex, child age at diagnosis, years after diagnosis, genital ambiguity, father occupation, cognitive disorientation, and emotional distress). However, cognitive confusion, rather than emotional distress, predicted PTSS in this study. Consequently, the researchers speculated that direct cognitive therapies would be useful.

In the current study adds to the growing body of evidence supporting the necessity for multidisciplinary teams, particularly in developing countries with limited resources, to provide psychological assistance not just to children with DSD but also to their parents. A recent review emphasizes the importance of multidisciplinary care in preventing psychological injury, as well as the use of validated quality of life measures and systematic, regular monitoring of psychosocial outcomes to promote mental health. [6, 25] In addition, the Consensus Statement on Intersex Management and related guideline documents called for an integrated, interdisciplinary healthcare team that included qualified mental health providers who could help families understand and address early emotional reactions, explore current and future worries, adjust to the period of uncertainty during the diagnostic process and facilitate the shared decision-making process. Positive psychological and social adaptation for the patient and family are among the positive results, which extend beyond medical and surgical ones. [6, 26]

These findings should be seen in the context of some limitations. For example, because the study was cross-sectional, causal correlations could not be established. There was no real comparison group; therefore, it is impossible to say whether the symptoms reported are directly related to having a child with DSD. Due to the nature of the disease, there is a lack of homogeneity in the clinical characteristics of both groups, which acts as a confounding factor that affects the interpretation of the data. Finally, being recruited from a tertiary care institution clinic limited generalization. Future prospective studies are recommended to support the current study's findings.

CONCLUSION AND RECOMMENDATIONS

The current study concludes that XX, DSD had a greater negative impact on the parents than XY, DSD, especially in the worry domain. Additionally, having siblings with similar conditions exhibited a strong correlation to creating a negative influence. For optimal treatment of children with DSD, family support through a multidisciplinary team is essential.

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CONFLICT OF INTEREST

The authors declare no conflict of interest

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