Indexed by : DOAJ Google Crossref Dimension sinta GARUDA

J.Biomed.Transl.Res
e-ISSN: 2503-2178

JOURNAL OF BIOMEDICINE AND TRANSLATIONAL RESEARCH

Original Research Article

The Evaluation of Parental Acceptance Towards Children with Sex Chromosomal Disorders of Sex Development Using A Mixed-Method

Iit Fitrianingrum[1], Annastasia Ediati [2], Tri Indah Winarni [3], Sultana MH Faradz[3*]

[1]Departement of Biology and Pathobiology, Faculty of Medicine, Universitas Tanjungpura, Pontianak, Indonesia
[2]Faculty of Psychology, Universitas Diponegoro, Semarang, Indonesia
[3]Division of Human Genetics, Center for Biomedical Research, Faculty of Medicine, Universitas Diponegoro, Semarang, Indonesia

Article Info
History
Received: 29 Mar 2021
Accepted: 21 Apr 2021
Available: 30 Apr 2021

Abstract

Background: Sex chromosomal Disorder of sex development (DSD) is an atypical abnormality of external genitalia which is mismatched with its sex chromosome traits. The condition of children with DSD affects the dynamics in the family. Parents' reactions after discovering this health problem vary greatly, such as being in a state of shock, confusion, or self-blame. However, parents' acceptance is extremely important for better quality of caring, to the healthy social and emotional child development, and to make the best decisions regarding gender assignment.

Objective: To describe the acceptance process of parents that have children with sex chromosomes mosaicism DSD.

Methods: This study used a mixed-method with a sequential explanatory approach, which was preceded by quantitative data collection followed by qualitative. The total respondents consisted of 14 mothers and 12 fathers of 14 sex chromosome mosaicism DSD patients with XX/XY, X/XY, XYY or XXY variants. Quantitative data were collected using the Indonesian version of the Parental Acceptance-Rejection Questionnaire (PARQ), and interviews were conducted to determine the acceptance process.

Results: Most acceptance cases were based on the surgical stage completion in which a higher number of mothers (71.43%) than fathers (50%).

Conclusion: It is uneasy for parents to accept children with sex chromosome mosaicisms DSD, hence the fathers struggle more than mothers in accepting those affected. To the best of our knowledge this is the first study in Indonesia to help parent understand and accept their child condition.

Keywords:
Mosaic sex chromosome; Disorders of Sex Development; parental acceptance
Permalink/ DOI: https://doi.org/10.14710/jbtr.v7i1.10710

INTRODUCTION

Commonly, after giving birth, the baby's gender is often confirmed by simply examining the external genitalia.[1] However, this becomes more complicated in baby with disorders of sex development (DSD) cases, which is a congenital disorder characterized by genital atypical development at the chromosome, gonad, or anatomy level.[2]

* Corresponding author:
E-mail: sultanafaradz@gmail.com
(Sultana MH Faradz)

Sex chromosome mosaicisms are included in the classification of sex chromosome abnormalities. These occur when one or more cell populations experience an addition or reduction of the sex chromosome, either X or Y, which affects individual sexual development. Sex chromosome mosaicisms incidence is rare, only around 1.5 -1.7 per 10,000 births depending on the variant type.[3-4] Children born with DSD experience abnormalities in their sex organs, causing ambiguity, as a male or female.[5]

This condition's diagnosis requires complex medical procedures and this not only affects the patients but also their parents and families. For parents, the birth of a child with DSD causes a stressful experience, often due to uncertainty about the sex and the need for more attention to physical, psychological, and reproductive health conditions at every developmental stage.[6] The affected parents faced difficult medical decisions, including child care based on the sex, genital surgery or hormone therapy and also psychosocial factor including stigma, in shaping patients' and families' psychosocial adaptation. Duguid et. al stated some parents experienced an increased stress level and decreased coping mechanism ability .[7]Previous studies reported that the stress conditions experienced by those parents are due to two things, such as uncertainty related to diagnosis and optimal treatment as well as conflicts between parents on whether to conceal a child's abnormality as privacy or be more open to disclosure.[8,9] Sanders et al identified three elements namely shock, protection, and anxiety, which influence parents in seeking harmony related to ambiguous genitalia in children with genital atypical and its surgical procedure.[10] Some parents experience higher stress levels and maladaptive childcare characteristics when they chose not to continue surgery at all or to continue the child's genital surgery in the early days of life.[11]

Parents go through many phases in dealing with the health condition before final acceptance. Their emotional response when DSD is diagnosed as reported by some qualitative studies includes feelings of shock, disbelief, guilt, and shame.[10] This is further aggravated because of the taboo culture in the society to discuss abnormalities related to sex, sexuality, and genitalia, hence parents often do not disclose the child's condition to everyone including other family members. Parents stop being exposed to the outside environment and keep the diagnosis confidential to protect children from possible negative stigma and emotional reactions.[8,12-13]

An understanding of the psychological impact of parents having children with DSD is important for healthcare providers and genetic counselors because they assist in dealing with all problems related to this condition. Limited studies on parental acceptance towards children with sex chromosome mosaicism DSD have been undertaken in Indonesia setting on this issue. Therefore, knowledge of the acceptance process helps to predict parental decision-making in continuing the management of these children. The goal of the present study was to identify the acceptance process of parents with sex chromosome mosaicism DSD.

MATERIALS AND METHODS

Design

This study used a mixed-method with the sequential explanatory approach by first collecting and analyzing quantitative data followed by qualitative data in the second stage which was based on the initial results.[14] Mixed method was used because quantitative approach was not sufficient to describe and understand data on parental acceptance. Therefore, a qualitative approach was needed particularly in understanding parents' dilemmas and struggles in accepting their children with DSD.

Participants and Study Settings

Data of sex chromosome mosaicism DSD patients were obtained from the Center for Biomedical Research (CEBIOR), Faculty of Medicine, Diponegoro University, Semarang. There were 74 cases with sex chromosomes DSD of which 22 met the inclusion criteria namely mosaic sex chromosomes with Y chromosome material, but only 14 agreed to participate in this study. The total respondents were 26 parents consisting of 14 mothers and 12 fathers of 14 affected children. Parents that were eligible to become participants included i.e. a biological mother and father, but when one of them had died, the person was replaced by another family member as a caregiver who spend a minimum of 6 hours/day together with the child for more than 6 months.[11]

Approval was obtained from the Health Research Ethics Commission of the Faculty of Medicine, Diponegoro University/Dr. Kariadi Hospital registered by No. 75/EC/FK-RSDK/II/2018. Initially, parents received an explanation of the objectives, benefits, and procedures of this study, and their willingness to participate was requested by writing an agreement and signing an informed consent form. Home visit was done for data collection in which the demographic data included parents' age, education, employment, income, and religion by filling out the questionnaire.

Measurements

The instrument used consisted of the Parental Acceptance-Rejection questionnaire (PARQ) by Ronald and Nancy Rohner[15] and interview guides were compiled by investigators and validated by two experts (judgmental experts).

Data were collected through structured interviews using PARQ to determine parental acceptance towards children with sex chromosome mosaicisms DSD. Furthermore, the questionnaire consisted of 4 subscales, i.e warmth/rejection, indifference/neglection, hostility and aggression and undifferentiated rejection. It consisted of 24 items from the results of the adaptation measurement previously conducted, which yielded Cronbach's Alpha values of 0.834 and corrected item-total correlation of 0.193-0.628.[14] The Indonesian version of PARQ obtained a license from Rohner Research Publications. This used a scale of 1 to 4, corresponding to the four available answer choices which included score 1 "almost never happens" score 2 "rarely happens", score 3 "sometimes true", and score 4 "almost always right ". The PARQ score ranged from 1-96, where the category ≤ 34 = accept, and > 34 = not accept.

Data Analysis

Quantitative data analysis was performed using the Mann Whitney-U test to determine the differences in the fathers' and mothers' acceptance. A thematic analysis was used to analyze the qualitative data by following six steps procedure for data analysis: familiarizing with data, generating initial codes, searching for themes, reviewing

Table 1. Parents Acceptance-Rejection Questionnaire (PARQ) index

PARQ dimensions	Mother (n = 14)	Father (n = 12)	*P*
Warmth	9 (8-14)	11.75 ± 2.80	0.09
Aggression	8.64 ± 1.91	8.5 ± 2.50	0.92
Neglected	9.36 ± 2.82	10 ± 2.76	0.57
Rejection	4 (4-9)	4,5 (4-9)	0.63
Total PARQ score	33 ± 6.12	35.58 ± 7.36	0.38

Table 2. Percentage of Parents' Acceptance of Children based on Characteristics of Parents

Characteristics	n (%)	Acceptance of Parents	
		Accept (n)	Not Accept (n)
Mother (n = 14)			
Age (38.92 ± 8.04)			
31-40	8	5 (62.5%)	3 (37.5%)
41-50	4	3 (75%)	1 (25%)
51-60	2	2 (100%)	-
Education			
Elementary school	4	3 (66.7%)	1 (33.3%)
Junior high school	4	4 (100%)	-
High school	4	3 (75%)	1 (25%)
Diploma	1	-	1 (100%)
Bachelor	1	-	1 (100%)
Employment			
Laborer	1	1 (100%)	-
Private sector	2	1 (50%)	1 (50%)
Farmer	2	1 (50%)	1 (50%)
Housewives	9	7 (77.8%)	2 (22.2%)
Income			
<1.5 million	14	10 (71.4%)	4 (28.6%)
Father (n = 12)			
Age (42.83 ± 8.61)			
31-40	6	1 (16.7%)	5 (83.3%)
41-50	4	3 (75%)	1 (25%)
51-60	1	1 (100%)	-
61-70	1	1 (100%)	-
Education			
Elementary school	4	3 (66.7%)	1 (33.3%)
Junior high school	1	1 (100%)	-
High school	4	1 (25%)	3 (75%)
Diploma	1	1 (100%)	-
Bachelor	2	-	2 (100%)
Employment			
Laborer	3	2 (66.7%)	1 (33.3%)
Civil servants	1	-	1 (100%)
Private sector	3	1 (33.3%)	2 (66.7%)
Entrepreneur	2	1 (50%)	1 (50%)
Farmer	2	1 (50%)	1 (50%)
Unemployed	1	1 (100%)	-
Income			
<1.5 million	8	5 (62.5%)	3 (37.5%)
> 1.5 million	4	1 (25%)	3 (75%)

themes, defining and naming themes, and writing the report.[18] Prior to analysis, the researchers (IF & AE) read the interview verbatims carefully to get familiar with the data, then generating initial codes, searching for themes, reviewing themes and defining and naming themes. Afterwards, all researchers (IF, AE, TIW, SMHF) reviewed the themes and wrote the reports after all authors agreed with the findings.

RESULTS

The warmth dimension had a higher score than the acceptance-rejection dimension of care (see Table 1). However, after testing the score differences between the father and mother using the Mann Whitney test (U = 0.38, $p < .05$) was not significant.

The older parents had a higher acceptance score towards their children (see Table 2). However, those with higher education levels had a lower acceptance score. Parents with monthly income below 1.5 million rupiahs were more accepting compared to those earning above 1.5 million.

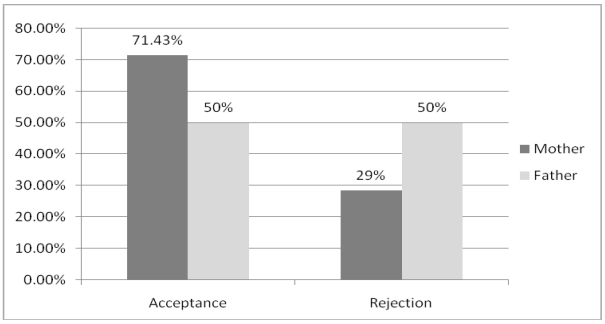

Figure 1. The percentage of parents' acceptance-rejection.

The acceptance results are illustrated in Figure 1, parents' acceptance score based on PARQ showed the percentage of mothers who accepted their children with sex chromosome mosaicism DSD were higher (n=10;71.43%) compared to the fathers (n=6; 50 %).

Qualitative Analysis

Three overarching and clinically salient domains of parental acceptance rearing affected children emerged from the thematic analysis: (1) sex announcement and timing diagnosis, (2) parents' support at each development stage, and (3) surgical decision-making and gender assignment.

Sex Announcement and Timing of Diagnosis

Investigation based on interviews identified that the beginning of discovering their children's abnormalities, almost all fathers experienced a denial (rejection) stage such as feeling shocked, confused, and asking why this happened. Afterward, they tried to find information related to these abnormalities by asking the doctor.

> *"When my child was born, I did not immediately tell my wife about her abnormalities, I waited for the right time and prepared myself to be able to tell her about our child"* (Father of child A)

The children's age was estimated with the mean being 5.09 ± 5.49 when establishing DSD diagnosis, from which twelve patients were presented with hypospadias and two with genital atypical.

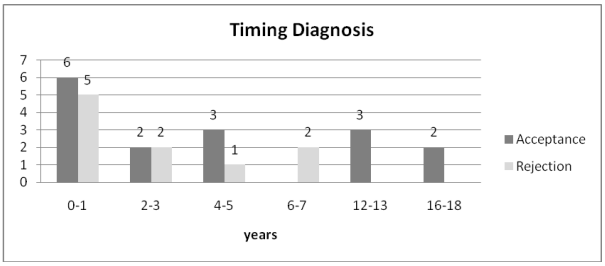

Figure 2. Parents' acceptance based on the children's age when the diagnosis was established

Children's age when the diagnosis was established also probably affects parents' acceptance. Based on Figure 2, five of the children diagnosed at age 0-1 years from 11 parents, only six parents accepted their children, but those who were diagnosed at 12-13 and 16-18 years were all accepted. When a child was diagnosed right after birth, parents experienced stress and took on blames.

> *"When I first discovered my child's abnormality, I was stressed, and experienced a baby blues, I did not want to be close to my child, I felt like I was carrying this disease until my child was 3 months old, I did not want to breastfeed my child" (Mother of child J)*

Parents' support at each developmental stage

Individuals with DSD face different problems at each developmental stage which also gives parents challenges to care for their children. This stage includes pre-school age (3-6 years), school-age (7-11 years), teenagers (12-18 years), and adults (19 years and above). Out of seven parents that had children at preschool age demonstrated four did not accept their health condition but had more acceptance as the children get older.

> "*I want the problem of determining the gender of this child to be resolved quickly, because when the child is more than 5 years old, I am afraid that he will feel inferior and shame to his friends"* (Mother of child R)
>
> *"Yes, this has been given by God, I just accept it. The important thing is I have tried, so that my boys can be like his friends. The doctor said that usually when my son at junior high school there is possibility of menses, that's why I noticed during junior high school while waiting for it. He was not menses, and men are sure."* (Father of child N)

Acceptance towards school-age children was shown through parents' support in all forms of activities with positive response and providing the best facilities for them.

> *"I always provide support to my child, provide facilities as long as it is in a positive direction, such as being active in karate sports or other men's activities such as an automotive workshop." (Father of child F)*
>
> *"Yes, the point is that there are advantages, besides there are shortcomings, I am sure that his shortcomings are not something he lacks, they are his strengths. Don't let your child be abused or stunted because of it"*

Surgical Decision-Making and Gender Assignment

However, based on the interview results of parents that had school-age children and adolescents, they had concerns about possible having gender reassignment, namely child gender change, which tends to cause deviant sexual behavior after growing into adulthood, for example, becoming transgender.

"My biggest fear is if later when he grows up into an adult, he changes their sex, then become transgender (Mother of child J)
"I have accepted it all as God's destiny, now, it is how I try to not make a wrong decision regarding the sex of my child, so I watch his personality and relationship closely."(Father of child E)

Children with sex chromosome mosaicism DSD who have decided their gender assignment undergo further care, such as hormonal therapy or surgery according to their gender assignment

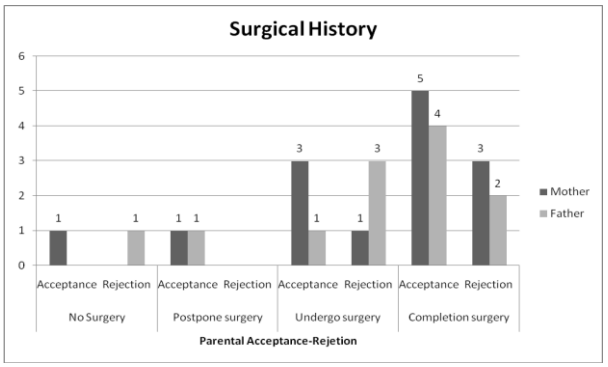

Figure 3. Parents' acceptance based on surgical histories conducted on the children with sex chromosome mosaicisms DSD.

Before the Gender Assignment Surgery

Based on Figure 3, the father did not accept their child that had never been treated surgically, while, the mother accepted without concerning it. From the interview results, the father did not agree on the surgery due to financial problems, and there was a feeling of self-pity for the little child (5 years) that had to pass through a series of surgical processes and the father expressed a fear of unsuccessful gender assignment surgery.

"The cost for the operation was quite high, so we collected the money first. Besides that, I could not bear to see my child being operated on because he was young, I am afraid that my child would not be able to succeed it" (Father of child A)

However, in children with postponed surgery, both parents accepted their abnormalities. Due to having a cordectomy earlier at the age of 3 years, the parents postponed further surgery in waiting for the child to decide the preferred gender assignment, hoping no regrets follow the choice later.

"I delayed the operation of my child because I waited until he could decide which gender to choose later, so there would be no regrets later. I always advised my child this was a fate that must be accepted" (Father of child E)

After the Gender Assignment Surgery

After completing the surgical procedure, out of the 14 parents, only 9 accepted their children (five mothers and four fathers), while 5 did not (three mothers and two fathers). The experience of parents of children who had completed gender assignment surgery before enrollment in formal education is stated below.

"Since the child was diagnosed, I planned for the child to be operated on immediately so that when entering school, the child clearly knew the sex so that the child did not feel inferior to the friends." (*Father of child N*)
"Even though it's not as perfect as other boys, let's try to be a boy who is like other boys, not as afraid if we don't try surgery or anything like that, we don't know in the future." (Mother of child J)

DISCUSSION

Based on the acceptance score, some parents' rejection possibly due to their concerns about future, such as children bearing potential problems such as sexual satisfaction for married couples or gender change problems at puberty or adulthood

The birth of a child with DSD causes anxiety and confusion for both parents because they are the first to know about the condition and also have crucial roles in handling this properly. The parenting pattern refers to the parent's behavior and belief towards their children. Furthermore, those having affected children are at risk of providing overprotective care and considering the children in vulnerable conditions. [19]

The percentage of mother's acceptance towards children with sex chromosome mosaicism DSD was higher compared to the fathers. The results are in line with previous studies conducted on mothers and fathers in four countries namely China, Italy, Sweden, and the United States, which showed that the mothers had higher acceptance than the fathers. The mothers were also reported to show more warmth compared to the fathers in China, Italy, Sweden, Philippines, and Thailand.[20] Also, the mothers having a higher acceptance level realistically see the children's condition and also realize their strengths and weaknesses. This realistic acceptance prevents the mother from being trapped in feelings of pity, but rises and takes various actions that are beneficial for the children's development.[17] Although the parental experience is probably the same as those having children with other chronic medical diagnoses. Those having children with DSD have special characteristics, such as rare cases of sex chromosome mosaicism DSD which creates confusion and feelings of isolation, because most parents do not have knowledge and understanding of the sexual differentiation process, as well as the accompanying abnormalities at the prenatal stage.[21] In addition, parents are concerned about the possibility of gender changes as adults, a physical appearance that is not in accordance with gender and physiological functions of reproduction may affect parental acceptance of children.[8,22]

Parents' acceptance towards children is described as resignation to the pre-outlined destiny that must be lived by the child and parents, while no one can change it. This is known as fatalism, while the parent's attitude is closely related to feeling helpless which is interpreted as a belief that humans are unable to change what has happened or is outlined. By holding the fatalistic belief, more parents accept abnormalities and future possibilities in the children. This includes both acceptance and rejection,

and it also has a positive aspect, the extent to which the abnormalities are accepted as a reality. Moreover, fatalistic belief is also interpreted as negative when parents feel they have no means of changing a reality that has occurred.[23] Income, social status, and education are strongly correlated with fatalistic beliefs which is why individuals with low income and social status tend to be more fatalistic. Conversely, education level is inversely proportional to fatalistic tendency, therefore playing an important role in the tendency of individuals to have fatalistic beliefs in the economy and social status.[22-23] This is in accordance with the results that showed mothers with primary to secondary education accepted their children compared to the highly educated ones that did not. The result suggests that mothers with low education are more likely to hold fatalistic worldviews, which could lead them to be over hopeful.

More than 50% of parents accept the children diagnosed in early life. Kohva et.al stated that patients with DSD are potentially diagnosed at the time of birth based on varied characteristics such as sexual ambiguity, family history, discrepancies between the sex chromosomes, and male or female genitalia appearance. But, in adolescents, the condition is identified with symptoms of an inguinal hernia in the female, late or incomplete puberty, virilization in the female, primary amenorrhea, breast growth in males, and a previously unknown ambiguous genitalia.[24] In cases of sex chromosome mosaicisms, the question is often about the gender assignment, the right time for surgery, and the potential for gonadal malignancy.[26] Enforcement of early DSD diagnosis is considered important, and diagnostic procedures efficacy often requires evaluation based on the child's developmental stages.[24] In Indonesia, this health condition is not widely known by both health workers and the common population, while clinical treatment is a challenge because of limited diagnostic and therapeutic facilities. Consequently, many patients experience delays in diagnosis,[27] but treatment in children reduces parental anxiety about their condition and possible stigmatization due to these abnormalities.

Gender also influences parental acceptance, but from this study, the phenomenon was not properly represented. Because there was only one female child observed, while the others were raised as males, of which the female was accepted by both parents based on the PARQ score. However, more than 50% of mothers accepted those males, while less than 50% of fathers accepted. Gender assignment to children is not only a doctor's decision but also involves parents. Children's development evaluation, for example, game choices, personal nature, and friendliness of the environment are all the parents' duty. Previous studies reported that in the first year of life, differences between babies are based on sound and facial shape, which later forms a stereotype of children. The children begin to acquire individual "labels" based on gender at the age of 17-24 months, and that awareness then develops towards gender seen from their daily behavior. Furthermore, most of them consistently answer callings as male or female at the age of 3 years.[28]

In Asian countries, cultural and religious factors play an important role in determining children's gender.[29] Moreover, in Indonesia, social life only accepts two gender categories, where the third choice becomes unpopular in contrast to the population in Western countries, especially in environments that strictly enforce religious rules. For example, in the Muslim community, it is very important to distinguish between male and female in life aspects such as how to dress, worship, and other religious activities, as well as in the educational environment. Parents and individuals with DSD need to deal with stigmatization in the family, among friends, colleagues, and strangers as well as on the media.[30] Social plays an important role in determining gender assignment and sex rearing which are often considered important as parental rights, bonds, and responsibilities. Strong social pressure is influenced by culture, tradition, and economic factors existing in the community, where the males have a dominant role in financial and social life.[31]

The acceptance based on the surgical stage completion in the child showed that almost 70% of parents accepted their children. The previous study stated that after a year of undergoing genitoplasty surgery, parents have good coping with their children's condition.[32] Surgical procedures performed in the first year of a child's life are easier, they provide good results and also reduce stigmatization against the child and family.[21] Currently, parents have the right to decide whether their children need to have genitals at infancy or childhood. It is important to adopt a respectful and non-blaming stance when considering parental decision to highly take care of their child in uniquely difficult circumstances.[28] From the acceptance score, some parents rejected the children possibly due to concerns about their future, such as children bearing potential, sexual satisfaction for married couples or gender change problems at puberty or adulthood.

Limitations of the study

This study used a cross sectional study, while the acceptance measurement of parents possibly has fluctuated results. In addition, there were variations in the time intervals when collecting the data with the beginning of the diagnosis, it probably affects the memories and emotions of parents that might change due to the length of the time interval. The number of respondents were limited, therefore, the result did not represent the acceptance of parents towards children with sex chromosome mosaicism DSD.

CONCLUSION

Parents need to proceed in order to accept children with sex chromosome mosaicism DSD. The main findings were that fathers struggle more than the mothers in accepting their children. Our finding suggests that timing of diagnosis in children will affect parents' acceptance of children, and its implications for parents' decisions about further management of children. It is important to investigate parents' acceptance starting from the beginning of a child's diagnosis and at each stage of child development. It is suggested to plan better interventions that can be provided by healthcare providers and genetic counselors to parents to increase the acceptance rate and decrease the rejection rate toward children with sex chromosome mosaicisms DSD.

ACKNOWLEDGMENTS

The authors are grateful to the patients and their families. The authors declare no conflict of interest.

REFERENCES

1. Abusaad FE, Yosr Mohamed Elmasri. Effect of counseling sessions on coping strategies and anxiety among parents of children with ambiguous genitalia. J Am Sci. 2011;7(5):674-682.
2. Risso R, Einaudi S, Crespi C, Caldarera A, Verna F, Merlini E, et al. Sex attribution, gender identity and quality of life in disorders of sex development due to 45,X/46,XY mosaicism: methods for clinical and psychosocial assessment. AIMS Genet. 2015;2(2):127-147.
3. Demaliaj E, Cerekja A, Piazze J. *Sex chromosome aneuploidies*. In Storchova Z, ed. Aneuploidy in health and disease. Croatia; Press INTECH. 2012 :p.121–140.
4. Rosa RFM, Bartel D'Ecclesiis WF, Dibbi RP, Rosa RCM, Trevisan P, Graziadio C, et al. 45,X/46,XY mosaicism: Report on 14 patients from a Brazilian hospital. A retrospective study. Sao Paulo Med J. 2014;132(6):328-334.
5. Chivers C, Burns J, Deiros Collado M. Disorders of sex development: Mothers' experiences of support. Clin Child Psychol Psychiatry. 2017; 22(4):675-690 http://journals.sagepub.com/doi/10.1177/1359104517719114
6. Sandberg DE, Gardner M, Callens N, Mazur T. Interdisciplinary care in disorders / differences of sex development (DSD): The psychosocial component of the DSD — Translational research network. Am J Med Genet C Semin Med Genet. 2017;175(2):279-292.
7. Duguid A, Morrison S, Robertson A, Chalmers J, Youngson G, Ahmed SF. The psychological impact of genital anomalies on the parents of affected children. Acta Paediatr. 2007;96(3):348-352.
8. Crissman HP, Warner L, Gardner M, Carr M, Schast A, Quittner AL, et al. Children with disorders of sex development: A qualitative study of early parental experience. Int J Pediatr Endocrinol. 2011;2011(1):10.
9. Ernst MM, Liao LM, Baratz AB, Sandberg DE. Disorders of sex development/Intersex: Gaps in psychosocial care for children. Pediatrics. 2018;142(2):e20174045.
10. Sanders C, Carter B, Goodacre L. Searching for harmony: parents' narratives about their child's genital ambiguity and reconstructive genital surgeries in childhood. J Adv Nurs. 2011;67(10):2220-2230.
11. Wolfe-Christensen C, Fedele DA, Kirk K, Mullins LL, Lakshmanan Y, Wisniewski AB. Caregivers of children with a disorder of sex development: Associations between parenting capacities and psychological distress. J Pediatr Urol. 2014;10(3):538-543.
12. Sanders C, Carter B, Goodacre L. Parents need to protect : Influences, risks and tensions for parents of prepubertal children born with ambiguous genitalia. J Clin Nurs. 2012; 21(21-22):3315-23.
13. Zair H, Ullah O, Khan WA, Khan F, Ali I. Sexual ambiguity — A social predicament: Case report. Dr Sulaiman Al Habib Med J. 2020;1(3-4):70-72.
14. Creswell JW. Research design qualitative, quantitative, and mixed methods approaches. Second Edition; 2003.
15. Listyasari NA, Santosa A, Juniarto AZ, Faradz SM. Multidisciplinary management of disorders of sex development in Indonesia, A prototype in developing country. J. Biomed. Transl. Res [Online]. 2017;3(1):17-22. https://doi.org/10.14710/jbtr.v3i1.1209
16. Rohner RP, Ali S. Parental acceptance-rejection questionnaire (PARQ). In: Zeigler-Hill V, Shackelford TK, eds. Encyclopedia of personality and individual differences. Cham: Springer International Publishing; 2016:1-4. https://doi.org/10.1007/978-3-319-28099-8_56-1.
17. Valentia S, Sani R, Anggreany Y. Hubungan antara resiliensi dan penerimaan orangtua pada ibu dari anak yang terdiagnosis autism spectrum disorder (ASD). J Psikol Ulayat. 2017;4(1):43-58.
18. Wisniewski AB. Psychosocial implications of disorders of sex development treatment for parents. Curr Opin Urol. 2017;27(1):11-13.
19. Braun V, Clarke V. Using thematic analysis in psychology. Qual. Res. Psychol. 2006;3:101–77.
20. Putnick DL, Lansford JE, Deater-deckard K. Agreement in mother and father acceptance-rejection, warmth, and hostility/rejection/neglect of children across nine countries. Cross-Cult Res. 2012;46(3):191-223.
21. Creighton S, Chernausek SD, Romao R, Ransley P, Pippi J. Timing and nature of reconstructive surgery for disorders of sex development- introduction. J Pediatr Urol. 2012; 8(6): 602-610. http://dx.doi.org/10.1016/j.jpurol.2012.10.001
22. Duberstein PR, Chen M, Chapman BP, Hoerger M, Saeed F, Guancial E, et al. Fatalism and educational disparities in beliefs about the curability of advanced cancer. Patient Educ Couns. 2018;101(1):113-118. https://doi.org/10.1016/j.pec.2017.07.007
23. Ruiu G. Is fatalism a cultural belief? An empirical analysis on the origin of fatalistic tendencies. Munich Pers RepEc Arch. 2012;(14).
24. Kohva E, Miettinen PJ. Disorders of sex development : Timing of diagnosis and management in a single large tertiary center. Endocr Connect. 2018;7(1):595-603.
25. Mouriquand PDE, Gorduza DB, Gay C, Meyer-Bahlburg HF, Baker L, Baskin LS, et al. Surgery in disorders of sex development with gender issues : If (why), when and how? J Pediatr Urol. 2016;12(3): 139-49. http://dx.doi.org/10.1016/j.jpurol.2016.04.001
26. Hemesath TP, de Paula LCP, Carvalho CG, Leite JCL, Guaragna-Filho G, Costa EC. Controversies on timing of sex assignment and surgery in individuals with disorders of sex development: A perspective. Front Pediatr. 2019;6(1):1-6.

27. Ediati A, Juniarto AZ, Birnie E, Okkerse J, Wisniewski A, Drop S, et al. Social stigmatisation in late identified patients with disorders of sex development in Indonesia. BMJ Paediatr Open. 2017;1:e000130. doi: 10.1136/bmjpo-2017-000130
28. Meyer-bahlburg HFL, Baratz K, Berenbaum SA, Cohen-Kettenis PT, Hines M, Schober JM. Gender assignment, reassignment and outcome in disorders of sex development : Update of the 2005 Consensus Conference. Horm Res Paediatr. 2016;85(2):112-118.
29. Deeb A, Khamis M, Sayed SA, Magdy Omar O, Odeh R, Ladjouze A, et al. Sex assignment practice in disorders of sexual differentiation: Survey results from paediatric endocrinologists in the Arab region. J Pediatr Endocrinol Metab. 2019;32(1):75-82.
30. Ediati A, Maharani N, Utari A. Sociocultural aspects of disorders of sex development. Birth Defect Res. 2016;108(4):380-383.
31. Kuhnle U, Krahl W. The impact of culture on sex assignment and gender development in intersex patients. Perspect Biol Med. 2002;45(1):85-103.
32. Ellens REH, Bakula DM, Mullins AJ, Scott Reyes KJ, Austin P, Baskin L, et al. Psychological adjustment of parents of children born with atypical genitalia 1 year after genitoplasty. J Urol. 2017;198(4):914-920. https://doi.org/10.1016/j.juro.2017.05.035