Open Access Maced J Med Sci **electronic publication ahead of print, published on May 13, 2019 as https://doi.org/10.3889/oamjms.2019.227**

ID Design Press, Skopje, Republic of Macedonia
Open Access Macedonian Journal of Medical Sciences.
https://doi.org/10.3889/oamjms.2019.227
eISSN: 1857-9655
Clinical Science

Functional and Cosmetic Urethroplasty Outcome, Emotional Stress after Genital Examination, Post Traumatic Stress Disorder, and Ages at the Time of Urethroplasty as Potential Risk Factor Causing Psychosocial Disorder of Hypospadia Children

Gede Wirya Kusuma Duarsa[1*], Dinar Ayu Pratiwi[2], Pande Wisnu Tirtayasa[1], Wayan Yudiana[1], Kadek Budi Santosa[1], Anak Agung Gde Oka[1], Sri Wahyuni[3], Tjokorda Gde Bagus Mahadewa[2]

[1]Department of Urology, Faculty of Medicine, Udayana University, Sanglah General Hospital, Bali, Indonesia; [2]Department of Surgery, Faculty of Medicine, Udayana University, Sanglah General Hospital, Bali, Indonesia; [3]Department of Psychiatry, Faculty of Medicine, Udayana University, Sanglah General Hospital, Bali, Indonesia

Citation: Duarsa GWK, Pratiwi DA, Tirtayasa PW, Yudiana W, Santosa KB, Oka AAG, Wahyuni S, Mahadewa TGB. Functional and Cosmetic Urethroplasty Outcome, Emotional Stress after Genital Examination, Post Traumatic Stress Disorder, and Ages at the Time of Urethroplasty as Potential Risk Factor Causing Psychosocial Disorder of Hypospadia Children. Open Access Maced J Med Sci. https://doi.org/10.3889/oamjms.2019.227

Keywords: PSC-17; HOPE score; GEDS; CRIES-13; PTSD

***Correspondence:** Gede Wirya Kusuma Duarsa. Faculty of Medicine, Udayana University, Bali, Indonesia. E-mail: gwkduarsa@yahoo.com

Received: 01-Jan-2019; **Revised:** 05-Apr-2019; **Accepted:** 06-Apr-2019; **Online first:** 13-May-2019

Funding: This research did not receive any financial support

Competing Interests: The authors have declared that no competing interests exist

Abstract

BACKGROUND: Children with hypospadias, being born with congenital abnormalities, having repeated genital examination, hospitalization, and undewent genital surgery, experienced psychological stress that may negatively affect their psychosocial life. Choosing the proper time of surgery as recommended is important, since it may have a positive impact on the psychosocial adaptation.

AIM: This study aims to find the risk factors causing psychosocial disorders in post-repair surgery on hypospadias children.

METHODS: This is a case control study, from 203 hypospadias patients underwent urethroplasty from 2009 to 2018. Subjets were screened for psychosocial disorders by Pediatric Symptoms Questionnaire 17 (PSC-17) questionnaire to find those with psychosocial disorders, score 15 or more (case group) and those without psychosocial disorders (control group), score 0-14. We traced back the data retrospectively from both group (history of utrethrocutaneous fistula and meatal stenosis, age upon urethroplasty) and collecting new ones (cosmetic outcome, emotional stress after genital examination, and the existence of PTSD). Fisher's exact test was performed to see the Odds ratio (OR) for each variable.

RESULTS: Some children with hypospadias show impaired on psychocosial. Functional and cosmetic outcome not significantly different as potensial risk factor psychosocial disorders, genital examination doesn't trigger psychological stress and also none children show PTSD symtomp after surgery. Comparison time of age urethroplasty did not differ significantly between two group

CONCLUSIONS: Twenty-nine children post urthroplasty show psychosocial disorders. Functional and cosmetic urethroplasty outcomes, emotional stress after genital examination, post-traumatic stress disorder were not risk factors of pshycososcial disorder of hypospadias patients. Ages at time of surgery did not differ significantly between two group and this is contradict to the previous recommendations.

Introduction

Hypospadias is defined as an abnormal opening of the urethra on the underside of the penis and is a frequently found malformation with an incidence varies from 0.04-0.06 per 100.000 births [1]. Previous study reported the incidence of hypospadias is 1.86 per 100.000 population [2].

This malformation affects the genital development and patients with severe hypospadias presenting as ambiguous genitalia have serious and potentially life-long consequences and can affect their psychosocial adaptation. The surgery itself, together with the genital examination, hospitalization, eventual separation from the parents in perioperative periods,

and postoperative pain, can be a stressful experience for the affected children; those could be mentioned as potential stress factors for these children and may negatively affect psychosocial adaptation [3].

Choosing the best time for surgery is important. It can have a positive impact on the psychosocial adaptation. It is generally agreed, that at certain ages, a child may be more vulnerable to psychological stress related to surgery [3], [4], [5]. Therefore, the American Academy of Pediatrics considers the age between 6 and 12 months to be the best time for surgery for hypospadias children [6]. Emotional development, acquisition of body image and sexual identity, as well as cognitive development begin in early childhood ages. Genital awareness and a period of special emotional risks occurs around the age of 18 months and therefore, it was suggested to complete surgery before this age [4], [6]. However, performing a hypospadias repair surgery is not always an easy task, especially in children with relatively smaller penis size.

The goal of this study is to find the risk factors causing psychosocial disorders in children with hypospadias.

Patients and Methods

This was a case-control study conducted with consecutive sampling. This research has been reviewed by the Committee of Ethical Research of our University. All participating subjects or their legal guardians have provided written consent to be included in this study. As many as 203 hypospadias patients underwent urethroplasty between 2009 and 2018 were contacted and they were screened for psychosocial disorders by PSC-17 questionnaire. After analyzing the PSC-17 results, we divided them into two groups, those without psychosocial disturbance (control group) and those with psychosocial disorders (case group).

Data collection were then implemented as we traced the history of each subject. Data includes the age when the subject underwent surgery, a history of urethrocutaneous fistula and meatal stenosis, surgery cosmetics outcome (measured by Hypospadias Objective Penile Evaluation/HOPE score), emotional stress after genital examination (measured by Genital Examination Distress Scale/GEDS), and the presence of post-traumatic stress disorder (PTSD) screening by the Children's Impact of Event Scale 13 (CRIES-13) questionnaire. Those with incomplete or undetermined data were excluded from this study.

Data analysis was conducted by SPSS 23.0 software for Windows. Fisher exact test was used to determine the odds ratio (OR) value for each variable. A p-value of < 0.05 was considered significant.

Results

From the total of 203 patients, only 29 subjects had the psychological disorder that can be enrolled in this study. Patients who underwent surgery at age of < 5 years old were 17 patients (58.6%), while those who underwent surgery over 5 years were 12 patients (41.4%), with mean age 5.9 ± 3.9 years in case group and 5.53 ± 2.54 years in the control group, it also assessed the level of parent education to describe the socio-economic condition of patients. In case group, 1 patient (10%) had a low socioeconomic level, while 9 patients (90%) had a high socioeconomic level, and in the control group it was observed that 3 patients (15.8%) had low socioeconomic status and 16 patients (84.2%) had a social level high level of education. Parenting education is very important in children due to the ability of parenting assistance in accompanying children and educate children, especially those with hypospadias that require special attention. Characteristics of the objects in this study can be seen in Table 1.

Table 1: Characteristics of research subjects based on the study group

Variable	Group	
	Case (n = 10)	Control (n = 19)
Age(years), mean ± SD	5.9 ± 3.9	5.53 ± 2.54
Sosioeconomic status, n (%)		
Low	1 (10%)	3 (15.8%)
High	9 (90%)	16 (84.2%)
HOPE[a] score, mean ± SD	47.5 ± 4.3	50 ± 4.2

HOPE: Hypospadias Objective Penile Evaluation.

To assess the risk factors psychosocial disorders of hypospadias who underwent urethroplasty, we conducted bivariate analysis to determine the odds ratio (OR) value for each variable with Fisher's exact test.

Table 2: Fisher's exact test results of independent variables

Variable	Group		OR		
	Case (n = 10)	Control (n = 19)		95%CI	p-value
HOPE[a], n (%)					
≤ 45	3 (30)	3 (15.8%)	2.28	0.367-14.253	0.331
> 45	7 (70)	16 (84.2%)			
Urethrocutan fistula, n (%)					
Yes	4 (40%)	2 (10.5)	5.66	0.818-39.267	0.086
No	6 (60%)	15 (89.5)			
Meatal stenosis, n (%)					
Yes	4 (40%)	2 (10.5)	5.66	0.818-39.267	0.086
No	6 (60%)	15 (89.5)			
GEDS[b], n (%)					
Negative	10 (100%)	19 (100%)			
CRIES-13[c], n (%)					
Negative	10 (100%)	19 (100%)			
Age, n (%)					
≤ 5 yo	6 (60)	11 (57.9)	1.091	0.23-5.185	0.615
> 5 yo	4 (40)	8 (42.1)			
Sosioeconomic status, n (%)					
Low	1 (10%)	3 (15.8%)	0.593	0.53-6.572	0.571
High	9 (90%)	16 (84.2%)			

HOPE = Hypospadias Objective Penile Evaluation; GEDS = Genital Examination Distress Scale; CRIES-13 = Children's Impact of Event Scale 13.

Poor HOPE score was found more in the case group compared to the control group, but it was not statistically significant (30% vs. 15.8%, OR = 2.28, p = 0.331). The results of the bivariate analysis of risk factors calculated in this study can be seen in Table 2.

Discussion

Most hypospadias children showed disturbance on internalization aspect. The patient tended to have a low self-esteem and did not have the confidence due to the genital abnormality. Recent epidemiological results show an increased prevalence of intellectual disability, autism spectrum disorder (ASD), attention deficit hyperactivity disorder (ADHD), and behavioral/emotional disorders in hypospadias compared to the normal population of the same age [3], [7], [8].

Three patients in this study showed symptoms of attention disorder in which the child was unable to focus on something and tended to appear hyperactive. One of them was diagnosed with ADHD and needed psychiatric help, but this was already diagnosed before the surgery.

Statistically, functional and cosmetic were not known to be a risk factor for psychosocial disorders in children with hypospadias. Both case and control group had more subjects with satisfactory HOPE score (70% vs. 84.2%). The severity of the hypospadias type does not affect the cosmetic outcome, as shown in the previous studies [9], [10].

Fistulas tend to develop into psychosocial disorders. It has been reported that 30% of minor fistula cases after hypospadias repair recovered themselves after 10-21 days [11]. The urethrocutaneous fistula usually occurs 7-10 after the surgery. The significant factors in the formation of urethrocutaneous fistula are infection and ischemia [11].

Several methods are available to reduce the risk of urethrocutaneous fistula formation, such as using subcutaneous sutures, the use of catheter after the surgery, avoiding repeated surgical wound care, and using fibrin glue [11]. In addition, fistula incidence can also be minimized by post-urethroplasty cystostomy. The incidence of post-urethroplasty fistula with cystostomy was 2.9% while those without cystostomy were 17.6% [12].

American Academy of Pediatrics stated that the age of 6-12 months as the ideal age for the implementation of hypospadias surgery. This recommendation is based on considerations of surgery, anesthesia, and child psychology factors, such as cognitive development, genital awareness, emotional development, and psychosexual development. Additionally, the next ideal age group or the second window period is above the age previously mentioned for urethroplasty between the ages of 2.5-5 years [6], [13], [14]. Patients at this age are considered not able to remember the trauma of a surgery and not aware of the body's perception and sexual identity. This was the reason why we took the 5-years old cutoff point. In Indonesia, hypospadias surgery are done mostly at the age of above 5 years old due to differences in penis size of Indonesians [7].

The results of this study denied the previous recommendations. The age at the surgery was not statistically significant in influencing the occurrence of psychosocial disorders. Theoretically, children with a genital abnormality such as hypospadias tend to have psychological effects that may lead to post-traumatic stress disorder [3], [15], [16]. From the GEDS questionnaire we discovered that no patients in our study experienced emotional stress. This is different from some studies that showed genital examination had a negative impact on children compared to the examination of the ear and oral cavity [17]. Other study reported that children who underwent surgery in the genitourinary area had a high level of emotional distress compared with children who underwent throat ear surgery [3].

As many as 33% of children with hypospadias who underwent surgery experienced emotional disturbance, with 17% of those came to psychiatric to seek help [3], [14], [16]. We screened for PTSD with a CRIES-13 questionnaire that can be used in children younger than 5-years old [18], [20]. In this study, none of the children experienced PTSD symptoms. When asked what if they had to undergo the surgical procedure again, they did not show fear. The history of surgery does not interfere their daily lives. Parental care and guidance are important in the perioperative period. Parents with good education level may explain and provide a proper understanding to the affected child.

In conclusion, children with hypospadias show psychosocial impairment. They mostly suffer from internalization and attention problem. Age at the time of surgery did not differ significantly between the two groups, and this is contradicting to the previous recommendations. Other functional and cosmetic outcomes were also not statistically different. Genital examinations did not trigger an emotional stress. In our study, none developed PTSD symptoms after undergoing urethroplasty.

References

1. Shih EM, Graham JM Jr. Review of genetic and environmental factors leading to hypospadias. Eur J Med Genet. 2014; 57:453-463. https://doi.org/10.1016/j.ejmg.2014.03.003 PMid:24657417

2. Bergman JEH, Loane M, Vrijheid M, et al. Epidemiology of hypospadias in Europe: a registry-based study. World Journal of Urology. 2015; 33(12):2159-2167. https://doi.org/10.1007/s00345-015-1507-6 PMid:25712311 PMCid:PMC4655014

3. Blotcky MJ, Grossman I. Psychological implications of childhood genitourinary surgery. An empirical study. J Am Acad Child Psychiatry. 1978; 17(3):488-97. https://doi.org/10.1016/S0002-7138(09)62303-7

4. Hadidi AT. Classification of Hypospadias. In: Hypospadias Surgery. 1st ed. Berlin Heidelberg: Springer-Verlag, 2004:79-82. https://doi.org/10.1007/978-3-662-07841-9

5. Sandberg DE, Meyer-Bahlburg HF, Hensle TW, Levitt SB, Kogan SJ, Reda EF. Psychosocial adaptation of middle childhood boys with hypospadias after genital surgery. J Pediatr Psychol. 2001; 26(8):465-75. https://doi.org/10.1093/jpepsy/26.8.465 PMid:11700331

6. Springer A, Tekgul S, Subramaniam R. An Update of Current Practice in Hypospadias Surgery. Eur Urol Suppl. 2017; 16(1):8-15. https://doi.org/10.1016/j.eursup.2016.09.006

7. Saksono YWS, Suryadhi NT, Duarsa GWK. Tipe hipospadia proksimal merupakan faktor risiko pancaran urin yang lemah pada pasien hipospadia pasca uretroplasti teknik tubularized incised plate. Medicina. 2017; 48(3): 220-223. https://doi.org/10.15562/medicina.v48i3.163

8. Duarsa G, Nugroho T, Mahadewa T, Oka A, Yasa K, Suryawisesa IM. Cosmetic outcome of tubularized incised plate depends on the type of hypospadias: A case control study. Bali Medical Journal. 2016; 5(2): 355-357. https://doi.org/10.15562/bmj.v5i2.238

9. Wilkinson DJ, Farrelly P, Kenny SE. Outcomes in distal hypospadias: A systemic review of the Mathieu and tubularized incised plate repairs. Journal of Pediatric Urology. 2012; 8(3):307-312. https://doi.org/10.1016/j.jpurol.2010.11.008 PMid:21159560

10. Snodgrass WT, Granberg C, Bush NC. Urethral strictures following urethral plate and proximal urethral elevation during proximal TIP hypospadias repair. Journal of Pediatric Urology. 2013; 9(6):990-994. https://doi.org/10.1016/j.jpurol.2013.04.005 PMid:23707201

11. Shehata S, Hashish M. Management of Post Hypospadias Urethral Fistula. In: Current Concepts of Urethroplasty. Rijeka: InTech; 2011:47-60. https://doi.org/10.5772/22653

12. Laura SF, Duarsa GWK, Mahadewa TGB. Correlation Of Cystostomy To Low Urethrocutaneous Fistula Incident In Hypospadia Surgery. Bali Medical Journal. 2014; 3(3):125. https://doi.org/10.15562/bmj.v3i3.88

13. Yildiz T, Tahtali IN, Ates DC, Keles I, Ilce Z. Age of Patient is a Risk Factor for Urethrocutaneous Fistula in Hypospadias Surgery. J jpurol. 2013; 9(6):900-903. https://doi.org/10.1016/j.jpurol.2012.12.007 PMid:23290687

14. Schulz JR, Klykylo WM, Wacksman J. Timing of elective hypospadias repair in children. Pediatrics. 1983; 71(3):342-351.

15. Butwicka A, Lichtenstein P, Landen M, et al. Hypospadias and increased risk for neurodevelopmental disorders. J Child Psychol Psychiatry. 2015; 56(2):155-161. https://doi.org/10.1111/jcpp.12290 PMid:25048198

16. Ortqvist L, Fossum M, Andersson M, et al. Long-term follow-up of men born with hypospadias: urological and cosmetic results. J Urol. 2015; 193(3):975-81. https://doi.org/10.1016/j.juro.2014.09.103 PMid:25268894

17. Gully KJ, Britton H, Hansen K, Goodwill K, Nope JL. A new measure for distress during child sexual abuse examinations: the genital examination distress scale. Child Abuse Negl. 1999; 23(1):61-70. https://doi.org/10.1016/S0145-2134(98)00111-2

18. Smith P, Perrin S, Dyregrov A, Yule W. Principal components analysis of the Impact of Event Scale with children in war. Personality and Individual Differences. 2003; 34(2):315-322. https://doi.org/10.1016/S0191-8869(02)00047-8

19. Smith P, Perrin S, Yule W, Hacam B, Stuvland R. War exposure among children from Bosnia-Hercegovina: psychological adjustment in a community sample. J Trauma Stress. 2002; 15(2):147-156. https://doi.org/10.1023/A:1014812209051 PMid:12013066

20. Verlinden E. Time does not heal all wounds: Identifying children suffering from psychological trauma. University of Amsterdam - Digital Academic Repository, 2014.