

Common Practice of Hypospadias Management by Pediatric Urologists in Indonesia: A Multi-center Descriptive Study from Referral Hospitals

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Abstract

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Competing Interests: The authors have declared that no competing interests exist **BACKGROUND:** Hypospadias is the second most common congenital anomalies among human congenital disabilities. There are over 300 surgery techniques being introduced to treat hypospadias. The successful of hypospadias repair is assessed by several outcomes as well as complications following surgery.

AIM: This study aims to show the multicenter hypospadias data in Indonesia descriptively.

METHODS: All the data were compiled based on questionnaires, which were distributed to Indonesian pediatric urologists. The questionnaire includes several questions containing demographic aspect, preferred techniques being used, and complications being found regarding hypospadias repair.

RESULTS: Eighteen Indonesian pediatric urologists from 12 centres involved in this study. The data were collected from June – September 2018 based on the surgeon's experience throughout 2017. From 591 cases based on the returned questionnaire, penile-type hypospadias was the most common type of hypospadias being treated (35.7%) followed by penoscrotal (28.9%) and scrotal-type (12.9%). Moderate severity of chordee was mostly seen among all cases (40.6%). Tubularised incised plate (TIP), + Thiersch Duplay, was the most common technique being used to treat hypospadias (44.3%), followed by onlay island preputial flap (14.9%) and two-stage technique (14%). The incidence of urethrocutaneous fistulae in this study was 13.9%.

CONCLUSION: This study showed how Indonesian pediatric urologists dealt with hypospadias cases. TIP + Thiersch Duplay procedure being the preferred technique used by most participants and the rate of urethrocutaneous fistulae as one of the complications was comparable with previous studies.

Introduction

among human congenital disabilities [1], [2]. The prevalence of hypospadias is around 1 of 250-300 live male births [3]. Hypospadias repair is being performed with some aims such as enable micturition in standing position, good cosmetic appearance, as well as

Several studies reported that hypospadias is the second most common congenital anomalies

effective insemination [4], [5]. Currently, there are over 300 surgery techniques being introduced as management of hypospadias, and some newer methods continue to evolve and introduce. However, there are no standard methods to treat all hypospadias. Over the last decades, the surgery is subtle, and it now has a decreased stage of operations as well as postoperative complications [1], [4].

Distal hypospadias has been treated as a one-stage procedure currently as reported by many studies internationally. Transverse incised plate (TIP), Mathieu and Onlay island flap are being some of the techniques most used to treat distal hypospadias. Nevertheless, for proximal hypospadias, the surgical treatment is still debatable into two groups based on the stage of procedures. One group favour one stage procedure such as inlay preputial flap while other groups choose to perform two-stage procedure [6], [7].

The successful of hypospadias repair is assessed by several outcomes, out of which, the complications being the most concerns of all surgeons. Some of the complications after hypospadias repair include urethrocutaneous fistulae, urethral strictures, infections, meatal stenosis and others fewer common ones [8]. Some factors could affect the outcome of hypospadias repairs such as the site of the meatus, the severity of chordee, adequacy of preputial skin and the existence of penoscrotal transposition.

Other factors, including the age of the patients and surgeon's experience, could be counted as well [9], [10]. Some technical factors such as the type of surgery, second layer usage, duration of antibiotic and stenting duration are also notable [2], [11].

Regarding our knowledge, this is the first descriptive multicenter study regarding the hypospadias repair data being performed by Indonesian pediatric urologists and this study aimed to show the multicenter hypospadias data in Indonesia descriptively in terms of demographics, techniques being used, and outcomes in 12 institutions from across Indonesia.

Material and Methods

All the data were compiled from selfconstructed questionnaires, which were distributed to Indonesian pediatric urologists who have been completed the pediatric urology trainee to ensure their competence as well as homogeneity handling techniques. The data were collected and analysed from June – September 2018 based on the surgeon's experience throughout the year of 2017. The questionnaire includes several questions containing demographic aspect, preferred techniques being used, and complications being found regarding hypospadias repair.

As this study was being held, there were 18 pediatric urologists in Indonesia. Ethical approval for this study has been granted by the local Ethics Committee.

Results

Eighteen Indonesian pediatric urologists from 12 centres took part in this study. Of the returned questionnaires and through final analysis, we included data from 591 cases.

However, some questionnaires were returned with incomplete answers. In this case, we were trying to collect data as much and as relevant as we can. The distributions of hypospadias data based on hospital centres are shown in Table 1.

Table 1: The distributions of hypospadias data based on	the
hospital centres	

Hospital Center (City)	n (%)
Sanglah (Denpasar)	83 (14)
Cipto Mangunkusumo (Jakarta)	59 (10)
Hasan Sadikin (Bandung)	44 (7.5)
Saiful Anwar (Malang)	30 (5.1)
Sardjito (Yogyakarta)	28 (4.7)
Wahidin Sudirohusodo (Makassar)	27 (4.6)
Harapan Kita (Jakarta)	48 (8.1)
Fatmawati (Jakarta)	119 (20.1)
M. Djamil (Padang)	41 (6.9)
Soetomo (Surabaya)	56 (9.5)
Adam Malik (Medan)	25 (4.2)
Persahabatan (Jakarta)	31 (5.3)
Total (%)	591 (100)

The distributions of the age group of hypospadias patients about the occurrence of urethrocutaneous fistulae after a repair, the techniques of hypospadias repair in relation to type of hypospadias, and the severity of chordee are shown in Table 2, 3, and 4, respectively.

Table 2: The distributions of age group of hypospadia	IS
patients in relation to the occurrence of urethrocutaneou	IS
fistulae after hypospadias repair	

	Urethrocutar	~ (0/)		
Age group (years)	Yes	No	n (%)	
0-1	-	15	15 (2.5)	
>1-2	3	36	39 (6.6)	
>2-3	3	58	61 (10.3)	
>3-4	14	101	115 (19.5)	
>4	62	299	361 (61.1)	
Total (%)	82 (13.9)	509 (86.1)	591 (100)	

The length of neo-urethra during hypospadias repair, the placement of percutaneous cytostomy during hypospadias repair, and the size of urethral splint are shown in Table 5, 6, and 7 respectively.

The complication founds after hypospadias

repair other than urethral fistulae was 14 cases (4%) from 350 cases.

Table 3: Techniques of hypospadias repair being used about the type of hypospadias

Urethroplasty	Glandula		Penile	Penoscrot	Scrotal	Perine	Chorde	Failed	Total
Technique	r	al	i crinc	al	ocrotai	al	e only	urethroplasty	(%)
Chordectomy and orthoplasty	-	14	-	-	-	-	3	-	17 (2.8)
MAGPI	13	12	-	-	-	-	-	-	25 (4.2)
Mathieu	1	9	6	-	-	-	-	-	16 (2.7)
TIP + Thiersch Duplay	10	29	130	56	25	3	-	8	261 (44.3)
Onlay island preputial flap		-	39	35	13	1	-	-	88 (14.9)
Duckett	-	3	10	25	7	1	-	-	46 (7.8)
Dorsal inlay preputial flap or graft	-	-	15	16	2	2	-	-	35 (5.9)
Koyanagi	-	-	5	4	1	-	-	-	10 (1.7)
Two-stage technique	-	1	6	35	28	10	-	3	83 (14)
Fistulae repair	-	-	-	-	-	-	-	10	10 (1.7)
Total (%)	24 (4.1)	68 (11.5)	211 (35.7)	171 (28.9)	76 (12.9)	17 (2.8)	3 (0.5)	21 (3.6)	591 (100)

Discussion

Hypospadias is being one of the common congenital anomalies of the penile [12]. Currently, there have been more than 300 surgery techniques in hypospadias repair [1].

Table 4: The severity of chordee

Severity of chordee	n (%)
Mild	118 (28.7)
Moderate	167 (40.6)
Severe	126 (30.7)
Total (%)	411 (100)

All of those techniques have the same goal, which is to achieve cosmetically appropriate penile with acceptable shaped of penile glans that has a meatus at the tip of the penile.

Table 5: The length of neo-urethra during hypospadias repair

The length of neo-urethra (cm)	n (%)
< 1	28 (8.4)
1-2	79 (23.7)
> 2-3	107 (32.1)
> 3-4	83 (24.9)
> 4	36 (10.9)
Total (%)	333 (100)

Nonetheless, high rates of complications still be an issue in hypospadias repair compared to other reconstructive surgeries. Moreover, until currently, there is no same consensus about the preference procedures to treat any hypospadias.

Table 6: The placement of percutaneous cystostomy during hypospadias repair

Placement of percutaneous cystostomy	n (%)
No	213 (61.1)
Yes	136 (38.9)
Total	349 (100)

The urethrocutaneous fistulae rate as a complication

in our study was quite comparable with previous studies. The rate of urethrocutaneous fistulae in our study was 13.9%. Results from other literature were quite varying from 4-60%.

Table 7: The size of the urethral splint during hypospadias repair

The size of urethral splint (Fr)	n (%)
6	127 (32.6)
В	181 (46.4)
10	45 (11.5)
12	27 (6.9)
14	9 (2.3)
16	1 (0.3)
Total	390 (100)

However, the higher rates of complications were commonly in the studies with more severe hypospadias [6], [7], [8], [13], [14], [15], [16], [17], [18], [19], [20], [21], [22]. We have also noticed in this study that higher complication rate was associated with severe chordee as well as proximal hypospadias. The prevalence of proximal hypospadias (penoscrotal, scrotal, and perineal) was very high in this study (44.7%) as compared to other studies [9], [22], [23], [24]. This issue might be due to some of the distal hypospadias patients did not seek medical advice. Also, all Indonesian pediatric urologists work in each hospital centre where lots of severe cases will be referred to them from all around the country.

Hypospadias repair is recommended to perform around age 6 - 18 months. Some studies even showing minimal complication of hypospadias repair in patients with age 4 - 6 months [4], [10], [20]. In our study, most of the cases being treated were older than 4-year-old (61.1%). One of the main reasons for this problem was most of the cases were came to a physician at an older age, and most of the patients were coming from a distant area.

Tabularized incised plate (TIP) procedure has become very popular since Snodgrass introduced his initial technique of TIP for hypospadias repair in 1994. This technique is very popular for the treatment of distal hypospadias [25]. The treatment of proximal hypospadias is more challenging. A surgeon must be ready to use some different techniques to deal with proximal hypospadias. The majority of Indonesian pediatric urologists prefer TIP technique (44.3%) as a management to treat hypospadias which is comparable to another study [6], [22], [23], [24], [25], [26]. Not only for distal hypospadias, but TIP technique was also used for proximal hypospadias which had very mild chordee and only minimal dissection required for correction. Nonetheless, it should be considered that the TIP technique has a greater risk to produce an unsatisfying cosmetic outcome as management of proximal hypospadias [27].

The previous study has shown the correlation of cystostomy placement to a low incident of urethrocutaneous fistulae following hypospadias surgery [28]. Nevertheless, most of the Indonesian pediatric urologists prefer not to use cystostomy in this study. It is understandable that the placement of percutaneous cystostomy in hypospadias surgery cannot be applied to all cases because it needs a thoughtful decision from one case to another. A large number of surgeons use urethral stenting following hypospadias repair. However, until currently, there is no agreement in terms of its need, size, or material to be used [29]. In this study, all participants use urethral stenting for their patients.

In conclusion, this multi-centre descriptive study showed how Indonesian pediatric urologists dealt with hypospadias cases which were comparable to international level practices. TIP procedure is the preferred technique used by most participants, and the rate of urethrocutaneous fistulae as one of the complications was comparable with previous literature as well.

References

1. Cimador M, Vallasciani S, Manzoni G, Rigamonti W, De Grazia E, Castagnetti M. Failed hypospadias in paediatric patients. Nat Rev Urol. 2013; 10(11):657-66.

https://doi.org/10.1038/nrurol.2013.164 PMid:23917119

2. Goel P, Bajpai M, Verma A. Recent advances in hypospadias. JIMSA. 2014; 27(2):95-100.

3. Baskin LS. Hypospadias, anatomy, embryology, and reconstructive techniques. Braz J Urol. 2000; 26(6):621-9.

4. Zavitsanakis A, Gougoudi E. Timing of elective hypospadias repair. In Hypospadias Surgery Springer, Berlin, Heidelberg, 2004:83-85. <u>https://doi.org/10.1007/978-3-662-07841-9_8</u>

5. Ziada A, Hamza A, Abdel-Rassoul M, Habib E, Mohamed A, Daw M. Outcomes of hypospadias repair in older children: a prospective study. J Urol. 2011; 185(6):2483-6. https://doi.org/10.1016/j.juro.2011.01.032 PMid:21555019

6. Cook A, Khoury AE, Neville C, Bagli DJ, Farhat WA, Pippi Salle JL. A multicenter evaluation of technical preferences for primary hypospadias repair. J Urol. 2005; 174(6):2354-7. https://doi.org/10.1097/01.ju.0000180643.01803.43 PMid:16280842

7. Springer A, Krois W, Horcher E. Trends in hypospadias surgery: results of a worldwide survey. Eur Urol. 2011; 60(6):1184-9. https://doi.org/10.1016/j.eururo.2011.08.031 PMid:21871708

8. Bhat A, Mandal AK. Acute postoperative complications of hypospadias repair. Indian J Urol. 2008; 24(2):241-8. https://doi.org/10.4103/0970-1591.40622 PMid:19468404 PMCid:PMC2684260

9. Hadidi AT. Classification of hypospadias. InHypospadias surgery 2004 (pp. 79-82). Springer, Berlin, Heidelberg. https://doi.org/10.1007/978-3-662-07841-9_7

10. Bhat A. General considerations in hypospadias surgery. Indian J Urol. 2008; 24(2):188. <u>https://doi.org/10.4103/0970-1591.40614</u> PMid:19468396 PMCid:PMC2684287

11. Djordjevic ML, Perovic SV, Slavkovic Z, Djakovic N. Longitudinal dorsal dartos flap for prevention of fistula after a Snodgrass hypospadias procedure. Eur Urol. 2006; 50(1):53-7. https://doi.org/10.1016/j.eururo.2006.04.014 PMid:16707207

12. Rynja SP, de Jong TPVM, Bosch JLHR, de Kort LMO. Testosterone prior to hypospadias repair: Postoperative complication rates and long-term cosmetic results, penile length and body height. J Pediatr Urol. 2018; 14:31. https://doi.org/10.1016/j.jpurol.2017.09.020 PMid:29174377

13. Jan IA, Mirza F, Yaqoot AM, Arian A, Saleem N, Ahmad KD. Factors influencing the results of surgery for hypospadias: experience at NICH. J Pak Med Assoc. 2004; 54:577-9.

14. Borer JG, Bauer SB, Peters CA, Diamond DA, Atala A, Cilento BG, et al. Tubularized incised plate urethroplasty: expanded use in primary and repeat surgery for hypospadias. J Urol. 2001; 165(2):581-5. <u>https://doi.org/10.1097/00005392-200102000-00075</u> PMid:11176441

15. Snodgrass WT, Bush N, Cost N. Tubularized incised plate hypospadias repair for distal hypospadias. J Pediatr Urol. 2010; 6(4):408-13. <u>https://doi.org/10.1016/j.jpurol.2009.09.010</u> PMid:19837000

16. McNamara ER, Schaeffer AJ, Logvinenko T, Seager C, Rosoklija I, Nelson CP, et al. Management of Proximal Hypospadias with 2-Stage Repair: 20-Year Experience. J Urol. 2015; 194(4):1080-5. <u>https://doi.org/10.1016/j.juro.2015.04.105</u> PMid:25963188 PMCid:PMC4575613

17. Tiryaki T. Combination of tubularized island flap and ventral skin flap techniques in single-stage correction of severe proximal hypospadias. Urol Int. 2010; 84(3):269-74. https://doi.org/10.1159/000288227 PMid:20389154

18. Braga LHP, Pippi Salle JL, Lorenzo AJ, Skeldon S, Dave S, Farhat WA, et al. Comparative analysis of tubularized incised plate versus onlay island flap urethroplasty for penoscrotal hypospadias. J Urol. 2007:1451-1456. <u>https://doi.org/10.1016/j.juro.2007.05.170</u> PMid:17706707

19. Spinoit A-F, Poelaert F, Groen L-A, Van Laecke E, Hoebeke P. Hypospadias repair at a tertiary care center: long-term followup is mandatory to determine the real complication rate. J Urol. 2013; 189(6):2276. <u>https://doi.org/10.1016/j.juro.2012.12.100</u> PMid:23306089

20. Perlmutter AE, Morabito R, Tarry WF. Impact of patient age on distal hypospadias repair: a surgical perspective. Urology. 2006; 68(3):648-51. <u>https://doi.org/10.1016/j.urology.2006.03.079</u> PMid:16979730

21. Hagos M. Hypoaspadias Repair: Review of Techniques and Treatment Outcomes in Mekelle Hospital, Ethiopia. Ethiop Med J. 2017; 55(2):115-120.

22. Mammo TN, Negash SA, Negussie T, Getachew H, Dejene B, Tadesse A, Derbew M. Hypospadias repair in Ethiopia: A five year review. Ethiop J Sci. 2018; 28(6):735.

23. Dipaola G, Spalletta M, Balducci T, Giacomello L, Camoglio FS, Bianchi S, et al. Surgical treatment of chordee without hypospadias. Eur Urol. 2000; 38(6):758-761. https://doi.org/10.1159/000020375 PMid:11111197

24. Khan M, Majeed A, Hayat W, Ullah H, Naz S, Shah SA, et al. Hypospadias repair: a single centre experience. Plast Surg Int. 2014; 2014. <u>https://doi.org/10.1155/2014/453039</u> PMid:24579043 PMCid:PMC3918360

25. Snodgrass W. Tubularized, incised plate urethroplasty for distal hypospadias. J Urol. 1994; 151:464-5. https://doi.org/10.1016/S0022-5347(17)34991-1

26. Duarsa GWK, Nugroho TD. Characteristics of hypospadias cases in Sanglah general hospital, Bali-Indonesia: A descriptive study. Bali Med J. 2016; 5(1):12-4. https://doi.org/10.15562/bmi.v5i1.185

27. Duarsa GWK, Nugroho TD, Mahadewa TGB, Yasa KP, Suryawisesa IBM, Oka AAG. Cosmetic outcome of tubularized incised plate depends on the type of hypospadias: A case control study. Bali Med J. 2016; 5(2):355-7. https://doi.org/10.15562/bmj.v5i2.238

28. Laura SF, Duarsa GWK, Mahadewa TG. Correlation of cystostomy to low urethrocutaneous fistula incident in hypospadias surgery. Bali Med J. 2014; 3(3):125-8. https://doi.org/10.15562/bmj.v3i3.88

29. Chalmers DJ, Siparsky GL, Wiedel CA, Wilcox DT. Distal

hypospadias repair in infants without a postoperative stent. Pediatr Surg Int. 2015; 31:287-90. <u>https://doi.org/10.1007/s00383-014-</u> <u>3647-y</u> PMid:25475503