

RISK FACTORS FOR URETHROCUTANEOUS FISTULAS FORMATION AFTER ONE STAGE HYPOSPADIAS REPAIR

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ABSTRACT

Objective: To evaluate risk factors that contribute to urethrocuteaneous fistulas formation after one stage hypospadias repair. **Material & method:** A case control study was performed on hypospadias patients that underwent one stage hypospadias repair. We analyzed the correlation of urethrocuteaneous fistula formation with patient age, hypospadias classification, chordee severity, other urogenital anomalies, history of hormonal therapy, suture size, duration of operation, type of dressing, type of stent, duration of stenting, and three types of operation technique, which are TIP, Duckett, and Onlay Island Flap. **Results:** There were 116 patients with mean age $5,7 \pm 3,9$ years old (4 months - 19 years old). Urethrocuteaneous fistula occurred in 12 patients (10,3%). From the data analysis, we didn't find any significant correlation between urethrocuteaneous fistula formation and patient's age ($p = 0,426$), hypospadias classification ($p = 0,695$), chordee severity ($p = 0,564$), other urogenital anomalies ($p = 0,964$), history of hormonal therapy ($p = 0,739$), suture size ($p = 0,248$), duration of operation ($p = 0,856$), type of dressings ($p = 0,580$), type of stents ($p = 0,600$), and duration of stenting ($p = 0,796$). We also didn't find any significant correlation between urethrocuteaneous fistula formation and operation technique TIP vs Duckett ($p = 0,314$), and TIP vs Onlay Island Flap ($p = 0,644$). **Conclusion:** There were no significant correlation between urethrocuteaneous fistula formation and patient age, hypospadias classification, chordee severity, other urogenital anomalies, history of hormonal therapy, suture size, duration of operation, type of dressing, type of stent, and duration of stenting. There were also no significant correlation between urethrocuteaneous fistula formation and operation technique TIP vs Duckett, and TIP vs Onlay Island Flap.

Keywords: Hypospadias, one stage urethroplasty, urethrocuteaneous fistula.

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INTRODUCTION

One-stage hypospadias repair has recently been practiced by urologists, particularly for cases of glanular and distal hypospadias. Between 2002 and 2008, one-stage hypospadias repair has been performed in 116 (73,4%) patients at the Department of Urology, Faculty of Medicine/Indonesia University, Cipto Mangunkusumo Hospital, Jakarta. High success rate of one-stage hypospadias repair has been reported in various studies. One-stage hypospadias repair has more benefits than two-stage repair. In one-stage

hypospadias the skin used remains healthy and no scar tissue from previous surgical procedures is present, and blood supply remains normal and intact.^{1,2}

Urethrocuteaneous fistula is the most frequent complication of hypospadias repair. The incidence of urethrocuteaneous fistula has decreased in the last two decades along with the development of hypospadias surgery technique. In 1973, Horton and Devine estimated the incidence of hypospadias post-operative fistula of about 15 - 45%. Today, the incidence of post-urethroplasty urethrocuteaneous fistula is estimated to be 10 - 15%.³⁻⁹

A number of factors have been suggested as the causes of high incidence of urethrocutaneous fistula post-hypospadias repair. The most important factors are technical, which should have been prevented, such as rough tissue handling, the use of thin and fibrotic epithelium or skin, failure in making a water-proof anastomosis, the use of non-vital tissue, failed epithelial inversion in neoureter, the making of neourethra with poor vascularization, and the type and size of thread used. The use of a second layer and magnification has proved to reduce the incidence of fistula. The incidence of urethrocutaneous fistula also increases along with more proximal site of meatus, more complicated reconstructive procedure, the presence of meatal stenosis, urethral diverticula, stricture in distal urethra, and infection. Correlation between the use of urethral stenting and wrapping and the incidence of urethrocutaneous fistula remains debatable.^{2,6,7,10}

OBJECTIVE

To find correlation between various risk factors and the incidence of urethrocutaneous fistula in patients undergoing one-stage hypospadias repair.

MATERIAL & METHOD

This study was a case control study to find correlation between various risk factors and the incidence of urethrocutaneous fistula in patients undergoing one-stage hypospadias repair. The study was performed at the Department of Urology, Faculty of Medicine/Indonesia University, Cipto Mangunkusumo Hospital, Jakarta. Data were obtained from medical records of hypospadias patients who had undergone one-stage urethroplasty between January 2002 and December 2008. Samples were selected with consecutive sampling. All existing subjects who met the study criteria were enrolled. The inclusion criteria in this study were hypospadias patients undergoing one-stage urethroplasty, using PDS thread (size

according to age), using urine diversion with cystostomy/catheter, using urethral stenting, and using magnification. The exclusion criteria were previous hypospadias repair and incomplete medical records.

Data were collected from medical records/special status of the Department of Urology, Faculty of Medicine/Indonesia University, Cipto Mangunkusumo Hospital, Jakarta. Age at the operation, hypospadias classification, degree of chordee, associated urogenital abnormalities, operation technique, duration of operation, type of wrapping, duration of stenting, and the incidence of post-operative urethrocutaneous fistula were extracted. Data were analyzed descriptively and analytically using SPSS 16.0 program with chi-square and Fischer's exact tests. The results of analysis were presented in narration and tables. Analysis was carried out to find correlation between urethrocutaneous fistula and the patients' age, hypospadias classification, degree of chordee, accompanying congenital abnormalities, history of hormonal therapy, thread size, duration of operation, type of wrapping, type of stent, and duration of stenting. The authors also analyzed correlation between urethrocutaneous fistula with three operation techniques applicable for each hypospadias classification, the TIP (Tubularized Incised Plate), Duckett (Transverse Preputial Island Flap), and Onlay Island Flap.

RESULTS

There were 116 hypospadias patients who had undergone one-stage urethroplasty at the Department of Urology, Cipto Mangunkusumo Hospital, and met the inclusion criteria. Mean age of the patients was $5,7 \pm 3,9$ years (range 4 months - 19 years). Table 1 shows various classifications of hypospadias patients who had undergone one-stage urethroplasty. Complication of urethrocutaneous fistula was found in 12 patients (10,3%).

Table 1. Classification of hypospadias patients.

Classification	Notes	n	%
Age category	< 2 years	21	18,1
	2-15 years	92	79,3
	> 15 years	3	2,6
Hypospadias category	Glanular hypospadias	14	12,1
	Distal hypospadias	36	31,0
	Proximal hypospadias	66	56,9
Degree of chordee	Without chordee	6	5,2
	Mild	17	14,7
	Moderate	53	45,7
	Heavy	40	34,5
Accompanying abnormalities	Penoscrotal transposition	28	24,1
	Bifid scrotum	29	25,0
	Undescended testis	10	8,6
History of hormonal therapy	Yes	13	11,2
	No	103	88,8
Thread size	7.0	7	6,0
	6.0	97	82,8
	5.0	13	11,2
Type of wrapping	Tegaderm	78	67,2
	Tulle Gras	35	30,2
	No wrapping	3	2,6
Type of stent	Silastic	67	57,8
	Catheter	42	36,2
	NGT	7	6,0
Duration of stenting	< 7 days	25	21,6
	7-13 days	74	64,8
	> 13 days	17	14,6
Duration of operation	< 120 minutes	80	69,0
	>120 minutes	36	31,0
Operation techniques	Tubularization	7	6,0
	TIP	47	40,5
	Buccal mucosa graft	1	0,9
	Duckett (Transverse Preputial Island Flap)	26	22,4
	Onlay Island Flap	19	16,4
	Matthieu	7	6,0
	GAP	8	6,9
	MAGPI	1	0,9
Fistula formation		12	10,3

Notes: TIP = Tubularized Incised Plate, GAP = Glanular Approximation Procedure, MAGPI = Meatal Advancement and Glanuloplasty.

Table 2 showed no significant correlation between age ($p = 0,426$), hypospadias classification ($p = 0,695$), degree of chordee ($p = 0,564$), accompanying urogenital abnormalities ($p = 0,964$), history of hormonal therapy ($p = 0,739$), thread size ($p = 0,248$),

duration of operation ($p = 0,856$), type of wrapping ($p = 0,580$), type of stent ($p = 0,600$), and duration of stenting ($p = 0,796$) with the formation of post-operative urethrocuteaneous fistula.

Table 2. Correlation between various variables with the emergence of urethrocutaneous fistula.

Classification	Notes	Fistula		Statistical test	p
		No (%)	Yes (%)		
Age	< 2 years	18 (85,7)	3 (14,3)	Fischer	0,426
	2-15 years	84 (91,3)	8 (8,7)		
	> 15 years	2 (66,7)	1 (33,3)		
Classifications	Glanular hypospadias	13 (92,9)	1 (7,1)	Fischer	0,695
	Distal hypospadias	32 (88,9)	4 (11,1)		
	Proximal hypospadias	59 (89,4)	7 (10,6)		
Chordee	Without chordee	6 (100)	0 (0)	Fischer	0,564
	Mild	16 (94,1)	1 (5,9)		
	Moderate	48 (90,6)	5 (9,4)		
	Heavy	34 (85,0)	6 (15,0)		
Accompanying congenital abnormalities	No congenital abnormalities	70 (89,7)	8 (10,3)	x ²	0,964
	With congenital abnormalities	34 (89,5)	4 (10,5)		
History of hormonal therapy	No therapy	92 (89,3)	11 (10,7)	x ²	0,739
	With therapy	12 (92,3)	1 (7,7)		
Thread size	7.0	7 (100)	0 (0)	Fischer	0,248
	6.0	84 (87,5)	12 (12,5)		
	5.0	13 (100)	0 (0)		
Duration of operation	≤ 120 minutes	72 (90,0)	8 (10,0)	x ²	0,856
	> 120 minutes	32 (88,9)	4 (11,1)		
Type of wrapping	Tegaderm	71 (91,0)	7 (9,0)	Fischer	0,580
	Tulle Gras	30 (85,7)	5 (14,3)		
	No wrapping	3 (100)	0 (0)		
Type of stent	Silastic	59 (59)	8 (11,9)	Fischer	0,600
	Catheter	38 (90,5)	4 (9,5)		
	NGT	7 (100)	0 (0)		
Duration of stenting	< 7 days	16 (94,1)	1 (5,9)	Fischer	0,796
	7-13 days	66 (89,2)	8 (10,8)		
	> 13 days	22 (88,0)	3 (12,0)		
Operation techniques	Tubularization	7 (100)	0 (0)	Fischer	0,314
	TIP	42 (89,4)	5 (10,6)		
	Buccal mucosa graft	0 (0)	1 (100)		
	Duckett (Transverse Preputial Island Flap)	21 (80,8)	5 (19,2)		
	Onlay Island Flap	18 (94,7)	1 (5,3)		
	Matthieu	7 (100)	0 (0)		
	GAP	8 (100)	0 (0)		
	MAGPI	1 (100)	0 (0)		

The authors also compared three operation techniques applicable for each hypospadias classification, the TIP (Tubularized Incised Plate) as reference, compared to Duckett (Transverse Preputial Island Flap) and Onlay Island Flap techniques.

Apparently, data analysis revealed that there was no significant correlation between TIP technique vs. Duckett ($p = 0,314$) and TIP vs Onlay Island Flap ($p = 0,644$) with the formation of post-urethroplasty urethrocutaneous fistula.

DISCUSSION

Hypospadias is a congenital abnormality, in which the urethral meatus is located proximally from its normal position at the tip of glans penis.³ Hypospadias is commonly found in 1 from 250 - 300 male births.²⁻⁸ There are three anatomic abnormalities that can be found in children with hypospadias, (1) Abnormal urethral meatus may be located ventral of the glans penis to the perineum, (2) Abnormal penile curvature (chordee), (3) Abnormal distribution of preputium, in which there is excess in dorsal skin (dorsal hood), and deficit in ventral skin.^{3,9}

There are various classifications of hypospadias, according to Smith (1938), Schaefer (1950), Avellan (1975), Browne (1938), Duckett (1996), and the newest classification from 2003, which is commonly used today. In this classification, hypospadias is divided into three types, (1) Glandular hypospadias, in which the meatus is located on the glans and subcorona (50-70%), (2) Distal hypospadias, in which the meatus of the urethra is located distal or on the mid-shaft of the penis (30%), and (3) Proximal hypospadias, in which urethral meatus is located proximal, penoscrotal or perineal of the penis (20%).^{3,10}

In this study, from 116 patients, 14 were found with glanular hypospadia (12,1%), 36 with distal hypospadias (31,0%), and 66 with proximal hypospadias (56,9%). The frequency of these patients' distribution was different from that in the literature. In this study, most of the patients had proximal hypospadia. The characteristics of the patients' age in this study varied, with mean age was $5,7 \pm 3,9$ years (range 4 months - 19 years). In the literature, the optimum age advised for hypospadias repair is 6 - 18 months.^{1,9,11} In this study, there was no significant correlation between patients' age and the formation of post-urethroplasty urethrocuteaneous fistula ($p = 0,426$). The success of hypospadias repair can be achieved with penile curvature correction, the making of neourethra of adequate size, and the placement of the meatus on the tip of the glans penis,¹¹ and the primary principles that should be applied in hypospadias repair to prevent complication are the minimal cauterization, prevention of tension, the use

of tissue with good vascularization and adequate layers, single-stage with epithelial inversion, the use of loupe for magnification, and the use of fine thread material.^{2,7}

Urologists today have widely applied hypospadias repair, particularly for glanular and distal hypospadias cases. High success rate of one-stage hypospadias repair has been reported much in various studies. One-stage hypospadias repair has more benefits than two-stage repair. In one-stage hypospadias the skin used remains healthy and no presence of scar tissue resulting from previous surgical procedure, and blood supply remains normal and intact.^{1,2}

Urethrocuteaneous fistula is the most frequent complication of hypospadias repair. The emergence of post-urethroplasty urethrocuteaneous fistula may indicate the presence of meatal stenosis, urethral stricture on distal fistula, or urethral diverticula. In the past, the incidence of post-urethroplasty urethrocuteaneous fistula may reach 50%. However, along with the development of urethroplasty technique, the incidence of post-urethroplasty fistula is decreasing to 10 - 15%.^{1,2,4,12} There are several factors that contribute to high incidence of urethrocuteaneous fistula after hypospadias repair. The most important factors are technical ones, which are essentially preventable. These factors are rough tissue handling, the use of thin and fibrotic epithelium or skin, failure in making waterproof anastomosis, the use of non-vital tissue, failed epithelial inversion in neoureter, the making of neourethra with poor vascularization, and the type and size of thread used. The use of second layer and magnification has been proved to reduce the incidence of fistula.^{2,6,7,10}

Literature revealed that the incidence of urethrocuteaneous fistula also increases along with the more proximal site of meatus, more complicated reconstructive procedure, presence of meatal stenosis, urethral diverticula, stricture in distal urethra, and infection. Distal urethral destruction may result in increase of proximal urethral pressure, causing urinary leakage from the edge of the stitch. The presence of urethral diverticula may cause urine turbulence, increasing the incidence of fistula. The effect of the use

of urethral stenting and the type of wrapping with the formation of urethrocutaneous fistula remains controversial.^{2,6,7,10,13} Waterman et al (2005) evaluated various factors suspected to affect the success of hypospadias repair. Their result revealed no significant difference in the use of stent vs non-stent and the use of microscope vs loupe in the rate of success of the operation.¹⁴

In this study, urethrocutaneous fistula occurred in 12 patients (10,3%). The incidence of urethrocutaneous fistula in this study, which was only 10,3%, was acceptable and similar to the incidence rate of urethrocutaneous fistula in the literature (10 - 15%), since the patients in this study were predominantly those with proximal hypospadias (56,9%), in which the incidence of urethrocutaneous fistula in these patients is higher than other types of hypospadias. Several literatures also mentioned that fistular incidence is far higher (17 - 69%) after proximal hypospadias repair than after distal type (2,5 - 32%).^{4,5} Lower incidence of urethrocutaneous fistula in our center may be due to adequate chordee excision procedure that we applied. In the previous study at our center, post-urethroplasty persistent chordee was only found in three patients (6,1%).

Urethrocutaneous fistula in post-hypospadias repair may not only present as single and simple fistula, but also as multiple and complex one. Pinpoint fistula may also emerge, even though a skilled surgeon has applied the appropriate and fine technique. Fistula may emerge anywhere along the neourethra, but the most frequent location is at the meatus pre-operatively, and the corona or glans penis. Fistula at the corona and glans penis is harder to manage and has a high recurrence rate. Multiple and complex fistula rarely emerges, and it is generally due to technical factors, such as ischemia, infection, distal stenosis, and inadequate chordee excision. Several reports also mention that persistent chordee is one of the primary causes of failure in hypospadias repair.^{6,15}

Several operation techniques are apparently more susceptible to induce fistula compared to other techniques. In a study by Durham Smith (1990), the incidence of post-MAGPI (meatoplasty and

glanuloplasty) urethrocutaneous fistula is ranging between 0,5 - 10%, post-flip flap repair 2,2 - 35%, post island pedicle tubed repair 4 - 33%, and post free graft tubed repair 15 - 50%.⁶ In this study, the authors compared three operation techniques that can be applied to each hypospadias classification, the TIP (Tubularized Incised Plate) as reference, compared to Duckett (Transverse Preputial Island Flap) and Onlay Island Flap. From the result of data analysis, it was found that there was no significant correlation between TIP vs Duckett ($p = 0,314$) and TIP vs Onlay Island Flap ($p = 0,644$) with the formation of post-urethroplasty. In this study, all operations used magnification. Unfortunately, recording in medical record/urological special status on the use of second layer was incomplete, preventing further and deeper analysis.

A study in Cairo evaluated the role of wrapping in the emergence of fistula, with the result that the use of wrapping had statistically significant effect on the increase of fistula incidence. Several authors (Van Savage et al 2000; McLorie et al 2001) also suggested not using wrapping routinely after hypospadias repair. For surgeons who still intend to use wrapping, it is suggested to use materials, such as silicone foam or duoderm, that have been proved to be able in reducing fistular incidence.⁶ This study found no significant correlation between the type of wrapping and the emergence of urethrocutaneous fistula ($p = 0,580$).

There is still a debate on the use of catheter/stenting in hypospadias repair. The primary objective of urine diversion is to keep the anastomosis location dry. The problems are spasm and contraction of detrusor muscle due to the irritation of catheter tip to bladder wall. Some surgeons have overcome this problem by using stenting, where the tip of the stent is located distal from external sphincter. The experts recommend to use stent for 7-10 days in proximal hypospadias cases.⁵ This study did not find significant correlation between the type of stent used and the duration of stenting with the emergence of urethrocutaneous fistula ($p = 0,600$; $p = 0,796$).

The analysis performed in this study was univariate. Studies using multivariate analysis or

cohort studies are expected to provide better results. Evaluation to other potential factors for the emergence of urethrocutaneous fistula should also be examined to improve the outcome of hypospadias repair.

CONCLUSION

There is no significant correlation between age, hypospadias classification, degree of chordee, hypospadias classification, degree of chordee, accompanying urogenital abnormalities, history of hormonal therapy, thread size, duration of operation, type of wrapping, type of stenting, and duration of stenting with the incidence of post-operative urethrocutaneous fistula. There is no significant correlation between TIP vs Duckett, and TIP vs Onlay Island Flap with the formation of post-urethroplasty urethrocutaneous fistula.

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