



Open Access

## ORIGINAL ARTICLE

Operational Andrology

# Complications of proximal hypospadias repair with transverse preputial island flap urethroplasty: a 15-year experience with long-term follow-up

Chao-Xu Wang, Wei-Ping Zhang, Hong-Cheng Song

There is still debate regarding the optimal surgical approach for proximal hypospadias. This retrospective study aims to evaluate the long-term outcomes using transverse preputial island flap urethroplasty. A total of 320 patients were included, with a mean follow-up of 40.2 months (range: 1–156 months). Complications were encountered in 125 patients (39.1%), including fistulas in 53 (16.6%), urethral strictures in 31 (9.7%), and diverticula in 41 (12.8%). The mean timing of presentation with a complication was 15.8 months (median: 1.7, range: 1–145), of which 79.2% were early complications and 20.8% were late complications. In all, 20.8% of the patients with complications presented after  $\geq 1$  year, and 12.8% presented after  $\geq 5$  years. Univariate analysis revealed that age at the time of surgery, flap length, and location of the urethral meatus were not correlated with complications. A stricture was present in 31.7% (13/41) of those with diverticula ( $P < 0.001$ ), while late urethral diverticula were accompanied by urethral strictures in 11.1% (1/9) of cases ( $P = 0.213$ ). These results indicate that transverse preputial island flap urethroplasty still has a high incidence of complications, even when performed by highly experienced physicians. Most complications of hypospadias are diagnosed within 1 year postoperatively, while fistulas and urinary strictures generally occur within 2 months and diverticula tend to be present by 1 year. *Asian Journal of Andrology* (2019) 21, 300–303; doi: 10.4103/aja.aja\_115\_18; published online: 15 March 2019

**Keywords:** diverticulum; fistula; follow-up; hypospadias; late complications; urethral stricture

## INTRODUCTION

Hypospadias is one of the most common congenital anomalies in males, occurring in 1 of 300 live births. The prevalence of hypospadias in China is approximately 90.3 per 10 000 births.<sup>1</sup> Hypospadias surgery aims to create a straight urinary stream from a meatus with a normal anatomical position, a straight penis, and a normal circumcised penile appearance.

Despite the evolution and improvement of surgical techniques, proximal hypospadias is one of the most challenging issues faced by pediatric urologists. Several recent studies have found higher complication rates than previously reported for proximal hypospadias, ranging from 30% to 68%, which cautions us about the timing of urethroplasty complications, given that it is clear that more complications are encountered with longer follow-ups.<sup>2–5</sup> Better understanding of the time to presentation of hypospadias complications is important to allow for the diagnosis of complications because a significant number of complications present late.<sup>6,7</sup> However, there are little data published on the long-term outcome of the transverse preputial island urethroplasty, although the Duckett technique was originally described in 1980.<sup>8</sup>

In the present study, we reported our long-term outcomes of proximal hypospadias over a 15-year period with long-term follow-up. The specific aim of the study was to describe the incidence of urethroplasty complications and report timing of the presentation of urethroplasty complications.

## PARTICIPANTS AND METHODS

We retrospectively evaluated the medical records of patients who underwent repair of proximal hypospadias using transverse preputial island flap urethroplasty (Duckett technique) from January 2003 to January 2018. Exclusion criteria included patients who underwent repair at another hospital or distal hypospadias and those who underwent proximal hypospadias repair with 2-stage repairs. One surgeon performed all cases, and the surgical technique was the same for all patients. Due to the retrospective nature of the study, informed consent was waived, and the Ethics Board of the Beijing Children's Hospital approved the study.

Data regarding age at the time of surgery, location of the meatus, length of the flap, duration of follow-up, and timing of the presentation of complications were collected. Proximal hypospadias was categorized as proximal penile, penoscrotal, or perineal based on the location of the urethral meatus. The flap length was determined as the distance from the retracted meatus to the glans tip after straightening the curvature. The complications were divided into early (complications within 1 year after initial repair) and late (complications 1 year after initial repair).

### Urethroplasty complications

Urethroplasty complications were defined as any problem that required surgical correction. Urethroplasty complications included fistula, urethral stricture, and diverticulum. Urethral stricture was defined as

obstructive voiding symptoms and calibration <8 Fr. Diverticulum was defined as visible ballooning of the neourethra while voiding.

#### Follow-up protocol

All patients were required to visit our clinic at 3 months postoperatively, then 12 months after the surgery, and, if possible, at puberty.

#### Surgical techniques

The Duckett technique is briefly described.<sup>8</sup> A circumferential incision was made proximal to the corona that reached the depth of the Buck fascia, allowing for degloving and chordee release. Subsequently, we transected the urethral plate if the penis could not be straightened owing to severe chordee and dropped back the meatus to the penoscrotal junction or the proximal shaft. Dorsal plication was performed if the chordee was still present. The flap length was determined as the distance from the retracted meatus to the glans tip after straightening the curvature. A 12-mm wide rectangular flap was harvested from the inner prepuce, and the mobilized foreskin was rolled into a tube over a catheter. The glans channel was then created, and the neourethra was transposed ventrally and brought through the channel. Additional fixation of the transverse preputial island flap (TPIF) to the midline of the corpus was performed. The neourethra was anastomosed with the native urethra, and the distal meatus was attached to the top of the glans with interrupted fine sutures.

#### Statistical analyses

Statistical analysis was performed using logistic regression analysis and Fischer's exact test (IBM SPSS Statistics for Windows, version 19.0.; SPSS, Armonk, NY, USA). Univariable analysis was done to determine factors predictive of complications. Factors evaluated included initial meatus location (proximal penile, penoscrotal, or perineal), length of the flap, and age at the time of surgery.  $P < 0.05$  was considered statistically significant.

#### RESULTS

This study included 320 patients with a mean follow-up of 40.2 (range: 1–156) months. In all, 198 (61.9%) patients were followed for more than 1 year and 39 (12.2%) patients were followed for more than 5 years after the repair. Of the 320 patients who underwent the Duckett technique, 90, 151, and 79 had proximal penile, penoscrotal, and perineal hypospadias, respectively. The mean age of the patients at surgery was 15.1 months (range: 11–64). The mean length of the flap was 3.7 cm (range: 2.5–4.8).

A total of 125 patients (39.1%) developed complications, 23 of whom (18.4%) had two or more complications. There were three main

types of complications: 53 (16.6%) fistulas, 31 (9.7%) urethral strictures, and 41 (12.8%) diverticula.

The mean timing of presentation with a complication was 15.8 (median: 1.7, range: 1–145) months, of which 79.2% were early complications and 20.8% were late complications (Table 1). In all, 20.8% of the patients with complications presented after  $\geq 1$  year, and 12.8% presented after  $\geq 5$  years. The ratios of early versus late cases of fistulas, urethral strictures, and diverticula were 77.4% versus 22.6%, 83.9% versus 16.1%, and 78.0% versus 22.0%, respectively.

A stricture was present in 31.7% (13/41) of those with diverticula ( $P < 0.001$ ), while late diverticula were accompanied by urethral strictures in 11.1% (1/9) of cases ( $P = 0.213$ ).

On univariable analysis, age at the time of surgery, length of the flap, or meatus location (proximal penile, penoscrotal, or perineal) did not predict complications (Table 2).

Fistulae were repaired by simple excision in 53 patients 6–12 months after initial surgery, with 45 (84.9%) undergoing 1 repair, 5 (9.4%) undergoing 2 repairs, and 3 (5.7%) undergoing 3 repairs. Urethral strictures were found in 31 patients, with 12 (38.7%) treated successfully with one or more dilatations. Dilatation provided temporary improvement, and open surgical reconstruction was performed in 19 patients. Strictures usually occurred at the proximal anastomosis, and the stricture segment was opened, with urethroplasty subsequently performed 6–12 months later. Diverticula were asymptomatic in four patients who needed no surgical intervention. Diverticula were treated with resection of redundant tissue and multilayer closure in 36 patients and repair of a subsequent fistula in five patients.

#### DISCUSSION

Although delayed complications from the surgical repair of hypospadias are common, the time at which complications are encountered is uncertain because some complications may not be discovered until long after surgery.<sup>9,10</sup> In the present study, 77.3% of fistulas occurred within 1 year, and 18.9% of fistulas developed 5 years after initial surgery. One reason for the delayed appearance of urinary fistula may be that small fistulas are only diagnosed after patients are toilet trained.<sup>6</sup> Delayed complications may have already manifested earlier and gone unrecognized, particularly if their appearance was elusive, as is often the case with fistulas. Other reports have found similar late-presenting fistulas. Wood *et al.*<sup>6</sup> described their experience with 26 patients and found that the average time of urinary fistula presentation was 3 months, although it required 8 years and 20.5 years to diagnose 90% and 99% of all fistulas, respectively. As noted above, patients with

**Table 1: Complications and initial hypospadias severity**

Urethral complications	Proximal penile hypospadias	Penoscrotal hypospadias	Perineal hypospadias	Total
Fistula, <i>n</i> (%)				
Total	18 (5.6)	26 (8.1)	9 (2.8)	53 (16.5)
Early	10 (3.1)	22 (6.9)	9 (2.8)	41 (12.8)
Late	8 (2.5)	4 (1.2)	0 (0)	12 (3.7)
Urethral stricture, <i>n</i> (%)				
Total	6 (1.9)	13 (4)	12 (3.7)	31 (9.6)
Early	5 (1.6)	11 (3.4)	10 (3.1)	26 (8.1)
Late	1 (0.3)	2 (0.6)	2 (0.6)	5 (1.5)
Diverticulum, <i>n</i> (%)				
Total	9 (2.8)	18 (5.6)	14 (4.3)	41 (12.8)
Early	7 (2.2)	14 (4.4)	11 (3.4)	32 (10)
Late	2 (0.6)	4 (1.2)	3 (0.9)	9 (2.8)

**Table 2: Univariable analysis of complication predictor**

Variable	Complication		P
	No	Yes	
Number of patients, n (%)	195 (60.9)	125 (39.1)	
Meatal location			
Proximal penile	57 (29.2)	33 (26.4)	0.71
Penoscrotal	94 (48.2)	57 (45.6)	
Perineal	44 (22.6)	35 (28.0)	
Age at surgery (month), mean (range)	14.8 (11–64)	15.6 (12–58)	0.08
Flap length (cm), mean (range)	3.6 (2.6–4.5)	3.8 (2.5–4.8)	0.19

late-presenting urethroplasty complication usually have a lengthy follow-up interval.<sup>10</sup>

Urethral stricture after Duckett technique has an incidence of 6%–22%, which is mainly due to the proximal anastomosis.<sup>11,12</sup> In the present study, 31 (9.7%) patients developed urethral strictures. However, Castagnetti *et al.*<sup>13</sup> recently published a systematic review of 20 years of publications on the repair of proximal hypospadias, and the preputial island tube demonstrated a urethral stricture rate of 12.5%. The explanation is that the experienced surgeons, who had specialized in pediatric urology for more than 15 years, performed all surgeries in the present study. Moreover, 61.3% of patients had significant strictures requiring reurethroplasty, which is consistent with the severe hypospadias and long follow-up time reported in the present series. The time of presentation of urethral strictures ranged from 1 month to 124 months postoperatively, with 16.1% (5/31) presenting after 1 year. Given that the length and width of genital tubercles double between the ages of 1 year and 10 years, the risk of delayed urethral stricture may be present if the tissues used in urethroplasty during infancy grow at uncoordinated rates.<sup>14</sup> From a clinical point of view, the fact that some urethral complications presented late is a sign of late urethral deficiency.<sup>9</sup>

Diverticulum is not uncommon after the Duckett technique. There may be a causal relation between diverticula and strictures. The present study found that 31.7% (13/41) of patients with a diverticulum had a stricture ( $P=0.032$ ), while only 1 of 9 patients with a late diverticulum had a stricture ( $P=0.213$ ). The reason for this is unclear, but perhaps the formation of diverticula is multifactorial and may be associated with the presence of a distal stricture and poorly supported tissue for interpositional grafting. Early detection of stricture resulted in treatment of the stricture before the serious long-term effects increased voiding pressure and, subsequently, the late formation of diverticulum.

Our present results found that only 20.8% of complications presented 1 year postoperatively. However, Nuininga *et al.*<sup>15</sup> reported that 75% of the complications were diagnosed 5 years after the Duckett technique. One explanation is that our study included more patients, while more complications are diagnosed with the extension of follow-up. This finding coincided with the retrospective study of Cambareri *et al.*<sup>7</sup> who reported that 54.5% of the complications of surgery developed more than 1 year postoperatively and that 31.8% of the complications presented more than 3 years postoperatively with follow-up extending to 35 months.

It is noteworthy that our experiences with the Duckett technique demonstrated that new complications were found 10 years after surgery in 5 (4%) patients, including two fistulas and three urethral strictures. Two studies reported that new complications may occur long after primary hypospadias repair. Long-term follow-up of onlay flaps reported complications after a minimum of 10 years with 5 (17%)

patients, including two fistulas and three meatal stenoses.<sup>16</sup> Lam *et al.*<sup>17</sup> reviewed 44 patients with a follow-up of at least 10 years after primary hypospadias and found new complications in 2 (8%) cases, including a fistula and stricture. Long-term follow-up is important in patients with hypospadias, given the knowledge that complications can occur long after the initial surgery. We believe that these results have important implications when counseling families about further potential operations because lack of scrutiny may contribute to later presentation of complications. In the findings of Snodgrass *et al.*<sup>18</sup> and Faasse and Liu,<sup>10</sup> approximately 30%–50% of their patients with late-presenting complications had no encounters beyond 6 months postoperatively.

The precise time at which complications occur is unknown. A retrospective study of 70 patients who underwent various techniques of repair followed during or after adolescence found that 54.2% required a second operation.<sup>19</sup> However, we would not expect patients to have regular follow-up indefinitely. Based on the time when complications were discovered in our study, the recommended assessment schedule was initiated at 2 months postoperatively and then 1 year later, finally at puberty. Wood *et al.*<sup>6</sup> suggested that detecting fistulas up to 20 years after surgery would require continuous follow-up to eventually diagnose fewer complications. Our assessment schedule targets achieving a rational balance between diagnosing as many complications as possible and the impracticality of ongoing follow-up.

In the current study, we did not measure uroflow and postvoid residual volume in all patients. Outcome assessment was mainly based on evaluation of micturition by the surgeon and parents. Uroflowmetry was once recommended to detect asymptomatic strictures. In a recent long-term follow-up of patients who underwent proximal hypospadias repair, the obstructive urinary flow pattern observed in childhood is frequent. However, there is spontaneous and progressive improvement with age and during adolescence; hence, a watchful waiting approach was proposed in order to avoid unnecessary intervention.<sup>20</sup>

The main limitation of this study is its retrospective nature, which may have caused a certain degree of selection bias. The choice of surgical technique was largely determined by surgeon preference and practice. Most patients suffering delayed complications usually return only after symptoms occur. In addition, as the data used in our study were obtained from the medical records, there were lower complication rates compared with other studies because some patients suffering from problems may have received medical attention at other hospitals.

## CONCLUSION

Our long-term follow-up study indicates that complications of transverse preputial island flap urethroplasty generally develop within a short time period. Most complications of hypospadias are diagnosed within 1 year of surgery, where fistulas and urinary strictures generally develop within 2 months postoperatively, while diverticula tend to be present by 1 year.

## AUTHOR CONTRIBUTIONS

WPZ conceived of the study, participated in its design, and drafted the manuscript. CXW participated in data acquisition. HCS made critical revisions to the manuscript regarding important intellectual content. All authors read and approved the final manuscript.

## COMPETING INTERESTS

The authors declared no competing interests.

## ACKNOWLEDGMENTS

This work was supported by grants from the Beijing Municipal Administration of Hospitals “Dengfeng” Talent Training Plan (No. 20151102) and the

Beijing Municipal Administration of Hospitals “Yangfan”: Pediatric Urology (No. 201709).

## REFERENCES

- 1 Li Y, Mao M, Dai L, Li K, Li X, *et al*. Time trends and geographic variations in the prevalence of hypospadias in China. *Birth Defects Res A Clin Mol Teratol* 2012; 94: 36–41.
- 2 Stanasel I, Le HK, Bilgutay A, Roth DR, Gonzales ET, *et al*. Complications following staged hypospadias repair using transposed preputial skin flaps. *J Urol* 2015; 194: 512–6.
- 3 McNamara ER, Schaeffer AJ, Logvinenko T, Seager C, Rosoklija I, *et al*. Management of proximal hypospadias with 2-stage repair: 20-year experience. *J Urol* 2015; 194: 1080–5.
- 4 Pippi SJ, Sayed S, Salle A, Bagli D, Farhat W, *et al*. Proximal hypospadias: a persistent challenge single institution outcome analysis of three surgical techniques over a 10-year period. *J Pediatr Urol* 2016; 12: 21–8.
- 5 Long CJ, Chu DI, Tenney RW, Morris AR, Weiss DA, *et al*. Intermediate-term followup of proximal hypospadias repair reveals high complication rate. *J Urol* 2017; 197: 852–8.
- 6 Wood HM, Kay R, Angermeier KW, Ross JH. Timing of the presentation of urethrocutaneous fistulas after hypospadias repair in pediatric patients. *J Urol* 2008; 180: 1753–6.
- 7 Cambareri GM, Yap M, Kaplan GW. Hypospadias repair with onlay preputial graft: a 25-year experience with long-term follow-up. *BJU Int* 2016; 118: 451–7.
- 8 Duckett JJ. Transverse preputial island flap technique for repair of severe hypospadias. *Urol Clin North Am* 1980; 7: 423–30.
- 9 Grosos C, Bensaid R, Gorduzza DB, Mouriquand P. Is it safe to solely use ventral penile tissues in hypospadias repair? Long-term outcomes of 578 Duplay urethroplasties performed in a single institution over a period of 14 years. *J Pediatr Urol* 2014; 10: 1232–7.
- 10 Faasse MA, Liu DB. Early vs. late-presenting urethroplasty complications after hypospadias repair: a retrospective analysis of patient follow-up. *J Pediatr Urol* 2017; 13: 354.e1–4.
- 11 Barraza MA, Roth DR, Terry WJ, Livne PM, Gonzales ET, *et al*. One-stage reconstruction of moderately severe hypospadias. *J Urol* 1987; 137: 714–5.
- 12 Elbakry A. Complications of the preputial island flap-tube urethroplasty. *BJU Int* 1999; 84: 89–94.
- 13 Castagnetti M, El-Ghoneimi A. Surgical management of primary severe hypospadias in children: systematic 20-year review. *J Urol* 2010; 184: 1469–74.
- 14 Damon V, Berlier P, Durozier B, Francois R, *et al*. Study of the dimensions of the penis from birth to adult age and as a function of testicular volume. [Article in French]. *Pediatrerie* 1990; 45: 519–22.
- 15 Nuininga JE, DE Gier RP, Verschuren R, Feitz WF, *et al*. Long-term outcome of different types of 1-stage hypospadias repair. *J Urol* 2005; 174: 1544–8.
- 16 Patel RP, Shukla AR, Snyder HR. The island tube and Island onlay hypospadias repairs offer excellent long-term outcomes: a 14-year followup. *J Urol* 2004; 172: 1717–9.
- 17 Lam PN, Greenfield SP, Williot P. 2-stage repair in infancy for severe hypospadias with chordee: long-term results after puberty. *J Urol* 2005; 174: 1567–72.
- 18 Snodgrass W, Villanueva C, Bush NC. Duration of follow-up to diagnose hypospadias urethroplasty complications. *J Pediatr Urol* 2014; 10: 208–11.
- 19 Prat D, Natasha A, Polak A, Koulikov D, Prat O, *et al*. Surgical outcome of different types of primary hypospadias repair during three decades in a single center. *Urology* 2012; 79: 1350–3.
- 20 Hueber PA, Salgado DM, Chaussy Y, Franc-Guimond J, Barrieras D, *et al*. Long-term functional outcomes after penoscrotal hypospadias repair: a retrospective comparative study of proximal TIP, Onlay, and Duckett. *J Pediatr Urol* 2016; 12: 198.e1–6.

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

©The Author(s)(2019)

