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Two-stage urethroplasty is a better choice for proximal hypospadias with severe chordee after urethral plate transection: a single-center experience

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It is still debatable whether single- or two-stage urethroplasty is a more suitable technique for treating hypospadias with severe chordee after urethral plate transection. This retrospective study evaluated these two techniques. A total of 66 patients of proximal hypospadias with severe chordee were divided into two groups according to the techniques they underwent: 32 and 34 patients underwent single-stage (Duckett) or two-stage urethroplasty, respectively. Median ages at presentation were 7.5 years and 11.0 years in single-stage and two-stage repair groups, respectively. Median follow-ups were 28.5 months (20–60 months) and 35 months (18–60 months) in the single-stage and two-stage groups, respectively. The meatus of the neourethra was located at the top of the glans in all patients. No recurrence of chordee was found during follow-up, and all patients or parents were satisfied with the penile length and appearance. Complications were encountered in eight patients in both groups, with no statistically significant differences between the two techniques. The late complication rate of stricture was higher after the single-stage procedure (18.75% vs 0%). The complication rate after single-stage repairs was significantly lower in the prepubescent subgroup (10.52%) than in the postpubescent cohort (46.15%). These results indicate that the urethral plate transection effectively corrects severe chordee associated with proximal hypospadias during the intermediate follow-up period. Considering the higher rate of stricture after single-stage urethroplasty, two-stage urethroplasty is recommended for proximal hypospadias with severe chordee after urethral plate transection.

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INTRODUCTION

Hypospadias is a common congenital malformation, occurring in about 1 in 300 live births, with proximal hypospadias being identified in 20% of cases.¹ Compared with distal hypospadias, the proximal type usually has a higher incidence of complications. Various surgical techniques have been used to repair proximal hypospadias, including tubularized incised plate and onlay island flap urethroplasty.² Urethral plate mobilization and dorsal plication have been used to correct the penile curvature of hypospadias with chordee. Sometimes the native meatus drops back to the proximal location when the urethral plate has to be transected for maximum release. In this situation, some surgeons prefer single-stage urethroplasty (e.g. the Duckett and modified Koyanagi technique), whereas others advocate two-stage repair to achieve better functional and cosmetic outcomes.^{3–6} Few studies, however, have attempted to compare outcomes between single-stage and two-stage repairs. We reviewed our experience of repairing proximal hypospadias with severe chordee, wherein we made the correction by urethral plate transection. We then compared single-stage with two-stage urethroplasty with regard to the outcomes and complications.

PATIENTS AND METHODS

Study design and patients

We retrospectively reviewed the records of consecutive patients referred for primary treatment of proximal hypospadias from January 2006 to January 2011. The accompanying severe chordee (30°–50°) was corrected via urethral plate mobilization and transection. The subtypes were classified according to the position of the meatus after the curvatures were corrected. We excluded patients without urethral plate transection during the chordee correction.

After penile straightening, a single surgeon made primary repairs in 66 patients who had hypospadias with severe chordee. In all, 32 (48.48%) patients underwent single-stage urethroplasty, and 34 (51.52%) underwent two-stage urethroplasty. The choice of technique depended on the surgeon's preference. The ages of patients in the single-stage group ranged from 2 to 35 years. Among them, 19 were prepubescent (0–10 years), and 13 were postpubescent (11 years and older). The ages of patients in the two-stage group ranged from 2 to 43 years. Among them, 16 were prepubescent, and 18 were postpubescent.

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Single-stage repair

The single-stage repair (Duckett technique) was performed according to the description in the literature.³ A circumferential incision was made proximal to the corona and reached a depth of Buck's fascia. The dorsal skin was degloved toward the proximal penis, and the scar fiber around the corpus spongiosum was excised to release the chordee. An artificial erection was induced to identify the site and degree of chordee. The urethral plate was transected to correct the accompanying chordee after the ventral dissection and urethral plate mobilization failed to release the curvature completely. After urethral plate transection, the meatus dropped back to the penoscrotal junction or the proximal shaft. If the curvature was not completely corrected, dorsal plication or a patch of tunica vaginalis free graft was used to correct the refractory penile curvature. The distance between the retracted meatus and the glans tip was measured to confirm the expected length of the neourethra. The rectangular flap was outlined at the inner aspect of the dorsal prepuce with methylthioninium chloride according to the length of the defect. The outlined foreskin was incised and rolled into a tube over a catheter and sutured with 6-0 polydioxanone sutures. The size of the catheter ranged from 8 to 14 Fr and depended on the diameter of the patient's urethra. The tubularized neourethra was transposed ventrally and anastomosed with the native urethra with mucosa-to-mucosa sutures. The glans was incised deeply, the neourethra was placed, and the new meatus was sutured on top of the glans.

Two-stage repair

The first steps of the two-stage repair (incision, degloving the penis, releasing the curvature) were identical to those described for the single-stage repair. A rectangular buccal mucosa graft or transverse inner dorsal preputial pedicle flap was dissected according to the length of the defect. The glans was then split to facilitate dissection of the glans wings. The grafts or flaps were quilted from the native plate to the glans to create a neourethral plate. A protective tie-over dressing was placed to reduce the chance of hematoma collecting under the graft.

The repair was completed 6 months later using the Snodgrass modification of the standard Thiersch–Duplay technique. The neourethral plate was deeply incised along its entire length, followed by tubularization over a 8-Fr to 14-Fr urethral stent.

In both the one-stage and two-stage techniques, the second layers of subcutaneous tissue were placed along the neourethra with a tension-free closure over the repair as far as possible. Urethral catheters were used to drain the urine for 8–10 days in patients with the single-stage repair. In those with a two-stage repair, the urethral catheter was removed on postoperative day (POD) 4 after the first-stage and on POD 8–10 after the second-stage. Antibiotics were administered intravenously for at least 5 days.

RESULTS

All of the surgical procedures went smoothly. The demographic data for the two groups are shown in **Table 1**. There were no statistically significant differences between the groups. The meatus of the neourethra was located at the top of the glans. No recurrence of chordee was found during follow-up, and all patients or parents were satisfied with the penile length and appearance.

In the one-stage group, seven patients underwent further dorsal plication to resolve the refractory penile curvature. In the two-stage group, five patients underwent the same procedure to correct refractory penile curvatures. One patient underwent tunica vaginalis free graft corporoplasty to make the correction.

Table 1: Complications in the two groups

	Group 1 (n=32)	Group 2 (n=34)	Total (n=66)	P
Patients with complications, n (%)	8 (25)	8 (23.53)	16 (24.24)	0.8892
Early complications, n (%)	6 (18.75)	8 (23.53)	14 (21.21)	0.6350
Fistula (n)	5	7	12	0.2201
Dehiscence and infection (n)	1	1	2	0.5072
Late complications, n (%)	6 (18.75)	1 (2.94)	7 (10.61)	0.0396
Stricture (n)	6	0	6	0.01
Meatal stenosis (n)	0	1	1	0.5152
Diverticula (n)	0	0	0	-
Hair growth or uroliths (n)	0	0	0	-
Median age (year)	7.5	11	-	-
Median urethra defect (cm)	4	5	-	-
Median hospital stay (day)	13	19	-	-

Group 1: single-stage group; Group 2: two-stage group

In all cases, the transurethral catheter was withdrawn 8–10 days postoperatively, and the patients were discharged following catheter removal. Median hospital stays were 13 and 19 days, respectively, in single- and two-stage groups.

Concomitant penoscrotal transposition was undertaken in 3 of 34 patients in the two-stage repair group. Patients were required to visit our clinic 6–12 months after the operation for outpatient follow-up examination. They were then followed through a combination of telephone interviews and outpatient follow-up visits. The median follow-up for the one-stage group was 28.5 months (20–60 months), and that for the two-stage group was 35 months (18–60 months).

No complications were observed during the first-stage in the two-stage group. However, 12 complications were observed in eight patients in the single-stage group and 9 complications in eight patients during the second-stage in the two-stage group (**Table 1**). There was no significant difference in the overall complication rates between the single- and two-stage groups ($P = 0.8892$).

Complications after urination on POD 7 were recorded as the early results.⁷ Any others were defined as late complications. The early complications included fistulas, dehiscence, and infection. Late complications, observed during the long-term follow-up period, included meatal stenosis, stricture, diverticula, and hair growth or uroliths. There was no difference between the two groups regarding the early complications (6/32 vs 8/34, $P = 0.6350$). Patients with a fistula underwent repair 6 months postoperatively and were cured with a single procedure. The late complication rate was higher in patients who had undergone the one-stage procedure ($P = 0.0396$). In that group, six patients with strictures were cured after 4- to 6-month repeated dilations. One of the patients with stenosis underwent metal incision 1 year later.

All patients were divided into two cohorts: prepubescent (0–10 years) and postpubescent (≥ 11 years) according to their ages at the time of surgery following the Chinese pubescent classification criteria.^{8,9} The complication rates for these two cohorts were significantly different in the single-stage group (2/19 vs 6/13, $P = 0.0279$). In contrast, no difference was observed between these two cohorts in the two-stage group (3/16 vs 5/18, $P = 0.2643$). There were no statistically significant differences between the two groups in the prepubescent or postpubescent cohort (**Table 2**).

After complete release of the chordee, patients were divided into two groups according to the location of the meatus. In all, 3 of 18 (16.67%) and 13 of 48 (27.08%) patients had complications in

the proximal shaft and the penoscrotal area, respectively. A slight higher complication rate was found in association with penoscrotal hypospadias, but there was no significant difference between the two subtypes ($P = 0.1840$). The complications are presented in **Table 3**.

DISCUSSION

Hypospadias with severe chordee is a challenge for most urologists and orthopedists. The associated severe chordee should be corrected first. Various methods are available, including resection of ventral fibrous tissue or dorsal plication.¹⁰ In cases of severe chordee, grafts such as a corporeal tunica vaginalis free graft, buccal mucosa graft, one created from small intestinal submucosa, and flaps have been used to achieve full correction of the penile curvature.^{11–15}

We used a step-wise approach that had been described in the literature¹⁶ for penile straightening. Initial steps were skin degloving, ventral dissection, and urethral plate mobilization. Later, the urethral plate was transected to correct a residual curvature of more than 30°. Ventral lengthening using a free graft or flap was the final step.

Dorsal plication can be used before or after urethral plate transection. A recent worldwide survey suggested that surgeons preferred plication to urethral division and ventral grafting techniques in this range.¹⁷ However, simple dorsal plication was reported to be associated with a recurrence rate of 37% compared with no curvature recurrence after simultaneous dorsal plication and urethral plate transection in patients who had proximal hypospadias with severe ventral curvature.¹⁸ Transection of the urethral plate may reduce the recurrence rate of chordee, especially in cases of proximal hypospadias with severe chordee. In our study, no recurrence of chordee was found after nearly 3 years of follow-up. Although the interval is not long enough to rule out a recurrent curvature, the results at least indicate that urethral plate transection is effective for correcting severe chordee in proximal hypospadias during the intermediate period. Furthermore, all of our patients or their parents were satisfied with the penile length after chordee correction and urethroplasty, which is consistent with Castagnetti's findings.¹⁹

After the urethral plates are transected, the options are the one-stage and two-stage procedures. Because the management has yet not been standardized, urologists choose a technique with which they are comfortable. One-stage repair has been a conventional treatment of hypospadias with its advantages of less extensive surgery and lower cost. Examples are the modified Koyanagi repair¹⁵ and the onlay repair.^{16,20} In severe chordee cases during which a poor urethral plate must be transected, these techniques usually cannot reconstruct the neourethra in one-stage. An exception is simultaneous buccal or bladder mucosa

graft implantation to repair the urethral plate defect.^{11,21,22} The tubularized transverse preputial flap (Duckett technique^{4,7,8}) has been used classically to repair proximal hypospadias with severe chordee, especially in cases that require urethral plate transection.

In recent years, there has been renewed interest in treating those severe cases with two-stage repair.^{15,23} Although the two-stage procedures are more time-consuming and more costly, they usually provide a healthier urethral bed and less scar tissue around the urethra. Springer *et al.*¹⁷ demonstrated that the two-stage repair was preferred by 43.3%–76.6% of surgeons for repairing proximal hypospadias, although this conclusion was based on personal experience. In theory, the two-stage repair should always result in better cosmetic and functional outcomes as the delay between grafting and closure allows all scarring and contractions to occur around the meatus before the second-stage, which provides a better chance of success.⁶ The two-stage procedure was performed for severe proximal hypospadias with satisfactory results and was considered a versatile technique.^{6,12} The second-stage, performed as a Thiersch–Duplay urethroplasty using the Snodgrass modification, was demonstrated to be an effective technique with excellent short-term outcomes for penoscrotal hypospadias.²⁴

The overall complication rates of one-stage and two-stage procedures were similar, although the types of complication were different. Stricture has been the main complication in patients undergoing the single-stage (Duckett) procedure. It occurs late at 20%–50% rate and usually leads to secondary procedures.²⁵ Compared with fistulas, stricture is more severe and difficult to treat, especially in infants. It sometimes requires salvage urethrotomy and even redo surgery for urethral reconstruction. Strictures are mainly caused by a circular anastomosis between the neourethra and the native urethra. Onlay repair could significantly decrease the stricture rate if a semi-circular anastomosis is created during one-stage procedures. In patients with urethral plate transection, the onlay procedure must be performed with simultaneous urethral plate substitution by graft implantation. In the two-stage group, the Thiersch–Duplay urethroplasty/tubularized incised plate has resulted in a lower stricture rate. Furthermore, with advances in the neourethral plate, more of the plate could be incised to minimize the incidence of strictures.²⁵

Complication rates in prepubescent and postpubescent cohorts in the single-stage group were significantly different, which we mentioned in an earlier study.⁷ The difference may be caused by spontaneous penile erection, which was more common in the postpubescent group. However, no difference was observed between the two cohorts in the two-stage group. We suspected that the two-stage procedure itself may be a key factor in weakening the role of age in the urethroplasty results. In two-stage repairs, free grafts and flaps could be harvested that were long and wide enough to reconstruct a well-vascularized new urethral plates after 6 months, which may reduce penile erection tension. It also allowed correction before tubularization and avoided neourethral stricture when the new urethral plate was considered poorly vascularized.²⁶

Regarding surgical techniques, the Duplay technique is not as complicated as the Duckett technique. MacGillivray *et al.*²⁷ suggested

Table 2: Complications and age cohorts

Age at operation (years)	Group 1	Group 2	P ^a
0–10	2/19	3/16	0.2950
11	6/13	5/18	0.1736
P ^b	0.0279	0.2643	

Group 1: single-stage group; Group 2: two-stage group. ^aP value is the comparative result between two groups in the same age cohort; ^bP value is the comparative result between two age cohorts in the same group

Table 3: Complications and meatal location

	Number of patients (n)	Number of patients with complication (n)	Fistula (n)	Dehiscence and infection (n)	Stricture (n)	Meatus stenosis (n)	P
Proximal shaft	18	3	2	0	3	0	>0.05
Penoscrotal	48	13	10	2	3	1	
Total	66	16	12	2	6	1	

that more complications develop as the procedure becomes more complicated or extensive. A simple surgical technique may reduce the complexity of urethroplasty, thereby offering better risk control.

We also roughly analyzed the relation between the meatus location and the appearance of complications in our study. Although a slight higher complication rate was encountered in patients with penoscrotal (27.08%) hypospadias than in those with proximal shaft (16.67%) hypospadias, there was no statistically significant difference between these two groups. Moursy²⁵ mentioned a similar result. Theoretically, a longer urethral defect may increase the difficulty of the surgery, leading to a higher complication rate. However, the different compositions of the one- and two-stage techniques in either subtype group or the small sample size may cause confounding interference in the statistical analysis.

The major limitation of our study was its retrospective nature, which leads to a less restrictive design than would be seen in a prospective comparative study. The follow-up is rather short for detecting diverticular formation or penile curvature recurrence. That there is incomplete information about voiding stream analysis, quality of life, sexual function, and penile cosmetic appearance evaluation also limits estimation of the final results. However, taking into consideration the continuing debate on the best approach to treating proximal hypospadias with severe chordee, especially whether single-stage or two-stage surgery is preferred, this study provides information to help the surgeon regarding treatment decisions. Our future research will focus on a prospective random controlled trial with different urethroplasties for treating proximal hypospadias with severe chordee.

CONCLUSION

Urethral plate transection can correct severe chordee associated with proximal hypospadias, at least up to and during the intermediate period of our study. Although the overall complication rates of the single-stage urethroplasty using the Duckett procedure and the two-stage urethroplasty were not statistically different, stricture, a rather severe complication, appeared at a higher rate in the single-stage group. Therefore, we believe that the two-stage urethroplasty is preferred for treating proximal hypospadias with severe chordee after correction via urethral plate mobilization and transection. In contrast, the Duckett technique should be used with caution in this situation. In the single-stage group, the complication rates between prepubescent and postpubescent cohorts were significant different, whereas no difference was observed between the two cohorts in the two-stage group. Future research should focus on a prospective random controlled trial with different urethroplasties for treating proximal hypospadias with severe chordee.

AUTHOR CONTRIBUTIONS

DCZ and HJY conceived of the study, participated in its design and coordination, and drafted the manuscript. ZKC performed the surgeries. DCZ and HJY conducted the outpatient service. DCZ, QC, JD, MXX, KZ, MJL, and YBC participated in data acquisition. DCZ, MJL, and ZW made critical revisions to the manuscript regarding important intellectual content.

COMPETING INTERESTS

All authors declare no competing interests.

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