


Original Article: Clinical Investigation**Efficacy of one- and two-stage segment urethroplasty for severe chordee with congenital short urethra**Guanglun Zhou,[†] Jianchun Yin,[†] Junjie Sun, Zhilin Yang and Shoulin Li 

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Abbreviation & Acronym

CSU = congenital short urethra

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Objectives: The aim of this study was to analyze the efficacy of segment urethroplasty to treat severe chordee with congenital short urethra and compared one-stage and two-stage segment urethroplasty. This procedure involved urethral transection to correct the chordee, and urethroplasty was performed to restore natural penile length.

Methods: We retrospectively studied a cohort of patients with severe chordee and congenital short urethra who underwent one- or two-stage segment urethroplasty at our institution between February 2006 and May 2020. We evaluated the efficacy of the procedures based on the incidence of complications.

Results: A total of 37 children were included in this study: 25 were treated with two-stage segment urethroplasty and 12 were treated with one-stage segment urethroplasty. The median length of neourethra in the one-stage repair group (3.21 cm) was similar to that in the two-stage repair group (3.23 cm; $P > 0.05$). Of the 37 patients, 32 (86.5%) were cured after urethroplasty. There were three patients with complications in the one-stage repair group (one urethral fistula and two urethral strictures) and two patients with fistula in the two-stage repair group. Among the five patients with complications, the three fistulas were successfully repaired through reoperation and the two urethral strictures were cured after urethral dilatation. No patient had diverticulum or recurrent chordee.

Conclusions: Segment urethroplasty achieved satisfactory outcomes in the treatment of severe chordee with congenital short urethra. This can restore natural penile length, and the recurrence rate of severe chordee is low. The overall success rate of the two-stage procedure tends to be better than that of the one-stage procedure.

Key words: children, segment, severe chordee, two-stage, urethroplasty.

Introduction

Congenital chordee is usually associated with hypospadias, and it is unusual in children with orthotopic meatus.¹ Chordee without hypospadias is a challenging condition because of the lack of clear agreement regarding its etiology and optimal surgical management.² Donnahoo *et al.*³ reported their experience managing pediatric chordee without hypospadias and developed a classification system according to the etiology of the chordee. They were classified into type I (skin chordee), type II (fibrotic Buck's and dartos fasciae), type III (corporeal disproportion), and type IV (CSU).³

Severe chordee with CSU is uncommon.³ Although surgical correction of severe chordee with CSU is the only therapy in these patients, none of the procedures have been satisfactory or standardized to date.³ Surgical correction of severe chordee with CSU may be difficult, and significant postoperative complications may occur due to the need for urethrotomy and urethral reconstruction.⁴

At present, there are few studies on the procedure for and outcomes of severe chordee with CSU. To establish a satisfactory procedure, this study aimed to retrospectively analyze the long-term results of segment urethroplasty for severe chordee with CSU and to compare the results between one-stage and two-stage segment urethroplasty.

Methods

Study design and patients

We retrospectively reviewed the records of severe chordee patients with CSU who were referred for primary treatment from February 2006 to May 2020. Photo documentation of the children's erect penis provided by their guardians showed the severe curvature from different angles before surgery. Severe chordee due to a short urethra persisted in all patients with curvature of $>30^\circ$ after degloving (the exact degree of curvature is generally determined at the time of surgery using an artificial erection test). A total of 38 boys underwent one- or two-stage segment urethroplasty at the Shenzhen Children's Hospital during this period. The patients were divided into one- and two-stage repair groups according to surgical technique. Severe chordee with CSU was defined as a meatus on the normal location, curvature $>30^\circ$ after degloving, and curvature due to a short urethra^{3,4} (Fig. 1a). Written informed consent was obtained from the guardians or parents before treatment. All patients underwent surgery by a single surgeon. The choice of one-stage or two-stage segment urethroplasty was based on the surgeon's preference. Data on patient demographics, surgical technique details, postoperative complications, and uroflowmetry were reviewed. One patient was excluded from this study due to missing data on operative details. Patients with a paper-thin hypoplastic distal urethra or a history of penile surgery were also excluded from this study. The study was approved by our institutional ethics review board (2021033) and conforms to the provisions of the Declaration of Helsinki.

Surgical technique

One-stage segment urethroplasty

Circumcision was made, and penile degloving was performed. After removal of the tethered tissue in the ventral region, the degree of curvature was reassessed. When severe chordee due to a short urethra persisted, the urethra was incised at the point of maximum curvature to promote penile

straightening. In a few patients, urethral transection was not enough to straighten the penis, and a single dorsal plication was performed. Artificial penile erection was induced to ensure penile straightening and measure the length of the urethral defect (Fig. 1b,c). Based on the length of the urethral defect, a transverse preputial island flap of suitable size was carefully designed, and then tubed over a urethral sound to create a new urethral segment. The segment was then anastomosed with the distal and proximal ends of the divided urethra, which was restored. Closure of the urethral plate was achieved with running sutures using single-layer 7/0 polydioxanone sutures. The fascia and vessel pedicle were draped over the reconstructed urethra, and ventral skin closure was performed. The urethral catheter was reserved for 10–14 days after surgery.

Two-stage segment urethroplasty

The first stage of the two-stage segment urethroplasty (penile degloving, removal of the tethered tissue, urethral transection, and release of curvature) was identical to those described in one-stage segment urethroplasty. Then, Byars' flaps were created, and they covered the ventral urethral defect and closed it along the midline (Fig. 2a).

The second stage of segment urethroplasty was performed at least 6 months after the first stage. The Johanson procedure was performed to reconstruct the urethral defect (Fig. 2b). A double U-shaped incision was made demarcating the neourethra, and a 12–15-mm-wide incision was designed along the ventral side of the penis that extended from the proximal meatus to the distal meatus end of the transected urethra. The Buck's fascia was dissected, and an 8- or 10-Fr silicone catheter was used for the tubularization of the segment neourethra. Closure of the urethral plate was achieved with running sutures using single-layer 7/0 polydioxanone sutures. The second layer of the fascia vessel pedicle was placed along the neourethra, and the skin was closed (Fig. 2c). The urethral sutures were only in the midline between the new segment with proximal and distal segments. The urethral catheter was reserved for 10–14 days after surgery.

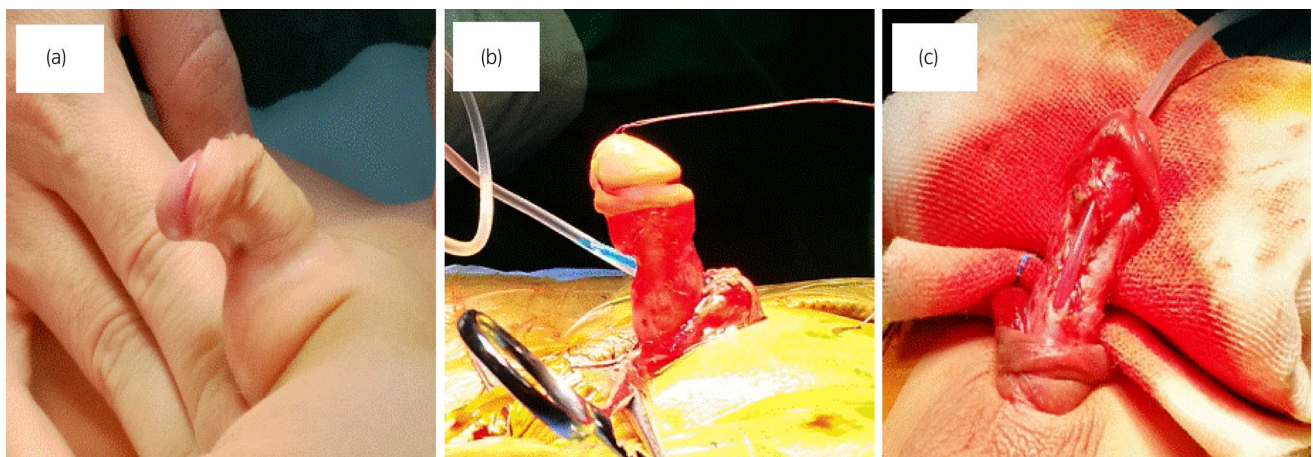


Fig. 1 Preoperative and intraoperative images of (a) severe chordee caused by the short urethra, (b) an artificial penile erection induced to ensure penile straightening after urethral transection, and (c) the length of urethral defect.

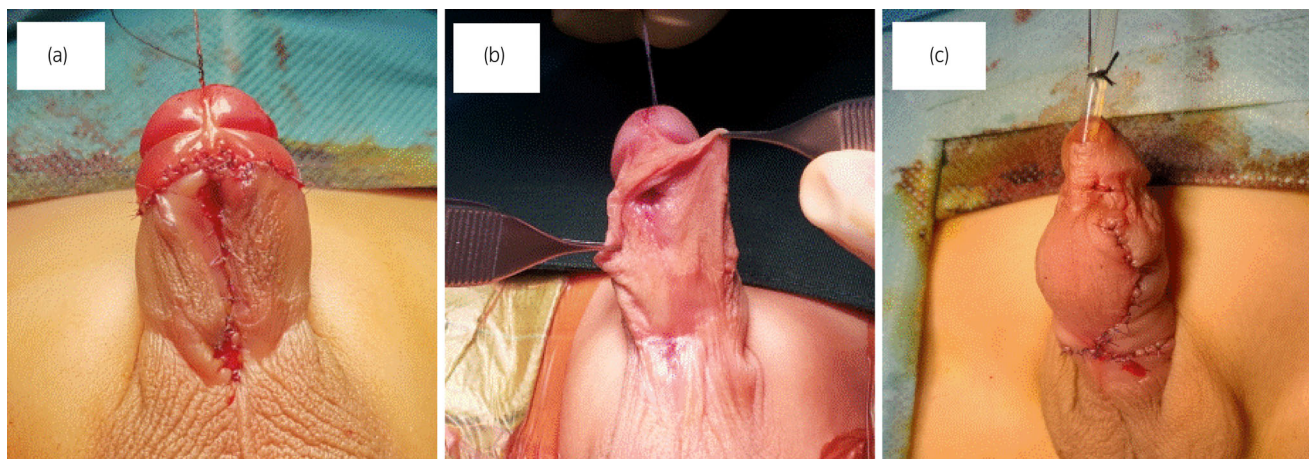


Fig. 2 Preoperative and intraoperative images of (a) reconstruction of new urethral plate with Byars' flap, (b) the length of urethral defect and preparing for segment urethroplasty, and (c) the appearance of penis after the second stage of segment urethroplasty.

Follow-up

All patients were reassessed at 1, 3, 6, and 12 months after normal urethral continuity was achieved. Thereafter, follow-up visits were made annually. The appearance of penile erection and micturition were evaluated, and uroflowmetry was performed during each follow-up visit.

Statistical analysis

Statistical analyses were performed using SPSS version 22.0 (IBM SPSS Inc., Chicago, IL, USA), and a P value of <0.05 was considered statistically significant. Continuous variables that followed a normal distribution are presented as mean \pm standard deviation and categorical variables as numbers and percentages. Fisher's exact test, the chi-squared test, and the t -test were performed for the study, as appropriate.

Results

A total of 37 children were included in this study; 25 in the two-stage repair group and 12 in one-stage repair group. The median age at surgery in the one-stage repair group (57.5 months, range 34–126 months) and the first surgery in the two-stage repair group (56.2 months, range 32–118 months; $P > 0.05$) were similar. The median length of urethral defect or neourethra in the one-stage repair group (3.21 cm, range 2.3–4.4 cm) was similar to that in the two-stage repair group (3.23 cm, range 2.3–4.5 cm; $P > 0.05$). There was no significant difference in the follow-up time between the two groups. The patients' demographic characteristics and outcomes are presented in Table 1. In 86.5% of patients (32/37), sufficient penile straightening was achieved based on penile degloving and transected urethra. Only 13.5% of patients (5/37) had persistent mild chordee after urethral transection (three cases in the two-stage repair group and two cases in the one-stage repair group), and they underwent additional dorsal plication (two patients underwent simultaneous dorsal plication and ventral corporotomies in the two-stage repair group).

The median follow-up duration was 60 months. Of the 37 patients, 32 (86.5%) were cured after urethroplasty. There were three patients with complications in the one-stage repair group (25%), including one patient with urethral fistula and two with urethral strictures, compared with only two patients with fistulas in the two-stage repair group. In the five patients with complications, three patients with fistulas were successfully treated through fistula repair 6 months later with a pedicled fascia coverage technique, two patients with urethral strictures were cured after urethral dilatation under anesthesia. During the follow-up period, there was no patient with urethral diverticulum or recurrent chordee. The overall cosmetic appearance of each penis was satisfactory.

The maximal urinary flow rate was lower in the one-stage repair group (6.6 ± 1.3 mL/s) than in the two-stage repair group (7.5 ± 1.1 mL/s; $P < 0.05$) at 1 month postoperatively. However, the maximal urinary flow rate was similar between the one-stage repair and two-stage repair groups at 6 months postoperatively (7.9 ± 1.7 vs 8.3 ± 1.8 mL/s; $P > 0.05$).

Discussion

The ultimate objective of any surgical technique used to correct chordee is to achieve penile straightening and corpora of

Table 1 Demographic and clinical outcomes of the one-stage and two-stage repair groups

Observations	One-stage repair	Two-stage repair	P -value
No. of patients	12	25	
Age at first surgery, months (range)	57.5 (34–126)	56.2 (32–118)	0.64
Length of neourethra, cm (range)	3.21 (2.3–4.4)	3.23 (2.3–4.5)	0.77
Follow-up, months (range)	60.5 (21–142)	59.3 (16–136)	0.85
Complications, n (%)	3 (25)	2 (8)	0.30
Urethral fistula	1 (8.3)	2 (8)	
Urethral stricture	2 (16.7)	0 (0)	

a similar size.⁵ In this study, the urethral corpus spongiosum were significantly shorter than the penile corpus cavernosum in patients with severe chordee caused by CSU. Segment urethroplasty can achieve satisfactory outcomes in the treatment of severe chordee with CSU.

Segment urethroplasty for severe chordee with CSU can avoid postoperative shortening of the penis. Various techniques are used to treat chordee, including simple penile degloving and dorsal plication, corporal rotation, and complete penile disassembly procedures.^{6,7} These methods can preserve the urethra, but cause postoperative shortening of the penis with an average loss of 2.5 cm.^{8,9} Although many patients can still maintain erectile function, loss of penile length may lead to physical and psychological disturbances in patients undergoing surgical correction.¹⁰ Severe chordee with CSU may reduce penile length after correcting the chordee using the aforementioned methods. Schneider *et al.*¹¹ reported that penile curvature after plication in a few patients can shorten the penis by as much as 5 cm. Moreover, Greenfield *et al.*¹⁰ pointed out that penile shortening is a complication after correcting chordee. Therefore, the above-mentioned methods are not suitable for severe chordee with CSU. On the other hand, Donnhoo *et al.*³ and Singh *et al.*¹² suggested that CSU required urethral division or excision of the distal urethral, followed by one-stage urethroplasty. In this study, segment urethroplasty restored natural penile length by reconstructing the urethra to lengthen the ventral side of the penile and avoid dorsal plication. Our results demonstrate that the average length of reconstructed urethra was more than 3.2 cm after correcting severe chordee, which maintains the length of the urethra consistent with the length of the penile corpus cavernosum and avoids unacceptable penile shortening after surgery.

Segment urethroplasty for severe chordee with CSU achieved low complication rates. To the best of our knowledge, there have been few studies on this disorder. In some small case series studied retrospectively, the complication rate was higher than that observed in our study. Singh *et al.*¹² reported that excision of the urethra from the meatus and one-stage urethroplasty (tubularized transverse inner preputial island flap) were performed in four patients with this disorder (mean age 5.30 years) and the incidence of complications was as high as 50% (follow-up 6 months to 9 years), including a fistula rate of 50% and stricture rate of 50%, and a reoperation rate for complications of 50%. Similarly, Donnhoo *et al.*³ reported that six patients with CSU (median age 14 months) requiring urethral division and one-stage urethral replacement (creation of a tubularized preputial graft) had a complication rate of 50% (follow-up 1 week to 72 months), including a fistula rate of 66.7% and chordee recurrence of 33.3%; the reoperation rate of complications was 50%. Tang *et al.*¹ reported that the distal urethra in nine patients (mean age 76.8 months) was resected and replaced with one-stage longitudinal island flap urethroplasty. The complication rate was 15.8% (follow-up 2 to 63 months), including a fistula rate of 5.3%, a stricture rate of 5.3%, and chordee recurrence of 5.3%; the reoperation rate for complications was 15.8%. In contrast, the segment urethroplasty procedure provided a success rate of 86.5%, and the reoperation rate for complications was only 8.1%. There are four possible reasons for this high success rate. First, severe

chordee with CSU can be corrected more thoroughly by transecting the urethra and avoiding chordee recurrence caused by excessive dorsal plication. Second, only the urethral segment was reconstructed in the ventral middle of the penis, and the distal urethral segment was preserved, avoiding the coronal fistula and meatal stenosis. Third, most patients had a decreased incidence of stricture owing to the use of semicircular anastomosis. Fourth, compared to other studies, most of the patients in this study underwent two-staged repair. In addition, segment urethroplasty provided a satisfactory penile appearance because it retained the natural shape of the glans and meatus.

When severe chordee is present with CSU, even if the meatus is normal, it should be incorporated into the hypospadias repair schedule.¹² The procedure for urethroplasty includes one-stage or two-stage surgery.¹³ Since 1980, one-stage transverse preputial island flap urethroplasty for proximal hypospadias has been increasingly used.¹³ However, it was found that this approach is associated with complicated steps, a high complication rate, and long learning curves in clinical practice.¹³ The one-stage repair was gradually converted to a two-stage repair based on improvement in complication rates and cosmetic results.¹³ Similarly, severe chordee with CSU was performed using a one-stage transverse preputial island flap of segment urethroplasty in the early stages of treatment, but we have mainly adopted two-stage procedures to treat this disease in recent years.

The main complications after correction of severe chordee with CSU were chordee recurrence, urinary fistula, and urethral stricture.³ In this study, we found no recurrence of penile curvature after urethroplasty, which may be due to urethral transection to completely release the curvature and the avoidance or reduction of dorsal plication. Measuring the degree of penile curvature after correction of the chordee is crucial; this helps to avoid persistent curvature.

In this study, the incidence of fistula in the two-stage repair group was similar to that in the one-stage repair group. All urinary fistulas were located in the ventral midpiece of the penis without coronal fistulas. These fistulas are usually repaired with a high success rate (100%) due to adequate subcutaneous fascial insertion and pedunculated flap coverage. In addition, we fulfilled the following conditions:¹⁴ (i) fistula repairs were performed at least 6 months after urethroplasty, when inflammation resolution and skin vascularization around the fistula; and (ii) during the surgery, we dissected the fistula tract up to its base and interposed the ventral dartos flap to the edge of the fistula so that it did not overlap with the suture line.

Uroflowmetry is a common objective index to evaluate micturition function, especially urethral stricture.¹⁵ An obstructive urinary flow pattern was common in the early period after urethroplasty. This obstructive pattern may resolve spontaneously with time.¹⁶ Similarly, our results demonstrated that the maximal urinary flow rate was low in the short term after urethroplasty, and improved progressively and spontaneously in the long term. This may be because the new urethra may lack the structural stability provided by the surrounding corpus spongiosum, and the urethral scar softened gradually.¹⁶ However, when a low urinary flow rate is present with symptoms of dysuria, further treatment is necessary.¹³ Although urethral stricture is less common than urinary fistula, the treatment is more complex, and severe urethral stricture may require

urethroplasty.¹⁷ Fortunately, in our study, the two patients with strictures were cured by urethral dilation. Interestingly, all strictures occurred in the single-stage repair group. Helmy *et al.*¹⁸ reported that neourethral stenosis is mainly caused by the circular anastomosis between the native urethra and neourethra, and it also occurs secondary to ischemia and tension. In this study, the neourethra was anastomosed using a circular anastomosis with the proximal and distal original urethra in one-stage segment urethroplasty. This may lead to an increased risk of urethral strictures, while a semicircular anastomosis technique was used in the second-stage segment urethroplasty. Moreover, one-stage transverse preputial island flap repair may be more prone to neourethral ischemia or tension than repair using the Byars' flap tubed to form the neourethra.

Our study has some limitations. It was a single-institution retrospective review of severe chordee with CSU. In addition, the number of cases in the single-stage group was relatively small because most patients undergo staged repair based on surgeon preference, which may have had an impact on the results of the statistical analysis of the data. However, the small sample size might also be explained by the fact that this disease is uncommon. Further, patients require longer follow-up periods to obtain puberty data.

Based on our experience, segment urethroplasty is suitable for severe chordee with a CSU. This technique can restore natural penile length, and the recurrence rate of chordees is low. The overall success rate of the two-stage procedure tends to be better than that of the one-stage procedure and has simple surgical steps. We recommend two-stage repair for this disease. The learning curve for the one-stage procedure for severe chordee with CSU is relatively long, and residents in training should be started with the two-stage procedure.

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Author contributions

Guanglun Zhou: Data curation; Investigation; Writing – original draft. Jianchun Yin: Data curation; Investigation; Methodology; Project administration. Junjie Sun: Investigation; Methodology; Project administration; Supervision; Validation. Zhilin Yang: Data curation; Investigation; Methodology. Shoulin Li: Investigation; Supervision; Validation; Writing – review and editing.

Conflict of interest

None declared.

Approval of the research protocol by an Institutional Reviewer Board

Shenzhen Children's Hospital ethics committee (2021033).

Informed consent

Written informed consent was obtained from the guardians or parents before treatment.

Registry and the Registration No. of the study/trial

N/A.

Animal studies

N/A.

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