Kurdi *BMC Urology* (2025) 25:23
https://doi.org/10.1186/s12894-025-01699-6

BMC Urology

RESEARCH Open Access

Chordee and hooded prepuce with no hypospadias; outcome of urethral preservation surgery with spongioplasty

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Abstract

Background Deficient ventral prepuce is a rare anomaly of prepuce with normally situated meatus. It is usually associated with this or hypoplastic urethra.

Methods A retrospective study included all infants diagnosed with a ventrally hooded prepuce. A penile degloving was the first step. Then reinforcement of the hypoplastic urethra by spongioplasty was performed. Sleeve resection of the hooded prepuce and reconstruction of sulcus corona was the last step.

Results The mean operative time was 44 ± 9.5 min. A hypoplastic urethra was reported in 35 cases (30.4%). A urethral stent was inserted at the beginning of the procedure and removed at the end. Twelve patients (10.4%) developed urethral cutaneous fistulas by the end of the first postoperative month.

Conclusion A hooded ventral prepuce with ventral chordee and normally situated meatus presents a challenge for paediatric surgeons. It is usually associated with a hypoplastic urethra. Attempts should be made to correct the chordee and preserve the urethra. The preserved urethra can be reinforced with spongioplasty. Unintended urethral injuries are common during dissection. Therefore, repairing the injury, reinforcing the dartos pedicle flap, and spongioplasty are suitable options. It is possible to preserve the urethra in patients having chordee without hypospadias. Utmost care should be taken to avoid urethral injuries which are common because shaft skin is stuck to the urethral skin. Urethral injuries can be repaired primarily; however, there is a high incidence of fistula.

Introduction

The prepuce forms a complete sleeve around the glans in healthy male children. In cases of hypospadias, it may be deficient on the ventral side. Regardless of the anatomical type, hypospadias is classically associated with ventral chordee, which may be caused by tethering of the ventral skin or disproportionate corporeal bodies [1].

Whatever the cause of the chordee, it remained a pathognomonic feature of hypospadias and should be addressed during the primary management [2].

Some male infants have a ventrally deficient prepuce without hypospadias. However, these cases may be associated with a ventral chordee and a thin transparent urethra [3].

Some complications might arise when addressing the management of this problem. These complications included urethral injury, conversion of the case to classic hypospadias case, glanular tilt, ventral tethering and development of urethrocutaneous fistula. However, these

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complications were developed in severe cases of long membranous urethra with severe chordee. [3.]

This situation presents a problem for which circumcision is planned for various reasons. Therefore, releasing the chordee, resecting the ventrally hooded prepuce, and reinforcing the hypoplastic urethra are challenging for paediatric urology surgeons. The aim was to assess the success of urethral preservation surgery by reconstruction of spongiosum over the urethra, which is devoid of spongiosum, and reconstruction of dartos.

Patients and methods

Study design

This retrospective study included all infants diagnosed with a ventrally hooded prepuce, ventral chordee, hypoplastic urethra, and normal external urethral meatus who underwent surgery between March 2018 and March 2024. We excluded patients with previous penile surgery, a small penis, no chordee, hypospadias, or patients with severe chordee (more than 30°) and long thin paper urethra. This long, thin, hypoplastic urethra was defined by the lesion that extended beyond the sulcus corona, according to Devine and Horton.

Ethics approval and consent to participate

An ethical approval was obtained under code HA-02-J-008 with reference No. 148 – 24 by the unit of biomedical ethics, research ethical committee, Faculty of Medicine, King Abdulaziz University.

The parents or caregivers of all participants in the current study signed an informed consent.

The ethical committee approval code HA-02-J-008 with reference No. 148 – 24.

Operative details

All patients underwent surgery under general anaesthesia with endotracheal intubation. A caudal block was used for postoperative analgesia in all cases. The surgeon re-examined the penis to confirm preoperative findings. Figure 1.

An artificial erection was induced to estimate the degree of chordee. A urinary stent was then inserted. Figure 2.

A circumferential subcoronal incision was made. The ventral skin was meticulously degloved, with special attention focused on the hypoplastic urethra. Urethral injury was avoided by starting the degloving laterally and proceeding medially with gentle skin dissection of the hypoplastic urethra. Figures 3 and 4.

Two cases had a risk of urethral injury. Therefore, the surgeon performed a hydrodissection with normal saline. This procedure helped elevate the thin ventral skin away from the urethra.

After complete degloving, the orthostatic appearance of the penis was rechecked using an artificial erection. The spongiosum tissues on both sides of the hypoplastic urethra was dissected. Then, the pillars of spongiosum tissue were approximated in the midline. Figure 5.

The last step was resecting the hooded prepuce. In this step, the inner perpetual strip was dissected and turned ventrally. The strip was then used to reconstruct the sulcal corona. Cosmetically, it helps separate the glans from the penile shaft. Circumcision was then completed. Figure 6.

The stent could be removed at the end of the procedure, after which the penis was wrapped in a sterile dressing soaked in broad-spectrum antibiotics. Figure 7.

Postoperative follow-up

All patients were followed up weekly at the clinic for the first month. They were followed up every month for the next six months. The recorded data included the development or recurrence of chordee, the occurrence of a urethracutaneous fistula, and the parents' satisfaction. Figure 8

Statistical methods

Statistical analysis was performed using SPSS statistical package v. 21 (IBM SPSS; NY, USA). Numerical data were compared using the independent samples t-test; categorical data were compared using the chi-squared test. Statistical significance was set at $P < 0.05$.

Results

This study included 115 consecutive patients diagnosed with hooded prepuce, ventral chordee, and hypoplastic urethra without hypospadias. These patients were admitted for routine circumcision or were diagnosed with distal penile hypospadias. The patients' mean age was 1.3 ± 0.9 years. No associated anomalies occurred, including undescended testis, congenital inguinal herniae, or hydrocele. The mean operative time was 44 ± 9.5 min. A hypoplastic urethra was reported in 35 cases (30.4%).

Hypoplastic urethras extend to coronal or subcoronal sites on the ventrum of the penis. A urethral stent was inserted at the beginning of the procedure and removed at the end. However, the stent was left for three days in 23 patients (20%). These infants had experienced an unintended urethral injury during the degloving step. Most patients were discharged on the next postoperative day. However, the patients with urethral injuries were discharged home on the fourth postoperative day. At follow-up, none of the patients had developed chordee. Unfortunately, 12 patients (10.4%) developed urethral cutaneous fistulas by the end of the first postoperative month. These patients had a previous urethral injury during surgery. The fistula sites were subcoronal. These

Fig. 1 Examination under general anesthesia

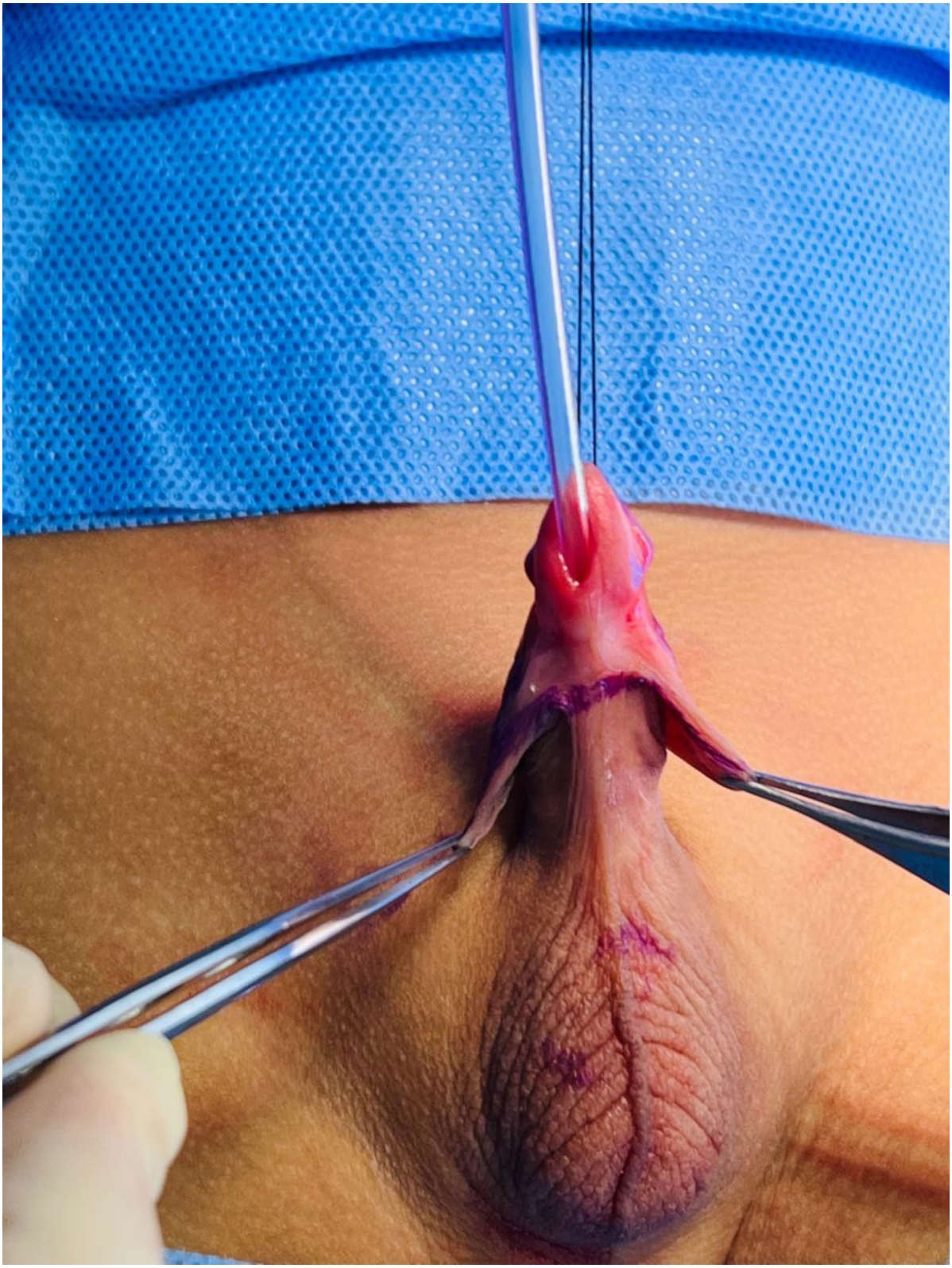

Fig. 2 Artificial erection and stenting of the urethra

Fig. 3 Mapping the incision

patients underwent surgery six months after the primary surgery. The fistulas were dissected and closed in two layers. The patients' conditions were completely normal during follow-up.

Discussion

Although chordee and hooded prepuce are pathognomonic features of hypospadias, the combination of these anatomical features without hypospadias is rare, accounting for 4–10% of hypospadias cases [4, 5].

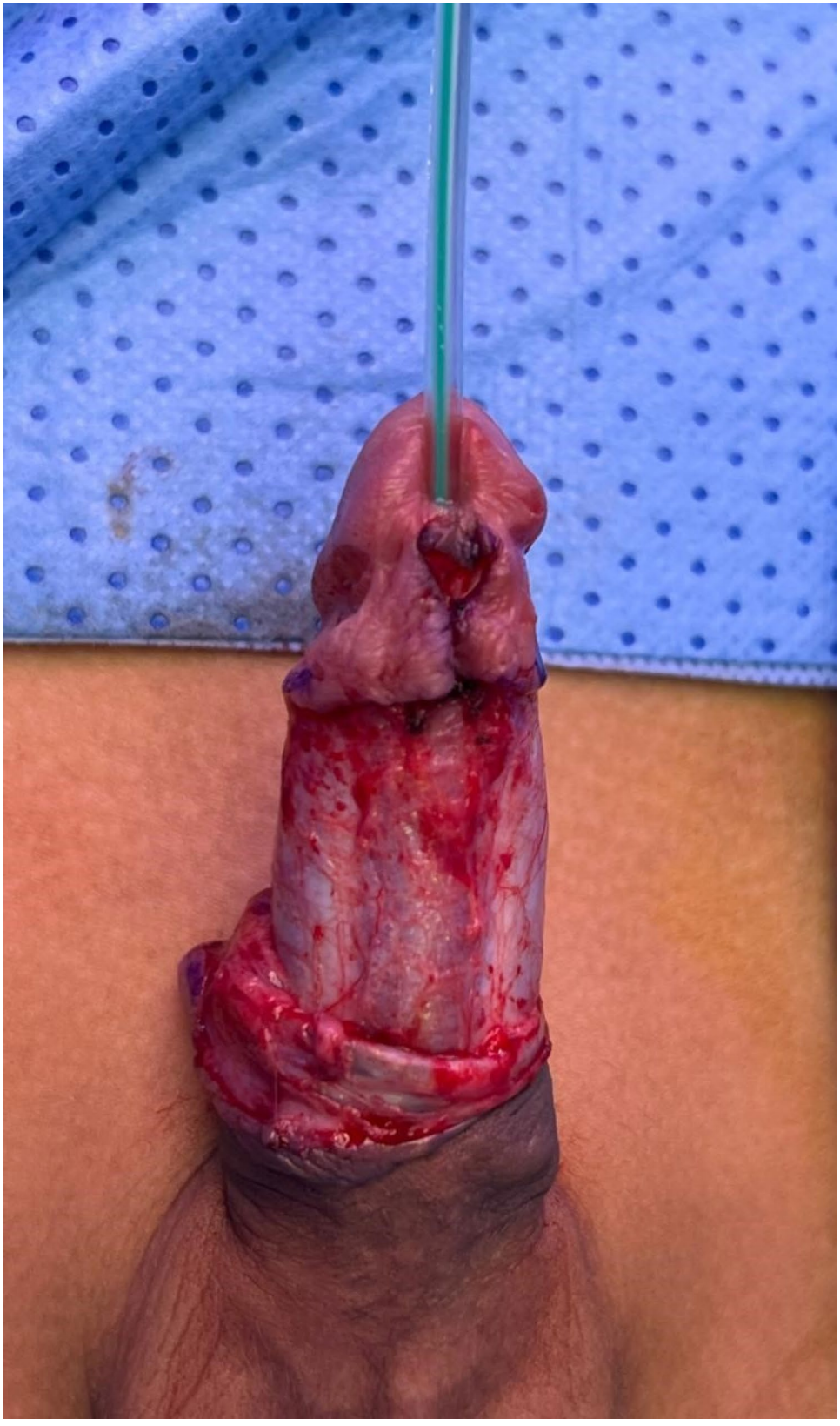

Fig. 4 Subcoronal incision and complete penile skin degloving

Fig. 5 Spongioplasty

Fig. 6 Resection of the prepuce and reconstruction of sulcus corona

Fig. 7 Immediate postoperative appearance

Fig. 8 Postoperative follow up appearance

The abnormal splitting and insertion of the spongiosum tissue causes diving and tethering of the urethra into the corpora, resulting in a characteristic shortening of the ventral part of the penis and possible underdevelopment of the urethra [6].

According to Devine and Horton, chordee is anatomically classified into three types. This division depends on the deficiency of the layers in the ventral part of the penis in the urethra [7].

This study characterised ventral chordee as a thinning of the ventral skin, deficiency of the dartos fascia, and splitting of the spongiosum tissue, clinically corresponding to a deviation of the median raphe from the side of the spongiosum tissue insertion. According to Devine and Horton, this presentation corresponds to type I.

The main concerns of our study were to resect the hooded prepuce and restore penis straightening. The first goal was achieved by anatomical dissection and resection of the hooded prepuce as part of circumcision. Chordee correction was performed by degloving the penile skin, reinforcing the urethra, and repairing the spongiosum tissue. This procedure enabled restoring the normal alignment of the penis layers on the ventral aspect.

Baskin et al., foetal studies described progressive zipping-up of the lateral structures in the midline, which

includes the urethral plate, spongiosum, bucks and dartos fascia, shaft skin and foreskin [8].

The patients operated on could be described as having failure of zipping up of the foreskin, and dartos fascia, buck's fascia, and spongiosum in the shaft with an intact urethra, and the procedure performed by the author could be described as completing the failed zipping-up procedure.

The management of a thin (hypoplastic) urethra, commonly associated with chordee and hooded prepuce conditions, has no consensus. Some authors consider this anomaly a variant of hypospadias; others do not [9].

Several factors directly affect management, including the surgeon's preference, the quality of available penile tissues, and the degree of curvature [10].

However, some reconstruction strategies are planned based on the position of the meatus. If the meatus is in its normal position (not hypospadias), the chordee is corrected, and the hooded foreskin is reconstructed [9].

In our study, these cases occurred among patients whose primary diagnosis was distal penile hypospadias. Among 556 cases with distal penile hypospadias, 17 cases had a hooded prepuce with chordee, thin transparent urethra, and normally situated meatus, accounting for 3% of all hypospadias cases.

While releasing the chordee and restoring the normal appearance of the penis, unintended urethral injury is the primary challenge. This issue was addressed by creating a lateral incision on the ventral penile skin and progressing gradually toward the midline. The skin adhering to the urethra at the midline was dissected. This approach effectively minimised urethral injuries.

To avoid injury, Yang et al. used hydro dissection to separate the ventral penile skin from the hypoplastic urethra. They injected a normal saline solution containing adrenaline (1/100000) into the sub coronal plane before beginning dissection. Although hydro dissection facilitated gentle and easy degloving, a urethral injury was reported in 3 of the 18 cases. The authors diagnosed these cases as hypospadias [11].

This study had 23 patients with iatrogenic urethral injury. The injuries were repaired using Vicryl 6/0. The repair was reinforced using the dartos pedicle flap of the dorsal prepuce before its resection. However, two patients developed a urethra-cutaneous fistula by the end of the first postoperative month. The patients were scheduled and managed six months after the first surgery.

Dipaola et al. treated patients with chordee and hypoplastic urethra with a hooded prepuce using urethroplasty. Mathieu's procedure was performed for patients with a distal hypoplastic urethra; Duckett's procedure was performed for patients with a long hypoplastic segment. A complication rate of > 50% was reported [12].

If this anomaly was left untreated, there will be a persistent chordee. This may troublesome during intercourse. Due to the deficiency of the ventral urethral support, this thin urethra is theoretically liable for the development of urethral diverticulum with associated recurrent urinary tract infection, and weak ejaculatory force. Parents might complain of a penile curvature at erection or a penile angle greater than 30° on examination under natural erection. Some authors proposed that the correction age of chordee should be after puberty [13].

Ali et al. noticed the dilatation of urethra in abducents and young adults who underwent distal hypospadias repair during childhood in the absence of a covering spongiosum. With the reconstruction of spongiosum tissue they could manage this problem [14].

This study was limited by a small sample size and its non-multicentricity. The inclusion of type 2 and type 3 might also be a cause of the study's limitations.

Conclusion

A hooded ventral prepuce with ventral chordee and normally situated meatus presents a challenge for paediatric surgeons. It is usually associated with a hypoplastic urethra. Attempts should be made to correct the chordee and preserve the urethra. The preserved urethra can be reinforced with spongioplasty. Unintended urethral injuries are common during dissection. Therefore, repairing the injury, reinforcing the dartos pedicle flap, and spongioplasty are suitable options.

It is possible to preserve the urethra in patients having chordee without hypospadias. Utmost care should be taken to avoid urethral injuries which are common because shaft skin is stuck to the urethral skin. Urethral injuries can be repaired primarily; however, there is a high incidence of fistula.

Author contributions

MK, designed the study. he wrote the original manuscript. he was the operator and data collector.

Funding

None.

Data availability

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

An ethical approval was obtained under code: HA-02-J-008 with a reference No. 148 – 24 by the unit of biomedical ethical, research ethical committee, King Abdulaziz University, Faculty of medicine.
An informed consent was signed by the parents or care givers of all participants in the current study.

Consent for publication

Author agreed for publication.

Competing interests
The authors declare no competing interests.

Received: 10 August 2024 / Accepted: 14 January 2025
Published online: 03 February 2025

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