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CASE REPORT

A case of successful repair of urethrocutaneous fistula post proximal hypospadias repair managed by Johanson's 2 stage urethroplasty

Ojas Vijayanand Potdar[1,*], Prashant Sarawade[1], Mohammed Ayub Karamnabi Siddiqui,[2] Kaustubh Vaidya,[3] Vivek Shaw[3] and Jay Bhanushali[1]

[1]*Senior Registrar in Urology, Grant Government Medical College and J.J. Group of Hospitals, Mumbai Central, Mumbai, Maharashtra-400008*

[2]*Head of Department of Urology, Grant Government Medical College and J.J. Group of Hospitals, Mumbai Central, Mumbai, Maharashtra-400008*

[3]*Assistant Professor in Urology, Grant Government Medical College and J.J. Group of Hospitals, Mumbai Central, Mumbai, Maharashtra-400008*

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Abstract

Urethrocutaneous fistula (UCF) after hypospadias repair remain a frustrating Problem for all the practicing urologists. Furthermore, with the improvement in suture materials and surgical techniques, complications if they may arise also present with litigations for the urologist. We present a case of a 18 year old boy with 46 XY mixed gonadal dysgenesis who had undergone proximal hypospadias repair with Duckett's tube urethroplasty with resultant complication of subcoronal urethrocutaneous fistula which failed first attempt at repair and resulted in two urethrocutaneous fistulas which was successfully managed by two stage Johanson's urethroplasty.

Keywords: Urethrocutaneous fistula, Johanson's urethroplasty, hypospadias, DSD

*Corresponding author: Ojas Vijayanand Potdar
Email: ojaspotdar@yahoo.com

Introduction

Urethrocutaneous fistula (UCF) after hypospadias repair remain a frustrating Problem for all the practicing urologists. Furthermore, with the improvement in suture materials and surgical techniques, complications if they may arise also present with litigations for the urologist. The occurrence of UCF precludes a goal of hypospadias surgery, i.e., an early one-stage repair of the defect. During the last decade, experts have proposed many principles of an ideal repairing technique. Delicate tissue handling, inversion of the urethral mucosa after excising the epithelialized tract of the fistula, a multilayer repair with well-vascularized tissues, avoiding overlapping sutures and nonabsorbable or thick suture materials, a tension-free closure, use of optical magnification and needle-point cautery for coagulation are currently considered mandatory [1]. However, unfortunately there is no single technique that can guarantee success in all cases. Also, UCF tend to recur again and multiple procedures again and again in the same patient cause all the potential harmful physical and psychological consequences [2]. We present a case of a 18 year old boy with 46 XY mixed gonadal dysgenesis who had undergone proximal hypospadias repair with Duckett's tube urethroplasty with resultant complication of subcoronal urethrocutaneous fistula which failed first attempt at repair and resulted in two urethrocutaneous fistulas which was successfully managed by two stage Johanson's urethroplasty.

Case report

A 18-year-old boy presented to the urology outpatient department of a tertiary care centre with complaints of three streams while passing urine. On enquiring about the past history, it was discovered that the patient had a proximal hypospadias with presence of uterus and fallopian tubes and left side streak ovary and right-side scrotal streak testis and on karyotype analysis, he was discovered to have 46 XY karyotype and hence, was diagnosed to have mixed gonadal dysgenesis. His previous medical records reveal that he underwent genitoscopy and Laparoscopic bilateral Hystero-salpingo-oophorectomy with excision of streak gonad on the right side with Duckett's tube urethroplasty. As per his previous clinical notes, at the time of infancy when he was examined, he had a proximal hypospadias with the meatal opening at the penoscrotal junction with the urethral plate well developed and evidence of ventral chordee and dorsal prepucial skin hood with the length of the phallus being 2 cm. The left side testis was not palpable and on the right side there was a streak gonad. On further evaluation by imaging, it was found that the child had a uterus and fallopian tubes and a left side rudimentary ovary. At the age of 2 years, child underwent Genitoscopy and Laparoscopic Hystero-salpingectomy with excision of left sided streak ovary and Duckett's tube urethroplasty for the management of proximal peno-scrotal hypospadias. Then the child developed a subcoronal urethrocutaneous fistula which was repaired by excision of the fistulous tract and flap closure from the surrounding area at the age of 3 years. However, the surgery failed and the patient developed two fistulous openings after which the patient was lost to follow up. Finally at the age of 16 years, patient presented to the urology department with the complaints of three streams while passing urine.

On local examination, penile shaft was of adequate length. There was evidence of puckered scarred skin over the ventral aspect of penile shaft with multiple buttonholes appreciated over it (Figure 1).

Figure 1: Clinical picture showing the ventral aspect of the penile shaft with multiple openings, puckered skin, empty left scrotum and right sided streak gonad.

The patient was asked to void and the three streams were documented (Figure 2).

Figure 2. Clinical picture of the patient showing three urinary streams while voiding.

The patient then underwent retrograde urethrogram to visualise the urethrocutaneous fistulous tracts (Figure 3).

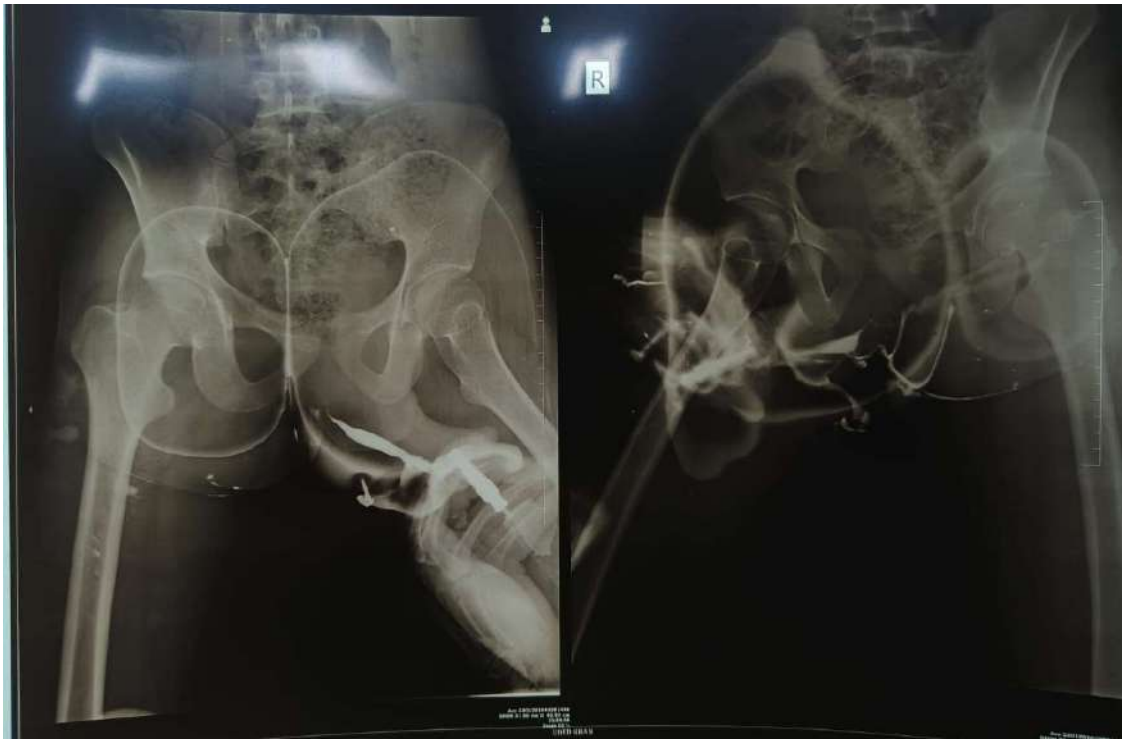

Figure 3. Retrograde urethrogram showing two urethrocutaneous fistulas along the proximal shaft of penis.

In view of the previous history of failed fistula repair and the scarred puckered skin over the ventral aspect of penile shaft, decision was made to go ahead with two stage Johanson's staged urethroplasty. A cystourethroscopy done using 4.5 F ureteroscope revealed a normal posterior urethra and bladder neck and evidence of prominent prostatic utricle. In the first stage, the urethra was laid open ventrally till the level of most proximal urethrocutaneous fistula opening and a buccal mucosal graft was harvested and anchored to the laid open urethra with quilting done to the underlying corpora cavernosa using PDS 4-0 sutures (Figures 4 and 5).

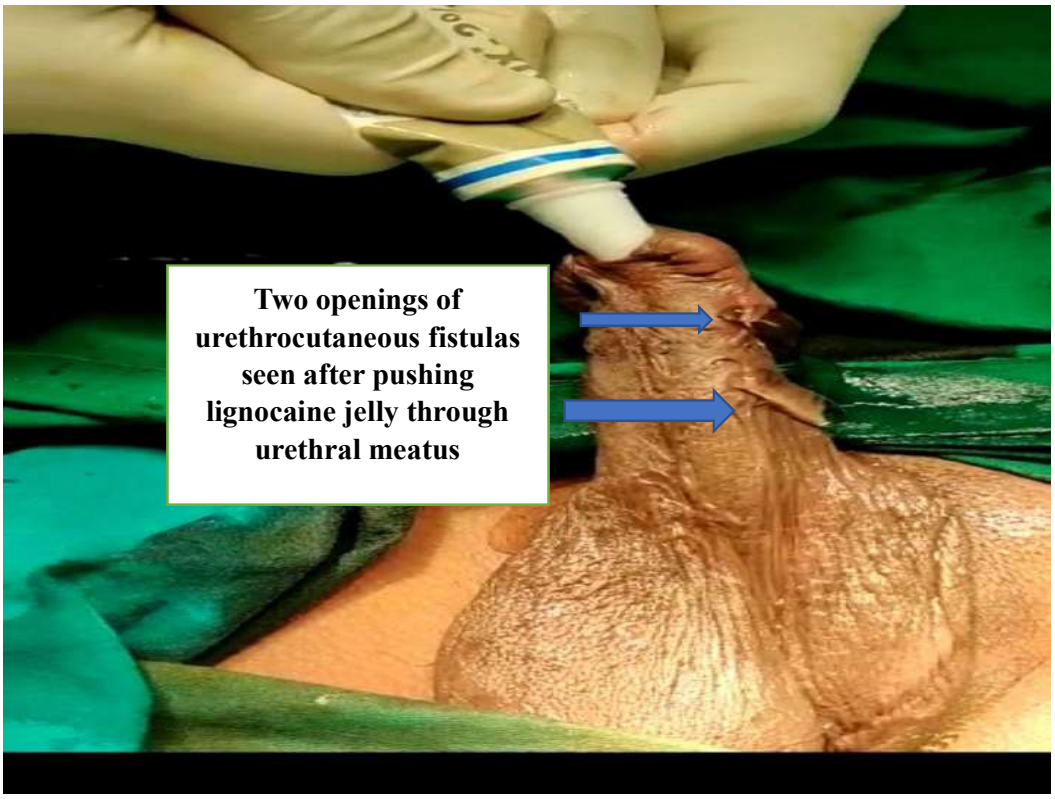

Figure 4. Clinical picture showing two openings of urethrocutaneous fistula seen after pushing lignocaine jelly through urethral meatus.

Figure 5. Clinical picture of patient after first stage of Johanson's first stage urethroplasty showing the buccal mucosal graft anchored to the graft bed.

Patient then underwent second stage of Johanson's urethroplasty where the buccal mucosal graft was tubularised and a second layer of tunica vaginalis flap from the scrotum was used to cover it. (Figure 6).

Figure 6. Clinical picture showing second stage of Johanson urethroplasty with additional coverage from tunica vaginalis from right scrotum.

After 4 weeks, the catheter was removed and the patient voided with a single stream. A follow up retrograde urethrogram was done which revealed a normal well healed urethra (Figure 7).

Figure 7. Follow up retrograde urethrogram revealed a normal well healed urethra.

Discussion

There are no perfect techniques for repairing UCF. Many variables could influence the surgical management and outcome, i.e. the time of occurrence after urethroplasty, the location (glanular, coronal, mid-shaft, etc.), size (pinpoint,large), the number and the conditions of local tissue [3]. However, Waterman et al. reported no significant difference in outcomes comparing some variables, e.g. the use or not of a stent or catheter, optical magnification, patient age and interval between surgery at time of fistula repair, type of original hypospadias procedure, and number of previous fistula repairs [4]. As no one technique is effective some failure rate is expected in every series [5-8]. In our case report, in view of the puckered skin and previous failed repair, decision was made to do a 2 staged repair and resulted in a successful outcome.

Conclusion

Thus, urethrocutaneous fistula is a challenging and frustrating complication to manage for a practicing urologist. The management of each case needs to be individualised. We put forth Johanson's two stage urethroplasty with use of buccal mucosal graft as an effective technique for the management of the same.

Ethics declarations

Funding This study did not receive any funding.

Conflict of interest

The authors declare that they have no competing interests.

Ethics approval, Consent to participate, Consent to publish, Availability of data and material, Code availability

Not applicable.

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