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Case Report

Congenital urethrocutaneous fistula in an adolescent male

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ABSTRACT

A urethrocutaneous fistula is a common complication after hypospadias repair, but congenital fistula is a rare anomaly. We present a 16-year-old boy with this unusual anomaly. Its etiology, embryology, and management are discussed in brief.

KEY WORDS

Adolescent; congenital fistula of penile urethra; urethroplasty

INTRODUCTION

Acquired urethrocutaneous fistula is one of the most common complications of hypospadias surgery, but congenital anterior penile urethral fistula is a rare condition.[1] It is commonly associated with anorectal malformation and several penile pathologies such as hypospadias and/or chordee, but isolated cases also have been reported.[1-4] We report a rare case of a 16 year old boy with isolated wide congenital anterior urethrocutaneous fistula with its treatment and review of the literature.

CASE REPORT

A 16-year-old uncircumcised boy was admitted with chief complaints of passing a single stream of urine from the undersurface of the penis, forcing him to micturate in squatting position. There were no complaints of a narrow stream of urine or pain while passing urine. He was the only child born out of non-consanguinous marriage. The antenatal period was uneventful.

On examination, a wide congenital urethral fistula was noted at the scrotal level [Figure 1]. The urethral plate measured 4.5 cm in length and 2.6 cm in width. The urethra distal to the fistula was normal with normal prepuce and glans without any chordee [Figure 2]. The external urethral meatus was situated normally at the tip. The stretched penile length and diameter were normal for his age. No previous penile surgery had been done. Urinalysis was found normal.

The patient was posted under general anesthesia. The anterior urethrocutaneous fistula was identified at the level of scrotum [Figure 3]. It was calibrated

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with 12 number foley's catheter passing down to the urinary bladder without any obstruction. The distal urethra was calibrated with the gradually increasing size of urethral dilator [Figure 4]. Meatotomy was done at the lower end of the distal urethra. The exposed urethral plate was marked at its periphery with ink [Figure 5]. The fistula was circumscribed and repaired in 2 layers over 12 number foley's catheter. Dartos fascia and skin were closed over it [Figure 6]. The post-operative course of the patient was uneventful. The catheter was removed on 14th post-operative day [Figure 7]. No recurrent fistula occurred. After 4 months of follow-up, patient is voiding normally without any recurrence.

Figure 1: Pre-operative photo with fistula visible at scrotal level

Figure 2: 16-year-old uncircumcised boy with congenital urethral fistula

Figure 3: Intraoperative photo with fistula identified at scrotal level

Figure 4: Distal urethra calibrated with urethral dilator

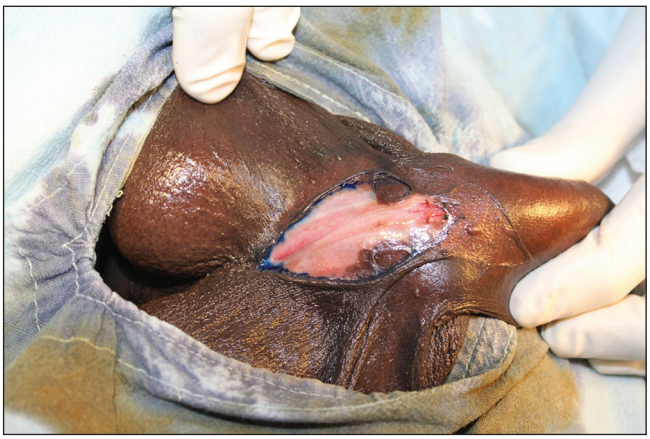

Figure 5: The exposed urethral plate marked at its periphery with ink

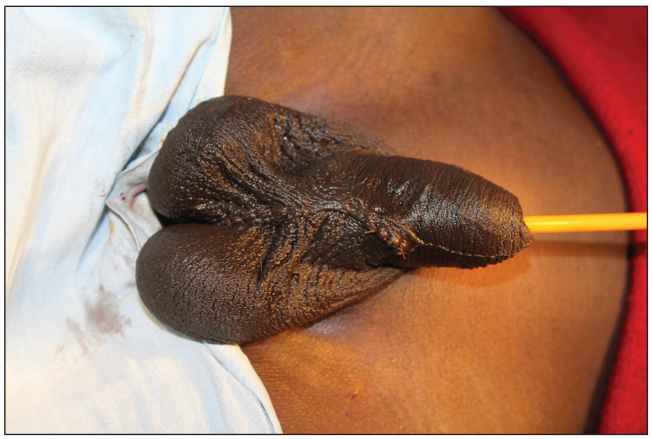

Figure 6: Intraoperative photo after skin suturing

Figure 7: Post-operative photo on day 14 after catheter removal

DISCUSSION

Congenital fistula of the penile urethra (CFPU) is a rare clinical entity characterised by a fistulous connection between urethra and skin.[1] They are often associated with anorectal malformations and are usually of posterior type. A congenital anterior urethrocutaneous fistula is most of the times an isolated deformity, but may be associated with ventral chordee or imperforate anus.[1-4] Revising embryology of genitourinary system, the external genital structures are identical in males and females until 8 weeks of gestation. The genitalia develop a masculine phenotype in males primarily under the influence of testosterone. As the phallus grows the open urethral groove extends from its base to the level of the corona. The classic theory is that the urethral folds coalesce in the midline from base to tip, forming a tubularised penile urethra and median scrotal raphe. This accounts for the posterior and middle urethra. Development of anterior urethra begins at 10 mm (4 weeks) stage when the urethral plate is recognised as thickening of the anterior wall of the endodermal cloaca. Subsequently primary and secondary urethral groove are established and at the 11th week, when Leydig cells increase in number and size, urethral folds begin to fuse ventrally in the midline to form urethra. Failure of fusion gives rise to this condition.[5]

CFPU is an extremely rare condition and the etiology is controversial. Although the embryologic mechanism of the fistula still remains obscure, several theories have been proposed. These include rupture of small diverticulum[6] or a focal defect in urethral plate[4] or it may be a variant of non-glanular hypospadias without glanular or preputial defect.[7,8] It may occur due to pressure necrosis from the heel of baby's foot[9] or by external compression.[10]

A number of techniques are described for CFPU repair like pedicle flap,[11] modified Denis-Browne urethroplasty[4] and proximal based skin flap.[12] The type of repair of CFPU is governed by the local findings like status of corpus spongiosum, skin and distal urethra. In our case the distal skin was normal and distal urethra was patent hence we repaired fistula by circumscribing it and closing it in multiple layers. In cases of deficient distal urethra or chordee, formal hypospadias repair is recommended.[4]

The primary complication of the repair is recurrent fistula formation.[4,13] The recurrent fistula may heal spontaneously, or a subsequent operation to close the fistula may be required.[4,13] In the present case, we did not encounter recurrence. It gave good cosmetic and functional results.

CONCLUSIONS

Congenital urethral fistula, though a rare entity requires meticulous examination to rule out other associated anomalies. The type of repair is governed by the status of skin, distal urethra and underlying structures.

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Conflicts of interest
There are no conflicts of interest.

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