A new indication for tubularized incised plate urethroplasty: Isolated congenital penile urethrocutaneous fistula

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ABSTRACT

Isolated congenital penile urethrocutaneous fistula is described as an unusual developmental anomaly in two infants who presented with an abnormal opening on the ventral aspect of penis with a normal foreskin and an absence of the chordee, hypospadias or other associated congenital anomalies. Tubularized incised plate urethroplasty augmented with dartos pedicle blanket wrap resulted in satisfactory recovery. Authors review their surgical experience with this new indication of tubularized incised plate urethroplasty in successful surgical repair of congenital penile urethrocutaneous fistula along with pertinent literature.

KEY WORDS: Urethrocutaneous abnormalities, urethrocutaneous fistula

INTRODUCTION

Isolated congenital penile urethrocutaneous fistula is an extremely uncommon developmental anomaly.[1,2] To the best of our knowledge, only 16 such cases have been discussed in English literature till date.[3-10] Moreover, a high recurrence rate, reported with its surgical repair, demands its recognition as a separate clinical entity. Although different surgical techniques have been proposed in repair of congenital penile urethrocutaneous fistula, yet the surgical experience with tubularized incised plate urethroplasty in its repair has rarely been reported till date. So among handful of reported cases, the present cases appears unique as dartos pedicle augmented tubularized incised plate urethroplasty was performed with the aim to reconstruct a near-normal caliber neourethra with minimal scarring of the penile skin. To the best of our knowledge, it appears to be the first surgical experience to be reported in the Indian literature. Thus, rarity justifies reporting of present cases with the aim to highlight the feasibility of tubularized incised plate urethroplasty in successful surgical repair of isolated congenital penile urethrocutaneous fistula so that an undue morbidity associated with not infrequent recurrence would be avoided.

CASE REPORTS

Case 1
An 8-month-old male child presented with an abnormal opening on the undersurface of penis, which had been present since birth. The patient was passing urine from both the terminal glandular meatus and the fistulous opening. There was no history of any trauma, stone impaction or surgery. On examination, an ovoid shape urethrocutaneous fistula measuring 2 cm by 1 cm was present on the ventral aspect of penile urethra approximately 2 cm distal to penoscrotal junction. The external urethral meatus appeared normal with normal foreskin and without any chordee or hypospadias. Ultrasonography and cystourethrography were normal. The patient was taken up for surgical repair, and tubularized incised plate urethroplasty was planned with the aim to reconstruct a near-normal caliber neourethra. After applying stay sutures, proposed circumferential and racket-shaped incisions incorporating the fistulous opening were marked [Figure 1a]. The penile shaft skin was degloved, fistulous margins were mobilized and urethral plate ‘relaxing incision’ was made in through fistulous opening [Figure 1b]. The margins of fistulous opening were marked [Figure 1c]. The neourethral suture line was reinforced with dartos pedicle flap as a blanket wrap [Figure 1d]. The penile skin was rotated ventrally to provide skin coverage. The post-op period remained uneventful, and presently patient is doing well for the last 9 months.

Case 2
A 10-month-old male infant presented with an abnormal
opening on the ventral aspect of penile urethra, which had been present since birth. The patient used to pass most of the urine from the fistulous opening and only a small amount through the normal external urinary meatus. On examination, an ovoid shape fistula measuring 1.5 cm by 1 cm was present in the mid-penile region. The external urethral meatus was normal, with an absence of chordee or hypospadias. Ultrasonography and cystourethrography along with other laboratory investigations revealed no abnormality. Surgical repair was performed by tubularized incised plate urethroplasty augmented with dartos pedicle blanket wrap. Postoperative period remained uneventful, and the patient recovered satisfactorily. Patient is on regular follow-up for the last 7 months and is doing well.

DISCUSSION

Congenital penile urethrocutaneous fistula is an uncommon clinical entity characterized by the presence of a fistulous communication between penile urethra and skin in the absence of associated congenital anomalies. It is considered as rarest among a spectrum of urethrocutaneous fistulae that run between penile, scrotal or posterior urethra to the skin and are commonly associated with congenital anomalies like anorectal malformations and urethrocystic duplication. The etiopathogenesis of congenital penile urethrocutaneous fistula still remains unclear. Although its etiopathogenesis has been explained by the theory of pressure necrosis of penile urethra by fetal parts, yet an absence of scarring in the present and a few reported cases supports the theory of focal developmental defect of urethral plate, which prevents the fusion of urethral folds, thus resulting in isolated congenital penile urethrocutaneous fistula. The management of isolated congenital penile urethrocutaneous fistula is primarily surgical, aimed at reconstruction of near-normal caliber neourethra with minimal recurrence rate. Although a number of different surgical techniques, including pedicle, prepuce based skin flap, are available, direct fistula closure or modified Denis Browne urethroplasty have been proposed in the repair of congenital penile urethrocutaneous fistula; however, recurrence remains a potential complication with a reported incidence of 30%. Although satisfactory results have been reported in a few cases undergoing primary fistula closure, yet the resulting scarring and a lack of penile skin make subsequent repair difficult if recurrence occurs. With the aim to reconstruct a near-normal caliber neourethra with minimal recurrence rate, we experienced satisfactory results with tubularized incised plate urethroplasty. As experienced in the present cases, the urethral plate ‘relaxing incision’ given through fistulous opening while performing surgical repair by tubularized incised plate urethroplasty facilitated tension-free tubularization of neourethra. Moreover, augmentation of neourethra with dartos pedicle blanket wrap further minimized the risk of recurrence by acting as a barrier between two opposing suture lines and also provided an additional support to neourethra. Although recurrence was not experienced in the present cases, yet our experience on long-term follow-up suggested that the ventral rotation of dorsal penile skin provided adequate skin coverage with minimal scarring, thus making subsequent surgical repair easy in cases with recurrence, and this appears to be an additional advantage of this surgical technique.

Thus we conclude that apart from hypospadias, isolated congenital penile urethrocutaneous fistula appears to be one of the indications for performing tubularized incised plate urethroplasty, which not only results in successful surgical repair of this rare anomaly but is also associated with minimal complications and recurrence rate.

REFERENCES


