ABSTRACT

Traumatic or iatrogenic urethro-cutaneous fistulas are uncommon and leads to gross morbidity to the patients. Improper treatment shows poor outcome, so these fistulas needs to be managed by urologist having vast experience in lower urinary tract reconstruction. We are presenting a case of urethro-cutaneous fistula due to an unusual etiology and initially treated by a non-urologist.

KEYWORDS: Penile Trauma, Iatrogenic Injury, Urethro-cutaneous Fistula, Management

INTRODUCTION

Genitourinary injuries constitute less than 10% of trauma and among them penile injuries are relatively uncommon which represents only about 1% to 1.6% of all genito-urinary injuries. Urethro-cutaneous fistula is one of the complication of severe penile trauma.1 Penile injuries complicating as urethro-cutaneous fistula may be due to blunt, penetrating or iatrogenic trauma.2 The various structures of the penile anatomy involved to a variable extent either singly or in combination in trauma. Injuries may include penile fracture penile amputation, contusion, laceration or degloving of skin. Unidentified urethral contusion or laceration at the time of injury may result in a urethrocutaneous fistula.1 The exact incidence of urethrococutaneous fistula complicating penile trauma in not known. Most cases reported in literature were due to iatrogenic trauma following hypospadias or penile surgery and among those cases the urethral defects are usually small.1,3 Congenital urethral fistula is a very rare anomaly as reported in literature. Among these meatus and urethra are normal and a urethrococutaneous fistula is typically present subcoronal or coronal.1 This is usually an isolated deformity, but may be associated with ventral chordee, hypospadias or imperforate anus. A few congenital urethral fistula has been reported and literature search shows only two cases of spontaneous ventral urethral fistula. Some unusual causes of spontaneous urethrococutaneous fistula were also reported. Wei et al reported a case of spontaneous ventral urethral fistula in new born due to retained IUD in uterus during the gestation.5 Danzinger reported spontaneous ventral urethral fistula accompanied by severe infection due to diabetes mellitus.6 A few cases of fistula as a complication of intra-cavernous injection (especially Papaverine) reported which occur after abscess formation.7 Similarly, Kumar et al has reported a unusual case of urinary retention with urethral fistula proximal to a urethral stricture and history of passage of urine and stones through a fistula at the root of the penis.8 Sancaktutar and colleagues reported urethra cutaneous fistula as a complication of circumcision.9 We are presenting a child with urethra cutaneous fistula due to unusual etiology.

CASE HISTORY

This 7 years old boy presented with painful swelling of shaft of penis and dribbling of urine from and abnormal opening over ventral aspect of shaft of penis since last seven months. He has recurrent episodes of urinary retention also. History revealed that his mother use to tie a piece of thread over shaft of his penis in order to control his bed wetting since he was four years old (Fig - 1). He also have recurrent attacks of urinary tract infection and hypogastric pain in past. He has no congenital abnormality of external genitalia as reported by his mother. Clinical examination shows intact thread tied over the penis producing a constricting deformity all around and a tiny urethra cutaneous fistula just proximal to the constriction. External urethral meatus was situated at normal position and of normal size. X-ray KUB (Fig - 2) and US scan shows multiple vesical calculi and urinary tract infection on urine examination. Thread was released, suprapubic urinary diversion and vesicolithotomy was done (Fig - 3). Subsequently, after six weeks urethroscopy was done to assess the urethra and easy passage of 16F Foley’s catheter passed through external urethral meatus into the bladder and open. Repair of fistula was planned subsequently.
DISCUSSION

Our index patient presented with a fully developed urethro-cutaneous fistula. He had no urinary diversion and no attempt was made to determine the extent of urethral involvement by the referring physician. Rather an attempt at primary repair of a severe penile crush injury was made by some doctor. This scenario of delayed referral and attempt to manage purely urological problems by non-urologists is not uncommon in a resource poor environment such as ours. At the time of presentation the urethral fistula was already well defined and we did not consider an urethrogram necessary at this stage. We did suprapubic urinary diversion to enable patient to void freely and also to allow for resolution of inflammatory reaction at the fistula site. At the time of definitive repair, urethroscopy was done to assess the urethra which shows easy passage of a size 16F Foley catheter through the external urethral meatus into the bladder.

Urethral fistula is the most common complication of urethroplasty. For urethral fistulas, no classification has been found in literature. Horton has classified these fistulas in acute (early) and mature fistulas. Penile injuries rarely leads to urethro-cutaneous fistula and as compared to erect penis, the flaccid penis is less prone to injury. Penis may be injured from penetrating trauma like stab injuries, gunshot wound, animal or human bites. Iatrogenic trauma commonly comprises of penile instrumentation, surgery or urethral catheterization. Blunt external trauma to penis include coital injuries, road accident, machine injuries and forcefully bending of the erect penis. Thermal injuries to penis have also been described but rarely leads to urethro-cutaneous fistula.

In literature, the incidence of urethro-cutaneous fistula due to complication of penile trauma is not given. Majority of reported cases are iatrogenic and as a results of hypospadias surgery. Urethro-cutaneous fistula is also reported after severe crush or penetrating injury due to the sloughing of dead and devitalized tissue. With respect to the mechanism of injury, the lax and loose genital skin has a protective role, by allowing the skin to slide away and deform from the potential point of contact during trauma.

Initial management of penile crush injuries at the time of presentation involves urinary diversion by suprapubic cystostomy followed by careful debridement of dead and devitalized tissue. To assess the extent of urethral involvement, preoperative or inta-operative urethrogram is useful. Early, acute fistulas are managed conservatively. In mature single fistulas, the surrounding tissue may be used for repair. These mature fistulas are subdivided depending upon the size of the fistula opening. Chronic, multiple or large lesions draining the urethra require tissue from a distance for repair. In severe surgical cripples, a total new urethral reconstruction will be required. Repair is individualized depending upon the site and size of fistula and may involves the techniques used for hypospadias repair or may involve simple multilayer fistula closure or more complex reconstruction of the distal urethra and glans.

Different surgical techniques are proposed to repair these fistulas which include primary closure via Thiersch-Dupley urethroplasty, turned down flap urethroplasty and pedicled island tube, or on lay urethroplasty. Repair with local flaps is preferred in case of an isolated urethral fistula with intact corpus spongiosum. However, if distal urethra from fistula is deficient or deficient spongiosum or there is associated hypospadias or chordee, then the formal hypospadias repair is recommended welch.

The major challenge for the reconstructive urologist is closing of a large urethral defect. This can be achieved by mobilizing a fascio-cutaneous flap from distal island of penis and it is used as an on lay ventral flap to close the defect over a stenting Foley catheter. The skin cover is achieved by mobilizing the rest of the penile skin to cover the penis. Quartey and McAninch describes the use of penile fascio-cutaneous flaps for urethral reconstruction which is based on the axial superficial blood supply in the penile dartsos fascia. These flaps are versatile and can be used as dorsal or ventral on lay to cover any size of the anterior urethral defects. These flaps can also be tabularized and sufficient length can be mobilized to replace the entire urethra with good functional and cosmetic results. To avoid diverticula formation the proper sizing of the flap is very important and width of the flap is meticulously determined by measuring the length of the urethral defect. Similarly, the height of the flap is determined by subtracting the width of the residual urethra from the expected circumference of a 22F urethra (approximately 23 mm). The complications may be expected after fistula repair include penile skin dehiscence, alterations in penile skin sensation, recurrence, stricture formation, diverticula formation associated with post micturition dribbling. The most
worrisome complications are recurrent fistula and stricture formation. By meticulous suturing and avoidance of overlapping suture lines can leads to recurrent fistula formation. Despite this if short stricture occur, then it can be managed adequately by direct vision internal urethrotomy (DVIU) or urethral dilatation. The longer strictures (approximately 1.0–2.5 cm) should be treated by urethroplasty. For urethral replacement, on lay repairs is more successful as compared to flap tabularization. Carney and McAninch has reported an initial success rate of 87% with on lay repairs and long term success rate of 98% after one additional procedure.

**CONCLUSION**

This unusual case speaks to the importance of a thoughtful surgical management with the urologist having experience in lower urinary tract reconstructive repair surgery. Awareness of public is also essential to avoid such incidence.

**Contribution of authors:**
Muhammad Salman: History collected data regarding patients.
Muhammad Akram: Operating Surgery, Patients follow up operating, Data and material
Ishtiaq Ahmed: Write Discussion and Final Formatting proof reading

**REFERENCES**