CASE REPORT

Urethrocutaneous fistula mimicking vesicocutaneous fistula: a rare entity

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SUMMARY

Urethral fistulas arising from the anterior urethra usually open on the ventral aspect and are located in the penile, scrotal or perineal region. A diabetic 65-year-old man presented with suprapubic urinary fistula that was communicating with a dorsally located internal urethral opening located in the bulbar region. Such unusual fistula has not been described before.

BACKGROUND

Urethral fistula is an uncommon condition and the external opening is usually located ventrally in the penile, scrotal or perineal region while its origin is usually from the penile or bulbous urethra.^{1 2} An unusual external location may delay the diagnosis and make the management complicated. We report a case of urine leakage from an unusual site, located on the suprapubic region, due to a fistulous communication from the dorsolateral part of the pendulous urethra in an adult man. To the best of our knowledge, this distinct location has not been reported.

CASE PRESENTATION

A 65-year-old diabetic man, who was also a chronic tobacco chewer, had thinning of urinary stream for last 2 years. There was no history of urethral trauma or urethral instrumentation. He developed severe dysuria along with development of a suprapubic tender lump about 15 days ago. Along with a failed attempt of urethral catheterisation, the patient underwent an emergent incision and drainage of suprapubically located abscess that drained about 400 mL pus at another primary care hospital. The patient started leaking urine from the suprapubic wound with cessation of urethral micturition. Examination revealed a normal urethral meatus while there was a 4×2 cm surgically created fistula at the suprapubic site (figure 1).

INVESTIGATIONS

Ultrasonography of abdomen revealed thickened bladder walls with normal upper tracts. Retrograde urethrogram revealed a stricture involving pendular and distal bulb (panurethral stricture) with contrast extravasation from bulbar urethra reaching up to the pelvic brim with leakage of contrast through the suprapubic site (figure 2). Micturating cystourethrogram was attempted, but the bladder could not be filled.

DIFFERENTIAL DIAGNOSIS

A diagnosis of stricture urethra along with urethrocutaneous fistula was made.

TREATMENT

Intraoperative cystoscopy with paediatric cystoscope revealed two fistulous openings located at 10 o'clock position dorsolaterally at the penoscrotal junction (figure 3A). A guide wire passed antegradely through the suprapubic fistula was seen cystoscopically through the internal dorsolateral fistula confirming the diagnosis of urethrocutaneous fistula (figure 3B). In view of panurethral stricture with urethrocutaneous fistula, the patient was subjected to urethral marsupialisation (Johanson stage 1) with the creation of perineal urethrostomy (figure 4).

OUTCOME AND FOLLOW-UP

Postoperative period was uneventful. After 3 weeks, the patient was voiding comfortably through perineal urethrostomy while the suprapubic fistulous opening had become dry and had shrunk considerably. Second-stage urethral repair is planned after 3 months.

DISCUSSION

Acquired urethral fistulas generally result from trauma, infection, urethral stricture, surgical complications, radiation therapy and neoplasm.³ Rare causes such as forgotten and amputated urethral Foley catheter have also been reported.⁴

To localise the fistulous tract, and to identify the causative factor investigations such as retrograde urethrography, micturating cystourethrography, cystoscopy, fistulography and MRI are needed. In our case, fistula site was diagnosed by retrograde urethrography and confirmed with cystoscopy.

It was challenging to define the cause of fistula in our patient and it resulted due to a stricture of anterior urethra. The patient had probably developed periurethral abscess due to infection in the periurethral glands which progressed in the face of uncontrolled diabetes through the retropubic space and extended into the suprapubic space. Infective fistulas usually open on the ventral position, which is explained on the basis of gravity-dependent drainage. Urethral glands, namely the bulbourethral glands (Cowper glands), empty into the bulbar region at the three o'clock and nine o'clock positions while the periurethral glands of Littre drain into the urethra dorsally.⁵

Rarely, some unusual external openings from urethral fistula have been described. In one report,

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Figure 1 Urinary fistula located at the suprapubic site.

Figure 2 Retrograde urethrogram demonstrating panurethral stricture (multiple white arrows) located in penile and bulbous urethra with extravasation of contrast at bulbar urethra (coloured arrow) reaching up to the pelvic brim with leakage of contrast through the suprapubic site.

Figure 3 (A) Cystourethroscopy showing guide wire in the urethra with additional two fistulous openings located dorsolaterally at 10 o'clock (arrows) in the penoscrotal junction. (B) Cystourethroscopy showing the guide wire (arrow) visible through the internal fistulous opening that was passed through the suprapubic fistula in an antegrade fashion.

a young insulin-dependent diabetic patient developed a dorsal urethral fistula after debridement of necrotising fasciitis of the penis.⁶ Patodia *et al*⁷ described another case where stricture along with long-term catheterisation caused a prostatic abscess that found its way through the retroperic fat into the right inguinal region to result in a prostatocutaneous fistula.

The usual initial treatment for infective urethrocuteaneous fistulas includes diversion with a suprapubic catheter. Definitive treatment in the form of flap/graft urethroplasty along with excision of the fistula is done at a later date after a thorough evaluation of urethra. In our case, urinary diversion through a suprapubic cystostomy was not feasible due to the presence of bowel loops (risk of bowel injury) and a large wound in that area. The patient was subjected to urethral marsupialisation with perineal urethrostomy due to the presence of concomitant panurethral stricture. Second-stage urethral closure is planned at a later date.

Figure 4 (A) Intraoperative view showing the ureteric catheter passed from urethral fistulous opening (long black arrow) to the suprapubic opening (short black arrow) along with a guide wire in the urethra (coloured arrow). (B) Intraoperative view: perineal urethrostomy proximal to the fistula (white arrow) with laid-open distal urethra.

Learning points

- ▶ In a man presenting with suprapubic swelling/abscess with lower urinary tract symptoms possibility of a urethral fistula should be considered.
- ▶ Imaging techniques such as retrograde urethrography and urethrocystoscopy are helpful in making a diagnosis of urethrocuteaneous fistula.
- ▶ Treatment of urethral fistulas should be individualised according to patient's age, co-morbid factors and local tissue factors.

To the best of our knowledge, this is the first of its kind of spontaneous dorsolateral fistula of pendulous urethra passing subcutaneously and opening into the suprapubic site.

Contributors RA and AG: concept, design, supervision, processing, writing manuscript and critical analysis. AKS: supervision, processing, writing manuscript and critical analysis. SG: concept, supervision, writing manuscript and critical analysis.

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