ORIGINAL RESEARCH PAPER
Volume - 14 | Issue - 10 | October - 2025 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

ISOLATED CONGENITAL ANTERIOR URETHROCUTANEOUS FISTULA: A CASE REPORT AND REVIEW OF LITERATURE

Paediatric Surgery

Dr. Ameya S. Muzumdar	Department of Paediatric Surgery, T.N.M.C & B.Y.L. Nair, Mumbai.
Dr. Suraj R. Gandhi	Department of Paediatric Surgery, T.N.M.C & B.Y.L. Nair, Mumbai.
Dr. Neha S. Shenoy	Department of Paediatric Surgery, T.N.M.C & B.Y.L. Nair, Mumbai.
Dr. Kshitij A. Mane	Department of Paediatric Surgery, T.N.M.C & B.Y.L. Nair, Mumbai.
Dr. Wafa I. Khatri	Department of Paediatric Surgery, T.N.M.C & B.Y.L. Nair, Mumbai.
Dr. Parth D. Barfiwala	Department of Paediatric Surgery, T.N.M.C & B.Y.L. Nair, Mumbai.
Dr. Shravya S. Shetty	Department of Paediatric Surgery, T.N.M.C & B.Y.L. Nair, Mumbai.
Dr. Hemanshi S. Shah*	Department of Paediatric Surgery, T.N.M.C & B.Y.L. Nair, Mumbai. *Corresponding Author

ABSTRACT

Background: Congenital anterior urethrocutaneous fistula is an exceptionally rare penile anomaly, with fewer than 70 cases reported worldwide. It is often misdiagnosed as hypospadias. **Case Report:** Patient presented with passing urine from the undersurface of the penis as well as the external meatus since birth suggestive of a congenital anterior urethrocutaneous fistula without any associated anomalies. Voiding cystourethrogram ruled out associated urethral stricture or diverticulum. The patient underwent anatomical repair with excellent results. **Conclusion:** Surgical management that considers the anatomical features of the fistula leads to excellent outcomes. This case contributes to the limited literature on this anomaly.

KEYWORDS

Congenital Urethrocutaneous fistula, Anterior urethra anomalies

INTRODUCTION:

Congenital anterior urethrocutaneous fistula (CAUF) is a rare and frequently misdiagnosed (as hypospadias) defect of the penile urethra. 63 cases have been reported.[1] It is usually solitary anomaly but can coexist with genitourinary or other abnormalities. The etiology postulated is a localized defect in the urethral plate that inhibits the union of the urethral folds. Operative management is based on the characteristics and location of the fistula.

CASE REPORT:

A 4-year boy presented with passing urine from the undersurface of the penis as well as the normally situated external urethral meatus. The boy had a ventral thin-walled cyst at the fistulous site at birth and a good urinary stream from a normally placed urethral meatus on the tip of the glans. The thin walled cyst burst spontaneously to form a fistula. There was no history of trauma or circumcision.

On examination, there was a 5 mm × 3 mm longitudinal defect on the ventral aspect of the penile urethra, a normally situated external urethral meatus with an intact prepuce and no chordee. (Fig.1) The urethra distal to the fistula was patent and of a good caliber. (Fig.2) Both testes were descended. Systemic examination was normal.

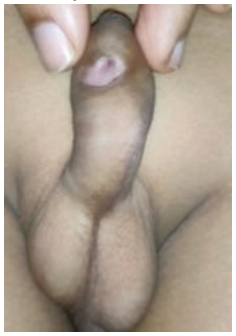

Fig 1. Congenital anterior urethrocutaneous fistula

Fig 2. Congenital anterior urethrocutaneous fistula with catheterisation of distal normal urethral meatus

A voiding cystourethrogram(VCUG) showed no diverticula or stricture along the urethra. Posterior urethra and bladder were normal with no vesicoureteric reflux.

A circumferential incision was made around the congenital anterior urethrocutaneous fistula, urethroplasty was done with a continuous subcuticular delayed absorbable suture over a 7 Fr feeding tube. The suture line was reinforced with a dartos flap and penile skin. The child voided normally postoperatively and has no complications after two years.

DISCUSSION:

The etiology of congenital urethrocutaneous fistulas is multifactorial because of the diverse appearance of congenital fistulas with or without hypospadias. According to a theory, congenital anterior urethrocutaneous fistula is a result of embryonal urethral blowout attributed to congenital distal urethral block.[1]Osbourne et al theorized that a focal deformity in the urethral plate results in arrested distal migration of the urethral plate or localized insufficiency of a portion of the plate.[2] In another theory, the testosterone or androgen receptors may be at fault giving rise to fistula. Misalignment of glandular and penile urethra may lead to development of a coronal type of fistula.[2] Presence of urethrocutaneous fistula may be explained by pressure

necrosis due to babies heel according to Cook and Stephens. [3] Occurrence of fistulas may also be triggered by growth of cysts along the mediogenital raphe during the antenatal period.[3]

Congenital anterior urethrocutaneous fistula (CAUF) usually presents as an isolated condition. Associations include imperforate anus, cryptorchidism, vesicoureteral reflux, and renal agenesis. [4] CAUF may coexist with chordee, scrotal anomalies (e.g., bifid scrotum) and hypospadias variants, suggesting overlapping developmental pathways. [5,6] Rare instances link CAUF to genetic syndromes, such as Pallister-Hall syndrome. [4]

An isolated fistula is associated with normal foreskin, no chordee and an intact distal urethra and spongiosum. Cases associated with hypospadias have features of chordee, dorsal hood and spongiosal deformity. Proximal urethrocutaneous fistula are a limb of the Y type of urethral duplication and may be associated with anorectal malformations. [6]

VCUG provides a detailed pathway of the urethra. Retrograde urethrography and cystoscopy enhance diagnostic accuracy.

The surgical procedure is tailored to the size and location of fistula and status of distal urethra and meatus. Local flap repair is adequate for isolated fistulas with an intact spongiosum. Cases accompanied by distal urethral deficiencies, chordee, hypospadias, or external meatal stenosis necessitate more extensive interventions, such as conventional hypospadias repair. [2,7] Various surgical techniques have been proposed, including Thiersch-Duplay urethroplasty, turned-down flap urethroplasty, and pedicled island tube or onlay urethroplasty, tailored to the specific anatomical characteristics of the fistula. [8]

CONCLUSION

Congenital anterior urethrocutaneous fistula is a rare anomaly often misdiagnosed as hypospadias. Treatment is individualized to the location and characteristics of the fistula.

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