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Congenital Distal Penile Urethrocutaneous Fistula Without Hypospadias or Meatal Stenosis in an Adolescent: A Case Report

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ABSTRACT: **Background**: Congenital anterior urethrocutaneous fistula (CAUF) is a rare developmental anomaly that may occur with associated genitourinary or anorectal abnormalities.

Case presentation: We report a 13-year-old male who presented with urinary leakage through an additional opening on the ventral aspect of the distal penile shaft while continuing to void through the orthotopic urethral meatus. The opening had first been noticed at approximately 10 years of age. There was no reported history of previous penile or urethral surgery, instrumentation, trauma, or preceding local infection. Examination demonstrated a distal penile urethrocutaneous fistula without clinical evidence of hypospadias or meatal stenosis. Both testes were descended, and the scrotum was normal. The diagnosis was established clinically. The fistulous tract was excised, the urethral defect was closed with interrupted absorbable sutures, and a second tissue layer was placed over the repair. The intraoperative saline leak test demonstrated no leakage. Estimated blood loss was 20 mL, and no intraoperative complications occurred.

Conclusion: This case describes an unusual distal penile urethrocutaneous fistula without hypospadias or meatal stenosis and without an identifiable acquired cause.

1. INTRODUCTION

Urethrocutaneous fistula is an abnormal communication between the urethral lumen and the overlying skin. Congenital anterior urethrocutaneous fistula (CAUF) is a rare developmental anomaly that may occur in association with other genitourinary or anorectal abnormalities. Its clinical presentation and anatomical location vary among reported cases. [1–4]

A systematic review published in 2018 identified 63 patients from 34 publications. The subcoronal region was the most frequently reported fistula location, accounting for 46.0% of cases, and associated genitourinary anomalies were identified in 30.2% of patients. [1] A more recent systematic review published in 2026 included 78 patients from 46 studies. The most frequent locations were subcoronal (42.3%) and mid-penile (39.7%), and the reported recurrence rate was 8.9%. [2] These findings emphasize the anatomical variability of CAUF and the importance of documenting associated abnormalities and postoperative outcomes.

We report an adolescent male with a distal penile urethrocutaneous fistula, a normally positioned orthotopic meatus, no clinical evidence of hypospadias or meatal stenosis, and no reported history of penile or urethral surgery, instrumentation, trauma, or preceding local infection. The case is notable for its delayed recognition and reported family history.

2. CASE PRESENTATION

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A 13-year-old male was referred to the pediatric urology service for evaluation of a urethrocutaneous fistula. He presented with urinary leakage through an additional opening on the penis while continuing to void through the orthotopic urethral meatus. The fistulous opening had first been noticed at approximately 10 years of age.

There was no reported history of previous penile or urethral surgery, urethral catheterization or other instrumentation, penile trauma, or preceding local infection or urethral discharge. The patient had been circumcised during infancy.

The patient's mother reported that a maternal cousin had reportedly been born with a similar urethrocutaneous fistula. However, medical records confirming the relative's diagnosis were unavailable. The patient's documented medical history included nasal deviation treated with nasal sprays and a history recorded as "ACL/corticosteroid treatment".

2.1 Clinical Findings

Genital examination demonstrated a urethrocutaneous pinpoint fistula on the ventral aspect of the distal penile shaft. The urethral meatus was normally positioned, with no clinical evidence of hypospadias or meatal stenosis. Both testes were descended, and the scrotum was normal. No additional fistulous opening was identified. (Figures 1 and 2)



Figure 1. Preoperative clinical photograph demonstrating the urethrocutaneous fistula on the ventral aspect of the distal penile shaft.



Figure 2. Preoperative clinical photograph demonstrating the urethrocutaneous fistula on the ventral aspect of the distal penile shaft.

2.2 Diagnostic Assessment

The diagnosis was established clinically based on the additional penile opening associated with urinary leakage and the findings on genital examination. Preoperative laboratory investigations were reported to be within normal limits. No radiological imaging was performed.

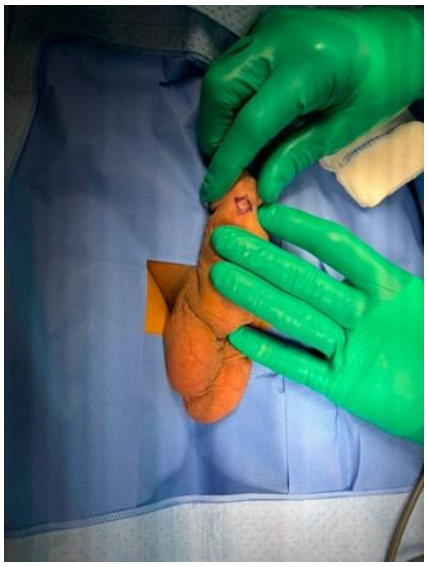


Figure 3. Intraoperative photograph demonstrating identification and surgical dissection of the distal penile urethrocutaneous fistula.



Figure 4. Intraoperative photograph demonstrating identification and surgical dissection of the distal penile urethrocutaneous fistula.



Figure 5. Intraoperative photograph demonstrating identification and surgical dissection of the distal penile urethrocutaneous fistula.

2.3 Informed Consent

The procedure, expected outcomes, alternatives, and potential complications were discussed with the patient and his guardian, and informed consent for the procedure was obtained.

2.4 Operative Technique

The patient was brought to the operating room and placed in the supine position. General anesthesia was induced, and the genitalia were prepared and draped in the usual sterile fashion.

An 8-Fr feeding tube was passed into the bladder. The fistulous opening was identified and circumferentially marked. A circumferential incision was made around the opening, and the fistulous tract was completely excised and carefully dissected from the surrounding tissues. The urethral edges were mobilized to permit tension-free closure. (Figures 3–5)

The urethral defect was closed using interrupted 5-0 absorbable Vicryl Rapide sutures. The repair was tested by instillation of sterile saline through the urethral catheter, with no leakage observed.

A second tissue layer was mobilized and secured over the urethral repair. with the second-layer suture line offset from the underlying urethral closure. Hemostasis was achieved, and the skin was reapproximated using interrupted absorbable sutures.

A 12-Fr urethral catheter was secured and left to free drainage, and a sterile dressing was applied. The estimated blood loss was 20 mL. No intraoperative complications occurred, and the patient was transferred to the recovery area in stable condition.

2.5 Post operative Technique

The patient was reviewed postoperatively and was awake, alert, and hemodynamically stable. Pain was adequately controlled, with no nausea or vomiting. (Figures 6 and 7)

The operative site was clean and dry, with the dressing intact. There was no active bleeding or significant swelling. The urethral catheter was in situ, patent, and draining urine adequately.

The postoperative plan consisted of monitoring vital signs and urine output, maintaining the urethral catheter on free drainage, and avoiding traction or manipulation of the catheter. Analgesia was prescribed, with antibiotics as indicated according to the operative plan and local protocol.

The patient was instructed regarding wound and catheter care and to monitor for bleeding, hematoma, infection, or urine leakage. Oral fluids and diet were encouraged as tolerated.

The urethral catheter was planned to remain in place for seven days, followed by pediatric urology follow-up for wound assessment and catheter removal. Further outpatient follow-up was planned approximately three weeks postoperatively to assess wound healing and evaluate for fistula recurrence.

At the time of preparation of this report, the patient was only two days postoperatively; therefore, definitive long-term outcome and recurrence data were not yet available.



Figure 6. Immediate postoperative photograph showing the penis following completion of urethrocutaneous fistula repair, prior to application of the postoperative dressing.

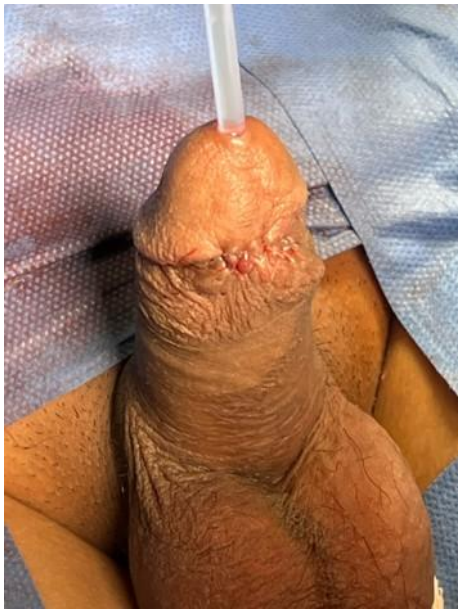


Figure 7. Immediate postoperative photograph showing the penis following completion of urethrocutaneous fistula repair, prior to application of the postoperative dressing

3. DISCUSSION

Congenital anterior urethrocutaneous fistula (CAUF) is a rare developmental anomaly that may occur in association with other genitourinary or anorectal abnormalities. Previous case reports and systematic reviews have documented considerable variation in fistula location, associated abnormalities, and surgical management. [1–13] In the present case, the fistula was located on the ventral aspect of the distal penile shaft, with a normally positioned orthotopic meatus and no clinical evidence of hypospadias or meatal stenosis.

The 2018 systematic review by Lin et al. included 63 patients from 34 publications and reported that subcoronal fistulas were the most frequent presentation, while associated genitourinary anomalies were identified in 30.2% of patients. [1] The 2026 systematic review by Sigumonrong et al. included 78 patients from 46 studies and found that subcoronal and mid-penile locations were the most frequently reported, with a recurrence rate of 8.9%. [2] These findings emphasize the importance of documenting the anatomical location of the fistula and assessing for associated abnormalities. In this patient, both testes were descended, the scrotum was normal, and no additional fistulous opening was identified. The available examination findings support an apparently isolated presentation, although the extent of the evaluation for associated anomalies should be stated accurately.

The fistulous opening was first noticed at approximately 10 years of age. The absence of a reported history of previous penile or urethral surgery, instrumentation, trauma, or preceding local infection raises the possibility of a congenital origin.

The patient had undergone circumcision during infancy. Although this history is relevant when considering possible acquired penile or urethral injury, no documented perioperative urethral injury or other evidence directly linked the circumcision to the fistula. Previous reports of CAUF have also documented patients with a history of circumcision, illustrating that circumcision history alone does not establish the cause of a fistula. [1] The available information does not support attributing the lesion to circumcision.

The patient's mother reported that a maternal cousin had reportedly been born with a similar urethrocutaneous fistula. This observation raises the possibility of familial occurrence; however, the relative's diagnosis could not be independently verified because medical records were unavailable. Consequently, a hereditary or genetic association cannot be established from this case. Further evidence would be required to investigate a possible familial predisposition.

The diagnosis in this patient was established clinically, and no radiological imaging was performed. The additional penile opening, reported urinary leakage, and examination findings supported the clinical diagnosis.

The principal objectives of surgical repair are complete excision or management of the fistulous tract, watertight closure of the urethral defect, tension-free approximation of the urethral edges, and coverage with an appropriate vascularized tissue layer when indicated. Various reconstructive approaches have been described in the CAUF literature, with technique selection depending on the anatomical location and extent of the defect. [1–13] In this case, the fistulous tract was excised, the urethral defect was closed using interrupted absorbable sutures, and the repair was tested with sterile saline without observed leakage. A second tissue layer was placed over the repair

A urethral catheter was planned to remain in place for seven days to provide urinary diversion during early healing. The immediate postoperative course was uncomplicated, with adequate catheter drainage and no significant bleeding or swelling. Nevertheless, the absence of immediate complications does not establish long-term surgical success. Follow-up is required to assess wound healing, urinary function, persistent leakage, and fistula recurrence.

4. CONCLUSION

A congenital distal penile urethrocutaneous fistula may present in an adolescent male without hypospadias or meatal stenosis and without an identifiable acquired cause. In such cases, a congenital or developmental origin may be suspected, but delayed recognition limits certainty regarding etiology. Careful documentation of associated anomalies, diagnostic assessment, operative anatomy, and surgical technique is important when reporting this rare presentation. In the present case, complete fistulous tract excision and urethral closure with a second tissue layer were followed by an uncomplicated immediate postoperative course. Further follow-up is required to establish long-term healing and exclude recurrence.

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AUTHOR CONTRIBUTIONS

Mais Al-Watban: prepared the manuscript, conducted the literature review, collected the data, and prepared the figures

Fawaz Al-Ibrahim: contributed to the operative details and description of the surgical procedure.

Mohammad Alabdulaaly: reviewed the manuscript and provided general feedback. All authors reviewed and approved the final manuscript.

CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

ETHICS STATEMENT

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying clinical information and images.

Consent for publication of images

Written informed consent was obtained from the patient for publication of the accompanying images and other relevant clinical information included in this manuscript.

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Clinical trials and ethical considerations

not applicable

DATA AVAILABILITY STATEMENT

All relevant clinical information supporting this case report is included in the manuscript. Additional patient-related information is not publicly available to protect patient confidentiality

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