Al-Azhar International Medical Journal

Volume 5 | Issue 6 Article 9

7-1-2024

Section: Pediatrics & its Subspecialty.

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How to Cite This Article

Salama, Ahmed; Daboos, Mohammad; Kamal, Ahmed; Zayed, Nader; and Hakiem, John (2024) "Role of Cysto-urethroscopy in the management of congenital urethral duplication in pediatrics. A case Report," *Al-Azhar International Medical Journal*: Vol. 5: Iss. 6, Article 9.
DOI: https://doi.org/10.58675/2682-339X.2469

CASE REPORT

Role of Cysto-urethroscopy in the management of congenital urethral duplication in pediatrics. A case Report

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Abstract

Background: ***Urinary tract duplication is an extremely unusual birth defect that can impact genito-urinary system. Two urethral tracts, either partial or complete, are the hallmark of this disorder. Numerous anatomical variations have been described, with various presentations. Voiding cystourethrography and intraoperative urethroscopic evaluation are important methods for diagnosis, determining the type of duplication, and providing a standard view for surgical operations.***

Aim: ***To demonstrate the importance of cysto-urethroscopy for intraoperative evaluation of urethral duplication.***

Case Presentation: ***Here we describe a case of full urethral duplication in a child who was 4 years old. During the operation, cystourethroscopy showed that the bladder and orthotopic urethra were both normal. Distal to the bladder neck, an opening of the accessory urethra was detected. So, the duplicated urethra was excised as proximally as possible.***

Results: ***The postoperative follow-up period was uneventful, and the patient had a satisfactory cosmetic penile shape, normal urinary stream, and free postoperative cystourethrogram.***

Conclusion: ***Intraoperative urethroscopic evaluation of urethral duplication is valuable to confirm the exact position of the internal opening and proper anatomical identification. Also, meticulous and complete separation of the accessory urethra is essential to obtaining a safe procedure and good post-operative results.***

Keywords: Pediatrics; Urethral duplications; Cysto-uretroscopy

1. Introduction

One of the rare congenital anomalies that primarily affects males is urethral duplications, while a few cases in females have been published. [1]

The clinical presentations and anatomical variations make diagnosis and classification challenging. Among the most common clinical indicators that patients may experience are urinary incontinence, recurrent UTIs, and oftenly a double urine stream. Even though some patients may not even show any symptoms .[2]

The diagnosis of urethral duplication may be challenging; it needs careful voiding history, physical examination, cystourethrography (VCUG), and recently, cystourethroscopy, used only in some centers. [3]

This study aimed to evaluate the effectiveness of intraoperative cysto-urethroscopy in managing cases of urethral duplication and report our experience with such anomalies.

2. Case presentation

The patient was a 4-year-old boy who had no prior history of urinary tract issues when his double urine stream and double urethral meatus were identified during a routine circumcision.

A larger, orthotopic urethral meatus was present at the glans in the patient, but a smaller, dorsally located meatus was present as well.

A voiding cystourethrogram revealed complete urethral duplication up to the bladder neck (Figure 1), while ultrasounds of the kidneys and urinary bladder were normal.

Accepted 21 June 2024.
Available online 31 June 2024

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https://doi.org/10.58675/2682-339X.2469

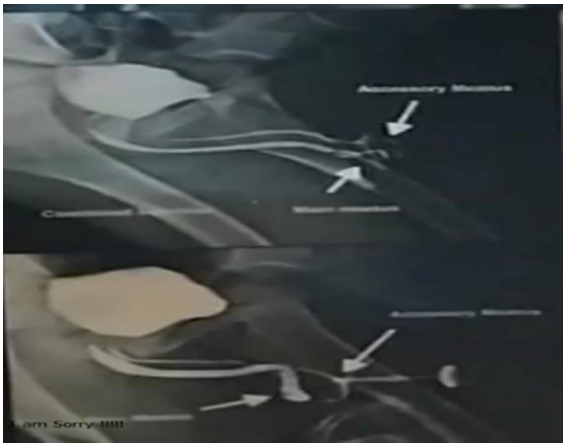

Figure 1. Two distinct lumens and apertures were revealed by the voiding cysto-urethrogram (VCUG), which revealed a fully replicated urethra that originated in the bladder.

2.1.Surgical procedure:

An orthotopic urethra and bladder were found during a cysto-urethroscopy that was carried out under general anesthesia following thorough sterilisation and draping. Further, an external, tiny ureteric catheter was used to catheterize the auxiliary urethra (Figure 2). An entry location for the ureteric catheter into the urinary bladder has been identified, which is located directly anterior to the bladder neck (Figure 3). Then, traction suture was applied at the glans, and penile skin was degloved up to the peno-pubic angle and was continued proximally to the symphysis pubis (Figure 4).

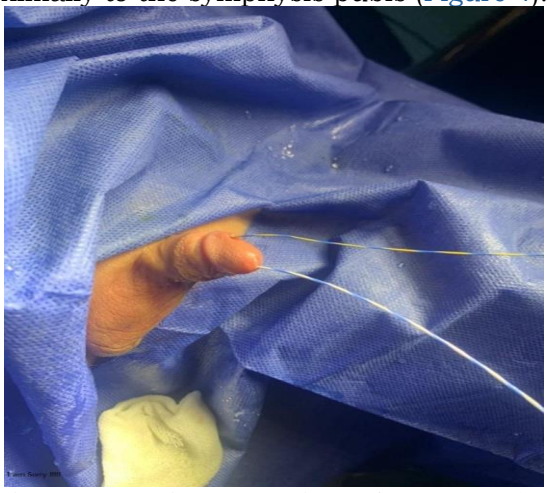

Figure 2. A tiny ureteric catheter (3-Fr) was used to cannulate the accessory urethra.

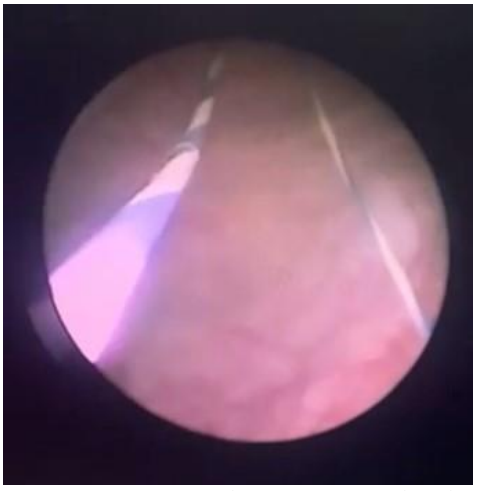

Figure 3. Intraoperative cystoscopic view after cannulation of both urethras.

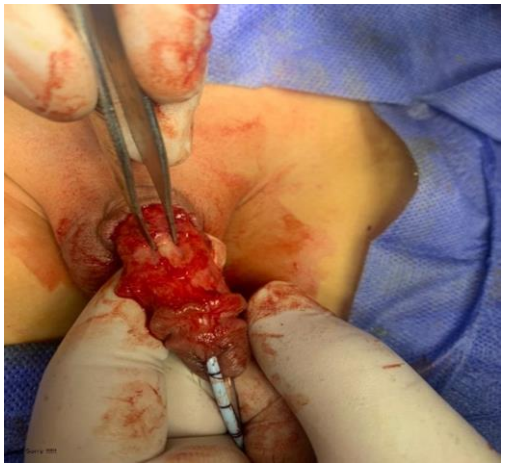

Figure 4. A full degloving of the penile skin was done, and the dorsal accessory urethra was shown.

To separate glandular part of the accessory urethra, a midline dorsal incision was created at the glans. Subsequently, the penile part was dissected and isolated from the corpora cavernosa's (Figure 5). Great care was given to the neurovascular bundles and external sphincter. Then, the accessory urethra was transfixed, ligated, and excised behind the symphysis pubis.

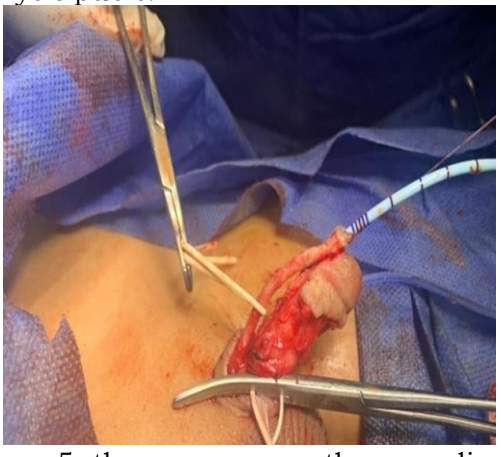

Figure 5. the accessory urethra was dissected off corpora cavernosa as proximal as possible to

the symphysis pubis.

3. Results

A Foley catheter was left for 6 days post-operatively, under antibiotic coverage (Amoxycillin 100 mg/Kg) and paracetamol (15 mg/Kg) for analgesia. The patient was followed for 9 months post-operatively, with uneventful follow-up period, he had normal urinary stream, very satisfied penile shape (Figure 6) and free post-operative cysto-urethrogram (Figure 7).

Figure 6. The penis after one month of the surgical correction.

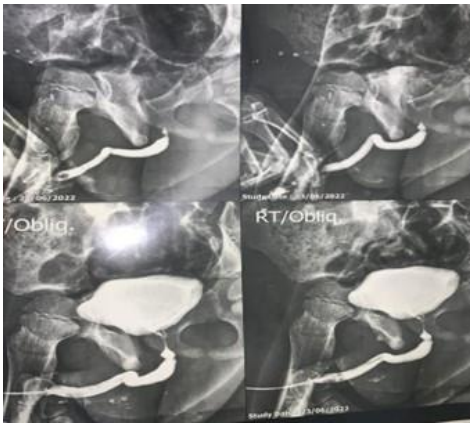

Figure 7. Cystourethrogram post operatively showing normal urethra without residual of duplication.

4. Discussion

A rare congenital urological defect known as urethral duplication (UD) is more commonly observed in males and can appear in children with a variety of symptoms. [4,3,5]

The clinical presentations include a double urine stream, recurrent infection of the urinary tract, and urinary incontinence .[6] Finding the right imaging test and arranging a suitable surgical procedure can be difficult, especially for patients who have coexisting conditions such as bladder exstrophy, epispadias, or hypospadias .[7]

The three types of urethral duplication identified by Effman et al. vary in relation to the position of the auxiliary urethra. In type I, the accessory urethra is blindly terminated; there are two subtypes of this disease. When the accessory urethra is Type I-A, it opens on the dorsal or ventral surface but doesn't connect to the main urethra. Type I-B urethral fistulas have an unplanned ending in the periurethral tissues after having started in the urethral channel. The second kind, Type II, is defined by an entirely patent accessory urethra; there are two subgroups within this variety, A and B, that are based on the quantity of meatuses. Two subtypes, IIA1 and IIA2, are recognized within Type IIA. The two urethras of type IIA1 come out of the bladder neck separately, but the second Y-shaped channel of type IIA2 comes out of the first and goes behind it into a second meatus. Two type II-B urethras join distally into one channel after emerging from the posterior urethra, also known as the bladder. An additional, separate urethra develops in cases of type III, which is a duplicate bladder.[8] The current case was diagnosed as urethral duplication type IIA1, the most prevalent type of urethral duplication described in the literature.[3,9]

The management of urethral duplication should be tailored for each patient according to clinical presentation and anatomical type. Although low-grade incomplete urethral duplication may go unnoticed and untreated, the higher-grade variants typically require complicated and repeated procedures. [2]

A genital examination is the first step in assessing a child with a double urethra in order to look into the distal orifices and determine whether both urethras are functioning or not. Radiological examinations, such as retrograde urethrography and micturating cystourethrography, are crucial for diagnosing and determining the anatomical type of duplication. From the perspective of this equipment, the current case has been identified preoperatively .[3]

Cysto-urethroscopy was performed in the current case prior to surgical excision of the accessory urethra to determine the location of the internal opening to avoid inadequate or excessive removal .[3] In addition, Guglielmetti et al. and Wani et al. believed that direct visualization by cysto-urethroscopy was important for preventing false results with radiological workups .[5,10]

Most documented cases, including ours, did not scope the accessory urethra because of its narrow lumen. However, Baid et al. reported a urethral duplication case when a cystourethroscopy was used to evaluate two urethrae .[11]

Every instance requires a tailored surgical approach that takes into account the specific form of urethral duplication as well as the

patient's present symptoms. .[9] Basic techniques involve separating the tissue, cutting out the extra urethra, and then reattaching it. Following procedures similar to those of Suoub et al. and Kang et al., we were able to successfully dissect the accessory urethra from the corporal bodies, transfix it, ligate it proximal to the symphysis pubis, and then remove it .[9, 12]

In our case, the surgical outcome was satisfactory; the patient had a single stream and no signs of urinary incontinence, the postoperative course was smooth, and the cosmetic results were very satisfactory. We agree with Suoub et al., and Kang et al. that cystourethrography postoperatively are valuable for demonstrating that the accessory urethra has been successfully and completely removed.[9,12]

4. Conclusion

Intraoperative urethroscopic evaluation of urethral duplication is valuable in confirming the exact position of the internal opening and proper anatomical identification. Also, meticulous and complete separation of the accessory urethra is essential to obtaining a safe procedure and good post-operative results.

Disclosure

The authors have no financial interest to declare in relation to the content of this article.

Authorship

All authors have a substantial contribution to the article

Funding

No Funds : Yes

Conflicts of interest

There are no conflicts of interest.

References

1. Thapa S, Awal S, Basukala S, Thapa N, Thapa BB, Thapa S. Duplication of urethra: A case report from Nepal. Clin Case Rep. 2023;11(2):e6925
2. Lopes RI, Giron AM, Mello MF, et al. Urethral duplication type influences on the complications rate and number of surgical procedures. Int Braz J Urol. 2017;43(6):1144-1151
3. Jahanshahi, A., Falaki, R. & Falahatian, M. Voiding cysto-urethrography of complete urethral duplication in a child, a case report. Egypt J Radiol Nucl Med .2022; *53*(1), 101.
4. Patel H, Watterson C, Chow JS. Case of urethral duplication seen by voiding urosonography. Clin Imaging. 2018;49:106-1101
5. Wani SA, Munianjana NB, Jadhav V, Ramesh S, Gowrishankar BC, Deepak J. Urethral Duplication in Children: Experience of Twenty Cases. J Indian Assoc Pediatr Surg. 2019;24(4):275-280
6. Lima M, Destro F, Di Salvo N, Gargano T, Ruggeri G. Fate of males with urethral "Y-duplication": 40-year long follow-up in 8 patients. J Pediatr Surg. 2017;52(8):1335-1339
7. Salle JL, Sibai H, Rosenstein D, Brzezinski AE, Corcos J. Urethral duplication in the male: review of 16 cases. J Urol. 2000;163(6):1936-1940
8. Effmann EL, Lebowitz RL, Colodny AH. Duplication of the urethra. Radiology. 1976;119(1):179-185
9. Suoub M, Saleem MM, Sawaqed F. Complete Urethral Duplication: Case Report and Literature Review. Res Rep Urol. 2020;12:15-20
10. Guglielmetti LC, Delcont M, Walker J, Wilcox D, Vuille-Dit-Bille RN. Urethral duplication-Epidemiology, diagnosis, and treatment in a case series of 19 patients. J Pediatr Urol. 2020;16(3):385.e1-385.e9
11. Baid M, Dutta A. Urethral duplication in a 15-year-old: case report with review of the literature. Rev Urol. 2014;16(3):149-151.
12. Kang SK, Kim J, Lee YS, Han SW, Kim SW. Urethral duplication in male children: A study of 12 cases. J Pediatr Surg. 2020;55(10):2216-2220