Urology 209 (2026) 90–92

Contents lists available at ScienceDirect

Urology

journal homepage: www.sciencedirect.com/journal/urology

Invited Pediatric Case Report

Urethral Duplication Mimicking an Interlabial Cyst in a Girl: A Diagnostic Challenge

Luciana Lerendegui[a,b,*], Juanita Velasquez[c], Daniel Tennenbaum[c], Miguel Castellan[c]

[a] Jackson Memorial Hospital, Miami, FL
[b] Universtiy of Miami, Miller School of Medicine, Miami, FL
[c] Nicklaus Children's Hospital, Miami, FL

A B S T R A C T

Urethral duplication is a rare urogenital anomaly, especially in females. We present the case of a preterm female with various congenital anomalies, hydrocolpos, and a persistent interlabial cyst, later found to be secondary to urethral duplication. At age 2, cystoscopy and VCUG confirmed a duplicated urethral tract. Surgical resection of the accessory urethra and genitoplasty were performed successfully. Postoperative recovery was uneventful, with excellent cosmetic and functional outcomes at follow-up. This case highlights urethral duplication as a rare but possible differential diagnosis for interlabial cystic lesions in females, particularly when associated with fluctuating size and underlying urogenital anomalies.

Urethral duplication is an uncommon urogenital anomaly, especially in female patients.[1] It is frequently associated with other anomalies and traditionally, diagnosis in girls is frequently made in the context of urinary incontinence that motivates further workup.[2] Usually, on physical exam, a secondary urethral meatus can be appreciated dorsally to the main urethra and contrast studies, such as voiding cystourethrography, can reveal the presence of the accessory urethra. However, this anatomical configuration can have variations, leading to diagnostic challenges if not included within the spectrum of differential diagnosis. We present a rare case of urethral duplication in a female presenting as an interlabial cyst.

CASE DESCRIPTION

We present the case of a preterm female infant, born at 30 weeks with a weight of 1.7 kg, who was consulted to our team due to hydrocolpos and an interlabial cyst Fig. 1a in the context of fused crossed renal ectopia, bicornuate uterus, atrial septal defect, vertebral anomalies (hemivertebrae) and tethered cord. On initial physical exam, limited by patient's size, the external genitalia looked predominantly with a female-type configuration but still an ambiguous appearance caused by the presence of a cystic structure between the labia. There were no palpable gonads. Identification of the urethral opening was difficult and there were concerns for a urogenital sinus. The anus was orthotopic. Decision was to start her on intermittent catheterization which was subsequently discontinued given the complete resolution of the hydrocolpos and lack of recurrence, with no need for further intervention. Additionally, a full workup for DSD was completed and results were consistent with normal XX karyotype, presence of both uterus and ovaries and no metabolic abnormalities.

She responded well to initial conservative treatment, demonstrating the ability to empty the bladder to completion with no recurrence of hydrocolpos and no episodes of urinary tract infection. However, the interlabial cyst persisted, showing cyclic fluctuations in size with voiding. Differential diagnoses were considered, including prolapsing ureterocele and paraurethral cyst among others. An attempt to perform a voiding cystourethrography (VCUG) was aborted due to difficulty catheterizing the urethra which motivated, at age 2, her being taken to the OR for assessment. Exam under anesthesia demonstrated a female configuration of the genitalia with a midline cyst, immediately ventral to the clitoris. Cystoscopy at that time revealed the urethral meatus to be on the anterior vaginal wall, 0.5 cm proximal to the introitus (female hypospadias). There was a single vagina and cervix that appeared normal. A Foley catheter was inserted under vision, and postprocedural VCUG demonstrated filling of the cyst during voiding Figure 1B. Decision was made to proceed with surgical treatment.

The surgery began with anterior incision of the cyst and subsequent passage of a hydrophilic wire through a small opening on its posterior wall, while simultaneous urethroscopy was performed Figure 2A,B,C. The wire was identified entering the main urethra just below the bladder neck, after passing through a long connecting tract Figure 2B,C. The cyst was carefully resected, with complete preservation of the neurovascular bundle and the clitoris. The communicating tract was

* Address correspondence to: 3200 60th Ct Sutie #104, Miami, FL 33155.
E-mail address: lerendeguiluciana@gmail.com (L. Lerendegui).

https://doi.org/10.1016/j.urology.2025.09.009
Received 1 July 2025; Accepted 5 September 2025

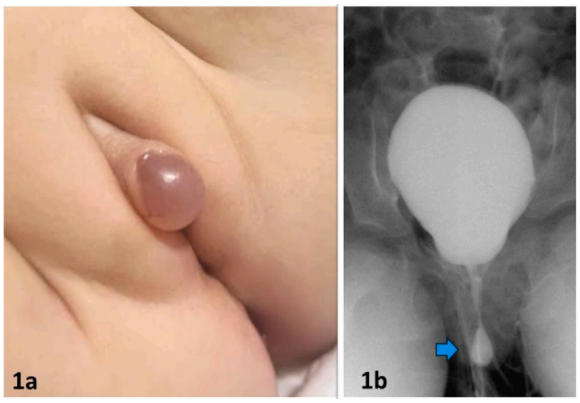

Figure 1. Initial assessment. **(A)** Physical exam: female configuration of the external genitalia with cystic structure on the ventral aspect of the clitoris. **(B)** Voiding Cystourethrogram: voiding phase of the study that shows contrast accumulation in a round structure (blue arrow) consistent with cystic lesion on physical exam.

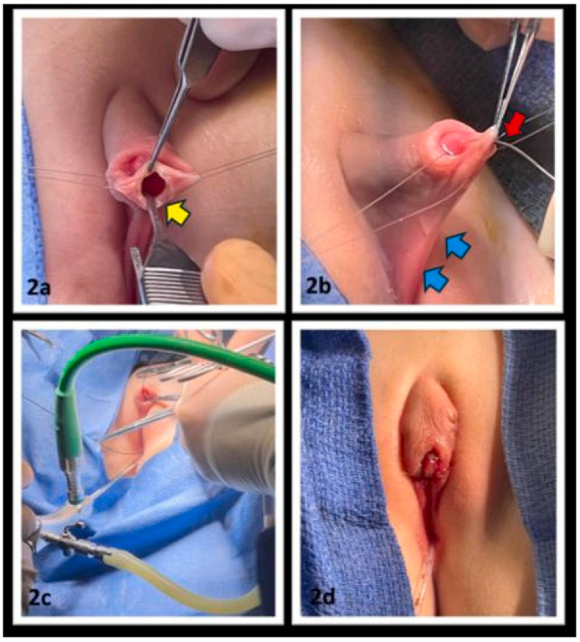

Figure 2. Surgical intervention. **(A)** Open cyst. **(B)** The wire (red arrow) is entering the open cyst. The direction of the accessory urethra can be externally predicted (blue arrows). **(C)** Simultaneous urethroscopy demonstrating the entrance of the wire from the cyst, through the accessory urethra into the main urethra. **(D)** Final result after completion of geitoplasty.

followed proximally and suture-ligated. Genitoplasty was subsequently completed Figure 2D. Final interpretation of the intraoperative findings was consistent with a female hypospadias with associated duplicated urethra connecting to a cyst Figure 3.

There were no intra or postoperative complications, with successful trial of spontaneous voiding and discharge on postoperative day 1. The final pathology report confirmed urethral duplication and associated cyst. At 5-month follow-up, the cosmetic result was excellent, with no evidence of cyst recurrence. The patient has continued to void without difficulty, keeps dry in between voids and has not had any episodes of UTI, upper tract dilation, or hydrocolpos, suggesting adequate voiding.

DISCUSSION

Urethral duplication is an extremely rare congenital anomaly, particularly in females, characterized by the presence of a complete or

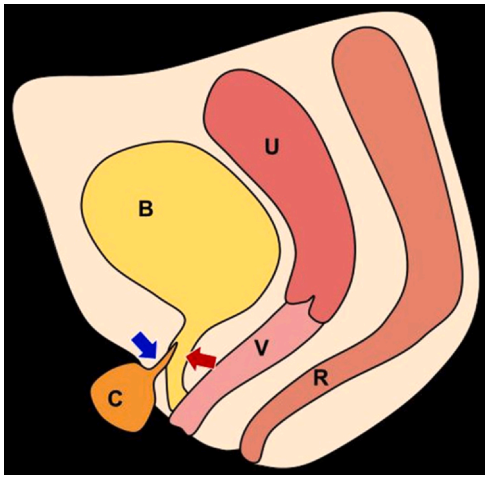

Figure 3. Diagram of intraoperative findings. Main urethra (red arrow) ending on the anterior vaginal (V) wall. Cystic structure (C) connecting with the duplicated urethra (blue arrow) that ends at the proximal main urethra, below the bladder neck.

partial accessory urethra that is typically located dorsal to the main urethra, following a sagittal plane.[1]

The etiology of this malformation remains unknown and, though different embryological theories have attempted to provide an explanation for its pathogenesis—such as urethral plate division secondary to inflammation or vascular insult or secondary to posterior displacement of the urogenital tubercule—none have gained widespread acceptance.[2-4]

In males, Effman's classification is most commonly used, including 3 distinct types with subcategories.[4,5] However, in females, 5 types have been described: (1) double urethra with a double bladder, (2) double urethra with a single bladder, (3) accessory urethra found posteriorly to the normal channel, (4) double proximal urethra with a single distal urethra, and (5) a single proximal urethra with a duplicated distal urethra.[6] In 2016, Bouty et al. published 3 cases of female urethral duplication cases in which the accessory urethra was epispadic and the main urethra had hypospadias.[5] This anatomical configuration is similar to what we observed in the present case. However, our patient had the additional feature of the accessory urethra draining into an interlabial cyst, different from that previously reported case series. Due to these particular anatomical features, the clinical presentation was not the classical form with urinary incontinence but rather required a careful analysis of differential diagnoses of pediatric female interlabial masses, which includes, among other paraurethral cysts, hymenal cysts, prolapsed urethra, botryoid sarcoma, and imperforate hymen with hydrocolpos.[7]

Previously reported presentations of urethral duplication include hydrocolpos, hydroureteronephrosis, incontinence, retrovesical masses, and even urethral diverticulum.[3,5] However, the presentation mimicking an interlabial cyst is even more rare, with only 2 more cases reported in the literature, to our knowledge.[8,9] Different from those cases in which the patients had large saccular cystic masses protruding dorsally to the urethra upon birth, our patient had a cyst that was anchored ventrally to the clitoris with progressive increase in size over time and cyclic fluctuations of its volume.

Management of urethral duplication varies depending on presentation and anatomical type. In the literature, surgical intervention is typically considered in cases of urinary incontinence, obstruction, and infections, double stream, and associated genitourinary malformations.[10] In our case, the presence of the interlabial cyst prompted surgical intervention, in a patient that was otherwise asymptomatic. Once anatomical features are properly delineated, the procedure can be executed safely with special consideration of preserving the clitoris intact while separating the accessory urethra proximally.

CONCLUSION

Urethral duplication is a rare congenital malformation, especially in girls. Though most cases will be diagnosed in the setting of incontinence due to the presence of an accessory tract, the present case illustrates that it should be included in the differential diagnosis of interlabial cystic lesions in females, especially when associated with intermittent size changes. Management includes resection of the cystic structure with closure of the communication between the accessory urethra, which will be dorsal and preservation of the main urethra. Timing for the procedure should be carefully decided based on each individual patient's characteristics.

Ethical Declaration

Informed consent was obtained from the patient's parents for publication of this case report. No patient identifying information has been included in this work.

Funding

The authors declare that they have no relevant financial interests.

CRediT Authorship Contribution Statement

Luciana Lerendegui: Writing – original draft, Investigation, Conceptualization. **Juanita Velasquez:** Writing – original draft, Investigation, Conceptualization. **Daniel M. Tennenbaum:** Writing – review & editing, Investigation, Conceptualization. **Miguel Castellan:** Writing – review & editing, Supervision, Project administration, Conceptualization.

Declaration of Competing Interest

The authors have no conflict of interest to declare.

Declaration of Generative AI and AI-assisted technologies in the writing process

The authors did not use generative AI or AI-assisted technologies in the development of this manuscript.

References

1. 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;16:385.e1–385.e9.
2. Ismail A, Al Rayes T, Alsalihi M, Abbas T. Female double urethra: a case report. *J Pediatr Surg.* 2012;47:29–31.
3. Arkan G, Kaya C, Turker LN, et al. Distal urethral duplication in a female child: a unique case report with diverticular component. *J Pediatr Adolesc Gynecol.* 2025;38:419–421.
4. Lima M, Destro F, Maffi M, et al. Practical and functional classification of the double urethra: a variable, complex and fascinating malformation observed in 20 patients. *J Pediatr Urol.* 2017;13:42.e1–42.e7.
5. Bouty A, Lefevre Y, Harper L, Dobremez E. Urethral duplication in girls: three cases associating an accessory epispadiac urethra and a main hypospadiac urethra. *J Pediatr Urol.* 2016;12:209.e1–209.e5.
6. Bhadury S, Parashari U, Singh R, Kohli N. MRI in congenital duplication of urethra. *Indian J Radiol Imaging.* 2009;19:232–234.
7. Willborg BE, Carroll VG, Meltzer J. Newborn with vaginal cyst. *Glob Pediatr Health.* 2021;8:1–2.
8. Kafando E, Moifo B, Mbouche L, Ndangoh D, Mah E, Mouafo Tambo F. Urethral duplication with a cystic phallic urethra associated with a uterus didelphys, partial agenesis of the tibia, and an equinovarus foot. *Case Rep Radiol.* 2018;2018:1–5.
9. Philippe-Chomette P, Zeidan S, Belarbi N, Van Der Meer G, Oury JF, El-Ghoneimi A. Female covered urethral duplication with urogenital sinus. *Urology.* 2012;79:e3.
10. Solanki S, Babu MN, Jadhav V, Gowrishankar, Ramesh S. Female urethral duplication: rare anomaly with unusual presentation. *J Surg Tech Case Rep.* 2015;7:1.