BJMS
www.batnajms.net

Batna Journal of Medical Sciences 2025;12(3):10
https://doi.org/10.48087/BJMSoa.2025.12310

ORIGINAL ARTICLE

Urethral duplication: from the careful examination to surgical repair

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ABSTRACT

Background. Urethral duplication is a rare lower urinary tract anomaly; less than 200 cases have been described in the literature, defined by two urethra, one orthotopic and the second ectopic. This study aimed to highlight the different spectrums or varieties of this rare anomaly to show the goal of careful examination and exploration to define the central urethra and the accessory urethra. **Materiel and methods.** retrospective study review of 10 cases of urethral duplication was operated between January 2018 and December 2022, age at the first consultation ranged from 15 months to 6 years. Urethral duplication is classified using Effmann classification. **Results**. Independent urethra was observed in 2 cases, urethral duplication type ⅡA2 in two instances, urethral duplication type ⅡA2, duplication in 1 case, urethral duplication with accessory epispadias urethra in 4 cases and blinded end duplication in two instances (type Ⅰ). The most common associated urinary tract disorder was VUR, which was present in 4 patients, and bilateral in 2 patients. **Discussion**. Urethral duplication is a rare anomaly, with poor sample cases described in the literature and not always identified in childhood. Classifications described malformations, but the most used classification was established by Effmann. The Evaluation included Ultrasonography, retrograde urethrography, and MRI. Surgical repair was indicated in all cases in cosmetic or symptomatic form. **Conclusion.** A rare condition that should be considered in cases of double stream by careful examination; associated malformations may be found and may be underdiagnosed. Surgical repair was defined according to anatomical findings, including excision of the accessory urethra and mucosal excision in Y duplication rather than excision.

Keywords: Congenital abnormalities, Urethra, Epispadias, Duplication.

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Received: 31 May 2025
Accepted: 01 Jul 2025

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1. INTRODUCTION

Urethral duplication is a rare condition of lower urinary tract anomalies [1]. Less than 200 cases have been described in the literature [2], defined by two urethra, one orthotopic and the second ectopic [3]. This study aimed to highlight the different spectrum or varieties of this rare anomaly to show that the goal of careful examination and exploration to define the primary urethra and the accessory urethra may be in dorsal position on epispadias or ventral position on hypospadias anywhere to the urethroperineal fistula. Several classifications described malformations, but Effman introduced the most used classification (Table 1).

The accessory urethra may be open near the anus, represented as a Y-type fistula, which is found in 6-30% of urethral duplications [1, 4]. The ectopic urethra is often expected and functional, and the main one is stenotic [5, 6]. With this different aspect of Y duplication, new terms are debated, like congenital perineal urethral-cutaneous fistula [7]

Urethral duplication may be in the sagittal plane, dorsal in epispadias, ventral, or coronal plane. Other malformations can be associated with urethral duplication, anorectal malformation, bladder extrophy, and posterior urethral valve [4].

Table 1. Effmann classification.

Type	Anatomical presentation
Type Ⅰ:	An accessory urethra or blinded incomplete urethral duplication.
Type Ⅱ:	complete duplication
Type ⅡA1:	separate bladder neck.
Type ⅡA2:	Y type duplication.
Type Ⅲ:	Complete bladder duplication.

2. MATERIAL AND METHODS

A retrospective review study analysed clinical presentation, evaluation and management of 10 cases of urethral duplication operated between January 2018 and December 2022 in the pediatric urology department of children hospital of Oran. The study was approved by the hospital's ethics committee. Inclusion criteria were all forms of urethral duplication; exclusion criteria were patients lost to follow-up or with incomplete medical records. The age at the first consultation ranged from 15 months to 6 years. Exploration studies included ultrasonography, retrograde cystourethrography, and lower endoscopy to better elucidate the anatomy of all patients. Urethral duplication cases were classified using the Effmann classification. All surgical procedures required general anaesthesia, and there were no dilatation attempts. Reconstructive surgery was indicated for all duplications.

3. RESULTS

Four patients were older than two years; the mean age at diagnosis was 2.5 years; no neonatal diagnosis was made. One case was diagnosed at the time of the urinary tract infection, and 1 case was diagnosed at the time of epispadias repair. Independent urethra was observed in 2 cases (type ⅡA1, case 1, figure 1), urethral duplication in two instances (type ⅡA2:1 case, type ⅡA2,Yduplication:1 case, figure8),), urethra duplication with accessory epispadias urethra in 4 cases (type Ⅱ, case 2, figure 2), and blinded end duplication in two instances (type Ⅰ). The most associated urinary tract disorder was VUR, which was present in 4 patients and bilateral in 2 patients. Two Surgical repairs were indicated in all cases, including excision of duplication in nine cases and mucosal excision in Y duplication rather than excision of duplications decided as a new technique for Y duplication in our department.

Case 1. A boy aged two years with epispadias. Clinical examination revealed a complete foreskin; two openings through which the child urine, a standard glandular apical hole, and another hole epispadias from joining forces are the root of the penis. The passage of two urinary probes confirms duplication (figure 1). Surgical repair included accessory urethral excision (Figure 1,2).

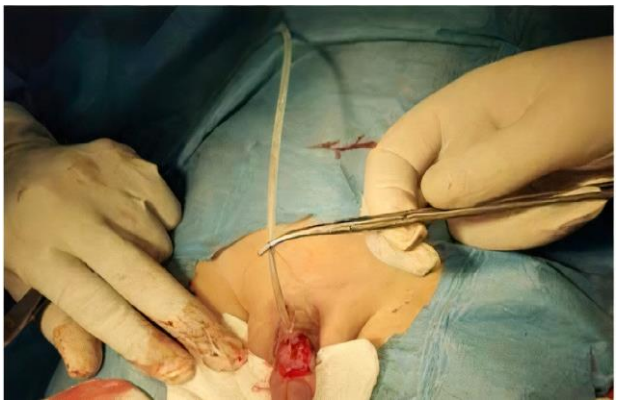

Figure 1. Urethral duplication dividing.

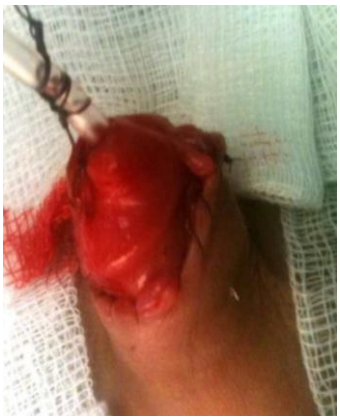

Figure 2. final aspect.

Case 2 A girl aged one year and a half was admitted for UTI. Examination revealed a distended bladder and a slightly hypertrophic clitoris. Accessory epispadias urethra (figure 3). Ultrasonography revealed bilateral hydronephrosis and large-capacity bladder-thick walls. Cystourethrography Showed proper unilateral vesicoureteral reflux. Endoscopy examination confirmed the accessory epispadias urethra .

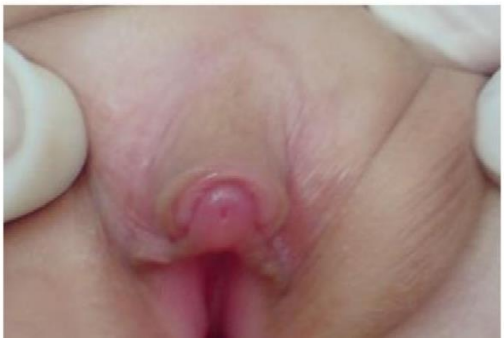

Figure 3. Urethral duplication with epispadias accessory urethra.

Case3. 1 The boy's perianal orifice was shown to be responsible for his limited urinary output through the posterior tract and his large urinary output through the orthotopic urethral meatus. Upon examining the youngster, we discovered a perianal hole causing problems with urine flow. Through contrast and endoscopic examination, a type Ⅱ A 2 urethral duplication (figure 4) was confirmed by comparison and endoscopy exam.

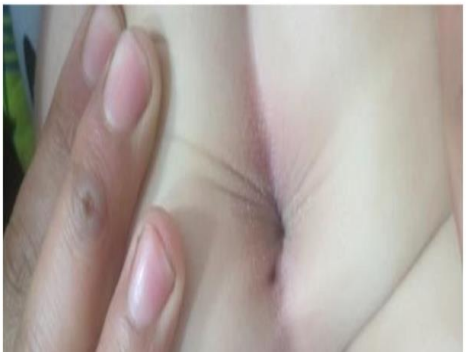

Figure 4. Urethra duplication , perianal urethrocutaneus fistula.

4. DISCUSSION

Urethral duplication requires a thorough clinical examination to determine the type and establish an appropriate therapeutic approach [1,4]. Urethral duplication is a rare anomaly, with poor sample cases described in the literature and not always identified in childhood, most commonly in boys; urethral duplication usually occurs in the sagittal plane rather than the frontal plane [4]. The disorder seems multifactorial, including ischemia abnormalities of Mullerian duct evolution [8], and no genetic evaluation has been identified in the literature. The most common classification for urethral duplication was established by Effmann [9]. It is functional and regroups all anatomical situations involving duplication. The Y duplication is different from other duplications by other severe malformations associated with the VACTERL association [10].

In our experience, using the most popular classification of Effman, we did not find complete bladder duplication (type Ⅲ),which is a more severe malformation. Type Ⅱ,with dorsal epispadias, is assimilated into complex extrophy epispadias[10], such as pubic bone diastasis and incontinence. Evaluation includes ultrasound, retrograde urethrography and MRI. The goal of preoperative assessment is to identify the anatomy of the duplication, associated anomalies, central urethra, and accessory urethra.

Surgical repair must provide a single, continent, and functional urethra for both voiding and sexual function. Retrograde cystourethrography is mandatory and can identify the enlarged and hypoplastic central urethra [11]. Removing the accessory urethra and epispadias urethra seems to be adequate repair for most urethral duplications. In the case of Y duplication, the central urethra is usually enlarged, with regular very montanum, diameter, and sphincter, and the accessory urethra is the ventral urethra with an atretic segment [7]. Mucosal excision is a simple treatment method without any functional complications and involves complex urethroplasty rather than duplication removal. Surgical repair was indicated in all cases for cosmetic reasons or symptomatic form.

5. CONCLUSION

In rare conditions, this anomaly can be missed but should be sought in cases of double stream by careful examination. Associated malformations may be found, and under-diagnosis and surgical repair are defined according to anatomical findings. Accessory urethral removal is the gold standard of surgical repair. The outcome in most cases was favourable, without any continence trouble or mechanical complications.

Competing interests: The authors declare that they have no competing interest.

Funding: This research received no external funding.

REFERENCES

1. Podesta ML, Medel R, Castera R, Ruarte AC. Urethral duplication in children: surgical treatment and results.*J Uro* 1998;160: 1830–3
2. Lukman O. Abdura-Rahman, Abur-Rasheed A Nasir, Agboola OJ. Penile shaft sinus: A sequelae of circumcision in Urethral duplication. Indian J Urol 2009;25:134-6.
3. Magalini SC, Magalini SIeds. VATER. In*Dictionary of Medical Syndromes*, 4th ed in: Philadelphia: Lippincott-Raven,1997: 816–7
4. Salle JL, Sibai H, Rosenstein D,Brzezinski AE, Corcos J.Urethralduplication in the male: review of 16 cases.*J Urol*2000;163: 1936–40
5. Wagner JR, Carr MC, Bauer SB, Colodny AH, Retik Ab et al. Congenital posterior urethral perineal fistulae. A unique form of urethral duplication.*Urology*1996;48: 277–80
6. Psihramis KE, Colodny AH, Lebowitz RL, Retik AB, Bauer SB. Complete patent duplication of the urethra. *J Urol*1986;136: 63–7
7. Bates DG, Lebowitz RL.Congenitalurethroperineal fistula.*Radiology*1995;194: 501–4
8. Belman, A. B.Hypospadias and other urethral abnormalities. In: Clinical Pediatric Urology, 3rd ed. Edited by P. P. Kelalis, L. R. King and A. B. Belman. Philadelphia: W. B. Saunders, vol. 1, pp. 619–663, 1992.
9. Effmann, E. L., Lebowitz, R. L. and Colodny, A. H.: Duplication of the urethra. Radiology, 119: 179, 1976.
10. Woodhouse, C. R. and Williams, D. I.: Duplications of the lower urinary tract in children. Br J Urol, 51: 481, 1979.
11. Psihramis, K. E., Colodny, A. H., Lebowitz, R. L. et al: Complete patent duplications of the urethra. J Urol, 136: 63, 1986