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Type IIB urethral duplication in young adult—A case report☆,☆☆,★,★★

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ABSTRACT

Urethral duplication is an extremely rare condition discovered in adults where most of them are diagnosed in childhood. Overall, it has 3 types according to Effman et al. The authors presented a case of an adult male complaining of dysuria, who was diagnosed with urethral duplication type IIB after performing retrograde urethrography and micturating cystourethrography. This is an extremely rare type of duplication of the urethra (type IIB) with late presentation. Further study may be required regarding the surgical management.

Background

Urethral duplication, a rare lower urinary tract congenital anomaly found in males (men and boys) [3]. Aristotle was the first who described urethral duplication [4]. It can present with other congenital malformations of the gastrointestinal tract, heart, and bones [1]. It is described as closely positioned 2 or more tracts of smooth musculature covered by the mucosal lining [2]. Multiple theories have been proposed to the mechanism of development of the urethral duplication such as partial mesodermal fusion, abnormal Mullerian ducts, ischemic insults during embryogenesis, and developmental defects of the urogenital sinus[1] however, the exact mechanism is still unclear [3]. The duplication can be partial or complete. It can occur in a sagittal plane as well as in the coronal plane [2]. For the diagnosis of urethral duplication, a genital examination is performed and confirmed by micturating cystourethrography (MCUG) and retrograde urethrography. Urethral duplication has various types that bring a therapeutic challenge for pediatric surgeons [1,2]. The selection of surgical technique depends on symptoms and the anatomy of the urethra and the urinary bladder neck [4]. We present a case of late presentation of urethral duplication, a rarer type (IIB), in an adult male.

Abbreviations: MCUG, Micturating Cysto-Urethrography; USG, Ultrasonography; IVU, Intravenous Urography; MRI, Magnetic Resonance Imaging.

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Fig. 1 – (A–C) Retrograde urethrogram (positive and negative fluoroscopic images) shows the opacification of both anterior and posterior urethras revealing two separate channels at the junction of anterior and posterior urethra reaching up to the neck of the urinary bladder.

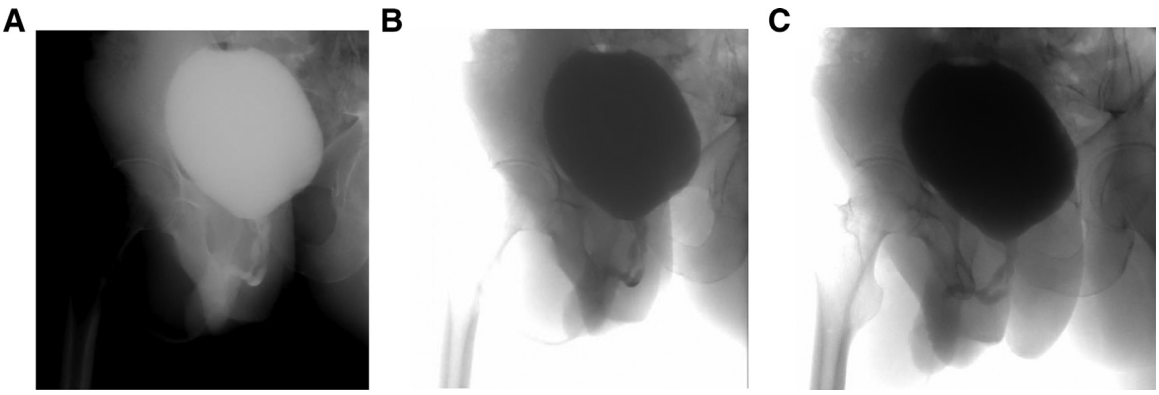

Fig. 2 – (A–C) Micturating cystourethrogram (positive and negative fluoroscopic images) shows two separate urethral channels originating at the neck of the urinary bladder during micturition phase and reuniting back at the junction of anterior and posterior urethras.

Case presentation

A 24-year-old adult male complaining of dysuria was referred to the radiology department for the retrograde urethrography procedure. No previous medical, family, psycho-social history, or relevant genetic information. No history of previous surgery. The patient had no abnormal findings during the physical examination. Retrograde urethrography was performed showing normal anterior urethra. When contrast passed to the posterior urethra, 2 distinct channels were opacified until reaching the urinary bladder (Fig. 1A,–C). The urinary bladder was filled with the contrast for voiding cystourethrography via video-fluoroscopy. During the micturition phase, double posterior urethral channels were opacified from the neck of the urinary bladder ending with single meatus at the junction of anterior and posterior urethra, classified as type IIB according to Effman et al. classification (Fig. 2A–C). Abdominal ultrasonography showed no abnormality in the urinary system. (kidneys, bladder). The patient had no other complaint that would implicate renal disorder nor had any gross abnormality in the external genitalia. After the radiological diagnosis, the patient went abroad for further management and was lost to follow up.

Discussion and conclusion

Urethral duplication was first described by Aristotle. It is a rare congenital anomaly, occurring mostly in males [2] frequently associated with the gastrointestinal and genitourinary anomalies [1,8] It is also termed as supernumerary urethra [10]. There are overall 300 cases present in the literature regarding urethral duplications [4–5. The frequency of this anomaly is mostly in males, however, few cases in females (women and girls) have also been reported [3]. In our case, an adult male patient presented with dysuria. The definite mechanism of its development is not well understood yet, however, various theories exist regarding the embryogenesis of the urethral duplication [4,5]. The most accepted theory is proposed by Patten and Barry as faulty communication

between lateral folds of the genital tubercle and ventral part of the cloacal membrane [4]. But this theory does not cover other types of duplications [5]. Often, urethral duplications are accidentally discovered in newborns while performing a physical examination of the external genitalia and such patients are asymptomatic. In contrast, symptomatic patients, most commonly present with recurrent urinary tract infection (UTI), double urinary stream, dysuria, and urinary incontinence. Usually, the age of diagnosis of urethral duplication is before 12 months of age [2]. Our patient was a 24-year-old male presenting only with dysuria. The upper urinary tract anomalies are rarely associated with urethral duplication [2]. Various classifications are present for urethral duplications, however, Effman et al. suggested a classification that has been used widely. In this classification, urethral duplications are subdivided into 3 major types (I, II, III) as follows [3,4,6–8]: Incomplete duplication of the urethra with a blind end is classified as type I (IA as distal; most common type [11], IB as proximal). Complete duplication of the urethra is referred to as type II AI specified by its patent second conduit which arises from the urinary bladder [3], and it is considered the most common type reported in the literature [2]. Type IIAII is characterized as a patent second conduit arising from the first urethra running through the second meatus. Type AII (Y type) is when a patent second conduit arises from the first urethra and ends up in the perineum. If a second conduit originates from the urinary bladder or a posterior urethra and subsequently joins the first urethra and ends up as a single meatus, thus, this type is recognized as type IIB [3] Our patient had the type IIB urethral duplication. In type II, functional urethra merges with perineum where the dorsal one usually remains hypoplastic. There are some cases in which orthotopic urethra is functional [4]. The urinary bladder, urethral and penile duplications are accounted as type III [3]. Type of the anomaly, clinical symptoms, and form of severity are the deciding factors to determine the surgical treatment of urethral duplications [1]. Patients with urethral duplication are mostly diagnosed before adolescence [12], while in our case the patient is adult. In all types of urethral duplication, retrograde urethrography is considered the initial ideal radiologic examination [13]. The main procedures used in the diagnosis of the double urethra can be MCUG; retrograde urethrography; intravenous urography; ultrasonography (USG); and magnetic resonance imaging [7]. Usually, urethral duplication is diagnosed by MCUG indicating 2 different conduits [4]. In cases where MCUG fails to show accessory urethra, then retrograde urethrography is performed [14]. In epispadias, intravenous urography would show widened pubic symphysis. For the evaluation of double urethra and surrounding soft tissues, magnetic resonance imaging is considered the winning choice. In case imaging studies fail to reveal desired results then urethro-cystoscopy is performed [9]. In our patient, retrograde urethrography was performed with additional MCUG and revealed the diagnosis. IIB type urethral duplication is a rare type of urethral duplication [14] and our case adds to it. Surgical treatment should be planned based on the anatomical structure of the duplication [8, 11]. No treatment is suggested by Salle et al. for types IB and IIB [9].

Overall, urethral duplications are rare congenital urethral anomalies, and type IIB makes a rarer subtype with additional late presentation. Further studies need to be carried out for the surgical management of urethral duplications, more specifically for IIB type.

Declarations

Ethics approval and consent to participate

The manuscript has got an ethical review exemption from the Ethical Review Committee of the authors' institution (French medical institute for Mothers and Children- {FMIC}) as case reports are exempted from review according to the institutional ethical review committee's policy. Written consent is obtained from the participants for publishing the case.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

Availability of data and materials

Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study (as this is a case report).

Authors' contributions

All of the authors have participated sufficiently in the submission and take public responsibility for its content. NF: writing and editing the manuscript, selecting the images, and corresponding with the journal. MJK: Selecting the case and revising the manuscript. All of the authors have read and approved the final manuscript.

REFERENCES

[1] Roshanzamir F, Mirshemirani A, Ghoroubi J, Mahdavi A, Mohajerzadeh L, Sarafi M. Complete urethral duplication in children: a case report. Iranian J Pediatr 2016;26(2):1–4.
[2] Bakkaly AE, Lafia T, Bouljrouf J, et al. Urethral duplication in male children's: report of four cases. Open Access J Trans Med Res 2017;1(4):106–9. doi:10.15406/oajtmr.2017.01.00022.
[3] Baid M, Dutta A. Urethral duplication in a 15-year-old: case report with review of the literature. Rev Urol 2014;16(3):149.
[4] De Carvalho BC, Talini C, Antunes LA, Camargo MH, Del Valle P, Junior AA. Our experience on urethral duplication in children: 4 case reports and literature review. Pediatr Urol Case Rep 2017;4(5):378–82.
[5] Ramareddy RS, Alladi A, Siddappa OS. Urethral duplication: experience of four cases. J Indian Assoc Pediatr Surg 2012;17(3):111.
[6] Salle JL, Sibai H, Rosenstein D, Brzezinski AE, Corcos J. Urethral duplication in the male: review of 16 cases. J Urol 2000;163(6):1936–40.

[7] Cicek T, Kizilkan Y, Coban G, Kosan M. Urethral duplication: a rare cause of recurrent urinary infection. J Pak Med Assoc 2015;65(1):90–2.
[8] Mori K, Shin T, Tobu S, Noguchi M, Sumino Y, Sato F. A case of urethral duplication arising from the posterior urethra to the scrotum with urinary stone in a 6-year-old male. Case Rep Pediatr 2014;2014.
[9] Maitama HY, Mbibu HN, Tella UM. Urethral duplication: case report and literature review. Ann Afr Med 2012;11(3):186.
[10] Gozar H, Prișcă RA, Derzsi Z. Two cases of dorsal urethral duplication. Acta Medica Marisiensis 2013;59(1):52–4.
[11] Davis NF, O'Connor KM, Quinlan DM. Incomplete urethral duplication in an adult male. Ir J Med Sci 2012;181(3):345–7.
[12] Chen CH, Chen JH, Hsieh JT. Epispadiac urethral duplication in an adult dorsal deviated penis. Urology 2008;71(2):352–3e1.
[13] Frankel J, Sukov R. Diagnosing urethral duplication including a novel radiological diagnostic algorithm. BJR| Case Rep 2017 20150506.
[14] Coleman RA, Winkle DC, Borzi PA. Urethral duplication: cases of ventral and dorsal complete duplication and review of the literature. J Pediatr Urol 2010;6(2):188–91.