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Case Report

Type IIB urethral duplication in adult: A case report☆,☆☆

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ABSTRACT

Urethral duplication are rare anomalies of the lower urinary tract consisting of partial or complete development of an accessory urethra that common in males than female. The exact embryological mechanism for this condition remain unclear, it is thought to relate to a disruption of development of the lateral folds of Rathke during hindgut development. There are 3 types based on Effman's classification. Diagnosis of urethral duplication, a genital examination has to performed and confirmed by micturating cytourethrography and retrograde urethrography. The authors presented a case of 67-year-old man complaining of dysuria, who was diagnosed with urethral duplication Effman Classification type IIB accompanied by bulbar urethral stricture and diverticula and multiple bladder diverticula 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.

Introduction

Urethral duplication is rare anomalies of the lower urinary tract consisting of partial or complete development of an accessory urethra [1]. It is more common in males, occurring usually in the sagittal plane. In females, the anomaly is rare and most often associated with bladder duplication [2]. Urethral duplication are reported 300 case in the literature. Although the exact embryological mechanism for this condition remain unclear, it is thought to relate to a disruption of development of the lateral folds of Rathke during hindgut development [3]. There is diversity of clinical manifestations, diagnosis is difficult as well as its classification. Patients can be either asymptomatic or symptomatic, most common clinical findings being incontinence, obstruction, recurrent urinary infection and double urinary stream [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

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that bring a therapeutic challenge for urologic surgeon. Selection of surgical treatment depend on symptoms and the anatomy of urethra and the urinary bladder neck [4].

Case presentation

A 67-year-old man came to our institution with complaints of dysuria since about 1 week ago. The urine is yellow, not accompanied by blood and the patient does not have a fever. The patient had undergone urethral surgery because of previous urethral stricture followed by cystostomy procedure and urinary catheter placement since approximately 3 months ago. About 1 week ago the urinary catheter was removed and the patient complained of dysuria. The patient had a history of recurrent urinary tract infections approximately 1 year ago. The patient had no abnormal findings during the physical examination.

Retrograde urethrography was performed, 50 cc of contrast is inserted through the external urethral orifice using Foley catheter. Contrast filling bulbar urethral, membranous urethral and prostatic urethral. There was a narrowing of the lumen in bulbar urethral accompanied by diverticula in bulbar urethral. Contrast appears to fill the bladder through the bifurcated urethra with a separate opening to the bladder.

Then a voiding cystourethrography with water soluble contrast was carried out by infusion of ± 340 cc by drip into the urinary bladder through Foley catheter. On the appearance of the urinary bladder is fully filled and no vesicoureteric reflux is seen. There are multiple additional shadows, firm boundaries, regular edges, on the bladder wall. At the time of urination, the bladder neck appears open, the contrast exits through the urethra. Two separate proximal urethra were found that came out of the urinary bladder and merged distally, classified as type IIB according to Effman Classification. Figures 1 and 2.

The treatment planning for urethral duplication itself must be individualized for each patient depending on the type of deformity, the severity of the symptoms and other associated anomalies. In this case, after the patient was diagnosed with urethral duplication Effman Classification type IIB accompanied by stricture and bulbar urethral diverticula and multiple bladder diverticula, the patient was planned to undergo perineostomy which was indicated because of his urethral stricture.

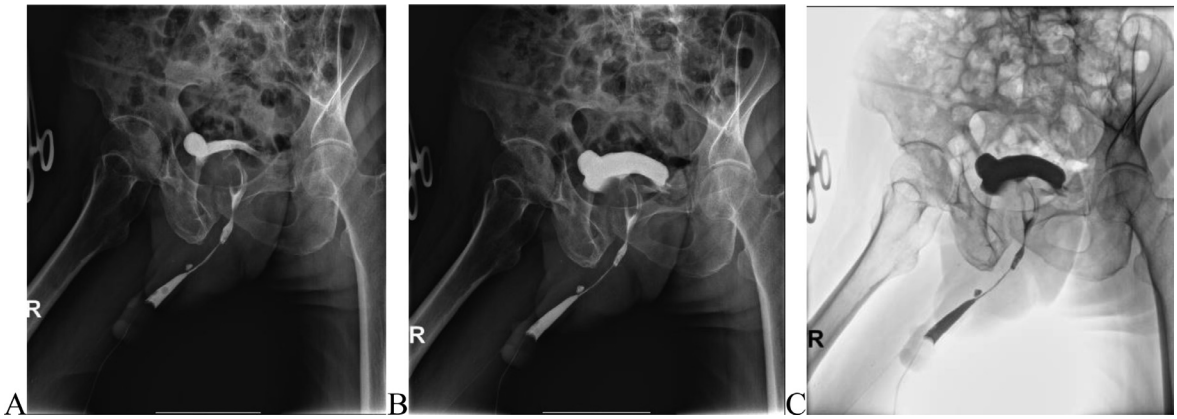

Fig. 1(A–C) – Retrograde urethrogram (positive and negative fluoroscopic images). There was bulbar urethral stricture accompanied by bulbar urethral diverticle. Contrast appears to fill the bladder through the bifurcated urethra with a separate opening to the bladder.

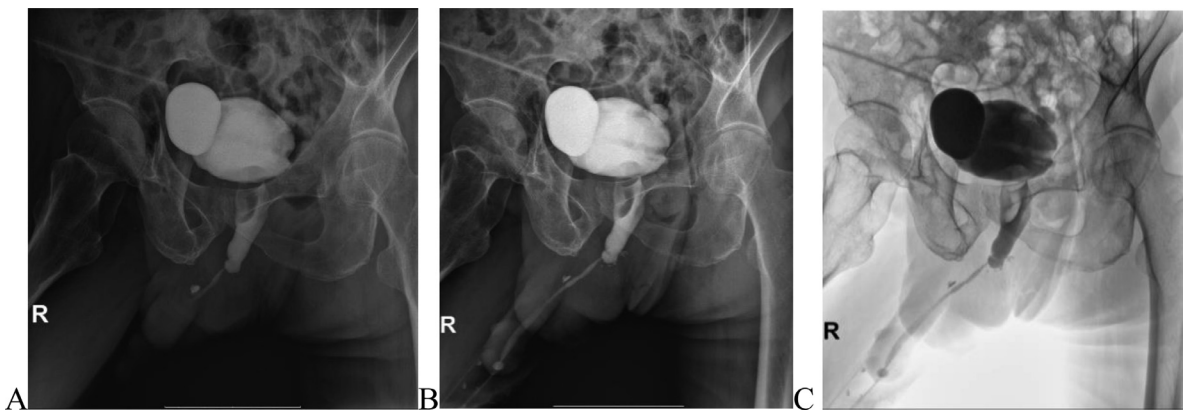

Fig. 2(A–C) – Voiding cystourethrogram (positive and negative fluoroscopic images). There are multiple diverticle on the bladder wall. Two separate proximal urethra were found that came out of the urinary bladder and merged distally.

Discussion

Urethral duplication is a rare congenital abnormality. Embryogenesis is not well understood and various hypotheses exist, but no one can explain all type of presentation, the common pathological process is supposed to result from an abnormal relationship between the lateral folds of the genital tubercle and the central end of the cloacal membrane and the duplication commonly occurs in the sagittal plane with 1 urethra located centrally and the other dorsally [5]. The frequency of this anomaly is mostly in males, and few cases in females [4]. Being congenital, mostly the diagnosis was established in childhood or adolescence rarely in adult age. In this case, adult male patient was coming with dysuria. The urethral duplication has multiple presentation, with a lack of specificity going from the asymptomatic, dysuria, deformed penis, twin streams, urinary tract infection, symptoms of bladder outlet obstruction to various other signs like a renal failure as one of the worst [5]. In this case, chief complain of patient is dysuria after catheter removal.

Effmann et al, classified urethral duplication into 3 types. Type I: Blind-ending accessory urethra (incomplete urethral duplication) IA. Distal-duplicated urethras opening on the dorsal or ventral surface of the penis but not communicating with the urethra or bladder (the most common type) IB. Proximal-accessory urethra opening from the urethral channel but ending blindly in the periurethral tissues (rare). Type II: Completely patent accessory urethra. It is divided into 2 parts: A (2 meatuses) and B (1 meatus) IIA1:Two noncommunicating urethras arising independently from the bladder IIA2 : Second channel arising from the first and coursing independently into a second meatus (Y-type) IIB : Two urethras arising from the bladder or posterior urethra and uniting into a common channel distally. Type III: Accessory urethras arising from duplicated or septated bladders [6].

Radiological investigation is mandatory to establish a diagnosis, identify the type of the anomaly and rule out associated with other anomalies [7]. In this case, after the patient carried out a supporting examination such as voiding cystourethrography, it was found 2 proximal urethra that came out of the urinary bladder and merged in the distal body which was suggestive of urethral duplication based on Effman classification type IIB.

The treatment of urethral duplication should be individualized for each patient according to the type of UD and the clinical presentation. Higher-grade types usually require complex multiple surgeries, while low-grade incomplete UD may remain untreated.

Conclusion

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

Patient consent

Written informed consent for publication of their case was obtained from our patient.

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