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Case Report

Congenital Effman type 2A II urethral duplication with hypoplastic ventral glandular urethra

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ABSTRACT

Urethral duplication is a rare congenital abnormality with varied clinical manifestations; less than 300 cases were reported in the literature. Urethral duplication (UD) commonly seen in male than female and is a very rare anomaly. Urethral duplication can present with double urinary systems, urinary tract infection and lower urinary tract systems. Retrograde urethrogram will be helpful in diagnosis. Treatment depends upon clinical presentation but there is lack of consensus on best managment. We present images of 6-year-old child presented with two streams due to congenital Effman type 2A II urethral duplication.

Keywords: Urethral duplication, Double urinary stream, Dorsal accessory channel

INTRODUCTION

Urethral duplication was first described by Aristotle and Vesalius. It is rare congenital anomaly and is mainly seen in children and rarely in adulthood. Urethral duplication is more commonly seen in males than females. Effmann et al. described classification of urethral duplication into three types and are currently mostly reported classification in the literature.[1,5] Numerous hypotheses have been put forward; however, there is ambiguity in regard to its embryology as the same explanation does not apply to all subdivisions of urethral duplication.

Presentation of urethral duplication can be deformed penis, twin streams, urinary tract infection (UTI), urinary incontinence, urethral discharge and asymptomatic and differs according to the anatomical variant of duplication. It is rarely associated with other congenital anomalies. The diagnosis of urethral duplication can be done by voiding cysto-urethrography (VCUG). Management of urethral duplication depends on the clinical presentation and type of duplication. If surgery is necessary, functional urethra should be preserved.[2,6-8]

CASE REPORT

A 6-year-old boy presented with double urinary stream since birth without any associated urological symptoms. On genital examination he had uncircumcised penis without chordee with two urethral meatus. One was normal in size representing accessory dorsal channel in mid glans and other was orthtopic but stenotic, located ventral to dorsal meatus. His main urine flow was through dorsal accessory channel and weak stream through hypoplastic ventral glandular urethra (Figure 1a and b). His basic biochemical evaluation and ultrasonography was normal with insignificant post void residue. Cannulation through normal ventral urethra was not possible due to pin point meatus, hence retrograde urethrogram (RUG) through dorsal urethra shows patent accessory dorsal urethra and retrogradly filled dilated ventral urethra up to glans area (Figure 1c).

Non-contrast pelvic magnetic resonance imaging (MRI) shows accessory dorsal and dilated ventral urethra with bifurcation in sphincteric zone. Diameter of ventral urethra in glans area is 1.79 mm. and distance between dorsal and ventral urethra in glans area is 2.23 mm (Figure 2a-d).

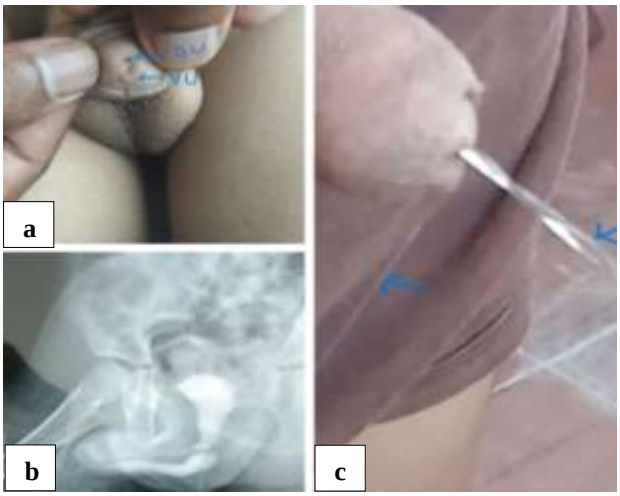

Figure 1: (a) Clinical picture showing opening of dorsal urethra (DV) and ventral urethra (VU) with stenotic opening, (b) clinical picture showing two streams of urine, and (c) retrograde urethrogram showing narrowed patent dorsal urethra and retrogradly filled dilated ventral urethra up to glans area.

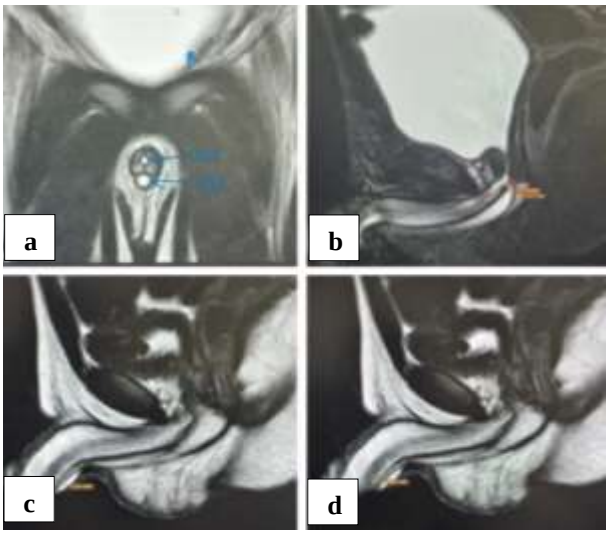

Figure 2: (a) Coronal view of T2 weighted images of MRI showing B- bladder, DU-dorsal urethra, and VU-dilated ventral urethra; (b) mid sagittal view of T2 weighted MRI image showing bifurcation of urethra in sphincteric zone; (c) mid sagittal view of T2 weighted MRI image showing diameter of ventral urethra in glans area is 1.79 mm; and (d) mid sagittal view of T2 weighted MRI image showing distance between dorsal and ventral urethra in glans area is 2.23 mm.

DISCUSSION

Urethral duplication (UD) usually seen in male which is a very rare anomaly and reported only in less than 300 cases, often associated with genitourinary and gastrointestinal anomalies. Embryogenic development of UD is not well understood and various hypotheses exist, but none can explain all types of presentations.[1,9]

Congenital UD can have different clinical manifestations, such as deformed penis, double urinary streams, urinary tract infection, urethral discharge, and out-flow obstruction or can be asymptomatic. In most previously reported cases, the accessory urethra ends most commonly with an epispadic opening but can also end in mid glans and continence of it is maintained due to intact sphincteric function.[2,3,6]

Radiologic evaluation is mandatory to establish a diagnosis, define anatomy, type, complexity of the UD and planning of the surgical approach. Retrograde urethrogram is required for better visualization of the two urethras but the advent of MRI has provided a powerful tool to study these anomalies in depth and on multiple planes.[3,4]

Cysto-urethroscopy may provide a benefit when performed before surgery to identify the functional urethra and find the opening of the accessory urethra.[3]

Goals of management of UD are to preserve the continence and physiological micturition. Surgical intervention is not an emergency as long as proper bladder drainage is ensured.[2]

The treatment of UD should be individualized as there is lack of consensus on the best surgical treatment. Treatment plan is based on type of urethral duplication and the clinical presentation. Higher-grade types usually require complex multiple surgeries, while low-grade incomplete UD may remain untreated.[3,4,6,7]

CONCLUSION

Reporting of every case will help in raising awareness about various type, diagnostic work-up, planning of complex reconstruction and will help in forming future guidelines for management of UD which are still lacking.

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