CASE REPORT

Hypospadiac Duplication of Anterior Urethra—a Rare Congenital Anomaly

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Abstract Duplication of the urethra is a complex and rarely seen congenital anomaly with three anatomic variants: epispadiac (dorsal), hypospadiac (ventral), and Y-type. We report here a case of hypospadiac duplication of anterior urethra with dorsal blind ending urethra in a 9-year-old boy who presented with complaint of passing urine from the ventral aspect of penis.

Keywords Hypospadiasis · Urethral duplication · Ventral aspect · MIP—megameatus with Intact prepuce · Pyramid procedure

Urethral duplication is an extremely rare, yet very well-defined, congenital malformation, with about 200 cases described in the specialized literature [1]. This anomaly is most commonly seen in males with few cases reported in females Figs. 1 and 2.

Case Report

We report here a case of 9-year-old boy who presented with chief complaints of passage of urine from the opening at the ventral aspect of penis since birth. Physical examination revealed a normal healthy boy with no other associated abnormality. Local examination shows a dorsal urethral meatus

situated in proximal glans ending blindly at 3 cm. Another urethral meatus situated on the ventral aspect which was wide, covered with semi lunar fold of penile skin (MIP variant) and which was communicating with the urinary bladder. On retrograde cystourethrography, the dorsally situated meatus showed approximately 3 cm long blind ending tract. While through the opening at the ventral aspect, the contrast has passed and opacified the anterior and posterior urethra and subsequently passed freely into the urinary bladder. No communication between these two opacified tracts seen. The diagnosis of hypospadiac duplication of anterior urethra (type 1A according to Effman et al. classification) with dorsal blind ending urethra with megameatus intact prepuce (MIP) of the communicating ventral urethra was made. The patient then underwent crushing and division of common septum between two urethras with urethroplasty by pyramid procedure.

Discussion

Duplication of urethra presents with three anatomic variants: epispadiac (dorsal), hypospadiac (ventral) and Y-type [2]. Most duplications lie in the same sagittal plane one above the other. Less commonly, the openings of the duplication lies laterally in the coronal plane [3]. The most widely accepted classification of the different types of urethral duplication was developed by Effman et al. [4]. This classification is the most functional representing all clinical aspects of urethral duplication.

Type 1: Blind incomplete urethral duplication

1a (distal): opens at the distal or ventral surface of the penis but does not communicate with the urethra or bladder (most common).

1b (proximal): opens from urethral channel and ends blindly in the periurethral tissue (very rare)

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Fig. 1 RGU showing opacification of the dorsally placed blind ending tract and opacification of ventrally placed tract which is communicating with urinary bladder

Type 2: Complete patent urethral duplication

2A1: two non-communicating urethras arising independently from the bladder or coursing independently to two different meati

2A2: a second channel arising from the first the first and coursing independently to the second meatus

2B (one meatus): two urethras arising from the bladder or posterior urethra and uniting to form a common distal channel

Fig. 2 Showing double meatus after catheterization

Type 3: Urethral duplication as a component of a partial or complete caudal duplication.

Many theories have been proposed to describe the embryology of this condition including an irregularity of the ingrowth of the lateral mesoderm of the cloacal [5] or abnormal termination of the Mullerian ducts [6]. In spite of various theories proposed, no single theory explains all the various types of anomalies.

The clinical significance of this condition is variable. Patients may be asymptomatic or may present with double urinary stream. Other presentations include urinary incontinence, recurrent UTI, or outflow obstruction. The treatment depends upon the type of duplication and clinical presentation. Surgery should be considered in patients with disturbing symptoms or correction of cosmetic deformity (as in our case).

According to Effman et al. classification, our case belongs to type 1A. The patient underwent crushing and division of common septum between two urethras with urethroplasty by pyramid procedure.

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