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Duplication of the Urethra in Boys: A Case Report

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Abstract

Supernumerary urethra in boys is a very rare anatomical entity. Sagittal urethral duplications are classified into four groups: epispadias, hypospadias, fusiform and Y-shaped urethra. The most widely used classification is that of Effmann and Lebowitz, which describes 6 types, one of the rarest being the 'Y' subtype IIA2, which corresponds to a duplicated urethral path from the vesical neck to an ectopic perineal or anal outlet. We report here the case of a 4-year-old child presenting with subtype IIA2 with a perineal orifice and no other urinary disorders.

Keywords

Duplicity, Ureter, Malformation

1. Introduction

Urethral duplication (UD), also known as supernumerary urethra (SU), is defined by the juxtaposition of two ducts with a smooth muscular structure and an excreto-urinary mucosal lining [1] [2] [3] [4]. In reality, there is an accessory urethra and a main urethra (**Figure 1**), in both the sagittal and frontal planes. In the sagittal plane, depending on whether the external meatus of the accessory urethra terminates above or below the external orifice of the main urethra, we speak of UD epispade or hypospadias. It is a rare congenital malformation, most often asymptomatic [1] [2] [3] [4] [5]. In symptomatic forms of UD, whether complete or incomplete, the clinical symptomatology is varied, and the most

commonly reported signs are: a double urinary stream, urinary incontinence, recurrent urinary infections, and penis curvature, to which must be added urogenital and gastrointestinal malformations [1] [2] [3] [4] [5]. To date, many theories have been put forward in support of embryopathological mechanisms, but many unknowns remain [1] [2] [3] [4] [5].

The aim of this paper was to present the first case of UD in our institution, and to discuss the diagnostic approach and management.

2. Observation

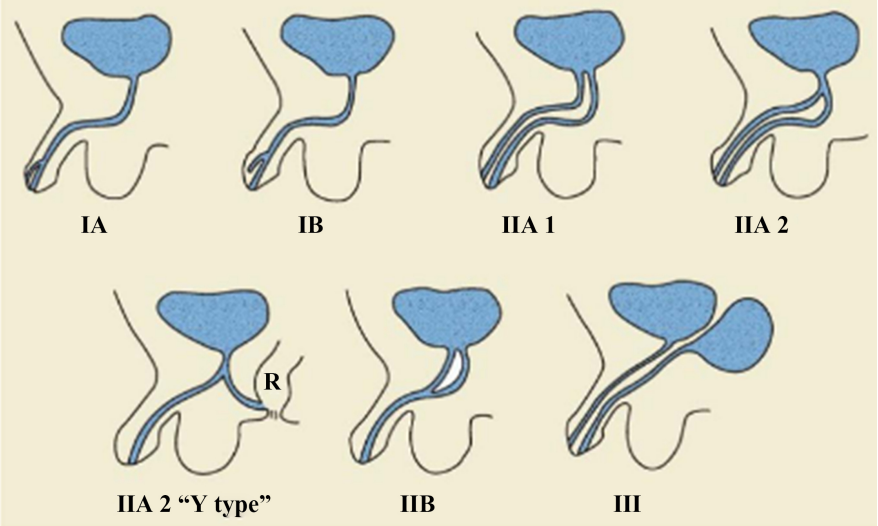

Figure 1. Urethral duplicity classification according to Effman.

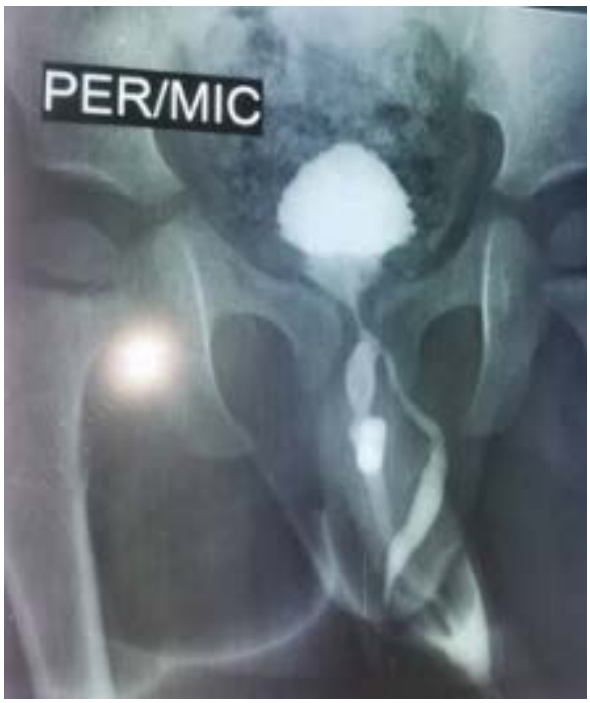

Figure 2. UCRM urethral duplicity.

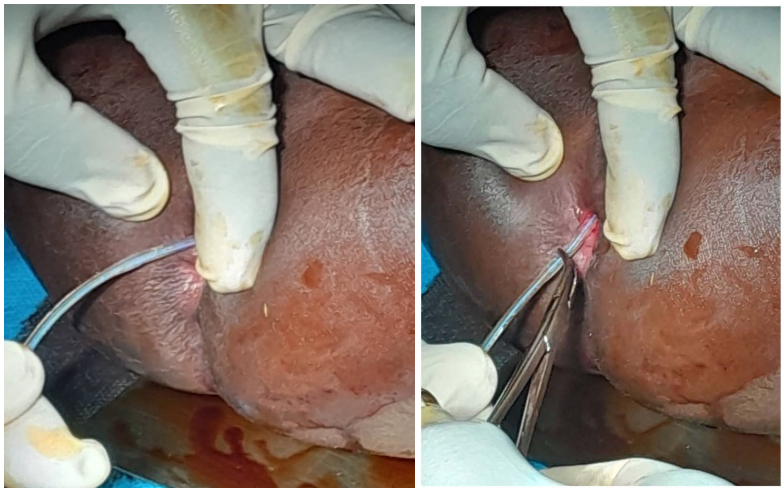

Figure 3. (a) Catheterization of the supernumerary urethra; (b) catheterization with a tube-assisted intubation probe.

Figure 4. Normal urethral catheterization.

M.S. was a 4-year-old male child who consulted his parents accompanied by his parents for a double micturition stream. His history revealed neither trauma to the penis nor circumcision. The parents attested to the existence of an erection without curvature of the penis. He had a history of urine leakage during micturition and post-micturition. Clinical examination revealed a perineal disruption initially suggestive of a urethral fistula. A retrograde and micturition uretrocystography (**Figure 2**) was performed after catheterization of the perineal orifice, confirming the presence of a well-systematized tract (**Figure 3(a)**, **Figure 3(b)**). Surgical exploration consisted of excision of the duplicated urethra after catheterization with a hydrophilic guide to its abutment at prostatic level (**Figure 4**, **Figure 5**). Study of the surgical specimen confirmed the histological nature of a supernumerary urethra. Retrograde and voiding urethrocystography performed after surgery was normal (**Figure 6**).

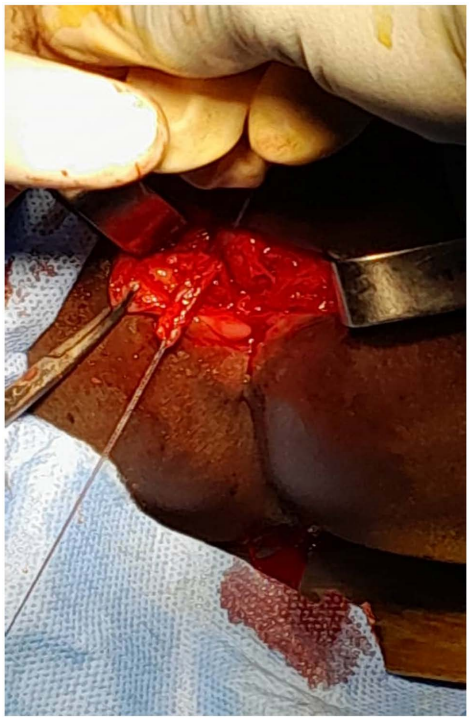

Figure 5. Dissected supernumerary urethra.

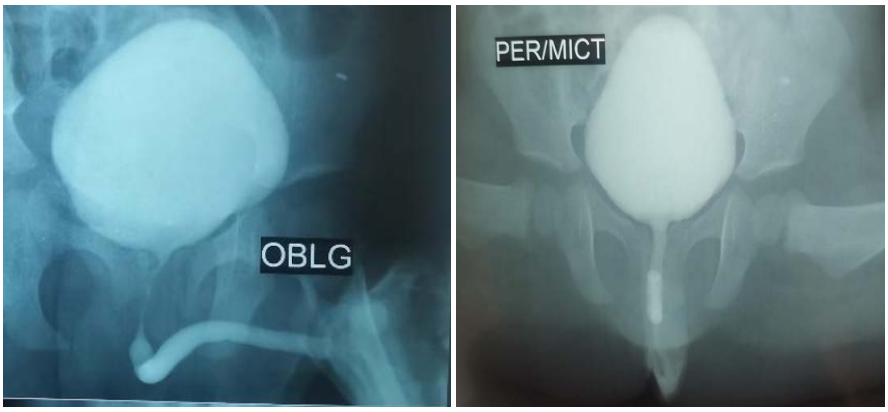

Figure 6. Retrograde uretrocystography control.

3. Discussion

UD is a rare congenital malformation. It mainly affects males, although cases have been reported in girls [2]. As of 2008, fewer than 500 cases had been reported in the literature [1] [3]. Many theories have been put forward to explain the embryopathological mechanisms of UD, but there are still many grey areas due to the multiplicity of anatomical variations [1] [2] [3] [4] [5]. Some authors have referred to the delayed formation of the balanic lamina in relation to the portion of the urethra originating from the urogenital sinus, which first reaches the dorsal part of the genital tubercle [3]; Mollard, quoted by Nsir et al. [3], has argued that this anomaly results from an embryological disturbance identical to that of bladder exstrophy and true epispadias. Other authors, in particular Wil-

liams and Kenawi, have blamed an anomaly in the median fusion of lateral mesoblastic flows at the cloacal membrane [3]. Anatomically, the UD has two urethras, one main and one accessory. It can be sagittal, and depending on whether the external orifice of the accessory urethra opens above or below the external orifice of the main urethra (whose jet is much stronger), a distinction is made between epispade and hypospadias UD. It can also be collateral or even posterior [4].

The case reported here is an example of a Y-shaped UD. Numerous classifications of this malformation have been published, the most widely used being that of EFFMAN (1976) [2]. For Effman, group I corresponds to incomplete urethral duplications:

- Type IA: distal blind accessory urethra with accessory meatus on the dorsal or ventral surface of the penis. No communication with the urinary tract;

- Type IB: blind accessory urethra, proximal in nature, leading into the main urethra and terminating intra-tissularly.

Group II corresponds to complete urethral duplications:

- Type IIA1: two independent urethras arising from the bladder;

- Type IIA2: one urethra starting from the bladder and splitting downstream;

- Type IIB1: two urethras originating in the bladder and merging downstream to form a single meatus termination;

- Type IIB2: urethra split over a given distance.

Retrograde and micturition uretrocystography was used to diagnose urethral duplicity in our case. Urethrocystoscopy may be useful in the case of group II anomalies to search for a second intravesical meatus [1], but is of little interest in group I cases. It remains, however, an easily accessible examination in the office. Imaging plays an important role in the assessment of this pathology, to define its type, look for possible complications and associated malformations, and guide the therapeutic procedure.

- Suprapubic cystography with voiding films, coupled with retrograde urethrography, can confirm the anomaly, analyze its anatomical configuration and, above all, look for communication with the urinary tract.

- Ultrasound scans of the kidney, bladder and prostate region are useful for detecting associated malformations and describing the structures surrounding the urethra.

Today, we can suggest replacing them with an MRI examination. Not available in our country (Mali). This type of imaging enables a much more precise study of the penis, perineum, prostate region and lower urinary tract in a single examination, which has the advantage of not requiring intravenous injection of contrast medium, and of presenting objective imagery that can be used as a reference for subsequent examinations [6] [7].

UD is a malformation that should be diagnosed early in life. The clinic was dominated by a double urine stream in our case. Most cases are asymptomatic, discovered by chance and require no treatment [1] [2] [3] [4] [5]. Symptomatic

forms, on the other hand, are managed surgically or endoscopically [1] [2] [3] [4] [5], and most often manifest as a double stream of urine, urinary incontinence, recurrent urinary tract infection and penile crush. Exceptionally, urethritis, orchi-epididymitis and prostatitis have been reported [1] [3]. Treatment of urethral duplications is the subject of a consensus [1] [2] [6] [8]: only symptomatic duplications should be treated surgically. The variability of anatomical forms conditions the procedure itself, and the choice should only be made on a case-by-case basis after a complete morphological assessment. The treatment of these urethral duplications is the subject of a consensus [1] [2] [6] [8].

In our patient's case, the demand for treatment was high because of urethral discharge. In our case, excision of the supernumerary urethra was more logical than outright ligation, which risks causing possibly superinfected retention in the proximal accessory urethra. At one month, the patient complained of no symptoms. The child is monitored for one month, six months and one year. Physical checks are carried out at one month, 6 months and one year.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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