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Urinary Lithiasis Secondary to Urethral Duplication: A Case Report

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Abstract

Urethral duplication is a rare congenital anomaly, mainly involving boys, although cases in girls have been reported. The majority of duplications of the urethra are asymptomatic and the discovery of this malformation can be done at any age. Diagnosis and determination of its type are based on urethrocystography with mid-void images, which helps to guide the surgical approach. The treatment is not yet well codified and the therapeutic attitude varies from one author to another. We report a clinical observation of a urethral duplication with a calculus in a six-year-old boy who underwent a partial urethrectomy of the supernumerary urethra in whom a lithotomy and a urethrectomy were performed via a suspension approach. This observation illustrates the possibility of urinary stone formation after partial urethrectomy of the supernumerary urethra.

Keywords

Urethra, Duplication, Calculus, Child

1. Introduction

Urethral duplication is a rare congenital malformation [1] characterized by the juxtaposition of two or more ducts with a smooth muscular structure and an excreto-urinary mucosal lining [2] [3]. It may be complete or incomplete and usually occurs in a sagittal plane [4], and the ventral urethra is most often functional and contains a sphincter mechanism [5]. Most urethral duplications are asymptomatic, with mild episodes of urinary incontinence in the most frequent cases [6], and can be discovered at any age. Diagnosis is made after careful ex-

amination. Treatment is not yet well codified, and therapeutic attitudes vary from one author to another. Asymptomatic forms are generally respected [7].

We report a clinical observation of a urethral duplication with a calculus embedded in the supernumerary urethra in a six-year-old boy who underwent partial urethrectomy of the supernumerary urethra and circumcision at a hospital in the Labé region.

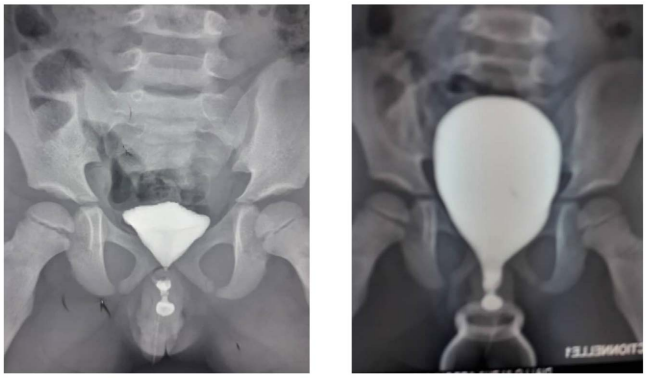
2. Observation

6-year-old male boy who consulted for suspensory pain and urinary leakage evolving for 14 days with the history of a circumcision and an excision of the urethra epispade complete February 08, 2018 in a hospital structure in the region of Labé.

The clinical examination found a good general condition, well colored, afebrile with a temperature of 37.4˚C. Locally, penis circumcised, presence of a glandular scar on the dorsal surface, the meatus is apical, there is a small mass at the root of the penis, of firm circumstance, well circumscribed and mobile in relation to the deep plane, urination possible with slight pain. The testicles in place are normal in size and appearance.

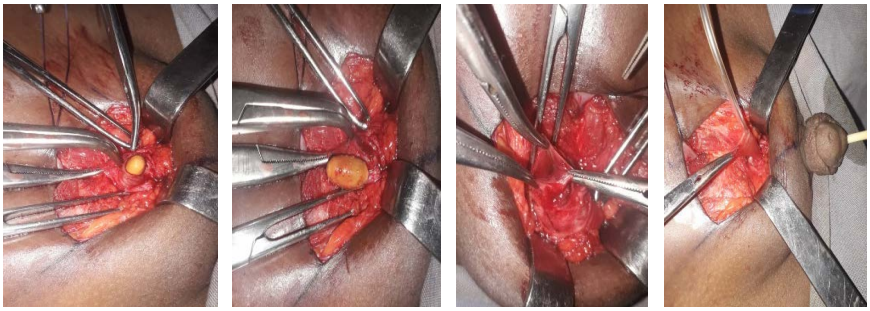
The cytobacteriological examination of the urine carried out was negative, then the retrograde and voiding urethrocystography (UCRM) revealed a stone wedged in the supernumerary urethra (**Figure 1(a)** and **Figure 1(b)**) and finally an ultrasound carried out to suggest the presence of a lithiasis at the uretho-vesical junction.

The diagnosis of a lithiasis on a urethral duplication was retained and the indication of a lithotomy and a uretrectomy were posed and carried out under general anesthesia by a suprapubic approach by a pfanenstielle incision, dissection and individualization of the mass under the pubic symphysis, dissection and identification of the bifid urethra followed by its opening which allowed us to extract a large stone (**Figure 2(a)** and **Figure 2(b)**), let's continue the dissection of the bifid urethra (**Figure 2(c)** and **Figure 2(d)**) on a length of 3.5 cm to the base followed by its ligation and resection with PDS 4/0. Closing of the different planes and the urethral probe kept for 24 hours.



(a) (b)

Figure 1. UCRM image showing duplicity and enclave calculus.



(a) (b) (c) (d)

Figure 2. (a) and (b): Demonstration of the calculus in the supernumerary urethra, (c) and (d): Supernumerary urethra and urethral sound.

The immediate post-operative follow-up was simple with removal of the probe at J1 and the exit was J3.

3. Discussion

Urethral duplication or accessory urethra is a rare congenital anomaly mainly affecting boys, although cases in girls have been reported [1]. In the literature worldwide less than 500 cases have been reported [6] [7]. It is defined by the juxtaposition of two or more canals with a smooth muscular structure with an excreto-urinary mucosal coating [2]. The age of discovery is early most often before the age of 1 year [8]. The age of discovery in our patient is 18 months. This malformation results from a disorder of the organogenesis of the penis. Normally, the urethra develops from the urogenital sinus whose pelvic segment will form the posterior urethra and the phallic segment of the anterior urethra [9].

A number of hypotheses have been put forward to explain the embryopathogenesis of urethral duplication, but none of these alone can explain all the anatomical forms of this malformation. Some authors have suggested a delay in the 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; for Mollard, cited by several authors, for Mollard, cited by several authors, this anomaly results from an embryological disturbance identical to that of bladder exstrophy and true epispadias and, finally, for Williams and Kenawi, an anomaly of median fusion of lateral mesoblastic flows at the level of the cloacal membrane [4] [10] [11].

Several classifications have been proposed, but two are widely used: that of Williams and Kenawi and that of Effmann-Lebowitz. The Effmann classification has been widely adopted to classify the different types of urethral duplication. It is considered the most comprehensive classification from a clinical and functional point of view, but is based on male forms only and does not distinguish sagittal from coronal duplications [6] [10].

Type I: Incomplete urethral opening

- Type IA: Opening on dorsal or ventral surface of penis without communication with urethra or bladder.

- Type IB: Proximal communication with the urethra, but no opening on the surface of the penis.

Type II: complete duplication of the urethra

- Type IIA1: two totally independent urethras arising separately from the bladder.
- Type IIA2: the accessory urethra arises from the main urethra and runs independently to its own meatus.
- Type IIA2-Y: a special form of type IIA2 with a ventral urethra opening into the perineum, known as congenital posterior urethroperineal fistula (PPUF).
- Type IIB: two urethras arise separately from the bladder and join distally to open into a single meatus.

Type III: Urethral duplication associated with a caudal duplication.

In our patient, it was a complete sagittal epispade duplication with two urethral orifices (IIA2), the accessory urethra was located above and anterior to the normal bladder neck. It passed anteriorly to the normal urethra, behind the pubic symphysis and terminated at the dorsal surface of the penis. After partial resection of the supernumerary urethra, it was transformed into a blind epispade duplication with a blind canal that terminates at the dorsal surface of the penis. This canal emerges from the bladder and terminates anterior to the symphysis, with no external connection. It should be noted that the presence of anatomical abnormalities can favour the appearance of a calculus, by impairing the flow of urine. They may be responsible for urinary tract infections, leading to the development of lithiasis. These anomalies promote lithogenesis through urinary stasis in a diverticulum, as in our patient's case, and allow microbial proliferation and crystallization [12].

The circumstances of discovery are variable and depend on the anatomical type of the duplication. Generally, the supernumerary urethra is asymptomatic; the most frequently reported clinical signs are represented by double urinary stream, urinary incontinence, recurrent urinary tract infections and curvature of the penis. However, fortuitous discovery represents a circumstance of discovery frequently reported in the literature, mainly concerning blind forms [6] [13].

Complementary investigations are essential for the diagnosis of the anatomical form of the accessory urethra. Retrograde urethrocystography with per mictional films (UCRM) is the diagnostic key, and was performed in our patient to confirm the diagnosis. UCRM is the complementary examination of choice, enabling a morphological study of the main urethra and possibly the accessory urethra [8]. Endoscopy represents a diagnostic complement to better appreciate the anatomical type and helps guide the therapeutic attitude [1], but may miss the orifice of the supernumerary urethra [14]. It should be noted that UCRM coupled with endoscopy enables a better appreciation of the anatomical aspect of urethral duplication [15]. It should not be forgotten that an unprepared abdominal X-ray can reveal any associated bony malformations, such as the pubic disjunction seen in these cases of episcleral duplication, and can also show the presence of a calculus in the lower urinary tract, as was the case in our patient.

Ultrasound examinations of the kidneys, bladder and prostatic region are useful for searching for associated malformations and describing the structures surrounding the urethra [7] [9]. Some authors suggest replacing these ultrasound examinations with MRI, which enables a much more precise study of the penis, perineum, prostate region and lower urinary tract in a single examination. This has the advantage of not requiring intravenous injection of contrast medium, and of presenting an objective image that can be used as a reference for subsequent examinations [7].

The treatment of the supernumerary urethra is not yet well codified and the therapeutic indication varies from one author to another; the only consensus is that the asymptomatic forms are respected [5] [11]. On the other hand, in the symptomatic forms, the treatment is essentially surgical, which consists of total excision of the supernumerary urethra, at the neckline of the penis if there is a dorsal curve of the latter. For our patient, the lithotomy was associated with excision of the supernumerary urethra approximately 3.5 cm in length. However, an incomplete uretrectomy can lead to complications such as diverticula (causes of repeated urinary tract infections), fistulas [2] [4] and stones in the urethral residue, as is the case in our patient whose formation can be done by urinary stasis in the blind urethra causing microbial pullulation and maintaining crystallization.

To prevent this, some authors suggest a double penile and transverse suprapubic approach, particularly in the treatment of complete urethral duplication [3]. In our patient, the approach was penile for the first operation where the urethrectomy was partial and the second was suprapubic in which a lithotomy was performed then the total uretrectomy.

4. Conclusions

Urethral duplication is a rare birth defect. This observation illustrates the possibility of urinary calculus formation after partial uretrectomy of the supernumerary urethra.

In this context, any unusual mass at the base of the penis after urethral resection can evoke the diagnosis, urethrocystography with mid-void images is the diagnostic key, and it is this that guides the surgical attitude.

Conflicts of Interest

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

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