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CASE REPORT

Two Cases of Dorsal Urethral Duplication

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Urethral duplication is a rare congenital anomaly. In addition to a normally urethra, there is an accessory urethra arising from the bladder or the proximal urethra which may open anywhere on the penis or even on the perineum. Many patients with this malformation are asymptomatic. Some patients may complain of intermittent discharge from the accessory urethra, incontinence, infection, double streams or stricture. The duplicated urethra may be complete or incomplete, may be located dorsal, ventral or in Y-type. The treatment of urethral duplication is individualized, depending on the type of malformation. We report two cases of dorsal urethral duplication discovered in two boys.

Keywords: urethral duplication, accessory urethra, stricture

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Introduction

Urethral duplication is a rare congenital anomaly, affecting mainly boys [1]. It is defined by the juxtaposition of two muscular channels with urinary tract mucosal lining [2]. In addition to a normally positioned urethra, an accessory urethra arising from the bladder or the proximal urethra may open anywhere on the penis, or even on the perineum [3].

The etiology of this malformation is not known. There are only hypotheses, which are not accepted in all cases [4,1]. Sometimes, this anomaly is associated with other, ano-rectal malformations [5] or other urinary tract anomalies [6].

Many patients with urethral duplication are asymptomatic. However, some patients may complain of intermittent discharge from the accessory urethra, incontinence, infection, double streams or stricture [7,8]. The treatment of urethral duplication is individualized, there is no surgical gold standard [8,9]. In most cases, the functional urethra is the preserved one.

Case report

We report two cases of dorsal urethral duplication, discovered in two boys. The first patient, a 3 year-old boy was brought to our clinic with an abnormal meatus on the dorsal side of the penis. He also had a significant dorsal curvature of the penis, simulating an epispadias (Figures 1 and 2). The second patient was a 1 year-old boy, with the same clinical appearance. Both patients were asymptomatic, and were brought to hospital by their mothers, who noticed the presence of the abnormal meatus and became worrisome.

The patients were first hospitalized in a Pediatric Unit of Urology. The following examinations were performed there: urine summary, urine culture, renal and bladder ultrasonography. Nothing pathological was found. Then, in the Pediatric Surgery Unit, both patients were examined under anesthesia. The examination revealed a normally located meatus, on the tip of the glans. The normal urethra was catheterized with a size 8 Ch catheter, and we did not find problems on that urethra. The catheterization of the abnormal urethra with a size 6 Ch and a size 4 Ch catheter revealed a stop after 2 cm in the case of the 3 year-old boy and after 3 cm in the case of the 1 year-old boy (Figure 3). Then, an urethrography with contrast solution was performed. We did not find a communication between the abnormal urethra and the bladder or the normal urethra. Beside this urethral duplication, the two boys had no other medical problems.

The treatment was surgical. We performed a circumferential incision of the abnormal meatus, on the dorsal side of the penis. Then we prepared and excised the duplicated urethra. In the case of the 3 year-old boy, the abnormal urethra stopped blindly at the dorsal basis of the penis. In the case of the 1 year-old boy the abnormal urethra also stopped blindly, but it continued with a fibrous tract to the bladder (Figure 4). We also excised that tract, which produced an important dorsal curvature, like a chordee. After the removal of the duplicated urethra and the fibrous tract, the penis became almost straight. We closed the two cavernous bodies of the penis with a couple of sutures on the dorsal side (they were partially separated, like in epispadias). Then we rebuilt the skin. We placed a Foley catheter.

The postoperative evolution of the patients was favorable. The function and the cosmetic aspect of the penis was good (Figures 5 and 6). The patients received antibiotics for three days and the bandages were changed every day. After three days, we removed the catheter and the patients were discharged. They were followed-up monthly during the first 6 months, then every 12 months. There were no complications after the surgical treatment.

Discussions and conclusions

Urethral duplication is an uncommon congenital anomaly, not often reported [5]. It is also called supernumerary ure-

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Fig. 1. Epispadic aspect of the penis in urethral duplication (lateral view)



Fig. 2. Urethral duplication (dorsal view)



Fig. 3. Catheterization of the abnormal urethra



Fig. 4. Preparation of the abnormal urethra



Fig. 5. Final aspect (dorsal view)

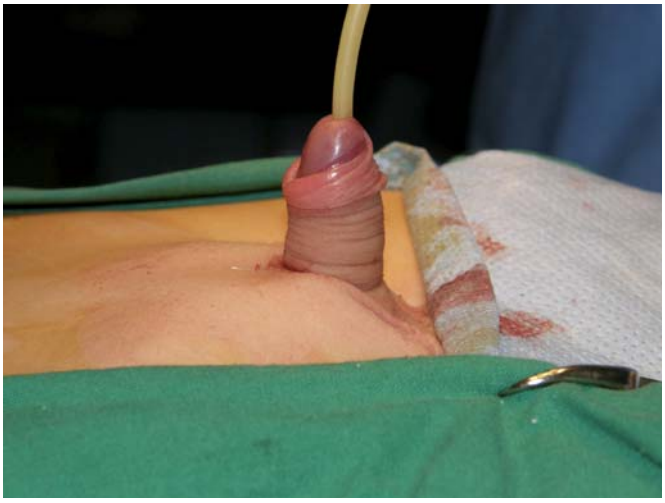


Fig. 6. Final aspect (lateral view)

thra [2]. Most urethral duplications are in the same sagittal plane, one on top of the other [10]. Less commonly, the duplicated urethra lies with the normal urethra in a frontal plane. These anomalies may be complete or incomplete [7]. A complete duplicated urethra emerges from the urinary bladder and opens somewhere on the penis or on the perineum. An incomplete duplicated urethra may not originate from the bladder and may not have an external opening. Incomplete urethral duplications are more common than complete urethral duplications [4]. There are

three anatomic variants of urethral duplication: epispadiac (dorsal), hypospadiac (ventral) and Y-type [11]. According to the Effman classification [12], there are also three types. Type I is a blind incomplete urethral duplication. Type I A (the most common type) is represented by a duplicated urethra which opens on the penis and does not communicate with the urethra. In type I B, the duplicated urethra opens like an extension from the urethra and ends blindly in the periurethral tissue (this is a rare type) [4]. In type II there is complete urethral duplication. In type II A-1 there are two independent urethras emerging from bladder and opening with two meatuses. Type II A-2 is represented by a single proximal channel opening in double streams and type II B is represented by two urethra opening in a single meatus [13]. In type III urethral duplication is a component of caudal duplication [4].

Both our cases were type I A. The duplicated urethra opened on the dorsal side of the penis, simulating an epispadias due to the significant dorsal curvature and the partial splitting of the cavernous bodies of the penis. The two boys were asymptomatic, because they had an incomplete urethral duplication. The symptoms mentioned in the introduction may be present in complete urethral duplications. The observation made by their mothers led them to a physician and made the diagnosis possible.

The treatment of urethral duplication depends on the type of malformation. In our cases, we performed the excision of the abnormal urethra. Hypospadias and epispadias are treated with standard techniques. It is important to investigate the type of urethral duplication, because an unknowing excision of the functional urethra may lead to urinary retention or incontinence.

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