Decision-making in the management of an incomplete urethral duplication in a young male

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Abstract
Objective: This is a case report of a 27-year-old Mexican man complaining of a double urethral meatus located at the tip of the glans.
Material and methods: An exhaustive physical examination was performed together with an intravenous excretory urography and retrograde urethrogram in order to evaluate the case properly.
Results: The patient presented an incomplete urethral duplication type 1B according to Effmann’s classification.
Conclusion: The lack of symptoms as well as the absence of significant clinical or functional repercussion in the patient led us to recommend therapeutic abstention for the time being.

Introduction
Urethral duplication is an extremely rare lower urinary tract anomaly (more frequent in males) that was first described by Aristotle, and includes a wide spectrum of anatomical variants in which the urethra is partially or completely duplicated. The most frequent anomaly occurs in the sagittal plane, in which the duplicated urethra is in either the dorsal or ventral position in relation to the orthotopic urethra. The therapeutic management of these conditions is complex and depends on the presence of symptoms as well as the type of anomaly.

Case report
A 27-year-old Mexican man without personal or family medical history of interest, attended the Urology Clinic...
complaining of a double urethral meatus located at the tip of the glans, one in the orthotopic position and another in the ventral or hypospadic position, respectively (Fig. 1). The patient informed us that micturition and ejaculation occur just through the hypospadic meatus. He reported no voiding problems or difficulties such as ejaculatory abnormalities, urinary incontinence or urinary tract infections. An integral physical examination of the patient confirmed the presence of a ventral urethral duplication, a retractable foreskin, and the testicles were normal to palpation without any evidence of mass or tumors. The intravenous excretory urography of the upper urinary tract and the bladder was unremarkable. On the other hand, the retrograde urethrogram revealed a unique origin urethra at the vesical level, which presented an incomplete proximal duplication in its anterior section with a short stenotic segment in the penile urethra of approximately 2.5 cm (Fig. 2). Because of the fact that other anomalies were not present, and due to the absence of functional repercussions, therapeutic abstention was advised.

Discussion

Urethral duplication is an infrequent congenital malformation with an estimate of 150 reported cases worldwide. Embryogenesis of this phenomenon is uncertain and likely multifactorial. In this sense, many hypotheses have been proposed to explain this unusual condition, including ischemia, abnormal Müllerian duct termination and growth failure of the urogenital sinus. However, a universal etiology or embryological explanation cannot be applied to all subtypes of urethral duplication.

Clinical relevance of urethral duplication is diverse. Patients may have a double stream, urinary incontinence, outflow obstruction, recurrent urinary infection or be completely asymptomatic. In the case presented, the patient did not suffer any associated problem, remaining asymptomatic to date. A proper clinical examination, voiding cystourethography, retrograde urography, urethrocystoscopy and intravenous excretory urography orientate in the diagnosis of these anomalies. The retrograde urography, as well as the intravenous excretory urography, were particularly useful to diagnose the condition of the patient.

Several classifications distinguish between complete and incomplete urethral duplication. However, the most exhaustive and widely used classification, based on radiological findings, has been offered by Effmann et al. as it is functional and represents all clinical aspects involved (Fig. 3). Effmann's classification divides urethral duplications into incomplete (type 1), complete (type 2) and coronal (type 3), the last one being the one usually associated with bladder duplication. Type 1 is the most common variant of urethral duplication and is usually asymptomatic. Based on the clinical findings obtained in this case and according to Effmann's classification, the patient presented a urethral duplication type 1b due to the proximal localization of the accessory urethra, which originated from the principal urethra and had a blind end in the periurethral tissue.

The treatment of these conditions must be personalized, taking into consideration the anatomic variant and functional outcome, as well as the coexistence of other malformations. The treatment criteria ranges from therapeutic abstention, to the excision of the accessory urethra. The surgical approach is not always required, as patients are at risk of developing a variety of postoperative complications, such as urethrocutaneous fistula, recurrent meatal stenosis and urethral diverticulum with calculi. According to the Salle et al. recommendations for the management of each urethral duplication subtype, in this particular case, the lack of symptoms as well as the absence of significant clinical or functional repercussion in the patient led us to advised therapeutic abstention for the time being.
Conflict of interest

The authors declare that they have no financial or non-financial conflicts of interest related to the subject matter or materials discussed in the manuscript.

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References


