A case of coronal urethral duplication with no other abnormalities

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Urethral duplication is a very rare congenital anomaly. Urethral duplication is seen most commonly in the sagittal plane. We report a rare case of complete urethral duplication in the coronal plane with no other associated anomalies. Surgical correction of this coronal urethral duplication resulted in a normal-appearing penis and good functional outcome with a single midline urethral meatus and urinary stream.

Key Words: urethral duplication, coronal

Case report

An otherwise healthy 11-month-old male was referred to the pediatric urology clinic for an abnormal meatus. The child had an unremarkable birth history with no hormonal exposure during gestation. The abnormality was only noticed following circumcision. There was no history of urinary tract infections. There was no family history of hypospadias.

Physical examination revealed bilaterally descended testes with no hernias or hydroceles. The shaft of the penis had no abnormalities. There appeared to be a barrel-like two hole opening at the tip of the glans penis. These openings were laterally displaced in the coronal plane with approximately 4 mm of intervening glanular tissue, Figure 1. The opening on the right appeared to be patent and moist mucosa was easily visualized. The opening on the left however was smaller and no moist mucosa was visualized. The remainder of the physical examination was unremarkable.

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Consent was given for exploration and potential surgical correction. Initially, the two openings were explored. An 8-Fr pediatric feeding tube was passed easily into the bladder through the right opening and presumed dominant urethra. The opening on the left initially did not appear to be patent until a glidewire was passed through it and into the bladder. The glidewire was able to be passed approximately 6 cm into the left opening. A plain x-ray with the glidewire in the left meatus of the presumed accessory urethra confirmed a clear communication between the left opening and the bladder. Retrograde urethrogram via the right meatus demonstrated a normal urethra without evidence of bladder duplication. After consideration of the patient's unremarkable history and these findings, cosmetic surgical correction for the glans penis was performed and the accessory urethra was left intact. The two openings were then closely approximated to give the appearance of a single urethral meatus and to enable a single urinary stream. A deep wedge of glandular tissue was excised between the two openings and a rotational closure was performed resulting in the two meatuses immediately adjacent in the sagittal plane. The procedure resulted in a normal-appearing glans penis with a single midline meatus, Figure 2.

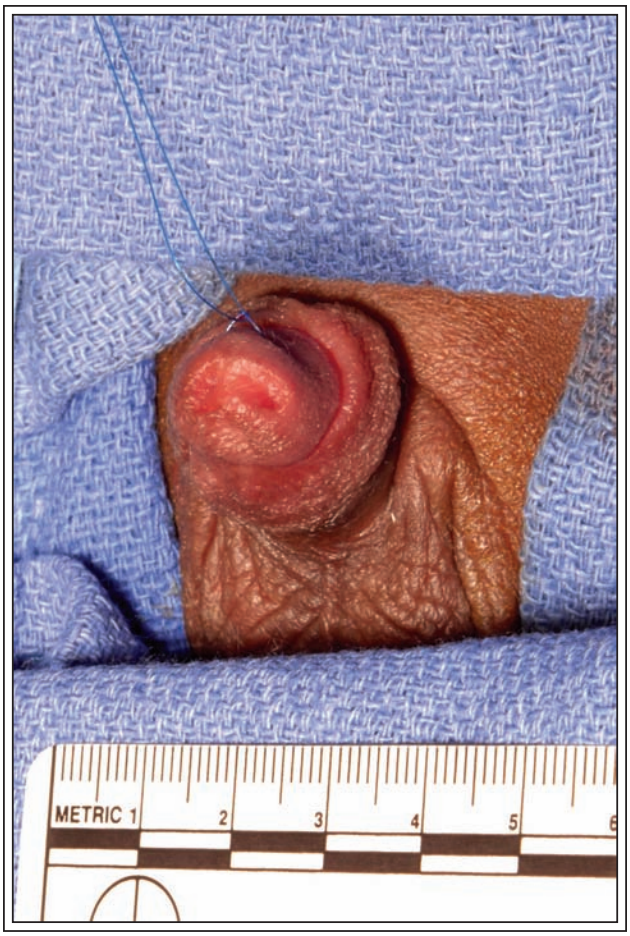

Figure 1. Preoperative photograph of penis with duplicated urethra in coronal plane.

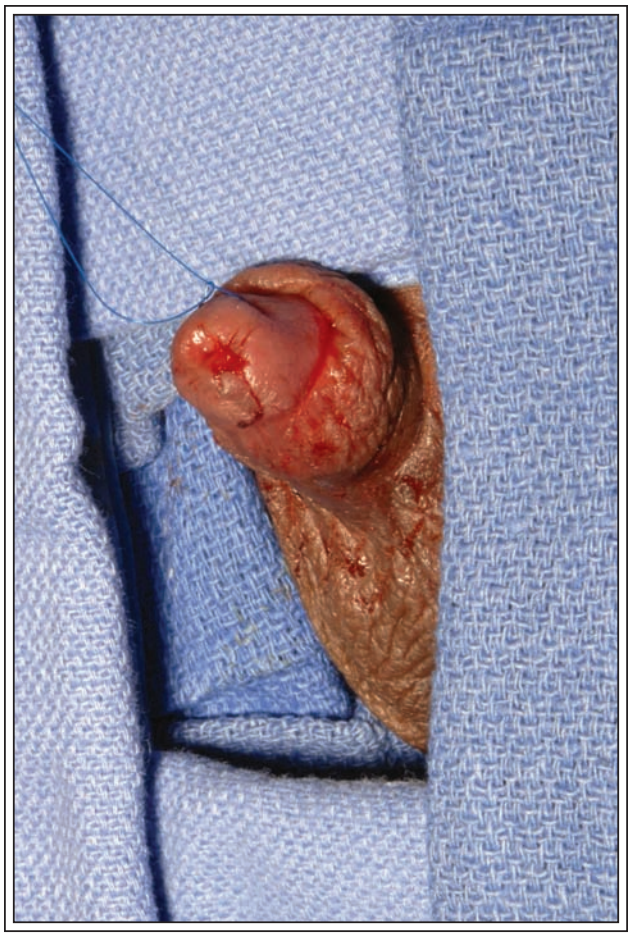

Figure 2. Postoperative photograph of penis following excision of intervening glanular tissue and rotational closure to place the two meatuses immediately adjacent to one another in sagittal plane resulting in appearance of single meatus.

Discussion

Duplication of the urethra is a rare congenital abnormality, which can vary in its clinical presentation due to variations in the duplications. The duplications may be complete or incomplete with multiple different anatomic variations. Effmann described a classification system for these abnormalities which has been well explained elsewhere.[1] Salle added to this system by providing a distinction for urethral duplication in the coronal plane,[2] which would classify this diagnosis into a coronal type IIA1 urethral duplication.

The pathogenesis and embryology of urethral duplication is unclear. There have been many proposed theories but no single theory explains the many variations of urethral duplication.[3] Urethral duplication is most common in the sagittal plane resulting in a dorsal and a ventral urethra. Coronal duplication is much more rare and has only been reported in the literature a handful of times.[2,4-7] Coronal duplication does have a higher association with other abnormalities. These abnormalities include bladder duplication, complex caudal duplication, and various gastrointestinal abnormalities.[2,5-9] Of note, our patient did not have any of these associated abnormalities.

Surgical management of these different anomalies varies from simple meatoplasty to excision of the accessory urethra. Salle et al proposed various surgical options based on the Effmann classification. These classifications and surgical options are helpful but the surgeon's judgment plays a key role in addressing this uncommon problem. Considering the patient's unremarkable history and the physical findings, a meatourethroplasty was appropriate in this case as the accessory urethra did not present any functional problems. The result has been an excellent cosmetic appearance and normal function with a single midline meatus and urinary stream. □

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EDITORIAL COMMENT

Re: A case of coronal urethral duplication with no other abnormalities

Urethral duplications are rarely encountered in clinical practice of which duplication at the coronol level is even rarer and is associated with higher incidence for genitourinary abnormalities especially concomitant bladder duplication. Proper planning by studying the anatomy of the urethra with VCUG preoperatively will help in planning appropriate surgical intervention. It is not apparent if the authors have done that prior to definitive surgery in their case report. Surgical treatment of patients with urethral duplication should be individualized based on the anatomy of the urethra as currently no gold standard surgical approach exists. In this case report the patient has excellent functional and cosmetic results with this simple approach of excising the intervening glanular tissue between the duplicated urethra.

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