

Radiological Findings of Male Urethral Duplication Associated with Bladder Duplication: Case Report¹

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Urethral duplication or accessory urethra is a rare congenital anomaly. Even rarer, is its association with bladder duplication. We report a case of urethral duplication associated with bladder duplication in a seven-year-old boy who underwent retrograde urethrography, sonography and magnetic resonance (MR) imaging. While retrograde urethrography can demonstrate the extent of the duplicated urethra, MR imaging and sonography can provide detailed information on the anatomy of the adjacent tissues as well as urethral duplication.

Index words : Urethra, abnormalities

Urethra, radiography

Urethra, US

Urethra, MR

Urethral duplication associated with bladder duplication is a rare congenital anomaly (1, 2). Physical examination undoubtedly remains the primary step in the initial evaluation of the patient and is usually sufficient for defining abnormal external structures. Traditional retrograde urethrography demonstrates the extent of the anomaly. However, it cannot provide detailed anatomic information of the adjacent tissues surrounding the duplicated urethra. Magnetic Resonance Imaging (MRI) and sonography show pelvic and urethral anatomy and are helpful for planning corrective surgery. To our knowledge, MR imaging and sonographic findings of the urethral duplication associated with bladder duplication have not been previously reported in the literature.

Case Report

A seven-year-old boy presented with low abdominal

pain during micturation, which developed 10 days before his visit to the hospital. The patient had no history of incontinence or urinary tract infection. Lower abdominal tenderness was found on physical examination. Furthermore, a small opening was noted at the dorsal aspect of the proximal penile shaft. There was no evidence of infection at the opening. Transabdominal ultrasonography revealed a thick walled cystic mass anterior to the urinary bladder. Subsequent endorectal sonography revealed a cystic mass and tubular low echoic structure below the cystic mass (Fig. 1A). Sonography using high frequency linear transducer identified a tubular structure at the dorsal aspect of the penile shaft, running parallel with the penile urethra (Fig. 1B). In the sagittal T2-weighted image, the cystic mass showed heterogeneous high signal intensity with an ill-defined margin. The bladder wall that contacted the soft tissue mass was thickened due to submucosal edema (Fig. 1C). Gadolinium-enhanced coronal T1-weighted image showed a thick walled cystic mass, which connected with the fistulous tract running through the dorsal subcutaneous layer of the penile shaft. The penile urethra originating from the urinary bladder showed no abnormality (Fig. 1D, E). Retrograde urethrography was per-

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formed through the small opening at the dorsal aspect of proximal penile shaft. It showed a fistulous tract and a small pouch at distal end of the tract (Fig. 1F). At surgery, the long tubular structure was dissected away from the top of the corpora cavernosa, which then led through the abdominal wall, running dorsal to and cranially around the symphysis pubis, to eventually end at a thick-walled cystic mass anterior to the urinary bladder. Yellowish pus was found within the cystic mass and *Escherichia coli* was cultured. In the pathologic specimen, the fistulous tract and cystic mass was lined by epithelium strongly resembling transitional cell ep-

ithelium. The cystic mass also showed a muscle layer. Our case was diagnosed as an accessory urethra associated with bladder duplication.

Discussion

Urethral duplication is a rare anomaly. To the best of our knowledge, only 188 cases have been described in the literature (1, 2). Urethral duplication unassociated with bladder duplication occurs exclusively in males whereas urethral duplication in females is almost always associated with bladder duplication (3). Although

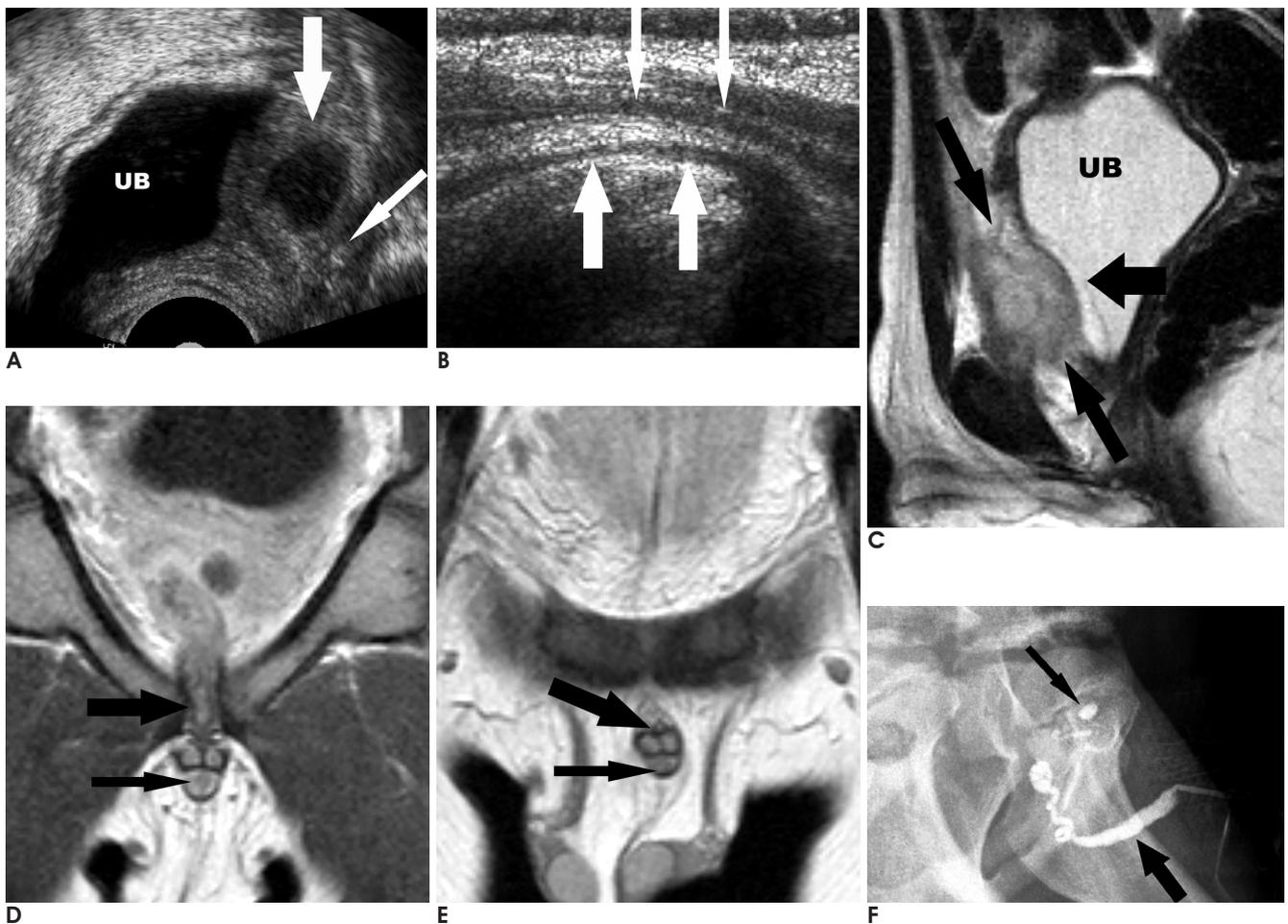


Fig. 1. Urethral duplication associated with bladder duplication in a seven-year-old boy.
A. Endorectal sonography shows the urinary bladder (UB) and nonfunctioning anterior bladder (thick arrow). Hypoplastic accessory urethra is seen as a tubular low echoic lesion (thin arrow) at the inferior aspect of nonfunctioning anterior bladder.
B. High frequency linear transducer is applied to the ventral aspect of the penile shaft. Hypoplastic accessory urethra (thick arrows) and functional urethra (thin arrow) are clearly defined.
C. On sagittal T2-weighted MR image, the nonfunctioning anterior bladder shows heterogeneous high signal intensity anterior to the urinary bladder (UB). A submucosal edema of anterior bladder wall (thick arrow) and mild infiltration (thin arrows) adjacent to the nonfunctioning anterior bladder indicate the inflammatory change.
D, E. Coronal contrast-enhanced T1-weighted MR images show functional urethra (thin arrow) and accessory urethra (thick arrow) separately.
F. Retrograde urethrography through the opening of proximal penile shaft shows the extent of an accessory urethra (thick arrow) and the nonfunctioning anterior bladder (thin arrow).

a number of theories about the cause of urethral duplication have been proposed, no single theory explains all of the various types of this anomaly. The most widely accepted theory is that it may occur if the genital tubercles lie more posterior than their normal position or if the cloacal membrane extends more ventrally than usual; a part of the membrane remains in front of the tubercle and interferes with its subsequent growing, thereby causing the urethral duplication (4).

There have been several attempts to classify urethral duplications (5, 6). The classification proposed by Effmann et al. is the most commonly accepted one. They classified male urethral duplication into three main types: type I or a blind-ending accessory urethra; type II or a patent accessory urethra-type I and II are not associated with duplicated or septated bladders; and type III or accessory urethras arising from a duplicated or septated bladder. In type III urethral duplication, bladder duplication may exist in the coronal or sagittal plane. Coronal bladder duplication is more common and each duplicated bladder receives the ipsilateral ureter (6). Sagittal bladder duplication, on the other hand, is rare; the posterior bladder receives both ureters, whereas the anterior bladder is nonfunctional (2). According to the classification of Effmann et al., our case was a type III.

The clinical presentation varies according to the anatomic types of urethral duplication and the presence or absence of infection. A wide range of symptoms, such as double stream, incontinence, urinary tract infection, or outflow obstruction may occur (2, 7). However, many cases are asymptomatic and some cases are found incidentally during repair of epispadia or hypospadias (2, 3).

Management of urethral duplication may be complex and depends on the duplication subtype. In practice, it is important to differentiate the functional urethra from the hypoplastic accessory urethra for surgical planning. The functional urethra usually has a ventral position, wide caliber to empty the bladder, good sphincteric mechanism and normal verumontanum (2, 3, 5, 8). In our case, the hypoplastic accessory urethra had a dorsal position and originated from the small non-functioning anterior bladder that had no connection with the ureter.

In the radiologic evaluation of urethral duplication, diagnostic imaging should provide anatomical delineation of the abnormality, recognition of the functional ure-

thra, and detection of other anomalies. Both voiding cystourethrogram and retrograde urethrogram have been used for demonstrating the two channels of urethral duplication. They can demonstrate the length, shape, and proximal extent of the anomaly. Sonography can demonstrate the urethra and its surrounding corporal bodies; however, it has limitations on showing the full extent of the anomaly and the inflammatory change of adjacent tissues, if present. The advantages of MRI in the evaluation of genitourinary anomalies include superb soft-tissue contrast, direct multiplanar imaging capability, and high sensitivity in the detection of fluid (9). With MR imaging, not only were the length, shape, and extent of the anomaly accurately assessed, but also the relationship between the urethra and its surrounding corporal bodies and inflammatory change of adjacent tissues were shown.

In conclusion, male urethral duplication associated with bladder duplication, a rare congenital anomaly, can be detected by some radiologic examinations, as in our case report. Retrograde urethrography renders the diagnosis and shows the extent of this anomaly. Sonography allows further assessment by demonstrating the anatomical configuration of the anomaly. MRI can also render accurate diagnosis and provide detailed anatomy of urethral duplication required for preoperative planning.

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