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Voiding cysto-urethrography of complete urethral duplication in a child, a case report

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Abstract

Background: Urethral duplication is a very uncommon congenital anomaly that predominantly affects males. This condition is distinguished by the presence of two urethras, which might be incomplete or complete. The diagnosis of urethral duplication is very important, and missing this diagnosis leads to the delayed surgical correction of patients. Voiding cysto-urethrography is an important diagnostic method to diagnose the urethral duplication that can provide a standard view for surgical operation and determine the type of duplication.

Case presentation: In the present study, we report a 5-year-old boy with complete urethral duplication diagnosed via voiding cysto-urethrography in our medical center. We reviewed the classification of this anomaly, indications of surgery, the importance of voiding cysto-urethrography, and the role of other imaging modalities to diagnose the urethral duplication.

Conclusion: For children with a double urinary stream, a complete genital examination and radiological assessment are required not only to diagnose urethral duplication but also to determine the kind of urethral duplication and rule out related anomalies. These findings guide surgeons in planning the best treatment for the patients to prevent future complications. Voiding cysto-urethrography is one of the most useful and beneficial procedures to diagnose the urethral duplication which can be used as a first imaging modality, especially when there are double functional urethras.

Keywords: Urethral duplication, Voiding cysto-urethrography, Congenital anomaly

Background

Urethral duplication is known as a rare congenital urogenital anomaly which is mostly observed in the male gender and is diagnosed in the pediatrics by various clinical conditions [1]. A deformed penis, twin streams, urinary tract infection (UTI), urinary incontinence, and urethral discharge are the main clinical manifestations of urethral duplication [2, 3]. Various types of urethral duplication include complete or incomplete, dorsal or ventral opening, or coronal/collateral urethral duplication [3].

The diagnosis of urethral duplication may be difficult which could be conducted via voiding cysto-urethrography (VCUG) [4]. We present a 5-year-old boy with double urinary stream in his history and double meatuses in the physical examination which urethral duplication was diagnosed for him by VCUG in our medical center. We aimed to compare the clinical and imaging findings of presented case with previous studies.

Case presentation

A 5-year-old boy was referred to our medical center with the complication of a double urinary stream and urinary incontinence. The parents had noticed double urinary stream after the circumcision within a few weeks after birth, but no imaging or managements were conducted. We asked for the full history of the patient from his parents but no other urinary symptoms, including UTI were

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reported. No family or genetic history of genitourinary abnormality was noted in the patients 'family. A circumcised penis with a normal urethral opening at the glans and another 1.5 cm proximal to the normal urethra was discovered during genital examination. The rest of the physical examination was routine and ordinary.

A VCUG was conducted using an 8-Fr feeding tube by cannulating the normal urethra, then normal saline with iodinated contrast media (250 cc) was injected slowly into the bladder. Results of VCUG showed normal-shaped and normal-sized bladder with a smooth border. Vesicoureteral reflux was not seen also. Urethral duplication was observed that initiated from the bladder with two separate lumens and two separate meatuses distally (Fig. 1). Ultrasound of both kidneys did not show any hydronephrosis or renal stone.

The patient left our medical center by parent's will and did not allow any interventions. The patient did not have any symptoms other than a double stream in voiding at 6 months follow-up. Urine analysis and urine culture tests were normal in the follow-up.

Conclusions

The exact mechanism of urethral duplication remains still unknown, but the main theory is based on the abnormal relationship between the lateral Anlagen of genital tubercle and the ventral end of the cloacal membrane. Diagnosis and proper management of this anomaly are important in terms of the prevalence of UTIs among these patients. VCUG is the fundamental diagnostic tool for urethral duplication diagnosis which can provide a suitable surgical plan for the patients [3, 4].

Various studies have claimed that urethral duplication can be easily underdiagnosed in terms of associated genital anomalies [5–7], but we reported a 5-year-old boy with isolated urethral duplication. Previously, Effmann and colleagues have divided urethral duplication into 3 types and some subtypes based on radiological findings as below (Fig. 2).

Type I: This type is incomplete urethral duplication. Type IA (more common) is a distal Blind-ending accessory urethra which duplicated urethra's opening is on the dorsal or ventral surface of the penis, but there is no communication with the urethra or bladder. In type IB, proximal-accessory urethra's opening from the urethral channel ends blindly in the periurethral tissues (rare).

Type II: There is a complete patent accessory urethra in this type. It is divided into 2 subgroups: A (2 meatuses) and B (1 meatus). In type IIA1, there are two non-communicating urethras arising independently from the bladder. This type is the most common type in the literature. In type IIA2, second channel arises from the first and courses independently into a second meatus. Special type of IIA2 is Y-type, which second meatus ends up posteriorly in perineum. In Type IIB, two urethras arise from the bladder or posterior urethra and uniting into a common channel distally.

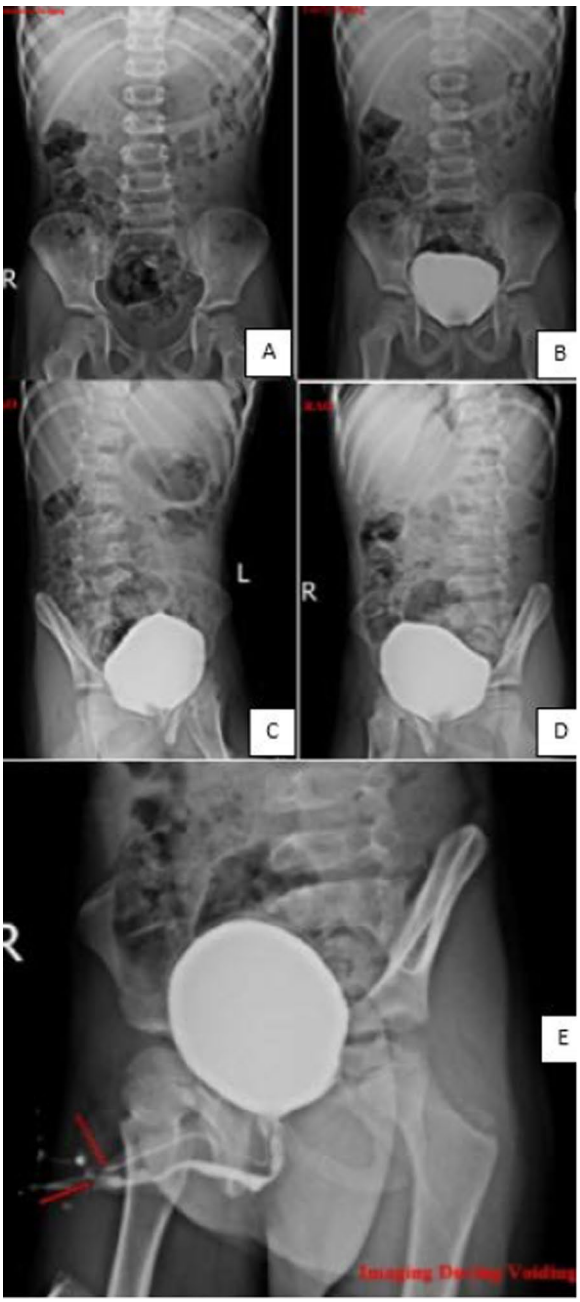

Fig. 1 VCUG of presented case. "**A**" is preliminary imaging, "**B**" is early filling image, "**C**" is left anterior oblique view(LAO) image, "**D**" is right anterior oblique view(RAO) image and "**E**" is voiding phase image. Dashed arrow (upper) shows distal opening of accessory urethra and solid arrow (lower) shows distal opening of main urethra. As shown in picture "**E**" there are two urethras arising from bladder with different proximal and distal orifices

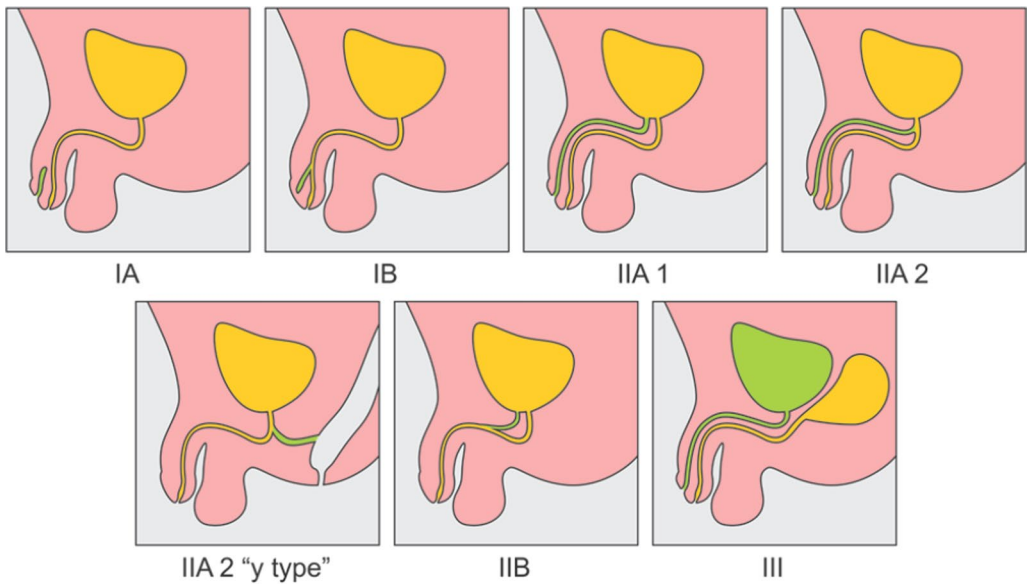

Fig. 2 Effmann's classification for urethral duplication

Type III: Accessory urethras arising from duplicated or septated bladders [8].

The main diagnostic tools used for the diagnosis of urethral duplication are VCUG, retrograde urethrography(RUG), intravenous urography(IVU), ultrasonography, and magnetic resonance urethrography (MRU), but the first step for the evaluation of a child with double urethra is the genital examination to see distal orifices of urethras and to determine the functional status of both urethras. Urethral duplication is generally detected by VCUG revealing two distinct urethras if both urethras are functioning. When VCUG fails to reveal the accessory urethra, RUG is performed. Because certain duplications have a non-dominant urethra that may be clogged with debris or have too much resistance for downstream flow to exhibit significant contrast opacification in VCUG, some authors advocate RUG as a first diagnostic modality. Intravenous urography can show widened pubic symphysis in epispadiasis and other urogenital anomalies like unilateral renal agenesis, ureteral duplication, and a duplicated bladder. Ultrasonography can show stricture, extra-luminal abnormal soft tissue or diverticulations of urethras. To evaluate surrounding soft tissues, complex fistulas and coexistent genitourinary or gastrointestinal anomalies and exact demonstration of sizes, shapes and positions of two urethras, MRU can be helpful. If this routine diagnostic modalities fail to reveal desired results, then direct visualization of urethras and bladder with urethro-cystoscopy can be done [4, 9–12].

The present case was isolated complete urethral duplication Effmann type II-A1 which had two distinct urethra with two meatuses, diagnosed by VCUG. More than half of cases of urethral duplication can be associated with other urogenital and gastrointestinal anomalies, such as vesicoureteral reflux, posterior urethral valves, bladder exstrophy, congenital urethral polyps, megalourethra, epispadias, hypospadias, cryptorchidism, anal stenosis and renal dysplasia [11, 12]. For example, Patel and colleagues evaluate a 3-month-old premature male infant with imperforated anus and hypospadias. They showed a distinct linear tract arising dorsally from the posterior urethra that partially opacified with contrast-enhanced voiding urosonography which was confirmed by VCUG as a urethral duplication [1].

The radiological type, the patient's symptoms, and the severity of the abnormality are all essential criteria in deciding whether or not to treat urethral duplications surgically. Urinary incontinence, urinary blockage, and the presence of other genital anomalies may all be signs that surgery is needed [10, 12]. In Type IB or IIB urethral duplication treatment is not recommended. Although the study by Lopes and others showed that the patients with incomplete duplication (type I A or I B) can totally be asymptomatic, with no need for surgical correction, Guglielmett and her colleagues in a case series of 19 patients noted that Type IA, IB duplications are also treated similar to Type IIA2 by excision of the accessory atretic urethra and reconstruction of functional urethra. It was reported that type IIA2-Y is the most complex form of duplication to correct, and multiple procedures might be required [3, 10, 13].

Hence, the radiological investigations are mandatory in these patients not only to establish a diagnosis but also to identify the type of urethral duplication and ruling out associated anomalies which these findings guide surgeons to make the best decision for the patients. VCUG is one of the most useful and beneficial procedures for diagnosing urethral duplication.

These results are in line with what we found in our research. We also looked at the relevance of various imaging modalities in identifying urethral duplication and the indications of surgery for this condition in this research. The research was limited by the fact that the patient refused surgery.

We conducted VCUG for a 5-year-old boy with a double urinary stream which his final diagnosis was urethral duplication type II-A1 according to Effmann's classification. VCUG can be used as a first-line diagnostic modality for similar cases.

Abbreviations
VCUG: Voiding cysto-urethrography; RUG: Retrograde urethrography; MRU: Magnetic resonance urethrography; IVU: Intravenous urography; UTI: Urinary tract infection.

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Author contributions
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Availability of data and materials
The datasets used during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
The study was approved by the ethical board of the medical-educational center in which it was performed. The patient's parents included in this study provided the written informed consent to participate in this research.

Consent for publication
Patient's legal guardian gave a written informed consent to publish the data contained within this study since the patient is a minor.

Competing interests
The authors declare that they have no competing interests.

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