Bilateral Orthotopic Ureteroceles Presenting with Primary Nocturnal Enuresis, a Case Report

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Abstract
An 8-year-old boy presented with primary nocturnal enuresis as the first clinical sign of ureterocele in the absence of dysuria, urinary incontinence or urinary tract infections. A prolonged history of bedwetting prompted subsequent clinical and laboratory evaluations, leading to the correct diagnosis of ureterocele. Primary and persistent nocturnal enuresis as an initial manifestation of ureterocele has not been reported previously. The present patient showed a feature not previously described in children with ureterocele. Although rare, ureterocele should be considered in the differential diagnosis of children presenting with primary nocturnal enuresis.

Keywords: Primary nocturnal enuresis; Simple orthotopic ureterocele; Single-system ureter.

Introduction
A ureterocele is a congenital cystic dilatation of the distal ureter within the bladder (1). Ureteroceles occur in about 1 in 600 to 1000 live births, affect girls more than boys, and are often associated with a high incidence of urinary tract obstruction or VUR (1-3). The most common presenting symptoms are those of the UTI followed by the detection of an abdominal mass.

Case report
An 8-year-old boy was referred to the pediatric nephrology clinic for evaluation of primary nocturnal enuresis. He was born to non-consanguineous parents and his family had no genetic renal diseases. He had two healthy siblings and his medical history was unremarkable except for bedwetting occurring approximately 2 hours after falling asleep, seven days a week. He had no day-time wetting, urinary incontinence, voiding disorders or recurrent urinary tract infections (UTI) and received no medications. His weight and height were both at the 50th percentiles and his blood pressure was 100/60 mmHg. Laboratory data showed an estimated creatinine clearance rate of 105/mL/min/1.72m² using the Schwartz formula. Serum electrolytes were as follows: sodium: 140 mEq/L, potassium: 4.0 mEq/L, Cl: 95 mEq/L, bicarbonate: 25 mEq/L, BUN: 20 mg/dL, creatinine: 0.5 mg/dL, and glucose: 95 g/dL. The hemoglobin level was 11.5 g/dL, hematocrit was 34%, white blood cell count was 8,500/mm³ with a normal differential, and platelet count was 340,000/mm³. A urinalysis revealed a specific gravity of 1.017 and pH of 6.0 but no blood or protein. A urine culture revealed no bacterial growth. An abdominal ultrasound demonstrated bilateral normal-sized kidneys without hydronephrosis. The right kidney measured 9.2 cm and the left 9.0 cm. The bladder wall was smooth without trabeculation and the bladder size was in the normal range with an acceptable post voiding volume. Bilateral filling defects were noted...
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within the posterior aspects of the bladder (Figure 1).

![Figure 1. Renal ultrasound showing intravesical bilateral ureteroceles.](image1)

An intravenous pyelogram (IVP) showed single system ureters with cobra-head findings characteristics of distal dilatation of the ureters (Figure 2).

![Figure 2. Intravenous pyelogram (IVP) showing single-system ureters with cobra-head findings characteristic of distal dilatation of ureters.](image2)

A voiding cystoureterogram (VCUG) showed grade 1 vesicoureteral reflux (VUR) on the left (Figure 3). Cystoscopy excluded posterior urethral valves (PUV). A Te-99m DMSA scan showed no cortical scaring.

![Figure 3. Voiding cystoureterogram demonstrating grade 1 reflux on the left.](image3)

**Discussion**

A ureterocele is a congenital cystic dilatation of the distal ureter within the bladder. The most common presenting symptoms are those of the UTI followed by the detection of an abdominal mass.

An ectopic ureterocele is the most common type and almost always develops on the ureter originating from the upper renal unit of a duplex kidney (4, 5). Ureterocele is often seen on excretory urography, either as a positive cobra-head dilatation or as a negative filling defect on voiding cystourethrogram. VCUG shows VUR in more than 50% of the patients. A cystoscopy usually confirms the diagnosis (6).

A simple orthotopic ureterocele is a ureterocele that develops on the distal portion of a single ureter where normally implanted into the bladder. This type of ureterocele is often bilateral and is often associated with hydronephrosis and renal dysplasia. Single-system ureteroceles are less common than duplex-system ureteroceles (7). Simple ureteroceles may be asymptomatic or may produce a wide range of clinical signs and
symptoms, including voiding disorders, foul-smelling urine, recurrent urinary tract infections, bladder outlet obstruction, and renal failure. Because of the obstructive nature of ureteroceles, the activity of the affected renal unit varies from a normal, well-functioning kidney to a nonfunctioning, dysplastic renal segment or kidney (1-5).

Our patient’s clinical presentation was unique as he manifested with primary bedwetting complicated by bilateral ureteroceles developing on the ureters origin from a single-system kidney in the absence of voiding disorders, UTI, and hydronephrosis. Although dysuria and urinary incontinence have been described as early clinical symptoms of ureterocele, persistent primary nocturnal enuresis presenting as the only manifestation of ureterocele in the absence of hydronephrosis or VUR has not been reported previously.

The management of a patient with ureterocele should begin with appropriate antibiotic treatment if infection is present, followed by excision or drainage of the involved renal segment(s) (8, 9). A pus-filled upper pole and ureter may need to be removed or drained urgently in some ill patients. Nephroureterectomy is performed when the kidney is non-functional. Surgical intervention is not needed in most cases of simple orthotopic ureteroceles in the absence of upper tract dilatation or VUR (10, 11).

Conclusion
This study described nocturnal enuresis as a clinical symptom of ureterocele and emphasized the importance of considering ureterocele when evaluating children presenting with persistent unexplained nocturnal enuresis. Parental awareness should be raised about nighttime wetting, so that it may be possible to prevent serious renal diseases.

Conflict of Interest
The authors declare no conflicts of interest.

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References