Rare late presentation of bilateral single system intravesical ureteroceles complicated with ureterolithiasis: Case report and literature review

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**ABSTRACT**

Bilateral intravesical ureteroceles is a rare condition where both ureters terminally end in cystic dilations in the urinary bladder. Herein, a 31-year-old male patient presented with severe right flank pain and gross hematuria. Upon computed tomography imaging, a right ureterocele with an entrapped stone was revealed. Direct visualization also showed a smaller ureterocele at the left ureterovesical junction. Both ureteroceles were unroofed using rigid resectoscope with cold knife resulting in rapid bilateral efflux. The treatment was well tolerated with no known complications. Albeit uncommon, this case highlights the need to consider ureteroceles in adult patients with urinary tract symptoms.

**Introduction**

A ureterocele is an atypical anatomical finding of cystic dilatation of the terminal ureter within the urinary bladder. It results from the persistence of Chawalla’s membrane during embryologic development. The incidence of ureteroceles is not well elucidated, with autopsy studies estimating a prevalence between 1 in 500 to 1 in 4000.\textsuperscript{1} Ureteroceles are 4–7 times more likely in females with an estimated 10\% of ureteroceles occurring bilaterally.\textsuperscript{2}

There are numerous classification systems for ureteroceles. The American Academy of Pediatrics accepted terminology dichotomizes ureteroceles as intravesical or ectopic.\textsuperscript{3} An intravesical ureterocele is defined as entirely within the bladder and can be associated with a single or completely duplicated urinary system. In contrast, an ectopic ureterocele is defined as a ureterocele with some component permanently in contact with the bladder neck or in the urethra. Further description of ureteroceles include sphincteric, stenotic, sphincterostenotic, blind, nonobstructive, and cecoureterocele.

Patients with ureteroceles can have a range of symptoms and sequelae, from entirely asymptomatic to unrelenting pain and chronic kidney disease due to prolonged ureteral obstruction. Herein, we present an adult male patient with bilateral ureteroceles complicated by a ureteral stone, illustrating an exceedingly rare presentation of classic pediatric pathology in adulthood and its management.

**Case report**

A 31-year-old male with a past history of urolithiasis, mixed hyperlipidemia, and tobacco use disorder presented with severe and unrelenting right flank pain, urinary frequency, dysuria, and gross hematuria with worsening symptoms. The patient was afebrile despite urinalysis on admission testing positive for several bacteria and leukocytes. Non-contrast computed tomography (CT) revealed a single collecting system and mild dilation of the distal right ureter with a 9mm stone present at the right ureteral orifice (Fig. 1 a-c). A thin membrane at the level of the bladder suggested a right ureterocele.

Under general anesthesia, a 22-French rigid cystoscope with a 30° lense wasatraumatically inserted through the urinary meatus and into the bladder. Panendoscopic view of the lateral walls, trigone, bladder neck, and dome were observed. The right ureterocele was visualized crossing the midline and encroaching on the left lateral wall of the bladder. (Fig. 2 a). A left ureterocele was also observed, albeit smaller in size (Fig. 2b).

The resectoscope was then assembled and the right ureterocele was unroofed at its most superior portion with a rigid resectoscope and cold-knife. The left ureterocele was then unroofed in the same fashion to prevent future obstruction in a stone-forming patient. Rapid urinary efflux was observed from both unroofed ureteroceles. The stone within the right dilated ureter was visualized (Fig. 3a). Using a Boston Scientific Zero Tip™ basket the stone was grasped and transferred to the bladder.
Manual cystolitholapaxy was then performed and the stone fragments were irrigated from the bladder. Hemostasis was achieved with Bugbee electrocautery, or spot coagulation. The bladder was filled and proper collapse of the ureteroceles was visualized (Fig. 3 a-b).

The patient tolerated the procedure well with no known complications. The patient was discharged the following day with antibiotics for urinary tract infection. On further studies, bladder stone was found to be predominantly calcium oxalate.

Discussion

Ureteroceles are often found incidentally on prenatal ultrasound or a few months after birth in workup for lower urinary tract voiding symptoms. In rare instances, however, patients with ureteroceles may become symptomatic later in life. Treatment is usually mandated upon discovery to prevent chronic kidney injury secondary to obstructive uropathy.

As a primarily pediatric pathology, there is limited comprehensive data on adult ureteroceles management. One retrospective study found that in 26 adult patients (2 with bilateral ureteroceles), the transurethral transverse-incision of the ureterocele produced symptom free results. The study concluded that the surgical approach is safe and effective but mild vesico-ureteral reflux may persist as confirmed with follow-up imaging. As ureteroceles encompass the entire spectrum of symptomatology, imaging modalities including renal-bladder ultrasound and computed-tomography should be utilized to prevent lasting complications of chronic obstruction.

Conclusion

Although a pathology rarely seen in adults, ureteroceles should be included in the differential of any patient with urinary tract symptoms. This case highlights the importance of a thorough workup, especially in the face of distracting concomitant pathology. It also emphasizes a...
traditional pediatric pathology that urologists may encounter in predominantly adult practices.

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Fig. 3. a–b: Endoscopic imaging revealing unroofed ureteroceles. Large right ureterocele unroofed with trapped 9mm stone therein (a) and unroofed, left ureterocele with efflux (b).