

Bilateral complete ureteral duplication with fixed dumbbell stone formation in the ureterocele

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CASE STUDY

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ABSTRACT

Bilateral ectopic ureteroceles are extremely rare. There are few reports of bilateral duplex system with ureterocele stone formation. Our case report represents a 35-year-old woman with bilateral complete ureteral duplication and stone in a right sided ureterocele. A Bugbee electrode was used to puncture the ureterocele at its most dependent part followed by fragmenting the stone using pneumatic lithotripsy. Management of the ureterocele stone depends mostly on stone size. Whereas smaller stones are amenable to endoscopic incision and lithotripsy, larger ones have required combination therapy with endoscopic incision, laser or pneumatic lithotripsy and even orificiotomy and ureterolithotomy.

Key Words

Ureteral duplication, ureterocele, stone

Implications for Practice:

1. What is known about this subject?

Although unilateral ureteral duplication is common, bilateral complete duplication is not. Combined with ectopic

ureteroceles and complicated by stone formation this represents a very exceptional case.

2. What new information is offered in this case study?

This is a very rare combination of bilateral ureteral duplication and ectopic ureterocele with a stone traversing the ureterocele orifice in an adult. The patient suffered no specific lower tract symptoms nevertheless, and would have been missed by relying on clinical triage and urine analysis alone.

3. What are the implications for research, policy, or practice?

When confronted by an immobile dumbbell shaped bladder stone, one must strongly contemplate ureteroceles in the differential diagnosis. Furthermore, an ureterocele complicating bilateral complete ureteral duplication can still be managed safely by endoscopic means.

Background

Unilateral ureteral duplication is a common congenital anomaly reported to occur in about 1 per cent of the population, but bilateral ectopic ureteroceles are less common.¹ Anomaly of ureteral bud growth has been proposed as a possible cause of these conditions.² Our case report represents bilateral complete ureteral duplication with stone in a right side ectopic ureterocele.

Case details

This 35-year-old woman presented with subtle sensation of incomplete voiding for more than four months. She had no flank pain or any irritative lower urinary tract symptoms. There was no previous history of recurrent urinary tract infection, nephrolithiasis, or gross hematuria. On physical examination, there were no specific findings. Serum creatinine was 1mg/dl, and the urine analysis was normal. Ultrasonography of the urinary bladder revealed an 18mm clearly *immobile* echogenic density on the right side of her bladder, and negligible post voiding residue. Abdominal and

pelvic computed tomography (CT) scan with and without intravenous contrast showed bilateral complete ureteral duplication and a bladder stone possibly within a right side ureterocele (Figure 1). Complete ureteral duplication could be seen on the CT images. There was no evidence of vesicoureteral reflux on voiding cystourethrography (VCUG) as shown in Figure 2.

In addition to revealing double ureteral orifices on each side, hence confirming complete duplex systems; cystoscopy immediately disclosed the cause of the aforementioned conspicuously fixed bladder stone. This became evident as the dumbbell shaped stone was found protruding beyond the ureterocele orifice into the bladder lumen (Figure 3a).

The suspicious papillary lesion at the base of the ureterocele (Figure 3b) was delicately resected, followed by using a Bugbee electrode to puncture the ureterocele at its most dependent part. We fragmented the stone using pneumatic lithotripsy through an 8 french semirigid ureteroscope and the fragments were removed by forceps. The patient was discharged on day one and pathology confirmed inflammatory nature of the papillary polyp.

Discussion

At estimated prevalence of 1 per cent, partial unilateral ureteral duplication seems to be a common congenital anomaly of the kidney and urinary tract. Bilateral complete ureteral duplication is six times less common than its unilateral form¹ and occurs more often in females.³ There are few reports of bilateral duplex system with ureterocele stone formation in the literature.⁴ Embryologically, development of two separate ureteral buds from a single mesonephric duct will lead to complete ureteral duplication.⁵ Whereas, branching of a single ureteral bud before reaching the metanephric blastema, may be the more likely cause for incomplete ureteral duplication or bifid ureter.⁶ Ureteroceles usually affect the upper pole moiety in a duplex system in children.⁷ Ureterocele related urinary stasis and infection may contribute to stone formation in 4 to 39 per cent of patients.^{4,8,9} Ureterocele stone formation is more common in adults, but there are rare case reports of its occurrence in children.¹⁰

Using antenatal sonographic monitoring, ureteroceles are diagnosed before or soon after the birth in more than 75 per cent of cases;⁷ otherwise, they may remain undetected until being incidentally discovered in adulthood.¹¹ Ureteroceles are asymptomatic in most cases,³ but can well

present with recurrent infection, flank pain, hematuria, stone formation and urinary incontinence in females.¹¹

In clinical practice, ultrasonography may reveal the duplex system and associated ureterocele. Currently the classic diagnostic tool of intravenous urography has been substituted by CT urography and magnetic resonance urography (MRU) which render further anatomical detail of the genitourinary tract and adjacent organs.¹² VCUG should be performed in all patients before intervention.¹ It can show the ureterocele, associated VUR and possibly signs of a weakened detrusor backing of the bladder wall, in which the ureterocele seems to evert during voiding similar to a bladder diverticulum.^{1,12,13} The function of involved upper pole moiety may best be evaluated using 99m-Technetium dimercapto-succinic acid (DMSA) scan.¹²

Symptomatic ureterocele causing flank pain, recurrent infection, stone formation, and obstruction with progressive loss of renal function on renal scan is absolute indication for surgical intervention.¹⁴ Otherwise, many patients can be managed conservatively.^{15,16} Surgery can be performed endoscopically or through the open approach with excision of the ureterocele and upper pole partial nephrectomy or common sheath reimplantation.¹⁷ Considering its success for intravesical ureteroceles, endoscopic intervention has become the initial choice for nearly two decades.¹⁸⁻²⁰ In case of failure, especially in ectopic ureterocele,²¹ repeat incision or puncture will be necessary.²² Choosing between endoscopic incision or puncture is still a matter of debate in the literature.⁹ Although it seems that both have comparable success rate,¹⁸ whenever an endoscopic puncture is chosen for a thick walled ureterocele, one should be mindful that a second puncture may become necessary.¹⁹ One major concern about endoscopic management is the possibility of causing iatrogenic reflux.

Management of the ureterocele stone depends mostly on its size. Smaller stones are amenable to endoscopic incision and lithotripsy, while larger ones have required combination therapy with shock wave lithotripsy (SWL), endoscopic incision, laser or pneumatic lithotripsy and even orificiotomy and ureterolithotomy.

Conclusion

Endoscopic treatment of duplex ureterocele stones is feasible, but diagnosing this problem correctly requires clinical vigilance and sufficient workup.

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PEER REVIEW

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CONFLICTS OF INTEREST

The authors declare that they have no competing interests.

PATIENT CONSENT

The authors, *Vahidi Serajoddin, Shadpour Pejman, Narimani Nima, Haghdani Saeid*, declare that:

- They have obtained written, informed consent for the publication of the details relating to the patient(s) in this report.
- All possible steps have been taken to safeguard the identity of the patient(s).
- This submission is compliant with the requirements of local research ethics committees.

Figure 1: CT scan with and without intravenous contrast

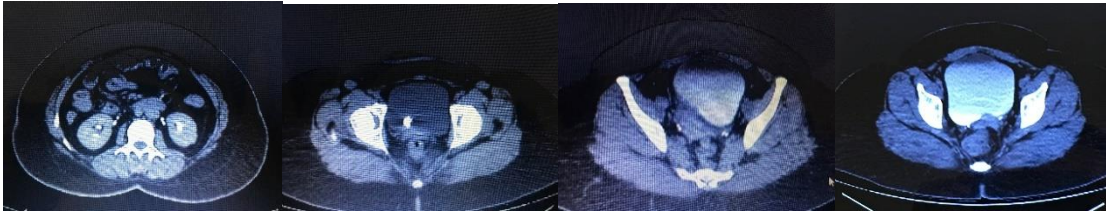


Figure 2: Voiding cystourethrogram displayed no reflux

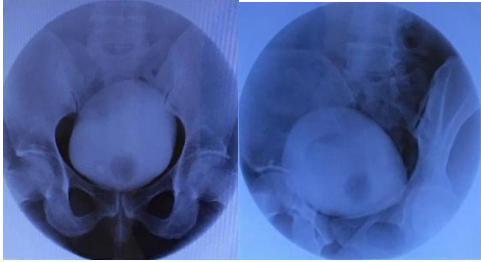


Figure 3: Cystoscopic view; a, ureterocele dumbbell stone protrusion; b, suspicious inflammatory polyp

