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CASE REPORT

Ureterocoele calculi masquerading as multiple vesical calculi with acute presentation: a case report

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Ureterocele refers to the cystic intramural ureteral out-pouching with variable presentation. Ureterocoele calculi are a recognised complication. In this case report, a patient presented acutely with this pathology. Other complications are urosepsis and uraemia. The ureterocele calculi can be unilateral or bilateral. It may be asymptomatic, requiring a high index of suspicion to diagnose, or symptomatic, and may masquerade as multiple vesical calculi during clinical evaluation. In asymptomatic patients with ureterocele calculi, symptoms may supervene due to stone impaction. This is a case report of a patient with single-system orthotopic ureterocele calculi and an impacted calculus manifesting with severe episodic and colicky suprapubic pain unabated by combined analgesic management. The patient had emergency bladder exploration, right-sided orthotopic ureterocele deroofing, and stone evacuation with complete symptom resolution.

This case highlights the importance of early consideration of a computerised tomography (CT) scan, if available and affordable, to minimise the diagnostic dilemma of urinary stone diseases and the relevance of ureterocele deroofing in selected patients based on clinical presentation.

Keywords: acute presentation, calculi, orthotopic ureterocele, open deroofing

Case report

The patient, a 37-year-old male farmer, presented with a history of intermittent passage of stones per urethra and dull but occasionally sharp suprapubic pain of one year's duration. The pain became progressively worse and more frequent. Three days before admission, he presented acutely with colicky right flank pain radiating to the medial aspect of the right thigh, with involvement of the suprapubic region. The pain was excruciatingly severe compared to earlier episodes, with the patient rolling on the floor, unabating, despite combined analgesic therapy, including opioids.

There was an associated history of storage lower urinary tract symptoms and haematuria. Before referral to our facility, the patient had visited several hospitals where he received antibiotics and analgesics with transient symptom relief. An earlier transabdominal ultrasound scan (USS) confirmed vesical calculus (Figure 1, A1). A physical examination revealed a young man, acutely ill-looking, in severe episodic painful distress. His temperature, respiratory rate, pulse rate, and blood pressure were 36.8 °C, 24 c/min, 96 b/min, and 130/90 mmHg, respectively. The abdomen was scaphoid with vague tenderness over the suprapubic region.

The urine cultured *Staphylococcus aureus*. Other laboratory tests were unremarkable. The transabdominal USS on admission at our facility reported normal kidneys with intravesical calculi (Figure 1, A2 and A3). The intravenous urogram (IVU) he had before acute presentation revealed normal kidney excretion with a cluster of multiple oval radio opacities in the region of the pelvic cavity, suggestive of multiple vesical calculi (Figure 1, A–C).

Given the persistent and unabating suprapubic pain despite analgesics, including opioids, the patient was counselled and had emergency bladder exploration, right-sided orthotopic ureterocele deroofing, and stone evacuation.

The intraoperative findings included a right dilated distal intramural ureter (orthotopic ureterocoele) with multiple (seven) calculi lodged inside the ureterocoele, with the largest impact at the vesicoureteric

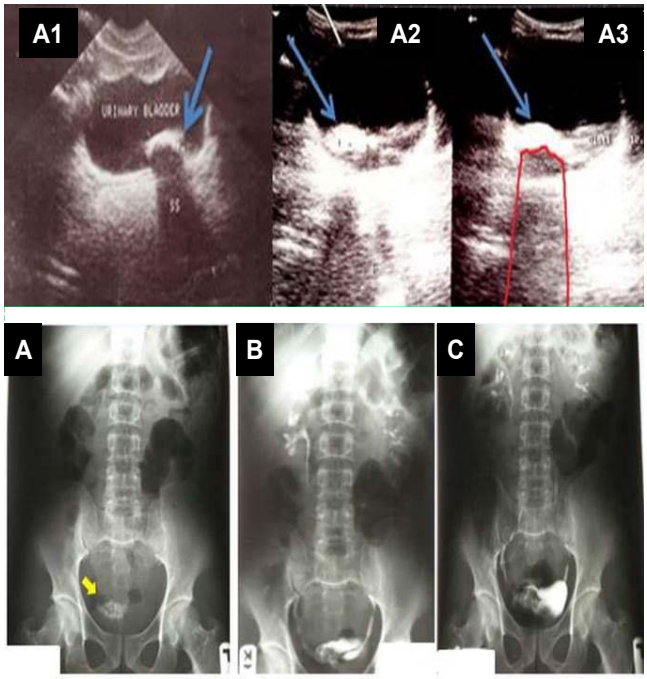

Figure 1: Abdominal USS and IVU findings
A1: Showing Intravesical calculus (blue arrow) extruded from ureterocele 6/12 earlier.
A2: Right ureterocele calculi (blue arrow) missed in earlier USS.
A3: Poorly defined posterior acoustic shadowing, area demarcated by pink line.
IVU Films - A: KUB showing stone clusters to the right of midline (ureterocele calculi). B and C: normal functioning kidneys (Image acquired at 10 & 30 minutes)
IVU – intravenous urogram, USS – ultrasound scan

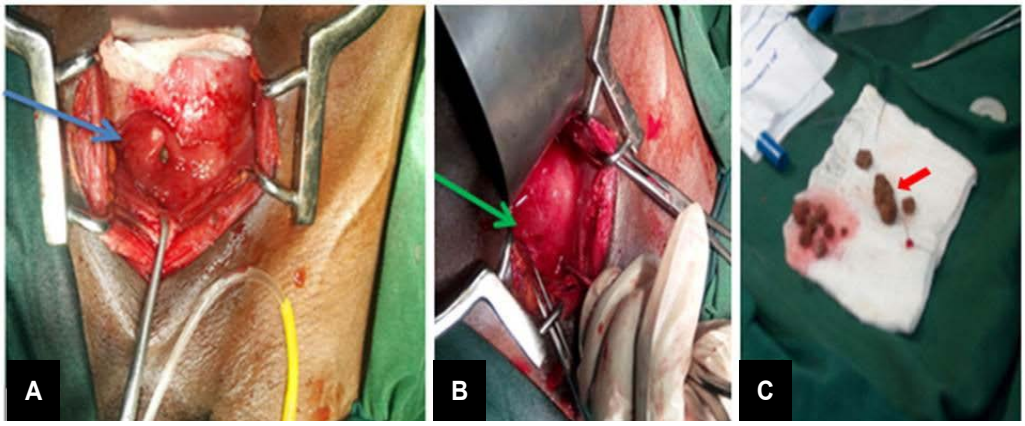

Figure 2: Intraoperative findings
A: Showing right uretrocele with a stone peeping from ureteric orifice (blue arrow)
B: Open deroofing of the ureterocele (green arrow)
C: Evacuated ureterocele & intravesical calculi with (impacted ureterocele stone - red arrow)

junction (Figure 2, A–C). Three more calculi were found within the bladder cavity with widespread inflamed bladder mucosa. The left ureter and orifice were relatively normal. The patient did well and was discharged home 10 days postoperatively with complete symptom resolution.

Discussion

A ureterocele is a cystic out-pouching of the distal ureter, primarily involving the intramural part. It is a rare congenital urologic anomaly that manifests in childhood and, occasionally, adulthood.[1] The index case is one of adult presentation at 37 years old. A ureterocele is variously classified based on its occurrence in a single or duplex pelvicalyceal system, location relative to the bladder cavity, among others, or a combination of these.[2] A single-system orthotopic ureterocele occurs in a ureter with a single pelvicalyceal system and is located within the bladder cavity.[3]

The ureterocoele diagnosis can be technically challenging and may be overlooked. The uncomplicated types are often asymptomatic and explain adulthood presentations of this pathology. The initial asymptomatic nature of the disease may be the reason for our index case presentation. The complicated type often presents early, irrespective of age. This condition, though congenital, remained largely asymptomatic in this patient from childhood to adulthood until it became complicated by calculi, a known complication of ureterocoele.[4] Despite the ureterocoele calculi, his initial visits to secondary health facilities led to considering bladder calculi from the transabdominal USS of the bladder, which confirmed bladder calculus and absent hydronephrosis. The absence of hydronephrosis could be due to the initial nonobstructive multiple right ureterocele calculi.

Moreover, this finding and the subsequent repeated per urethra passage of stones clearly indicated a non-contrast CT scan in the index patient. This is the gold standard imaging modality for diagnosing urinary stone disease. It is superior to the transabdominal USS in diagnosing stones in other difficult locations of the urinary tract, such as the ureter, which a transabdominal USS can overlook because of bowel gas. However, it is more expensive than the transabdominal USS and inaccessible or unavailable at most secondary health facilities in our setting.

A CT scan was not used in the index case because of the patient's poor socioeconomic background, compounded by out-of-pocket service costs. Similar problems have led to delayed presentation of vesical calculi from prolonged and neglected indwelling catheters, as reported earlier in our setting.[5] The repeated per urethra passage of stones and the additional development of intermittent mild suprapubic pain led to the patient's referral to our facility before his dramatic presentation following ureteral stone impaction. This further underscores the inadequacy of repeated transabdominal USSs in the diagnosis of ureterocele.

The tell-tale sign or Foley's sign of ureterocele on a transabdominal USS is the cyst-in-cyst appearance.[6] This sign was not classical or absent in the index patient's transabdominal USSs before the presentation. It is also interesting to note that the ureterocoele calculi visible on one of the transabdominal USSs were interpreted as multiple bladder calculi. This is not surprising because of the interobserver variation evident in USS reports. Also, the sonographer's experience cannot be overemphasised in clinching some difficult diagnoses.

Depending on the availability, other superior investigative modalities to transabdominal USS, such as IVU, CT urogram, and magnetic resonance imaging, may be employed to diagnose ureterocoele and ureterocoele calculi.[7] In the course of the patient's evaluation, he had an IVU, which characterised the pathology further and made the diagnosis of right-sided ureterocele calculi more apparent. Although not the obstructive type, the diagnosis of ureterocoele became clear from the cobra head sign on the contralateral lower ureter on the IVU image, supporting a bilateral type (Figure 1B).

The relative adynamic nature of the affected part of the lower ureter results in urinary stasis and the formation of ureterocoele calculi, as seen in the index patient. The other imaging modalities were not performed due to a lack of health insurance policy, more so due to the patient's dramatic presentation following ureteral stone impaction characterised by excruciating colicky suprapubic pain, which was unabating despite combined analgesics, including both nonsteroidal and steroidal types.

The patient's abdominal pain was dull to sharp and intermittent, initially localised to the suprapubic region, but later involved the right flank with radiation to the medial aspect of the right thigh and colicky shortly before surgery when one of the ureterocoele calculi became impacted and obstructive. Furthermore, ureteral inflammation caused by stones loaded with bacteria in the lower

ureter can produce referred pain, which may be dull or sharp to the suprapubic region according to a study on the distribution of ureteral pain conducted in 1938 by Ockerblad et al.[8] These authors described an area in the lower quadrant of the abdomen called the focal point of ureteral pain lying between McBurney's point and the midline, coinciding with the suprapubic region where pain from pathologies such as ureteral stones in the lower ureters are referred to.

Also, this ureterocoele was not obstructed when the initial IVU was done, as evidenced by the absence of a back pressure effect on the ipsilateral kidney (absence of hydronephrosis). Probably, if a repeat IVU or abdominal USS was done during the 72 hours of acute presentation with the severe colicky pain before admission, hydronephrosis may be evident because of the obstruction by the impacted stone.

Furthermore, a urethrocystoscopy is valuable in this patient's management because of its diagnostic and therapeutic benefits. These include diagnosing possible intravesical causes of bladder stones, like urethral stricture, and offering the advantage of assessing the feasibility of endoscopic ureterocoele stone deroofing. The facility for cystoscopy was available, but it was not done because, at the time this case was managed, we had no complete instrument set or equipment for endoscopic deroofing.

In addition, a urethrogram would be helpful to demonstrate the presence of urethral stricture but was not required because a size 18 FR silicone urethral catheter was successfully passed without resistance. Urethral calibration was done intraoperatively with bougies greater than 18 FR without resistance. This convinced us that the smaller stones seen earlier on abdominal USS were extruded into the bladder cavity from the right unobstructed ureterocoele.

The other causes of acute presentation can result from complications such as urosepsis and uraemia.[9] The presence of ureterocoele calculi is conducive to the development of urosepsis. In addition to analgesics, the patient had antibiotic treatment. There were no clinical, biochemical, or imaging features to suggest uraemia or its risk in the patient. Primarily, when symptomatic, the treatment of ureterocoele is mainly surgical and ranges from minimally invasive to invasive methods.[4] Treatment should be individualised and account for the surgeon's experience, patient and disease peculiarities, the treatment cost, and the availability of facilities for minimally invasive therapy. Based on these considerations, emergency bladder exploration, right-sided orthotopic ureterocoele deroofing, and stone evacuation were chosen for the index patient.

Notably, the index case stone analysis or 24-hour urine for stone studies of calcium is a part of standard care for his disease. Unfortunately, it was not done due to costs at our facility. This reason constitutes significant limitations to several required investigations during patient management. A key concern or complication of the treatment offered to this patient is vesicoureteric reflux and urinary tract infections. The patient had short-term follow-up, but there was no clinical evidence of these complications. He subsequently became inconsistent with follow-up visits due to the long distance and high cost of transportation to our facility. However, the patient was asymptomatic after surgery on several phone calls.

Conclusion

Ureterocele management may be attended with diagnostic and therapeutic challenges. The single-system orthotopic ureterocele calculi can be asymptomatic or symptomatic and may mimic ureteric colic or masquerade as multiple vesical calculi. The symptomatic presentation or aggravation of pain should raise suspicion of stone impaction at the vesicoureteric junction. Proper history, supported by imaging investigation, is essential, and open ureterocele deroofing in selected patients can be rewarding in such situations.

Conflict of interest

The authors declare no conflict of interest.

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Ethical approval

Ethical approval was obtained from the Health Research Ethics Committee, Usmanu Danfodiyo University Teaching Hospital, Sokoto (UDUTH/HREC/1466/V1).

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