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Case Report

Dual Challenge of a Cecoureterocele with Calculus: A rare case report $^{\bigstar, \bigstar \bigstar}$

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ABSTRACT

Ureteroceles are the most prevalent urinary tract malformations in humans. Only 5% of ureteroceles are predicted to prolapse and it usually occurs in childhood. We outline the clinical history, radiological results, and a potential course of treatment for this challenging condition. A 32-year-old female checked herself into our institution with complaints of burning urination and 20 years of complaints of urethral ballooning when urinating. Initial sonographic evaluation revealed that at the left vesicoureteric junction, a cystic lesion extends into the bladder, with a hyperechoic focus causing posterior acoustic shadowing. CT scan confirmed the diagnosis of an ureterocele with calculus. A voiding cystourethrogram revealed a left-sided ureterocele that descends down the urethra and into the interlabial region. CT cystogram verified the presence of a left-sided cecoureterocele with calculus. Cecoureterocele is a rare variant of ectopic ureteroceles. Girls experience this condition more frequently than boys, and they are predisposed to vesicoureteric reflux and recurrent infections. To prevent problems like renal function loss, recurrent urinary tract infections, and urinary incontinence, it is important to gain diagnostic confirmation of these circumstances. Less invasive surgical techniques like endoscopic ureterocele puncture or even nonoperative treatment appear to produce comparable functional outcomes. When a patient arrives with a urethral protrusion, one should be extra cautious. In this case report, a cecoureterocele that has prolapsed is presented in a rare way. It presents an important chance to evaluate the clinical and diagnostic characteristics of this urinary tract abnormality.

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Abbreviations: CT, computed tomography; IVU, intravenous urography; MRI, magnetic resonance imaging; UTI, urinary tract infection; VCUG, voiding cystourethrography; VR, volume rendering.

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Introduction

Ureteroceles are among the most prevalent urinary tract malformations in humans, occurring in 1/5000 to 1/12,000 births. Most often, urologists who serve mostly adults are less likely to diagnose them; instead, pediatricians who specialize in treating children are more likely to do so. Additionally, the anticipated prolapse rate for ureteroceles is just 5%, and after that, it usually occurs in the initial years of childhood, according to research [1]. Only a few case reports are available in literature like our current situation, in which a cecoureterocele with calculus was initially discovered in an adult patient.

Case report

For a urological consultation, a 32-year-old lady checked herself into our institution. She gave details about recent episodes of burning micturition and 20 years of complaints of urethral ballooning when urinating. During the examination, a reducible mucosa-covered mass that protruded from the urethra during the Valsalva maneuver or while micturating was found (Fig. 1). The initial sonographic evaluation revealed a cystic lesion at the left vesicoureteric junction that was extending into the bladder, with a hyperechoic focus seen within which was causing posterior acoustic shadowing, giving a diagnosis of ureterocele with calculus (Fig. 2). We performed a computed tomography scan of the patient's abdomen and pelvis to confirm our suspicion that the patient had an ureterocele. The presumed large left ureterocele with calculus was verified by the CT scan, along with a consequentially dilated left ureter and renal pelvis (Fig. 3). No overt indication of a duplex pelvic system was seen.

Fig. 1 – Reducible mucosa-covered mass protruding through the interlabial region during the Valsalva maneuver.



Fig. 2 – Ultrasonography showing a cystic lesion projecting into the bladder at left vesicoureteric junction, with a hyperechoic focus causing posterior acoustic shadowing (calculus).



Fig. 3 – Axial noncontrast CT of the pelvis shows left ureterocele with calculus.

Later, the patient underwent an intravenous urography (IVU) and a voiding cystourethrography (VCUG) on the same day. An initial IVU scout image showed an enlarged right transverse process of L5 vertebra with pseudo-articulation with the sacrum (Castellvi type IIa) and a well-defined radio opaque focus seen about 5.4 cm medial to left acetabulum in the pelvis (Fig. 4). Subsequent series of images taken revealed a dilated left pelvicalyceal system with a dilated and tortuous left ureter having a large rounded dilated distal part projecting into the lumen of the bladder with surrounding radiolucent halo (cobra head sign) causing upstream moderate



Fig. 4 – IVU scout image shows right sided lumbo-sacral transitional vertebra (Castellvi type IIA) and radio-opaque focus (likely calculus) in the region of pelvis.

hydroureteronephrosis. The right ureter was dilated and tortuous, with numerous degrees of luminal caliber changes, and right pelvicalyceal system appeared prominent (Fig. 5).

A properly executed voiding cystourethrogram (VCUG) made the issue quite evident. During the voiding phase, we were able to see the bladder neck within the pelvic brim and the round, contrast-filled, smooth, blind-ending saccular outpouching extending 5.7 cm beyond the lower margin of the pubic symphysis into the interlabial region (Fig. 6). The findings of a CT cystogram verified the presence of a left-sided ureterocele that descends down the urethra and into the interlabial region, where it manifests as a thin-walled, blind-ending



Fig. 6 – VCUG shows round, contrast-filled, smooth and blind-ending saccular outpouching extending beyond the lower margin of the pubic symphysis into the interlabial region.

cystic structure. Descent of the calculus which was seen within the ureterocele was also noted (Figs. 7 and 8).

Clinically asymptomatic and with renal parameters that were within normal limits, we decided to treat our patient with endoscopic excision after a cystoscopy and a transurethral incision. Parts were painted and draped while being handled with aseptic measures and spinal anesthesia. Cystitis with grade I trabeculations was seen, along with a large ureterocele that blocks the bladder outlet. Visualization



Fig. 5 - IVU shows left sided ureterocele with upstream moderate hydroureteronephrosis.



Fig. 7 – Axial CT cystogram (excretory phase) and 3D coronal VR images without Valsalva maneuver shows left sided ureterocele with calculus and hydroureteronephrosis.



Fig. 8 – Sagittal and coronal reformatted CT cystogram (excretory phase) and 3D VR images with Valsalva maneuver shows left-sided cecoureterocele with calculus (red arrow).

of the right ureteric orifice revealed mild ureterocele. Left ureteric orifice was visible on the medial wall of the cecoureterocele [2]. Ureteric catheter was inserted under C-ARM guidance, and coiling was observed inside the cecoureterocele. Moderate hydroureteronephrosis was seen after a retrograde pyelography. A laser-assisted inverted T- shape incision was made over the cecoureterocele, and a 10 mm calculus was basketed out completely before the cecoureterocele was completely excised and sent for histological analysis. A thin rim of cecoureterocele mucosa was left surrounding the intact ureteric opening. The procedure went smoothly. It is important to keep a check on renal function, symptoms, and whether these patients have vesicoureteric reflux, especially in those who were treated using an endoscopic procedure or a straightforward open incision.

Discussion

A cecoureterocele is a relatively uncommon variant of ectopic ureteroceles in which the affected ureter's orifice is inside the bladder but the urethral cavity of the ureteroceles extends past the bladder neck [3]. During embryological development, ectopic ureteral orifices may be neighboring with the Wolffian and Müllerian ducts as they move caudally. A point on or close to the vestibule may be chosen for the orifice. A cecoureterocele can be produced when the distal wall of an ureterocele is dragged caudally along the urethra when it is continuous with the Wolffian and Müllerian ducts during the time of migration. Girls experience ectopic ureteroceles more frequently than boys. They are predisposed to vesicoureteric reflux and recurrent infections. Ureteroceles may develop calculi as a result of urine stasis [4]. Given the rarity of prolapsed cecoureterocele, especially in adults, it is challenging to make a therapeutic suggestion based on scientific research [1]. Nonetheless, preventing problems like renal function loss, recurrent urinary tract infections, and urine incontinence should always be the main objective. Due to the anatomically complex situations that need to be addressed while treating a patient with an ureterocele, obtaining diagnostic confirmation of these situations should be the initial action in approaching the patient's care. Radiologic modalities including ultrasonography, CT/MRI scans, and common procedures like IVU and VCUG should be selected for each patient individually in order to accomplish this clearance. Post operatively, our patient has been asymptomatic in all of the follow-up exams thus far. We advise treating cecoureterocele instances in adults who have been asymptomatic for most of their lives conservatively in the beginning, after ruling out any aggravating factors. However, followups are necessary so that surgical intervention may be done as soon as there are any difficulties with conservative treatment. The paradigm of primary urinary tract reconstruction in ureterocele patients has just begun to change, even when it comes to young children and newborns. Less invasive surgical techniques like ureterocele puncture under an endoscope or even non-operative treatment appear to produce comparable functional outcomes. Therefore, it is important to consider each patient's unique diagnostic results while deciding on their specific course of treatment.

Conclusion

Females who have a prolapsed cecoureterocele may experience acute urinary retention and burning micturition. When a patient arrives with an accompanying urethral protrusion, one should be extra cautious. Only a few cases of cecoureterocele in adulthood describe patients who present with the cecoureterocele protruding through the external genitalia [5]. These accounts mostly include women, which is more likely due to the short female urethra. Endoscopic therapy is most frequently used to treat these patients. In this case report, a cecoureterocele that has prolapsed is presented in a rare way. It presents an important chance to evaluate the clinical and diagnostic characteristics of this urinary tract abnormality [1].

Declaration of generative AI and AI-assisted technologies in the writing process

During the preparation of this work, OpenAI was used in order to improve language and readability. After using this tool/service, the author(s) reviewed and edited the content as needed and take full responsibility for the content of the publication.

Patient consent

The authors certify that they have obtained all appropriate patient consent.

REFERENCES

- Florian L, Preusser S, Mark M. A prolapsed cecoureterocele in an adult treated conservatively: highly rare, but existent. Case Rep Urol 2016;2016:1–3. doi:10.1155/2016/5049072.
- [2] Parmar KM, Sharma P, Mandal S, Kumar S. Ureterocele with impacted stone. BMJ Case Rep 2020;13(7):e236301. doi:10.1136/bcr-2020-236301.
- [3] Keays MA, Keefe D, Braga LH. Chapter 1—definition, classification, and embryology. In: Leonard M, Guerra L, editors. Ureteroceles: contemporary diagnosis and management. Academic Press; 2020. p. 1–16. doi:10.1016/B978-0-12-817888-1.00001-X.
- [4] Blumenthal I. Vesicoureteric reflux and urinary tract infection in children. Postgrad Med J 2006;82(963):31–5. doi:10.1136/pgmj.2005.036327.
- [5] Czarniecki M, Barrett T, Thiruchelvam N, Wiseman O. Case 267: cecureterocoele manifesting in a man with infertility. Radiology 2019;292(1):263–6. doi:10.1148/radiol.2019162781.