Case Report

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Rare presentation of huge ectopic ureterocele in an adult female: a case report

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Introduction and importance: Ectopic ureteroceles are primarily found in children, often detected incidentally during antenatal ultrasonography or due to urinary tract infection (UTI) symptoms. However, they are rare in adults, with limited published cases.
Case presentation: This report details a case of a 24-year-old woman who experienced recurrent UTIs and sudden urinary retention, ultimately needing manual compression to urinate due to poor urine flow. Intravenous urography revealed a large right ectopic ureterocele that protruded through the urethra during urination. Cystoscopy confirmed extensive right-sided ureteroceles affecting the bladder and causing her urinary difficulties.
Clinical discussion: Although ectopic ureteroceles in adults can present with a range of symptoms, including obstruction and recurrent infections, the management approach is often individualized based on the clinical presentation and imaging findings. In this case, endoscopic incision was chosen for its minimally invasive nature, leading to full recovery without complications. Despite the risks of recurrence and potential scarring, the patient showed no recurrence at follow-up and remains symptom-free.
Conclusion: This case underscores the rarity of symptomatic giant ectopic ureteroceles in adult females and highlights the importance of considering this condition in women with recurrent UTIs.

Keywords: cystoscopy, ectopic ureterocele, recurrent urinary tract infections, retention of urine

Introduction

Ectopic ureterocele is an abnormal condition in which a tube carrying urine from the kidney does not reach the bladder. Instead, it drains into other parts of the urogenital system, such as the vagina, urethra, or even the abdominal cavity. The incidence of ureteroceles identified during autopsy is estimated to be as high as 1 in 500. Ureteroceles are notably more prevalent in females, with a ratio of four to seven times compared to males, and they occur bilaterally in ~10% of cases. Orthotopic ureteroceles are positioned at the normal location of the uretero-vesicular junction and tend to be less likely asymptomatic, often discovered incidentally in adults.

HIGHLIGHTS

- This case report describes an exceedingly rare instance of a giant ectopic ureterocele in a 24-year-old woman, a condition typically found in children.
- The patient presented with recurrent urinary tract infections (UTIs) and acute urinary retention, requiring manual abdominal compression to urinate due to poor flow.
- Diagnosis was confirmed via intravenous urography and cystoscopy, revealing a large ectopic ureterocele in the right ureter, which protruded through the urethra during urination.
- The patient underwent an endoscopic incision of the ureterocele with the placement of a double J stent, resulting in significant symptom improvement and resolution of the ureterocele.
- Postoperative follow-up at 6 months demonstrated complete recovery without recurrence, emphasizing the importance of considering giant ureteroceles in adult women with recurrent UTIs.

In contrast, children more commonly present with ectopic ureteroceles, which are frequently associated with secondary symptoms. These ectopic ureteroceles are typically linked to the upper pole moiety of a duplex collecting system. The most common complication arising from an ectopic ureterocele is a urinary tract infection, which often results from obstruction. Due to the rarity of the cases, the incidence in adults is not reported[1]. This condition can lead to frequent urinary infections, kidney damage, and kidney stones. It is more common in female fetuses and is typically associated with one of three variants of an accessory

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ureter, a partial double ureter, or a nondouble ureter[2]. The diagnosis of this condition is typically done via ultrasound ima ging and can be further confirmed via excretory urography, voiding cystography, or other imaging techniques[3]. In this study, we reported a case of an ectopic huge ureterocele in an adult female involving the whole bladder. An endoscopic incision of the ureterocele was performed, and the patient recovered uneventfully.

This work has been reported in line with the Surgical CAse REport (SCARE) 2023 criteria[4].

Case presentation

A 24-year-old female presented with a 6-month history of decreased urinary stream, urgency, and frequency. The patient was otherwise healthy and had no significant medical history. Her physical examination revealed suprapubic fullness. Initial laboratory tests, including complete blood count, liver function tests, renal function tests, and coagulation profile, were within normal limits; however, numerous pus cells were seen on complete urine examination. An ultrasound scan of her pelvis demonstrated a 7 cm cystic outpouching involving the right ureter that was filling the whole bladder lumen (Fig. 1A). Intravenous urography revealed similar findings (Fig. 1B). Cystoscopy revealed a huge ureterocele protruding into the urethra, and the ureteral opening was not located at its typical anatomical position; instead, it was positioned anomalously (ectopic) (Fig. 1C). Based on the imaging findings and cystoscopic examination, a huge ectopic ureterocele in an adult female was diagnosed. The decision to incise ureterocele endoscopically was planned on clinical judgment after thoroughly evaluating the patient's condition.

We engaged in detailed discussions with the patient about the potential risks and benefits, emphasizing the option for minimally invasive surgery by the patient and explaining the possibility of requiring further procedures. In managing the patient, we opted for an incision at the base of the ureterocele with a resectoscope and hook knife, followed by stabilization through the placement of a double J stent, which was subsequently removed 6 weeks later. This tailored approach allowed us to address the patient's unique circumstances while ensuring informed consent and prioritizing their overall well-being (Fig. 1D). Postoperatively, the patient was doing well, and her symptoms improved significantly. After 6 months of follow-up, cystoscopic examination showed complete resolution of the ureterocele with no evidence of residual or recurrent lesions. The patient was advised to undergo long-term follow-up to monitor for relapse. Her voiding pattern returned to normal without any significant urinary complaints. The patient was satisfied with the treatment received. This case demonstrates that with timely diagnosis and proper management, even large ectopic ureteroceles can be managed successfully in adults without any adverse effects on quality of life.

Discussion

The prevalence of ureteroceles is four times greater in women, especially on the left side, and they are often associated with other abnormalities, such as stenotic ureteric orifices or duplex upper tracts[5]. Despite speculation that it is congenital in children and acquired in adults, the causes of this condition are still unknown[6]. The distinction between a simple ureterocele and an ectopic ureterocele has been outlined in multiple studies, which emphasize that the defining characteristic is not solely reliant on the appearance of the cystic structure but also the anomalous drainage pattern of the ureter. They assert that any ureterocele associated with an ectopic ureter should be classified as such irrespective of its size or protrusion characteristics, provided there is supporting evidence of abnormal ureteral insertion[7].

Ureteroceles may be found either in the bladder (intravesical) or outside of it, in the bladder neck or urethra (ectopic)[8]. Generally speaking, ectopic types are more common than intravesical. In children, they often appear as part of a duplicated collecting system situated extravesical, while adult ones typically appear unilaterally and within the bladder[9]. In our case report, the ureteral opening was not located at its typical anatomical position; instead, it was positioned anomalously. This case pre sents an exception: an adult woman with a giant ectopic ureter ocele on the right side that protrudes through the urethra off and on. The imaging studies, including intravenous urography and cystoscopy, indicated that the ureteric orifice for the right ureter was positioned anomalously, contributing to the development of the ectopic ureterocele. Although the ectopic ureterocele was large and protruded through the external urethral orifice, its classification remains ectopic due to its origins and anatomical abnormalities discussed in the literature[1]. The fact that it pro trudes does not negate its classification as ectopic, as the under lying cause—a ureter draining from an abnormal location—remains. The upper renal moiety of the duplex system is typically affected by stenosis and obstruction of the ureter, resulting in hydroureteronephrosis ipsilateral to the ureterocele. As a result of vesicoureteral reflux, hydronephrosis occurs in the lower moiety, resulting in morbidities such as recurrent UTIs and chronic pyelonephritis[10]. As in our case, all these symptoms were present.

Ureterocele can vary greatly in clinical presentation. In children, they may present with recurrent urinary tract infections, urosepsis, incontinence, failure to thrive, urinary calculus, and abdominal masses. Although adults often have no symptoms, the diagnosis typically occurs by chance. Although infrequent, obstruction of the urinary tract for adult patients with duplexed kidneys is possible and may accompany intermittent pelvic pain, urinary tract infections, and calculi[11]. Our patient, a 24-year-old female adult, exhibited recurrent urinary tract infections, right flank pain, and an object emerging from their urethra during voiding, followed by retention of urine. Hydroureteronephrosis and a giant ectopic ureterocele were ultimately diagnosed.

Ultrasound is the primary imaging technique for the diagnosis of ureteroceles and can identify cystic enlargement of the bladder wall as well as duplication of structure[12]. If decreased renal function is suspected, an intravenous urogram should be performed to check for the absence or delayed excretion of contrast medium[6]. Renal scintigraphy is useful in detecting scar tissue, determining functionality at the upper and lower poles, and aid ing in deciding upon therapy. Ectopic ureteroceles are mostly linked to a nonfunctional upper pole in duplex kidney systems and appear darkly on radiographic images[12]. The 'drooping lily' sign may also be observed, indicating hydronephrosis has shifted the upper pole downwards and forced the lower, functioning pole laterally and inferiorly.

Depending on the patient's age, type of ureterocele, and other clinical factors that contribute to ureterocele management's best choice, ureterocele management should be individualized.

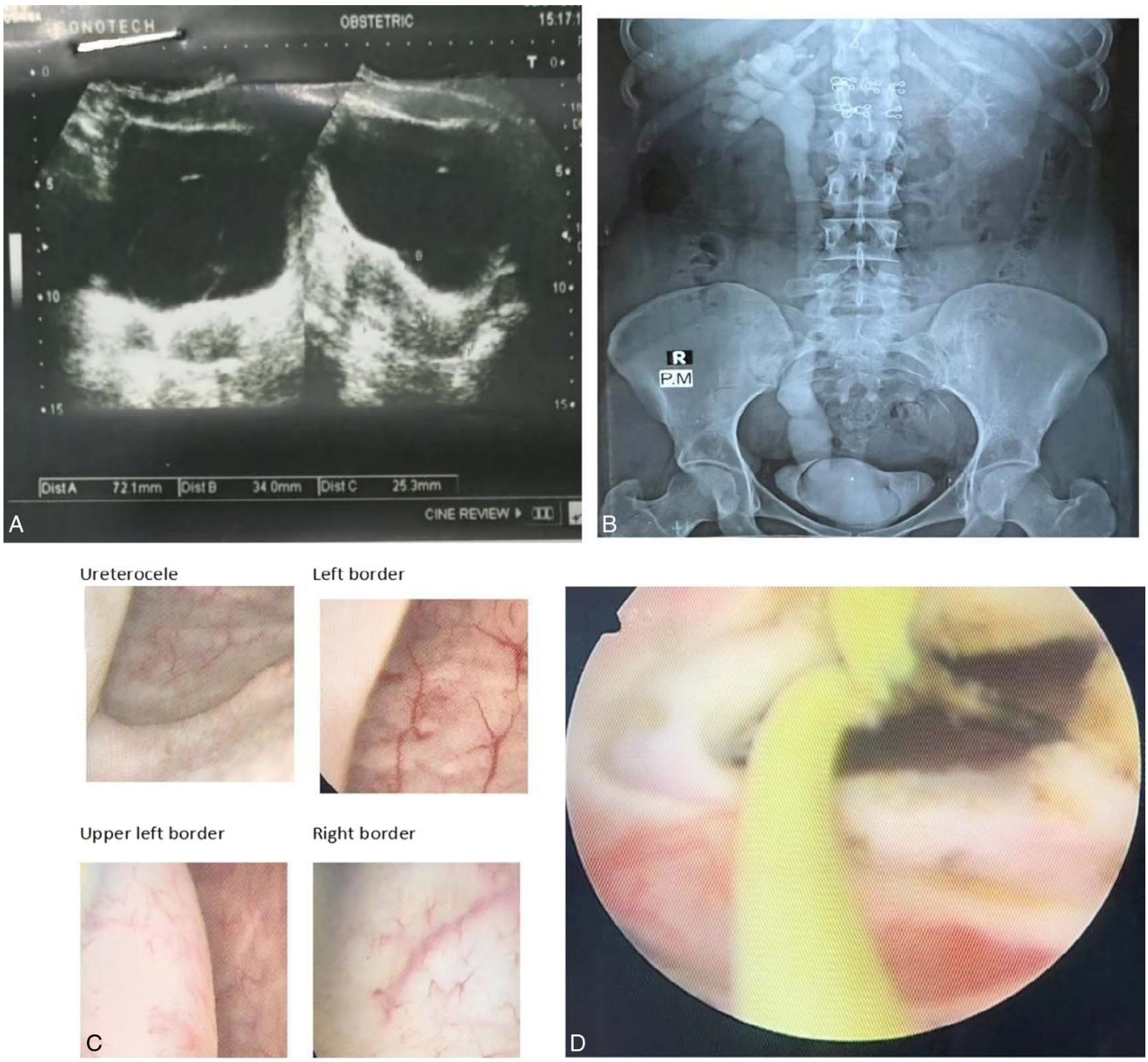

Figure 1. (A): Ultrasound KUB showing a huge 7 cm outpouching of ureterocele. (B): Intravenous urography showing a huge ureterocele on the right side. (C): Cystoscopic findings of ureterocele. (D): Incision of ureterocele.

Endoscopic treatment is simple and minimally invasive, but its effectiveness for treating ectopic ureteroceles remains unclear. After endoscopic treatment, 50–80% of ectopic ureteroceles require secondary surgery[12]. The latest findings showed that clinical success was achieved using the lower urinary tract reconstruction with no need for reoperation in 90.2% of patients with duplex system ectopic ureterocele[13]. Yoshimura *et al.*[14] reported a case of a 30-year-old male with an ectopic ureterocele of 3.5 cm that was managed by endoscopic excision successfully. In our case report patient, we opted for an endoscopic incision on the patient's consent for a minimally invasive procedure and complete resolution was achieved successfully.

Current studies have demonstrated an elevated risk of reoperation for extravesical ureteroceles compared to those intravesical. Despite this, many medical professionals concur that endoscopic puncture is mostly used to combat symptoms such as uncontrolled sepsis, azotemia, and obstruction of the bladder outlet, regardless of whether there is a prolapsed ureteroscope or not[8]. In our case report, the patient was having intravesical ectopic ureterocele, not azotemic, but was experiencing obstruction of bladder outlet. It is a rare condition in adults; to the best of our knowledge, this is the largest reported case in the literature. The patient had a previous history of recurrent urinary tract infections. She was being treated according to urinary tract infection guidelines until she presented with a protrusion of something out of the bladder during micturition. Imaging showed a huge ureterocele filling her entire bladder. She underwent surgery, and a ureterocele was incised endoscopically using a

resectoscope with a hook knife successfully. We acknowledge that the decision to incise the ureterocele may carry inherent risks, including potential scarring, recurrence of the ureterocele, and changes in bladder dynamics, but the patient made an uneventful recovery and was discharged home on the third postoperative day.

Conclusion

According to prior medical studies, ectopic types of ureteroceles are typically observed in children. This case report contributes to the literature by documenting a rare presentation of a giant ectopic ureterocele in an adult female, emphasizing the role of proper diagnosis and timely surgical intervention in achieving successful outcomes in affected individuals.

Ethical approval

Patient anonymity is maintained throughout this manuscript, and consent was obtained for publication from the patient.

Consent

Written informed consent was obtained from the patient's parent for the publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

M.D.J.: concept, manuscript edit and review, and guarantor; A.A.: manuscript preparation, edit, and review; H.A.: manuscript preparation, edit, and review; Z.W.: data collection and obtaining consent from the patient; M.A.: manuscript preparation, edit, and review; H.S.: manuscript preparation, edit, and review; M.H.A.: manuscript preparation, edit, review, and supervision.

Conflicts of interest disclosure

The authors declare no conflicts of interest.

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