Letter to the Editor

Unilateral complete ureteral duplication with ectopic ureteral opening inserting into urethra in a female patient without incontinence: a case description and review of the literature

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Introduction

Ectopic ureteral openings are often associated with duplicated kidneys (1). Conventional abdominal ultrasound, computed tomography (CT), and magnetic resonance imaging (MRI) (2) are useful for diagnosing duplicated kidneys. The identification of ectopic ureteral openings in adults is primarily achieved through cystoscopy, retrograde ureteropyelography (3), magnetic resonance urography (MRU) (4), and computed tomography urography (CTU) (5). Reports on the use of dual-plane transducers for diagnosing ectopic ureteral openings are scarce. This case report adds valuable information to the research in this area.

Case presentation

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this article and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Patient history

The patient, a 26-year-old female, presented to the hospital on 19 April 2023 following an artificial abortion 2 months prior. She reported recurrent urinary tract infections (UTIs), but maintained regular menstrual periods, with no similar family history of such conditions.

Physical examination findings

The patient's vital signs were normal, and she appeared in good general condition. Her abdomen was flat and soft, with no pressure pain on palpation.

Gynecological examination

Normal vulva, white vaginal douche-like discharge, a light cervix, and a normally sized uterus with no tenderness; the adnexa were unremarkable on both sides.

Laboratory findings

Renal function tests indicated normal urea and serum creatinine levels. The urine analysis results are detailed in *Table 1*.

Imaging findings

Transvaginal ultrasound revealed a tubular cystic mass on the posterior left side of the bladder, located between the

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Table 1 Urine analysis

Urine laboratory test	Patient level	Unit	Reference range
Color	Yellowish		Yellowish
Transparency	Turbidity		Clear and transparent
Urinary pH	6.0		4.5–8.0
U-NIT	(–)		(–)
U-GLU	(–)		(–)
U-PRO	+++		(–)
U-KET	(–)		(–)
U-BIL	(–)		(–)
U-SG	1.031↑		1.003–1.030
U-UBG	(–)		(–)
Urine leucocyte	+++		(–)
Urine latent blood	++		(–)
White blood cell	38,247.0	/μL	0–36
Red blood cell	360.8	/μL	0–27
Epithelial cell	105.3	/μL	0.4
Urine cast	42.97	/μL	0–3
Mirror-white blood cell	26,000	/μL	0–12
Mirror-red blood cell	50↑	/μL	0–9
Mirror-epithelial cell	5–10	/HighPowerField	0–4
Urine-urinary chorionic gonadotropin	(–)		

+, positive; –, negative; +++, strongly positive; ++, moderately positive. ↑, the number is elevated and increased compared to reference values. U-NIT, urinary nitrite; U-GLU, urinary glucose; U-PRO, urinary protein; U-KET, urine ketones; U-BIL, urinary bilirubin; U-SG, urine specific gravity; U-UBG, urobilinogen.

urethra and the anterior vaginal wall. This mass appeared tortuous, and hypoechoic, measuring approximately 5.3×3.0×4.7 cm (*Figure 1*). The initial diagnosis suggested the possibility of hydrosalpinx. After receiving anti-inflammatory treatment, the patient underwent a follow-up on 19 July 2023. The abdominal ultrasonography indicated an ipsilaterally duplicated left kidney with an enlarged volume, duplicated collecting systems, and duplicated renal vessels. The left upper collecting system appeared dilated (*Figure 2*), whereas the right kidney's parenchyma and size remained normal. Concurrently, transperineal ultrasound examination detected a cystic mass in the posterior left side of the bladder. Considering the patient's medical history, the diagnosis considered was a left-sided ectopic ureteral opening associated with a ureterovaginal fistula (*Figure 3*). To pinpoint the location of this ectopic ureteral opening, the patient underwent a rectal examination using a dual-plane probe on 1 August 2023. The examination showed that the left ureter descended posteriorly, then turned horizontally to the right, and finally opened above the sphincter of the posterior urethral wall (*Figure 4*).

Pelvic MRI indicated a convoluted tubular structure adjacent to the left side of the uterus, suggesting a benign lesion and supporting the possibility of left-sided hydrosalpinx (*Figure 5*). Further review of the patient's medical history revealed that, on 19 March 2022, she had undergone a CTU examination, which showed normal right ureter (*Figure 6*) with complete ureteral duplication on the left side. Additionally, the ureter that connected to the renal pelvis at a higher level was found to be dilated from the upper to the lower part (*Figure 7*).

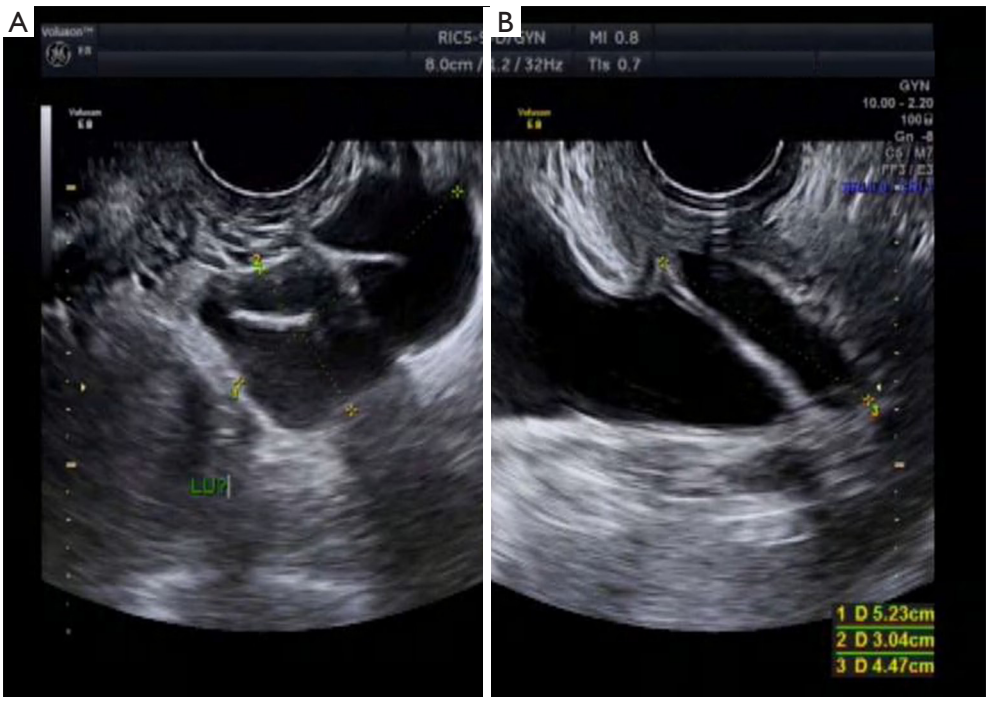

Figure 1 A cystically dilated duct travelling along the lower left side of the bladder with an opening posterior to the internal urethral. (A) Sagittal view, (B) transverse view. D, dimension.

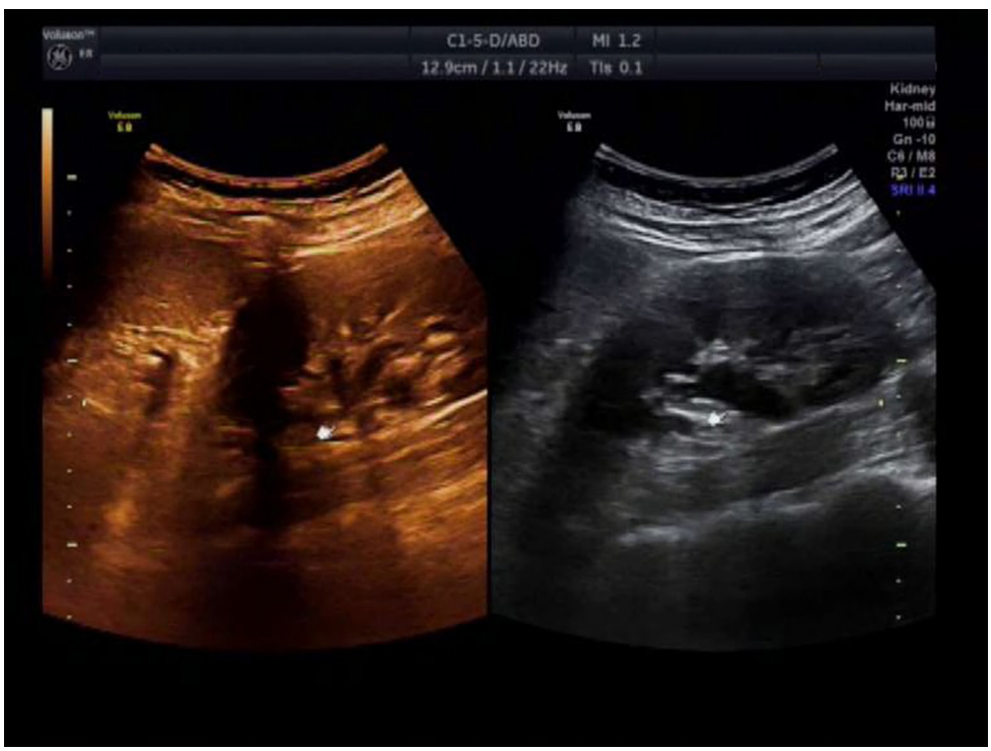

Figure 2 Sagittal ultrasound of the left duplicate kidney with 2 separate sets of ureters. The left image is pseudo colored yellow, and it shows a normal renal pelvis. The right image shows a dilated pyeloureteric junction.

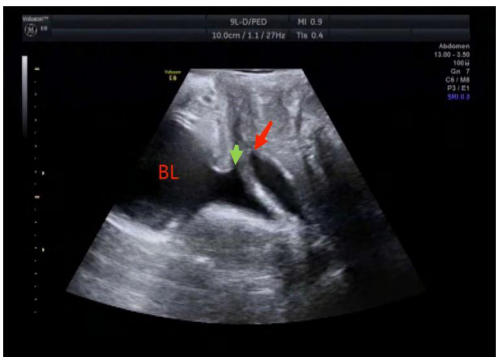

Figure 3 Sagittal transperineal ultrasound: the left ureter opening in the posterior urethral wall. The green arrow indicates the normal internal urethral orifice. The red arrow shows the end of the dilated ureter, suspected to terminate between the posterior urethral wall and the vagina. BL, bladder.

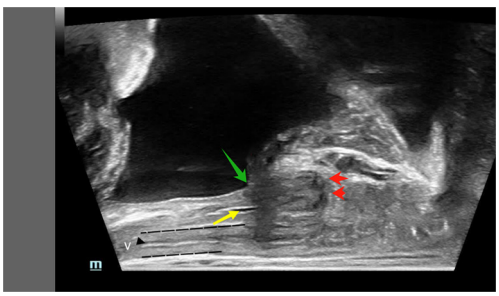

Figure 4 Transrectal ultrasound with a biplane probe showing the opening of the left ureter above the sphincter of the posterior urethral wall. Between the 2 black dotted lines: vagina. Black arrow shows the vaginal gas line. The red arrows show the urethral sphincter. The green arrow shows the internal urethral orifice. The yellow arrow shows the opening of the left ureter ectopically inserted into posterior urethral wall.

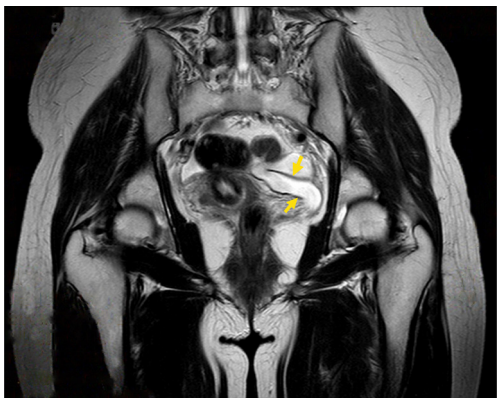

Figure 5 Coronal moderately T2-weighted magnetic resonance imaging showing dilation of the left ureter (shown by the yellow arrows).

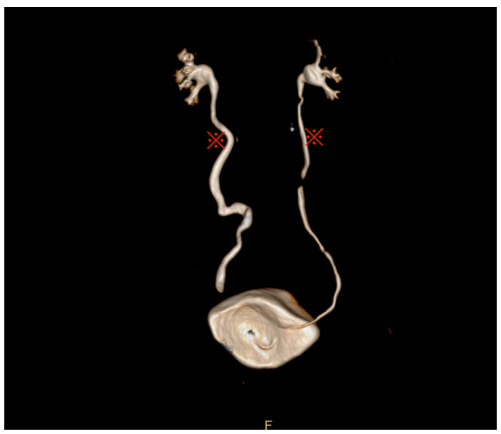

Figure 6 3D reconstruction of computed tomography urography showing the normally traveled ureters on both sides (※). 3D, 3-dimensional.

Treatment and follow-up

Recently, the patient expressed a desire to prepare for pregnancy. After discussing with the doctor, she opted to start conservative anti-inflammatory treatment, and planned regular follow-up visits every 6 months. The most recent follow-up on 4 March 2024 showed no abnormal findings. However, if her ureteral symptoms worsen or affect kidney function in the future, ureterotomy may be considered.

Discussion

Adult duplicated kidneys with duplicated ureters are uncommon urinary system anomalies in clinical practice. They occur in 0.7–4% of the population, predominantly affecting females (6). Normally, a single ureteric bud emerges from the primitive mesonephric (Wolffian) duct during fetal development and migrates to meet the metanephros, the embryological precursor of the kidney. Abnormal development of the middle renal tube during embryogenesis is believed to be the root cause of the disease (7,8). Some patients may experience persistent urine leakage and recurrent symptoms of UTIs (9). However, others often lack overt symptoms and are only identified during routine medical examinations (10,11). Additional rare anomalies may also be present. According to Spangler, when 3 ureteric

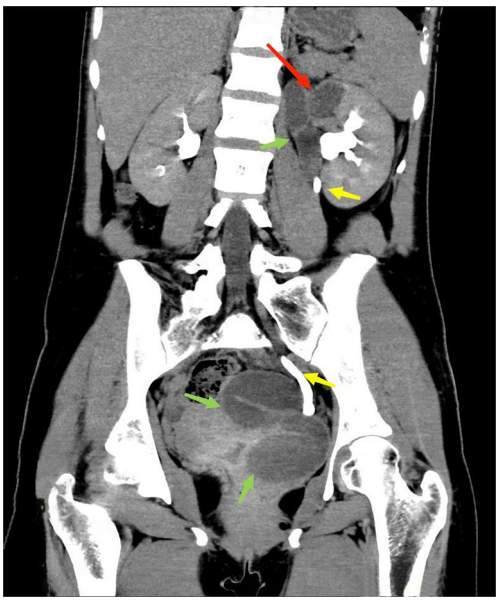

Figure 7 Coronal computed tomography shows a duplicated collecting system of the left kidney and complete ureteral duplication with ectopic ureteral insertion originating from the upper renal pole of the duplex system (shown by the red arrow). The left kidney is larger than the right. The yellow arrows suggest the normal ureter travels laterally to the left ectopic ureter. The green arrows show an ectopically dilated ureter.

buds meet the metanephros, a condition known as urinary tract triplication may develop. Patients often experience urinary incontinence (12), whereas others might develop renal hypertension and deterioration of renal function with increasing age (13). There are 2 types of duplicated kidneys: incomplete and complete. In incomplete duplicated cases, the upper ureters are distinctly separate but merge into a single ureter, forming a "Y" shape, which is less common (4). According to the Weigert-Meyer rule (14,15), complete duplicated kidneys have 2 completely separated ureters connecting to the upper and lower renal pelvis. The lower ureteral openings are typically located in the normal bladder trigone area, whereas the upper ureteral openings tend to be abnormally positioned near the bladder neck, urethra, vagina, or uterus. In this specific case, the ureter attached to the upper kidney parenchyma, dilated, and opened ectopically in the posterior urethral wall, which aligns with the aforementioned theory. Such openings often exhibit obstructions and narrowings, leading to poor urine drainage, ureteral torsion, and dilation, making it more likely to develop hydroureter or severe renal pelvis dilation (4). The Weigert-Meyer principle also indicates that patients with a distal opening of the sphincter usually experience leakage, whereas those with a proximal opening tend to experience UTI without leakage. In this instance, the patient exhibited a complete duplicated kidney anomaly with an ectopic upper ureteral opening proximal to the sphincter accompanied by a severe UTI and significant hydroureter. The vagina and the urethra were dry, supporting the clinical presentation described.

Ultrasound examinations often confuse hydroureter of the upper ureter in duplicated kidneys with hydrosalpinx. Initially, this case was misdiagnosed as hydrosalpinx. Therefore, for elongated cystic structures in the left side of the bladder, it is crucial to trace both the proximal and distal openings. If there is dilation of the upper renal pelvis and its connected ureter, the tracking must continue to the distal end. A high suspicion of an ectopic ureteral opening should arise if the distal end is below the level of the bladder trigone. Conventional ultrasound often fails to clearly show the distal ureter and the position of the opening. A dual-plane probe, used via a rectal examination, can provide a clearer view of an ectopic ureteral opening's path and position. Additionally, when examining a tortuous cystic mass in the pelvic region, expanding the scope of the examination to include closely related organs is essential to prevent misdiagnosis. In this case, the dilated ureter was initially misdiagnosed as hydrosalpinx due to the limited scope of the initial scan. When we searched upward through the abdominal probe, 2 sets of renal vessels in different sections were revealed, consistent with the diagnosis of a duplicate kidney, further confirmed by CTU and MRI. Thus, for larger abdominal masses, it is advisable to employ various methods (4) such as abdominal, transperineal, transvaginal, and rectal examinations to enhance the observation. Observing ectopic ureteral openings remains a challenge for CT (4) and MRI (16), as many are associated with hydroureter and blockages that prevent the contrast agent from traveling along the ureter. Both methods failed to locate the exact site of ectopic insertion of the ureter (*Figure 8*). This makes it impossible to directly observe the location of the ectopic ureteral opening. MRI is limited by the thickness of the scanning layers and cannot continuously track the disappearance of the ectopic ureteral opening (*Figure 9*). Although the location of the ectopic ureteral opening could be displayed by transperineal ultrasound (*Figure 3*), determining whether the opening is in the urethra or the vagina is challenging. In a previous case, Oliver Fannin III observed the ectopic insertion of the ureter

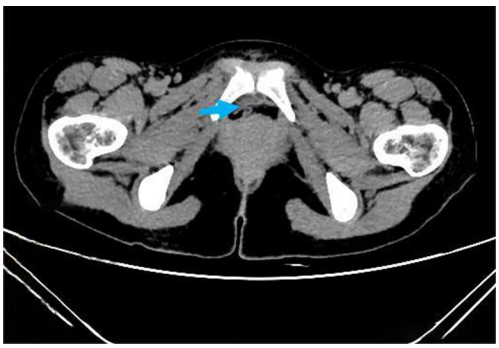

Figure 8 Computed tomography cross-sections fail to show the ureteral opening inferiorly connected to the urethra (shown by blue arrow).

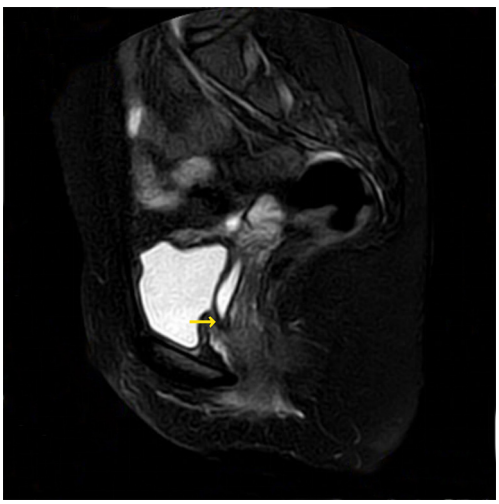

Figure 9 Sagittal moderately T2-weighted fat saturation image demonstrates the distal ectopic ureter inserts in the urethra bluntly (shown by yellow arrow).

into the prostate using a transrectal probe and concluded that transrectal observation could complement MRU (17). The dual-plane transducer features 2 beam emission modes: convex and linear array. The convex mode generates a fan-shaped image, displaying transverse tissue sections, whereas the linear array mode provides a rectangular image, depicting longitudinal tissue sections. Utilizing both modes enables a more comprehensive view of lesions. Additionally, the biplane probe offers higher frequency and resolution, making it suitable for initial diagnoses when ectopic urethral insertion is suspected. This probe complements MRU and CTU due to its convenience and feasibility. In this particular case, the dual-plane probe distinctly displayed the opening located above the urethral sphincter, and behind the urethra's posterior wall, along with the fibrous layer of the anterior vaginal wall and the muscular layer (*Figure 4*). The patient experienced recurrent UTIs but did not experience urinary incontinence. The clinical symptoms, combined with the dual-plane probe, led to the diagnosis of an ectopic ureteral opening above the urethral sphincter.

This patient experienced no urinary incontinence, similar to a case reported by Davis (18). Davis suggested that the ectopic opening of the duplicated ureter is located above the urethral sphincter and is innervated by it, thereby preventing incontinence. In his study, the patient was merely observed over time. Similarly, the patient in the current case exhibited no urinary incontinence, and the urologist recommended initial observation due to the patient's desire to maintain fertility. If pregnancy later exacerbates the condition, surgical removal of the ureter may be considered; patients without incontinence or other urinary symptoms, typically do not require treatment (18). However, those with symptoms may benefit from a partial nephroureterectomy, which has been shown to resolve incontinence (19). Ogawa detailed this surgical technique for removing a ureter inserted ectopically into the urethra (20).

Conclusions

The dual-plane probe is a prospective imaging technology in observing ectopic ureteral openings in the perineal region, and can be a complementary approach to CTU and MRU for the diagnosis of ureteral ectopic openings.

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Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://qims.amegroups.com/article/view/10.21037/qims-23-1736/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related

to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this article and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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