Scientific Research Publishing

Open Journal of Urology, 2026, 16(5), 231-238
https://www.scirp.org/journal/oju
ISSN Online: 2160-5629
ISSN Print: 2160-5440

Ureteral Ectopia, a Rare Cause of Uretero-Urethral Fistula in a Patient with Bilateral Duplex Kidneys: A Case Report and Review of the Literature

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How to cite this paper: Manda, E.K., Katapala, F.M., Mwamba, P.-C.K., Mabala, F.K. and Mbuyi, S.M. (2026) Ureteral Ectopia, a Rare Cause of Uretero-Urethral Fistula in a Patient with Bilateral Duplex Kidneys: A Case Report and Review of the Literature. *Open Journal of Urology*, **16**, 231-238.
https://doi.org/10.4236/oju.2026.165022

Received: February 9, 2026
Accepted: May 10, 2026
Published: May 13, 2026

Abstract

Introduction: Ureteral ectopia (UE), a congenital malformation characterized by the presence of a ureter opening outside the bladder trigone, manifests as a ureterourethral fistula (URF). The presence of bilateral duplication of the kidneys and ureters is a rarer condition in urology. **Methodology:** This is a descriptive study of a 17-year-old female patient treated at the HGR SENDWE in December 2024 for UE with UUF on a bilateral duplex kidney. **Case Presentation**: This is a 17-year-old patient who came for consultation for urinary incontinence since birth, characterized by constant involuntary loss of urine through the vagina associated with a feeling of needing to urinate at times, since the age of 3. She had consulted traditional practitioners and other doctors without success. On physical examination, her vulva was soiled with urine, we did not observe any vulvar fistula, and the methylene blue test was negative. After imaging tests, a diagnosis of complete bilateral duplex kidney with right ureteral ectopia was made. During surgery, we identified a bifid double collecting system. Using a methylene blue test via the ureters, we identified the ectopic ureter. After performing cystotomy and ureteral reimplantation, we left a JJ ureteral stent and a Foley urinary catheter in place. The postoper-

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ative period was marked by dry bedding the day after the procedure. **Conclusion:** Ureteral ectopia remains one of the rare causes of uretero-urethral fistulas, and its detection in cases of bilateral duplex kidneys is still very rare. The diagnosis is based on medical history, clinical findings, and imaging. Treatment remains surgical, with generally favorable postoperative outcomes allowing for successful social reintegration.

Keywords

Ureteral Ectopia, Uretero-Urethral Fistula, Bilateral Duplex Kidney

1. Introduction

Ureteral ectopia is a very rare congenital malformation of the urinary tract with an incidence of 0.05% to 0.025%, characterized by the presence of a ureter opening outside the vesical trigone [1]. It is most often associated with a duplex collecting system [2]. Depending on its location, the clinical picture may be asymptomatic or may lead to urinary incontinence, which is more common in women, or urinary tract infections in men. Ectopic insertion of a ureter into the urethra is even rarer.

Ureteral duplication is one of the most common congenital malformations of the urinary tract, but the presence of bilateral duplication of the kidneys and ureters is a rarer condition in urology [3].

One of the complications of complete ureteral duplication is ectopic ureteral opening, which, depending on the site of the opening, may be asymptomatic or cause urinary incontinence, which is more common in women and can have serious repercussions on their quality of life [4]. This ectopic insertion of the ureter is more rarely located at the level of the urethra, resulting in the clinical picture of a uretero-urethral fistula [5].

The aim of our study is to present a patient treated for ureteral ectopia presenting with a clinical picture of ureteral fistula, in whom a bilateral duplex kidney was identified.

2. Methodology

This is a prospective descriptive study of a 17-year-old female patient treated at the Jason Sendwe General Reference Hospital in Lubumbashi during December 2024 with a right ureteral ectopia on a bilateral duplex kidney who consulted with a clinical picture of a uretero-urethral fistula.

3. Patient Presentation

This is a 17-year-old patient who came to see us for involuntary urine leakage through her vagina since birth. According to her history, the problem began at the age of 3, when her mother noticed that her daughter's underwear was constantly

wet. At the age of 4, she consulted a traditional practitioner and doctors for several years without success. As a result, she dropped out of school at the age of 12. The persistence of her complaints prompted one of the obstetrician-gynecologists to refer her to us for treatment. It should be noted that the patient is a virgin. Regarding her urinary incontinence, she reports that, in addition to constant involuntary urine leakage, she sometimes feels the urge to urinate.

On clinical examination, her general condition was stable. Examination of the external genitalia revealed urine soiling. Upon removal, her vulva was soiled with urine, we did not observe any vulvar opening, and the methylene blue test was negative. In the paraclinical examination, we performed an intravenous urography (**Figure 1**), which revealed a double ureteral duplication with abnormal urine flow on the right side. The CT scan (**Figure 2**), meanwhile, revealed the presence of bilateral duplex kidneys with duplication of the ureteral tracts. This was confirmed by the angiogram (**Figure 3**), which revealed dual arterial circulation at the renal level.

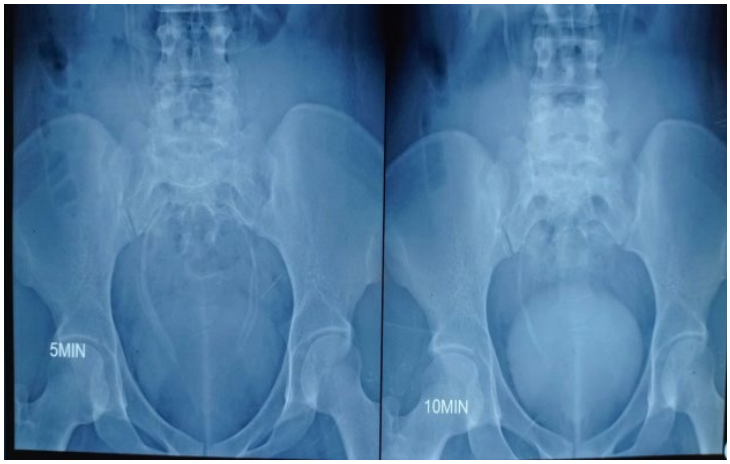

Figure 1. Intravenous urography showing double duplication.

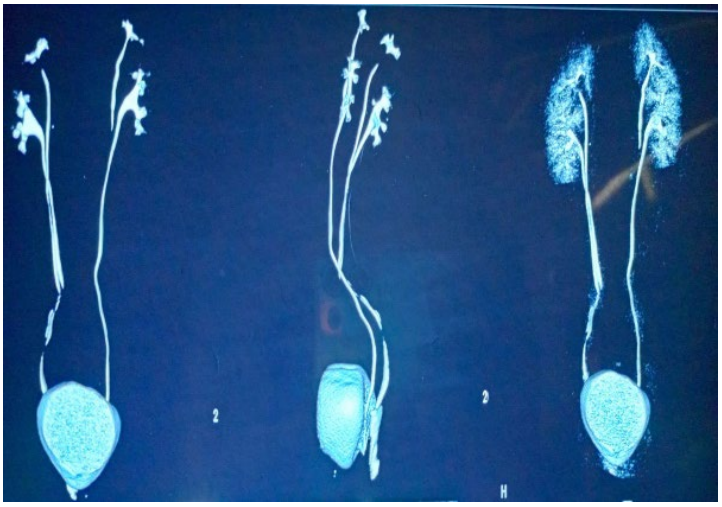

Figure 2. CT urography.

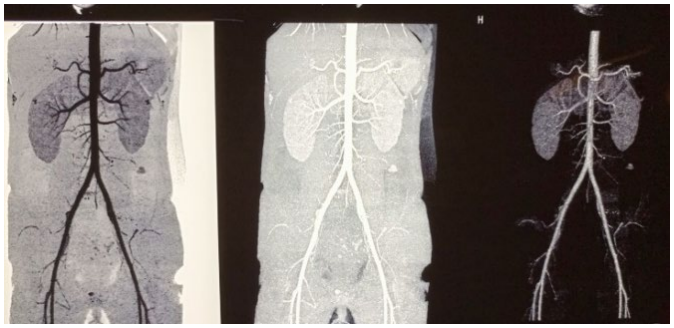

Figure 3. Angioscan.

After a pre-anesthetic consultation, ureteral reimplantation was scheduled.

Through a horizontal incision in the anterior abdominal wall, extending from the umbilicus to two finger-breadths above the pubis, we opened the retropubic space to expose the two ureters of each bifid kidney (**Figure 4**). After identifying the ectopic ureter using the methylene blue test and dissecting it from its attachments, we performed a cystostomy via a horizontal opening in the bladder wall. We made an incision in the bladder mucosa using a scalpel, then, using curved Kelly forceps, we created a zigzag path leading to the bladder dome. We reimplanted the ureter using heart-shaped forceps. We then sutured the ureter to the bladder wall using 4/0 PDS suture, thereby completing the ureteral reimplantation. We subsequently placed a JJ-type ureteral stent and a Foley urinary catheter.

We placed the patient on a 10-day course of beta-lactam antibiotic therapy (amoxicillin-clavulanic acid) and multimodal analgesia (1 gram of paracetamol infusion combined with a 100-milligram ampoule of tramadol). We recommended that she drink at least 3 liters per 24 hours.

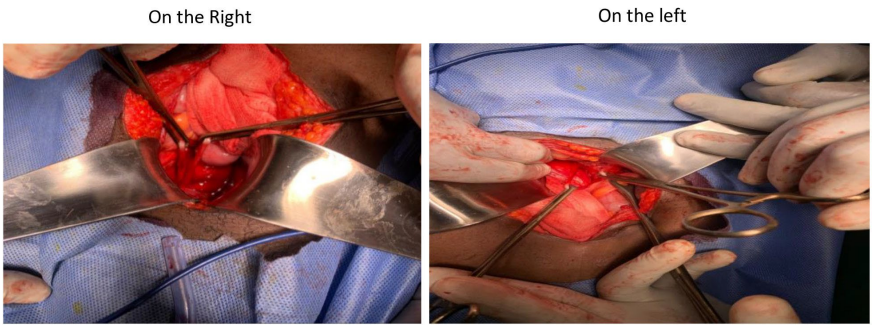

Figure 4. Highlighting of double ureters.

The postoperative period was characterized by the presence of dry urine as early as the day after the procedure. We removed the JJ stent and the urinary catheter on the fourteenth day after surgery. The patient experienced mild dysuria following catheter removal, but urination was uneventful. We discharged her from the hospital on the sixteenth day after the operation. A first follow-up was performed on the thirtieth postoperative day, with no abnormalities and good urinary flow, followed by another one month later. Our patient planned to resume her studies next year.

4. Discussion

Ectopic ureter is a rare congenital malformation usually associated with a duplicated kidney. It affects 0.7 to 4% of the population, mainly women [6]. The bilateral form of bilateral duplex kidney is extremely rare [7] [8].

There are two types of ureteral duplication: incomplete and complete. In incomplete duplication, there are two collecting systems and two ureters that join at any level between the kidney and the bladder to form a single ureter that drains normally into the base of the bladder. In complete ureteral duplication, there are two completely separate ureters and two renal pelvises [9].

Duplicated systems arise from an embryological anomaly. Since the ureter of the upper segment originates from a position above the mesonephric duct, it remains attached to this duct for longer and therefore migrates forward, establishing a lower medial position than the ureter that drains the lower segment (Weigert-Meyer law). As a result, the ureter draining the upper segment can migrate further in a caudal direction, becoming ectopic and obstructed.

Before the age of 4, it is generally difficult to identify urinary incontinence, given that daytime urinary continence in children is not usually achieved before the age of 4 and nighttime continence begins before the age of 5 [10]. Therefore, for our patient, the symptoms only became a cause for concern after the age of 3.

This condition has many consequences, including urinary incontinence, a disorder with multiple repercussions on the patient's health and well-being [4]. Our patient was therefore forced to withdraw from school life because of the constant embarrassment caused by the smell of urine, which prevented her from being around other people.

The key symptom in the diagnostic process is based on a thorough medical history and clinical examination. What alerted us was the fact that the patient reported feeling the urge to urinate at times and was able to hold it in, but despite this, she still felt wet from urine. Anatomically, this can be explained by the fact that urine is normally drained into the bladder by the properly inserted ureters, resulting in urinary continence and the urge to urinate at the time of micturition. However, urine flowing into the ectopic ureter is drained directly outside the bladder. This creates a picture of a uretero-urethral fistula, as was the case with our patient.

The differential diagnosis between ureterovaginal fistula and other conditions causing urinary incontinence can be established based on the patient's medical history and clinical examination. Indeed, the absence of involuntary urine leakage when coughing, for example, and the fact that the patient reported feeling the urge to urinate at times allowed us to rule out stress or urge incontinence. The fact that the patient has been wet both day and night since birth, with no history of trauma, and that she urinates normally at times while also experiencing urine leakage, led us to suspect a ureterovaginal fistula or a small vesicovaginal fistula.

The methylene blue test is a key examination in the clinical diagnosis of fistulas. A negative result proves that there was no communication between the urinary

and vaginal tracts.

Breen M. proposes various diagnostic approaches in cases of a negative methylene blue test. We indeed have two possibilities: either a very small vesicovaginal or uterine fistula with delayed passage of the blue dye into the vagina, or a urethrovaginal fistula [11].

In order to confirm our diagnostic hypothesis, we had to perform an intravenous urography and a uro-scan. The identification of ectopic ureteral openings in adults is mainly performed by cystoscopy, retrograde ureteropyelography, magnetic resonance urography [8], and computed tomography urography [12].

According to Zhang M *et al*. [6], the dual-plane probe is a promising imaging technology for observing ectopic ureteral openings in the perineal region and may be a complementary approach to CT and MRU for diagnosing ectopic ureteral openings.

In terms of treatment, ureteral reimplantation is the treatment of choice for ureteral ectopia. Some authors recommend ureterovesical reimplantation as the treatment of choice, as it offers the advantage of replacing the damaged part of the ureter with the well-vascularized bladder wall [13] [14]. The choice of the ureteral reimplantation technique was justified by its ease of performance in our setting. The lack of scintigraphy in our province prevents us from accurately assessing renal function in order to plan a potential partial nephrectomy. Nevertheless, the renal ultrasound performed revealed a normal renal structure. Blood tests, particularly measurements of urea and creatinine levels, help determine kidney function.

Other authors, however, recommend the laparoscopic approach because it offers certain advantages, including minimal invasiveness and rapid recovery, but its effectiveness has not been established by a large-scale evaluation of data [7]. Nishio H. *et al*. safely performed transvesicoscopic ureteral reimplantation for an ectopic ureter [15].

In the postoperative period, the results were positive, with dry bedding from the first two days after surgery. The abdominal drains were no longer productive on the fourth and fifth days and were removed on the fifth postoperative day. The urethral catheter was removed on the 14th day, as described by some authors [16].

5. Conclusion

Urethral ectopia remains one of the rare causes of uretero-urethral fistulas, which cause discomfort in everyday life due to urinary incontinence. It is generally associated with unilateral duplex kidney, but its detection in cases of bilateral duplex kidney is very rare. A thorough medical history, careful clinical examination, and appropriate imaging allow for a definitive diagnosis. The main treatment remains surgery, which generally has favorable postoperative outcomes and allows for successful social reintegration.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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