

Complete ureteral duplication with occult calculi: A case report

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Abstract. Duplication of the kidney and ureter is a relatively rare congenital malformation compared with other systemic malformations. It is even rarer for the duplicate to have stones inside; therefore, the present study reports the case of a patient with complete repetitive ureteral malformations combined with occult stones who showed marked improvement in symptoms and good recovery after surgical treatment. The patient received a timely diagnosis and achieved a good treatment outcome. The patient was satisfied with the treatment and had a good follow-up. Early and accurate diagnosis and timely treatment are crucial for patients with recurrent ureteral malformations combined with occult stones, and can effectively improve patient prognosis.

Introduction

Kidney and ureter duplication is among the most common congenital anomalies of the kidney and urinary tract, representing a frequent structural variant in clinical urology (1,2). Due to its diverse clinical manifestations, misdiagnosis or a missed diagnosis could cause difficulties in surgical treatment (3,4). Epidemiological studies have reported an overall incidence ranging from 0.3 to 2.0% in the general population, with autopsy series indicating incidence rates of ~1:125 to 1:150 live births (5-7). The condition displays a distinct female predominance, with a female-to-male ratio of ~2:1. Unilateral involvement is approximately six times more frequent than bilateral disease. Complete duplication accounts for ~40%

of all cases, while incomplete duplication constitutes the remaining 60% (1,8).

Embryologically, ureteral duplication arises from abnormal budding or premature bifurcation of the ureteric bud from the mesonephric duct during early fetal development, disrupting the healthy formation of a single collecting system for the kidney and ureter (9). Under healthy conditions, a single ureteric bud induces the differentiation of the metanephric blastema to form one renal pelvis and ureter. When developmental interference occurs, supernumerary or early-splitting ureteric buds lead to separate or partially fused collecting systems, resulting in duplex kidneys and duplicated ureters (10-12).

Stones complicating ureteral duplication remain relatively uncommon, with an incidence of 3-8% in patients with duplex collecting systems, and can be particularly elusive when located in communicating or blind-ending segments, mimicking bladder calculi on imaging (13). Early and accurate diagnosis along with timely intervention are critical to preserve renal function, relieve symptoms and prevent recurrence. To enhance clinical awareness and provide evidence for clinical practice, the current study presents a rare case of complete ureteral duplication with occult calculi successfully managed with endoscopic laser lithotripsy and stent placement.

Case report

A 50-year-old woman sought medical attention at the Second Hospital of Lanzhou University (Lanzhou, China) for intermittent hematuria for 1 month (symptoms appeared in February 2022). The patient reported gross hematuria 1 month prior, which worsened after physical activity. Ultrasound examination revealed a bladder stone and a stone in the left kidney in March 2022. However, cystoscopy revealed no bladder stones, confirming that there was a diagnostic error in the patient's case. At the Second Hospital of Lanzhou University (Lanzhou, China), a left ureteral double-J tube was inserted in March 2022. Later in that month, the patient visited the Department of Urology at Zhangye People's Hospital Affiliated to Hexi University (Zhangye, China). After admission, a physical examination revealed the absence of bilateral renal percussion pain, no obvious mass in the lower abdomen and no tenderness in the ureteral tract. Ultrasonography revealed an unclear upper urinary tract and strong echogenicity in the bladder. Intravenous pyelography (IVP) revealed postoperative

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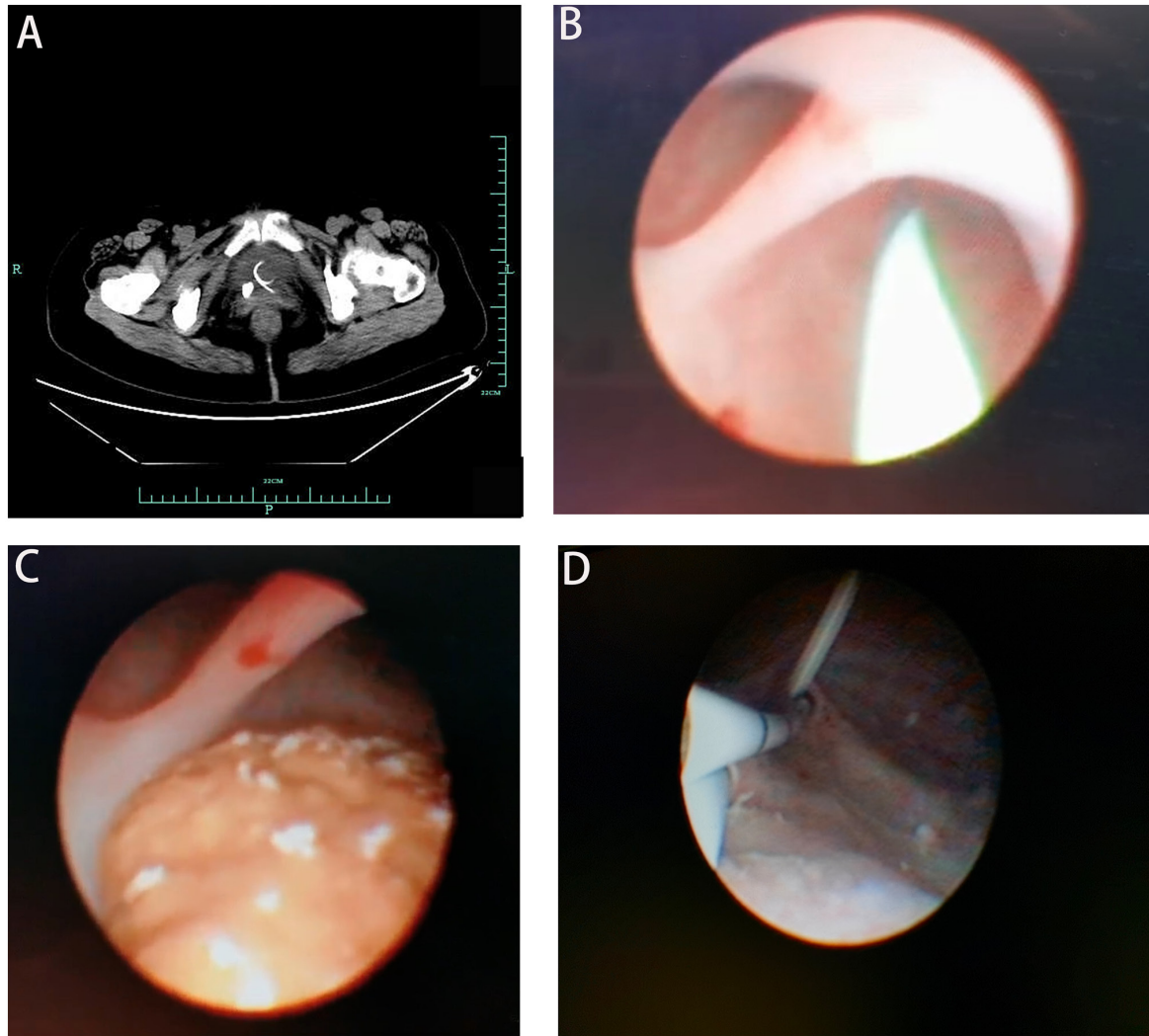


Figure 1. Hidden stone imaging and microscopic manifestations. (A) Computed tomography showing a right urinary tract stone. (B) Ureteroscopy showing that the urinary tract was divided into two parts. (C) Ureteroscopy showing a stone. (D) Ureteral stent tube in each of the two right ureters.

changes after left ureteral double-J tube insertion, including multiple dense nodular shadows in the left kidney and bladder and urinary tract stones. Computed tomography (CT) revealed bladder stones and the left ureteral double-J tube (Fig. 1A). Routine urine tests revealed the following: 1,269 white blood cells/ μl (normal range, 0-10 white blood cells/ μl); 5,134 red blood cells/ μl (normal range, 0-5 red blood cells/ μl); and positivity (++) for protein [normal value, (-) for protein]. Serum creatinine and BUN levels were within normal ranges.

Cystoscopy was performed 3 days after admission under general anesthesia in March 2022. The cystoscopy revealed the curled end of the double-J tube at the left ureteral opening in a good position. A 6.4/8.0 F ureteroscope was used to enter the bladder through the urethra, with a slit-shaped right ureteral opening and clear urine spraying. Under the guidance of a zebra guidewire, the ureteroscope was inserted into the lower segment of the right ureter. No stones were found. The urinary tract was divided into two parts (Fig. 1B), and an endoscopy was performed along each cavity. The renal nipple was flattened in the lower renal pelvis, and a blind end was observed at the junction of the upper renal pelvis, making it impossible for the endoscope to pass through. No stone shadows were

observed upon returning to the bladder for further exploration. Another fissure-shaped opening was observed ~2 cm from the ureteral opening of the right bladder wall. A ureteroscope was guided into this opening using a guidewire, and a stone was found in the ureter (Fig. 1C). The guidewire was removed, and a 200- μm holmium laser fiber, with a light energy of 1.0 W and a frequency of 20 Hz, was inserted. The holmium laser shattered the stones, and the ascension of the ureteroscope continued. This opening (fissure-shaped opening was observed ~2 cm from the ureteral opening of the right bladder wall) was fused and converged with the original ureteral opening (the normal opening of the right ureter). A ureteral stent tube was placed in each ureter (Fig. 1D), and an F18 urinary catheter was placed. Surgery was then completed. Blood creatinine and urea nitrogen levels were within normal limits. Postoperative retrograde pyelography revealed two kidneys and two ureters on the right side. The lower segments of each ureter were fused together but then opened separately into the bladder (Fig. 2). The patient was satisfied with the treatment outcome. The patient was discharged without complications and advised to continue follow-up observation as recommended by the physician. The patient underwent a follow-up CT scan of the urinary

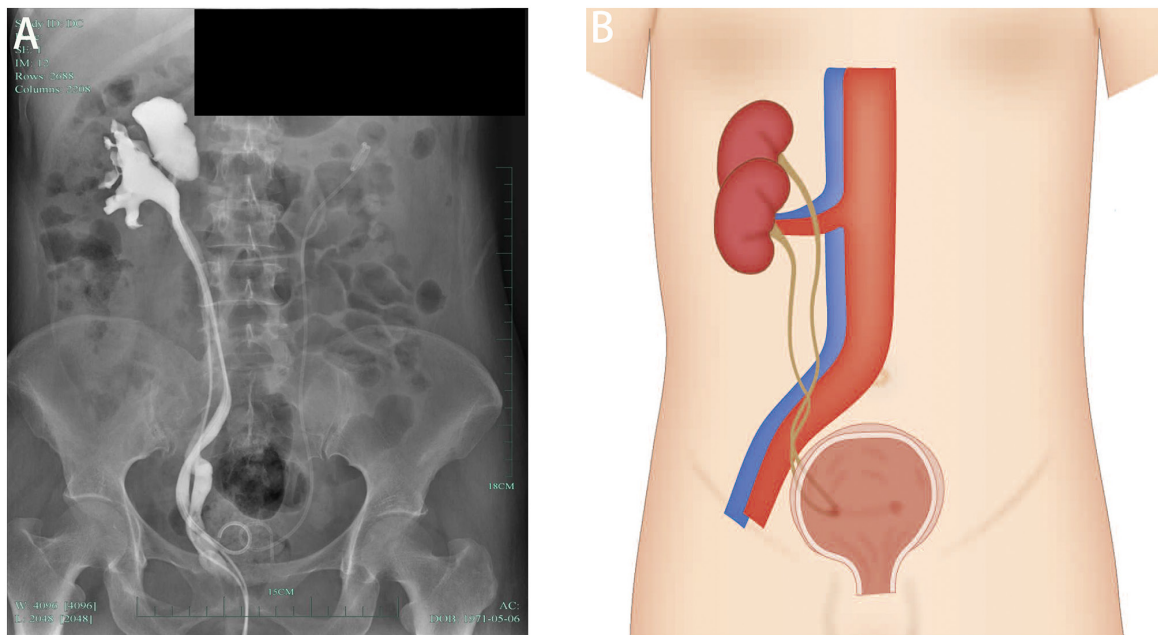


Figure 2. Retrograde pyelography and pattern diagram. (A) Retrograde pyelography showing a bilateral ureteral malformation and (B) a pattern diagram (drawn using Adobe Systems, Inc.).

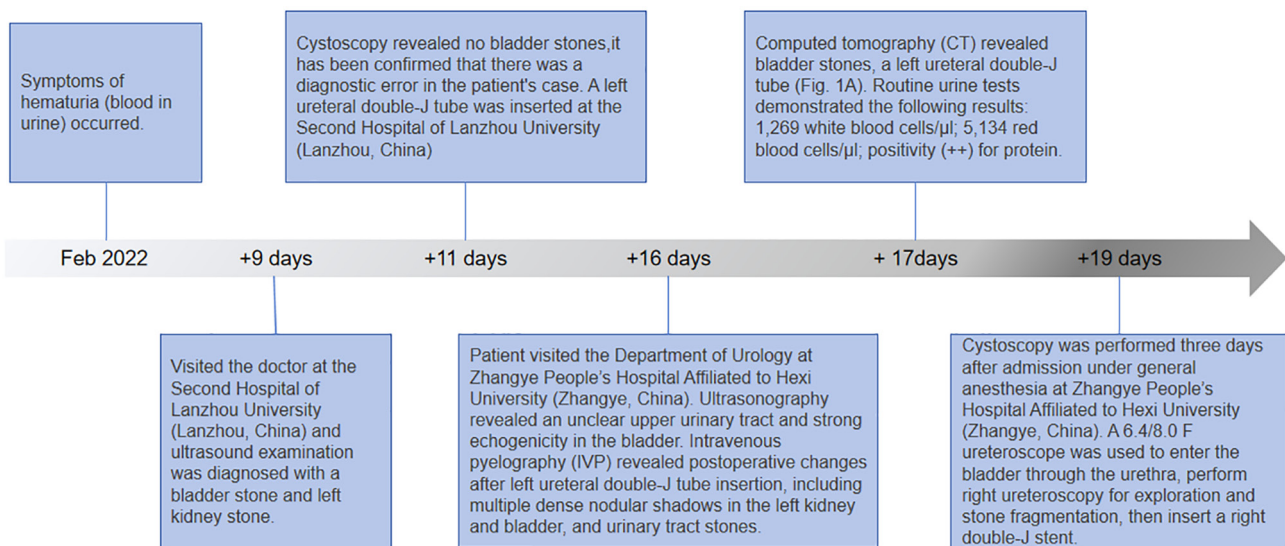


Figure 3. Timeline for the treatment and examination of a complete ureteral duplication with occult calculi.

system 1 month after surgery, and no residual stones were found. After discharge, a follow-up examination 6 months later indicated no abnormalities. Subsequently, a urinary system CT was performed once a year, revealing no recurrence of the urinary stones. Family members also underwent physical examinations to rule out any urinary system abnormalities. The patient timeline is presented in Fig. 3.

Discussion

Renal ureteral malformations mainly originate during early embryonic development, where ureteral buds emerge from the mesonephric duct and gradually grow and differentiate to form structures such as the ureters and renal pelvis (14-16).

Under normal circumstances, ureteral buds develop according to a specific pattern, ultimately creating a single functional collection system for the ureters and kidneys (9). However, when certain factors interfere with this normal developmental process, premature or excessive branching of the ureteral bud may occur, thereby causing repetitive ureteral malformations (17). Such abnormalities may be related to genetic, environmental or unknown factors during embryonic development (18). Although the specific genetic pattern is not yet fully understood, mutations in or abnormal expression of ACTA2, ACTG2, BNC2, CHRM3 and HNF1B, among others, may be associated with repetitive ureteral malformations (19).

Genetic factors are substantial in this condition. Ureteral duplication is recognized as part of the spectrum of congenital

anomalies of the kidney and urinary tract, with evidence supporting autosomal dominant inheritance with incomplete penetrance (20). Familial studies have revealed that individuals with an affected first-degree relative retain a markedly increased risk (21,22). Molecular investigations have linked duplication phenotypes to mutations or expression abnormalities in genes governing ureteric budding and outgrowth, such as roundabout guidance receptor 2, *GEN1*, *RET* and glial cell derived neurotrophic factor, and downstream mediators of the MAPK/ERK pathway.

These genes regulate critical steps in branching morphogenesis and tissue interaction, with dysfunction that could induce duplicated or ectopic ureteric buds (18,19). Environmental and maternal factors further contribute to developmental disruption. Adverse intrauterine conditions (including maternal infection, febrile illness, and exposure to tobacco, alcohol, toxic chemicals or certain medications) might interfere with mesonephric-duct and ureteric-bud development, increasing the likelihood of congenital urinary anomalies. Although the precise interactions between genes and the environment remain elusive, current evidence supports a multifactorial model combining genetic predisposition and environmental triggers (22). Future research with larger cohorts, familial analyses and molecular profiling is warranted to clarify causal pathways and identify actionable risk factors.

The patient described in the present case report was a middle-aged woman with recurrent gross hematuria as the primary clinical manifestation. The characteristics of the bladder stones were clearly displayed on imaging examination, but no stones were found during cystoscopy. Ureteroscopy revealed that the hidden location of the stones was in the communication section between the right repeated ureteral orifices, which was very shallow and difficult to distinguish from the bladder stones on imaging. The patient had stones in the lower segment of the ureter, but no symptoms of obstruction were observed. This was related to compensation caused by the mutual communication of the right repeated ureteral orifice; therefore, the patient did not present with clinical manifestations such as upper urinary tract obstruction and hydronephrosis. Ureteroscopy revealed a duplicated right kidney, with the upper renal pelvic ureteral junction at the blind end. Duplicate ureteral malformations cannot be visualized during the excretion period of CT urography, leading to difficulties in the clinical diagnosis (23).

In the present case, the diagnosis was difficult, with multiple examinations indicating bladder stones. However, cystoscopy did not detect the presence of stones, confusing the clinical diagnosis. The clinical manifestations of recurrent ureteral malformation are diverse. Some patients may be asymptomatic and the malformations may only be incidentally discovered during physical examination, there have been no reports of repeated kidneys, repeated ureters, repeated ureteral openings, mutual communication between repeated ureteral openings or hidden stones in the communication segment, both domestically and internationally. In terms of diagnosis, the combined application of imaging examination methods such as IVP and CT provided strong support for the accurate diagnosis of the present case, which is consistent with the comprehensive diagnostic methods emphasized in previous literature (24,25). However, the specific morphology and

degree of deformities may vary among cases, and treatment plans must be selected based on individual circumstances. Ultrasound, a non-invasive and convenient examination method, can first detect abnormalities in the renal structure and dilation of the ureter in most cases, providing clues for further diagnosis (26). However, ultrasound has certain limitations in displaying the entire ureter and observing subtle structures; therefore, it is necessary to combine IVP and CT (5). CT scans provide more detailed anatomical information, not only accurately displaying the structure of the kidneys and ureters but also detecting lesions in surrounding tissues, which is of great importance for the differential diagnosis and surgical planning. IVP can clearly display the morphology and course of the renal pelvis and ureter, which is of great value for clarifying the anatomical structure of the duplicated ureter and assessing renal function (23,27). In the present case, the IVP failed to detect the duplicated structures early. Postoperative retrograde pyelography was necessary to clearly display the dilation and tortuosity of the duplicated kidneys and ureters, which helped determine the location and degree of the lesion.

In conclusion, due to the diversity and lack of specificity in the clinical manifestations of recurrent ureteral malformations, they pose certain challenges to clinical diagnosis. Some patients may not have obvious symptoms in the early stages and the malformations may only be incidentally discovered during physical or imaging examinations for other reasons. In patients with symptoms, the symptoms may vary and include lower back pain, abdominal pain, hematuria, frequent urination and urgency (4,6). These symptoms are similar to those of other urinary system diseases and can easily lead to misdiagnosis or a missed diagnosis. Therefore, improving doctors' understanding of ureteral malformations and strengthening research on its clinical characteristics, diagnostic methods and treatment strategies have important clinical significance. The present case suggests that by comprehensively using multiple imaging examination methods and by proficiently using cystoscopy and ureteroscopy, diagnostic accuracy can be improved. In clinical practice, personalized diagnosis and treatment plans should be developed based on the patient's specific situation, and attention should be paid to postoperative follow-up observations.

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Availability of data and materials

The data generated in the present study may be requested from the corresponding author.

Authors' contributions

HZ, LW and JY drafted the manuscript and designed the study. RY, SN and FY contributed substantially to the conceptualization and design of the study. FY completed the surgery. FY and JY approved the final version of the manuscript for publication. FY and JY confirm the authenticity of all the raw data. All authors have read and approved the final version of the manuscript.

Ethics approval and consent to participate

Not applicable.

Patient consent for publication

Written informed consent was obtained from the patient for the publication of the data and images included in the present study.

Competing interests

The authors declare that they have no competing interests.

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