



Case Report

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Retrograde Intrarenal Surgery for Huge Ureteral Stones at the Y-Junction of A Bifid Ureter: A Case Report and Literature Review

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Abstract

Introduction

It is rare to hear of a huge ureteral stone in an incomplete, duplicated ureteral system. In this paper, we present our experience of staged flexible ureteroscopic management of such a case.

Case presentation

A 53-year-old man with no history of systemic disease experienced bilateral flank discomfort for 4–5 days with no fever. Abdominal computed tomography scanning revealed a 3.8 × 3.6 cm ureteral stone at the Y-junction of an incomplete duplicated ureteral system. We performed a multistaged operation with flexible ureteroscopy twice and follow-up rigid ureteroscopic management once for this case. One episode of fever with urosepsis was noted after the second operation, while the rest of the postoperative course was uneventful.

Discussion

Ureteral stones larger than 3.5 cm are rarely reported in bifid ureters. Extracorporeal shockwave lithotripsy play a role in urolithiasis in the case of a duplicated ureter. Patients with stone sizes of ≥3 cm in duplex systems seem to be better candidates for percutaneous nephrolithotomy (PCNL) or open surgery. However, PCNL may be limited due to anomalies of the ureter and kidney. In our case, we demonstrated a successful experience of staged endoscopic intervention to manage a massive ureteral stone at the Y-junction of a bifid ureter.

Conclusion

Flexible ureteroscopic management demonstrated a satisfactory and safe outcome even in the case of a massive ureteral stone at the Y-junction of a bifid ureter.

Keywords: ureteral duplication, ureteral stone, flexible ureteroscopic retrograde intrarenal surger (RIRS), percutaneous nephrolithotomy (PCNL), rigid ureteroscopic lithotripsy (URSL)

Introduction

Duplex renal collecting systems occur in 0.5% to 3.0% of the population. [1] The cause is the development of two separate ureteral buds from a single mesonephric duct. There are complete and incomplete types. [1, 5] The bilateral and complete types are more common in females. Urinary tract infections and urolithiasis are common in people with this malformation. There are other congenital anomalies associated with complete ureteral duplication. The partial duplication of the ureter, or the so-called bifid ureter, shows a Y-shape and converges into the bladder at the same orifice. [2,3] Rigid ureteroscopic lithotripsy (URSL) can manage ureteric calculus disease with success rates of up to 80%. A complete

survey of the patient's anatomy and coexisting anomalies should be performed carefully before surgical intervention. [4] However, there is little experience with retrograde intrarenal surgery (RIRS) with flexible ureteroscopy in such cases. In our patient, the left unilateral incomplete duplicated ureteral system with the formation of a massive stone larger than 3 cm was diagnosed with the help of abdominal computed tomography (CT). The stone was located at the Y-junction of the duplicated ureter. Multistaged surgery with both URSL and RIRS was performed smoothly and successfully in this case. The experience of using RIRS for stones >3 cm and the possibility of postoperative complications were also reviewed in this article.

Case Presentation

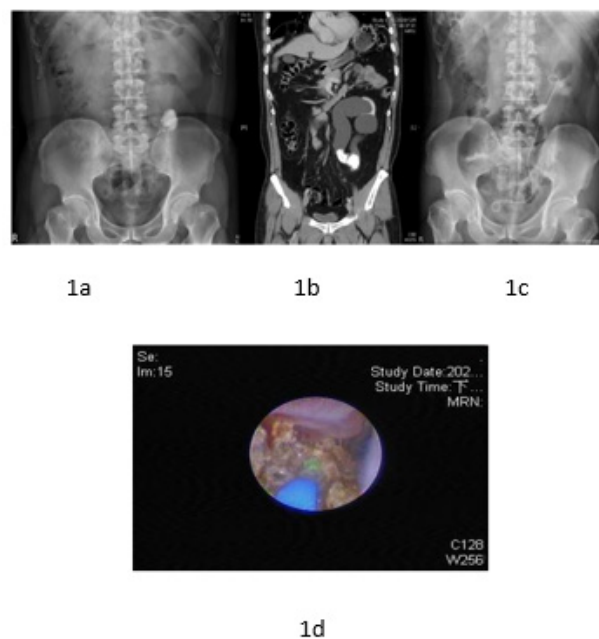


Figure 1a. the KUB before management of retrograde intrarenal surgery revealed a 3.8*3.6cm ureteral stone at middle ureter preoperatively.

Figure 1b. The computed tomography of abdomen without contrast enhancement revealed left incomplete ureteral duplication. The ureteral stone was stuck at the Y junction of bifid ureter.

Figure 1c. The KUB after the first RIRS with residual stone (3.0*2.0cm). Double-J stent was placed after management.

Figure 1d. The image of management of stone lithotripsy with flexible ureteroscopy.

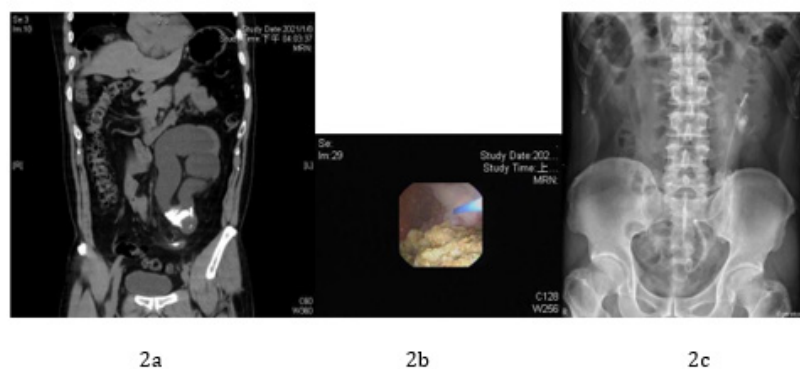


Figure 2a. The computed tomography after the first time of retrograde intrarenal surgery. Residual stone with size measured as 3.0*2.0cm was stuck at the Y junction of bifid ureter.

Figure 2b. The image of management of stone lithotripsy with flexible ureteroscopy for the second time.

Figure 2c. The postoperative KUB revealed that the stone was fragmented and some stone street was noted at left lower ureter. Double-J stent was placed after management.

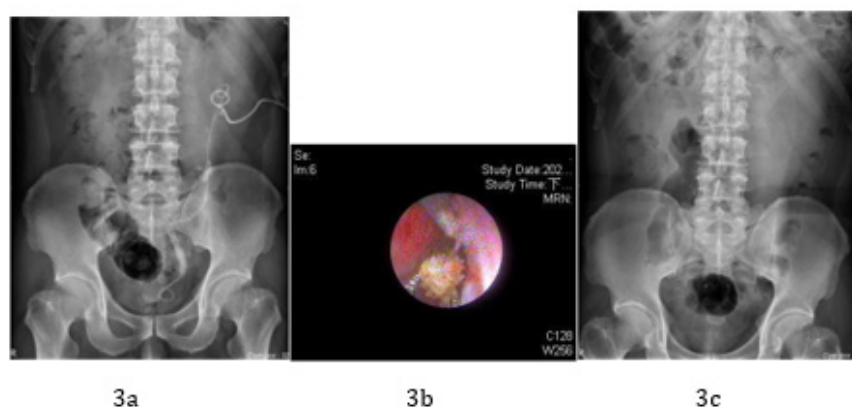


Figure 3a. The KUB after percutaneous nephrostomy drainage inserted due to postoperative acute pyelonephrosis. Much stone street was noted at left lower ureter.

Figure 3b. The image of management of stone lithotripsy with rigid ureteroscopy.

Figure 3c. The postoperative KUB revealed the lower ureteral stone street were removed.

A 53-year-old man with a history of a left renal cyst, incidentally, discovered through renal sonography during a routine examination, experienced bilateral flank discomfort for four days, with more intense pain on the left side. He initially visited the nephrology outpatient department (OPD) for help, where a renal ultrasound revealed multiple simple cysts of the left kidney and a suspected renal stone or angiomyolipoma. Abdominal CT revealed a left duplicate kidney with a huge stone measuring 3.8 × 3.6 cm formed over the Y-junction of the bifid ureter, with left hydronephrosis and dilated ureters in both limbs (Figure 1b). He was referred to the urology department for further management. The patient underwent an initial left ureteroscopic lithotripsy (URSL) with Homium-YAG laser and double J stent insertion (Figure 1d). The operation lasted 3 hours and 15 minutes. The follow-up kidney-ureter-bladder plain X-ray film (KUB) revealed residual stones in the left lower ureter and the Y-junction of the bifid ureter (Figure 1c). Unfortunately, the patient's left flank pain persisted after he was discharged from

the hospital, and he returned for the scheduled operation. The follow-up abdominal CT revealed a residual stone at the left middle and lower third of the ureter with marked hydronephrosis and hydroureter (Figure 2a). The residual stones measured 3.0×2.0 cm. Left retrograde intrarenal surgery (RIRS) with flexible ureteroscopy was performed smoothly at the second round (Figure 2b). This operation lasted 2 hours and 45 minutes. Postoperative laboratory data were unremarkable, and the patient was discharged on postoperative day 2. However, fever, urinary frequency, and dysuria were observed on postoperative day 4. During the scheduled OPD follow-up, renal sonography revealed left hydronephrosis. KUB revealed a residual left lower ureteral stone. Urinalysis revealed pyuria. He was admitted to the urological ward for treatment of suspected acute pyelonephritis. Empirical intravenous antibiotics (Ertapenem 1000 mg QD) were administered immediately after admission. However, fever persisted (temperatures of up to 40°C), and the patient experienced chills while he was on antibiotics. Emer-

gency left percutaneous nephrostomy (PCN) was performed, and purulent urine came out through the drain (Figure 3a). *Candida albicans* was identified upon the culture of this urine. Therapeutic antibiotics (Fluconazole 400 mg QD IVD) were given as the result of the report. After a one-week course of antibiotics, his fever abated, and his urine was clear and amber. With the patient in stable condition, he underwent left ureteroscopic lithotripsy (URSL) with Holmium LASER to remove the most residual stone street lodged in the lower ureter (Figure 3b). This intervention lasted for 2 hours and 15 minutes. There was no major discomfort after the intervention, and the patient was discharged on postoperative day 2.

Discussion

Ureteral duplication, one of the most common ureteral anomalies, occurs in 0.6% to 0.7% of the population. [1,7] Morphologically, this condition can be divided into partial or complete ureteral duplication. In complete ureteral duplications, two independent ureters link to the bladder. In ureteral duplications, many symptoms can be noted, such as vesicoureteral reflux, ureteropelvic junction obstruction, or pyelonephritis. [7] However, most of the patients remain asymptomatic and may be incidentally diagnosed during health examinations. In patients with renal stones, the preferred treatment method depends on the location and size of the stone, contraindications of certain procedures, the patient's physical condition, and preference. Extracorporeal shock wave lithotripsy (ESWL) is a noninvasive technique for managing stones that measure less than 2 cm. ESWL is also effective in patients with congenital urinary system anomalies. However, for patients whose stone sizes are more than 3 cm or whose stones are located in malformations of the urinary system such as horseshoe kidneys, malrotated kidneys, and ureteral duplications, percutaneous nephrolithotomy (PCNL) or open surgery might be the preferred treatment option. [6] PCNL, as an effective procedure, is often indicated in patients with stone sizes larger than 2 cm. [8] However, it is associated with a higher risk of hemorrhage and nephron damage, impacting renal function. There are contraindications that we need to consider, such as hemodynamic instability, bleeding tendencies, anticoagulant therapy, pregnancy, and severe cardiopulmonary disease. [6, 9] Rigid ureteroscopy (R-URS) for renal and ureteral stones smaller than 2 cm has been preferred to ESWL in recent years. It has also been suggested as a secondary treatment option after PCNL for stones that are larger than 2 cm. [10,11] Regardless of the proximal or distal location of the ureteral stone, R-URS has its role in both groups. [12] Still, we have to prevent ureteral wall injury, stone migration, or even ureteral perforation (which is a severe complication) during R-URS. Many patients with lower ureteral stones will choose to have R-URSL as their first attempt. However, flexible ureteroscopy (F-URS) shows more promising outcomes than rigid ureteroscopy (R-URS) in the efficiency of removing stones in the renal pelvis. According to recent studies, flexible ureteroscopic retrograde intrarenal surgery (RIRS) with holmium-YAG laser has proven more advantageous in treating renal stones even larger than 2 cm. [14] The risk of complications is also lower than that associated with PCNL. The associated stone-free rate of RIRS ranges from 65% to 92%. [13] However, the operation may last longer in patients with ureteral duplication.

[7] Most of these studies monitor the outcome of RIRS in stones in the renal pelvis or the normal ureter. The technique of RIRS is seldom described in patients with ureteral anomalies. In our case, there was a congenital ureteral anomaly with the incomplete type of ureteral duplication, the so-called bifid ureter. Moreover, one huge ureteral stone larger than 3.5 cm (3.8cm × 3.6 cm) obstructed the Y-junction of the left bifid ureter. Due to the stone size, ESWL was unsuitable. The obstruction was at the Y-junction of the bifid ureter, and the distance between the stone and the renal pelvis was more than 10 cm. PCNL was not considered. Ureterolithotomy was an option in this case, but the patient initially requested endoscopic treatment. The treatment options left were rigid ureteroscopy and flexible ureteroscopy. Due to the lack of previous experience of RIRS in a patient with a ureteral anomaly and a huge stone burden, we initially opted for rigid ureteroscopy for lithotripsy. However, due to the anomaly of a duplicated ureteral system, the alignment was not straight. There was a sharp angle at the diversion of the duplicated ureter, which resulted from incomplete stone removal due to the limited operative field. Then, we considered flexible ureteroscopy due to its good mobility at different angles. During the second stage of the operation, we used flexible ureteroscopy with holmium-YAG laser alternatively to solve the problem of large-angle diversion, and the result was satisfactory. Much of the stone was fragmented and removed. Postoperative KUB revealed some residual stones at the lower ureter. The stone-free rate was good, and there was no intraoperative ureteral injury or perforation. However, fever was noted on postoperative day 4, and the patient was diagnosed with left-sided acute pyelonephritis. Percutaneous nephrostomy (PCN) drainage was performed, and much pus urine came out from the catheter. We reviewed the whole course of the operation and postoperative care. The prolonged intervention and hydraulic pressurization might have caused the postoperative infection during flexible ureteroscopy. [14] Prolonged hydraulic pressurization could cause a higher incidence of bacterial translocation, leading to urosepsis or septic shock.

Conclusion

Retrograde intrarenal surgery (RIRS) with flexible ureteroscopy is associated with a satisfactory and safe outcome in the case of huge stones that measure more than 3.5 cm. It is practical in patients with obstructive ureterolithiasis at the Y-junction of a duplicated ureter whose anatomy is variable. However, a multistaged operation should be considered due to the possibility of bacterial translocation during prolonged interventions and the induction of infection. Conclusively, flexible ureteroscopy is an efficient and feasible intervention in urolithiasis in a bifid ureter.

Declaration

Ethics approval and consent to participate

The Institutional Review Board approved this study with certification number IRB2021030 (Approved on 2021.03.24)

Consent for publication

The patient gave written informed consent for the publication of this case report

Competing interests

The authors declare that they have no competing interests to declare

Author's Contributions

Dr. Pin-Jui Huang, Dr. Li Wei Chao, Dr. Hui Husan Chuang, Dr. Chi-Feng Hung, Dr. Chen-Huang Shen, Dr. Yeong-Chin Jou, Dr. Chang-Te Lin, and Dr. Ming-Chin Cheng conceived and planned the 18 12 research. Dr. Pin-Jui Huang, Dr. Li Wei Chao, Dr. Hui Husan Chuang, and Dr. Chi-Feng Hung 1 carried out the research. Dr. Yeong-Chin Jou, Dr. Chang-Te Lin, and Dr. Ming-Chin Cheng planned 2 and carried out statistical analysis. Dr. Pin-Jui Huang, Dr. Li Wei Chao, Dr. Hui Husan Chuang, Dr. 3 Chi-Feng Hung, and Dr. Chen-Huang Shen contributed to the interpretation of the results. Dr. Pin-4 Jui Huang, Dr. Li Wei Chao, Dr. Hui Husan Chuang, and Dr. Chi-Feng Hung wrote the manuscript. 5 All authors critically reviewed the manuscript and contributed to its final version.

Acknowledgments

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Conflict of Interest

No conflict of interest.

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