

Double The Trouble

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Abstract

Introduction: Bilateral cases of Duplex Kidneys and Ureters with Complete duplication are exceedingly rare. Herein, we report a rare case of Bilateral Complete Duplex Kidney and Ureter with Hydronephrotic right upper moiety complicated by ureteric stone and stricture in an adult male, managed successfully.

Case Capsule: A 44-year-old male patient with history of flank pain, fever, vomiting, raised creatinine and a complex past history of multiple interventions for Urolithiasis including ESWL, PCNL and RIRS, was diagnosed with Bilateral Complete Duplex system with gross hydronephrosis of right upper moiety, distal ureteric calculus in it and multiple right renal calculi on CT. Intraoperatively on Ureterorenoscopy revealed right upper moiety 5mm distal ureteric stone and stricture at L5 level. Stone fragmented with laser, with difficulty 5/24 DJ stent negotiated across the stricture. Patient’s

condition dint improved even after a month and was posted for definitive surgery. Right upper moiety Ureterorenoscopy and RGP revealed stricture at L3-L4 level. Right PCNL done revealed turbidity in the system and three 10mm lower calyceal calculi retrieved. Open Right side to side Uretero-Ureterostomy done of upper and lower moiety segments over 6/26 DJ stent for unhealthy, fibrosed, 3cm right upper moiety ureteric stricture at L3-L4 level. Surgery was uneventful. Right DJ stent removed after 6 weeks.

Conclusion: This article demonstrates a very rare incidence of Bilateral Complete Duplex system with ureteric stone and stricture in one moiety system that posed a surgical challenge. Thus double the problems of stones and stricture in double the systems caused actual trouble and was managed successfully.

Keywords: Duplex, Ureter, Kidney, Stone, Stricture.

Introduction

Duplex kidney and ureter represent a relatively frequent congenital anomaly of the urinary tract, characterized by the presence of two separate drainage systems within a single kidney. This condition may be discovered incidentally or in the context of urinary tract symptoms. The reported prevalence varies depending on the study population, with an incidence of approximately 0.8% among healthy adults, 0.3%-2.5% in autopsy series, and up to 4% in patients undergoing evaluation for urinary complaints¹.

Anatomically, a duplex system may involve either partial or complete duplication of the ureters. In partial duplication, also referred to as bifid ureter or incomplete duplication, the ureters join before entering the bladder. In complete duplication, each ureter drains independently into the bladder, resulting in two separate ureteral orifices⁴. Bilateral complete duplication is considered a rare variant of this anomaly, especially in adults¹.

Most cases of duplex renal systems remain asymptomatic and are identified incidentally through imaging or intraoperative findings. However, the anatomical variations associated with duplex kidneys can predispose individuals to certain urological complications. These include urinary tract obstruction, vesicoureteral reflux, ureterocele, and nephrolithiasis, which may occur in either moiety or be present bilaterally^{2,5}. Notably, the upper moiety is often at increased risk of obstruction, commonly due to ureterocele, while the lower moiety tends to be more susceptible to reflux-related pathology⁶. Although duplex systems are not inherently pathological, the presence of associated complications may necessitate medical or surgical intervention. The occurrence of bilateral complete duplex systems with concurrent complications such as stones and strictures is extremely rare in the literature⁷.

We present a rare case of a bilateral complete duplex collecting system in an adult male, in which the right upper moiety was hydronephrotic and complicated by a proximal ureteric stricture and calculus. The patient was managed successfully with a tailored surgical approach.

Case Capsule

A 44-year old male presented to the Emergency Room with history of sudden onset colicky right-sided flank pain associated with nausea, vomiting and fever with chills. No history of any lower urinary tract symptoms. Patient was known Diabetic and Hypertensive on medications since 3 years. Patient had a complex past history of multiple intervention for Urolithiasis: Bilateral PCNL; first right side and followed by left side 1 month later in 2001, left sided PCNL in 2011, Bilateral ESWL in 2014, Right sided PCNL in 2017.

Physical examination revealed right sided renal angle tenderness. Urine analysis showed 20-30 pus cells with microscopic hematuria and 2+ sugars. Blood investigations revealed raised serum creatinine-1.81 mg/dl, HbA1C-9%, rest other blood investigations were within normal limits. After optimization patient was subjected to CT KUB Plain. CT KUB Plain revealed Bilateral Duplex collecting system with hyperdense calculi in the upper moiety of right kidney (involving the mid pole region) sized 7.7 x 5.7mm and 6.4 x 6.1mm respectively. Another round hyperdense calculus measuring 3.1 x 2mm at the distal end of upper moiety ureter causing retrograde hydroureter. Moderate to severe dilatation of calyces and renal pelvis noted at the upper moiety of right kidney. Paper thin parenchyma of the upper moiety seen. Left renal scarring noted. Bilateral perinephric fat stranding seen. Bladder was normal (Figures 1A, 1B and 1C).

After stabilization and surgical fitness, patient was then shifted to the Operation Room. Under General

Anaesthesia and Antibiotic Prophylaxis, with the patient in lithotomy position, Rigid 22Fr 30-degree Cystourethroscopy done. It demonstrated four normally sited, ureteric orifices on the inter-ureteric ridge. Rest of the bladder was normal. Two 0.0338in. Terumo guide wires were negotiated retrogradely under fluoroscopy up to the renal pelvis of both the right ureter. It was able to negotiate guide wire with difficulty in the upper moiety. Right RGP showed stricture at L5 level (Figure 2 and 3). 6.5Fr semi-rigid Ureterorenoscopy done- Lower moiety appeared normal. Right upper moiety revealed a soft calculus approximately 5mm embedded in the distal ureter and a stricture at L5 level. Turbid urine with pus flakes was present. Holmium Laser Lithotripsy of the upper moiety ureteric calculus done and after multiple attempts and with difficulty 5Fr/24cm Double-J (DJ) stent negotiated beyond the stricture and placed in the right upper moiety (Figure 4). Patient was kept under regular follow up and good antibiotic cover with serial CBC and RFT assessment. Despite of the medications and surgery, creatinine remained to be high even after a month. Patient again had symptoms of fever with chills, flank pain and nausea. Patient was optimized again and then subjected to definitive surgery.

Under General Anaesthesia and Antibiotic Prophylaxis, Rigid 22Fr 30degree Cystourethroscopy done and the previous right DJ stent was removed. Right upper moiety ureter cannulated with guide wire and was able to negotiate it. Right 6.5Fr semi-rigid Ureterorenoscopy done in the upper moiety. Gripping was elicited and was unable to pass beyond the mid to upper ureter. Lower moiety check scopy was normal. Right RGP done revealed- stricture at L3-L4 level and even with Flexible Ureterorenoscopy of the right upper moiety same findings elicited (Figures 5 and 6). Two different ureteric catheters were the placed in the upper and lower moiety

respectively for PCNL and identification purposes. 16Fr Foleys placed. Patient was given prone position and Right sided Percutaneous Nephrolithotomy (PCNL) done by Postero-inferior calyceal puncture. 3 lower calyceal calculi measuring approximately 10mm removed and sent for stone analysis (Figure 7). As there was frank turbidity and pus flakes in the system, nephrostomy tube was placed. Then the patient was given Left Lateral position. Right sided 12th rib incision taken. After dissection, upper and lower moiety ureters traced. Unhealthy, pale, fibrosed around 3cm stricture of right upper moiety ureter at L3-L4 level confirmed. The stricture excised and the margin sent for Histopathology. Right Uretero-Ureterostomy done of the upper and the lower moiety segments in side to side, tension free, water tight fashion with Vicryl 4-0 over 6Fr/26cm DJ stent (Figures 8 and 9) and 20Fr drain was placed. Closure done. Surgery and the Post-operative recovery was uneventful.

Good antibiotic cover given. Serial blood investigations assessment done. Nephrostomy tube was removed on postoperative day 2. Foleys catheter was removed on post-operative day 3 and the drain was removed on post-operative day 4. Creatinine was showing a reducing trend and reached to a normal level of 1.1mg/dl. Patient reported no other symptoms and was getting clinically better. Patient was discharged on post-operative day 6 and was kept under close follow up. No complications encountered. Skin Staples removed on the 12th day. Stone analysis showed Calcium oxalate stones. Histopathology of the stricture revealed Granulomatous inflammation. Patient was then worked up for Genito-urinary Tuberculosis comprising of AFB smears and GeneXpert and was found to be negative. After 6 weeks, X-ray KUB done confirmed complete stone clearance. Right DJ stent was removed. Patient is doing well with normalisation of

all the blood parameters, and the creatinine after 6 weeks was in the normal range of 0.8mg/dl.

Discussion

Duplex kidney and ureteral anomalies are among the more frequently encountered congenital abnormalities of the urinary tract. These anomalies are categorized as either complete or incomplete duplications, depending on whether the duplicated ureters remain separate or join before entering the bladder. The estimated prevalence in the general population ranges between 0.2% and 2%, with unilateral presentation being more common and a higher incidence reported among females⁸.

Most duplex systems are detected incidentally during routine imaging. When symptomatic, they often manifest due to complications such as urinary tract obstruction, vesicoureteral reflux (VUR), stone formation, or associated anomalies including ureterocele, ectopic ureteral insertion, or other obstructive defects⁹. The unique anatomical configuration of these systems predisposes to urinary stasis and recurrent infections, which, in turn, increases the risk of urolithiasis. In the present case, multiple factors may have contributed to stone formation and the development of a proximal ureteric stricture. These include recurrent urinary tract infections, mechanical obstruction from ureteral narrowing associated with the duplicated anatomy, and potentially delayed stone passage due to impaired drainage or prior instrumentation¹⁰.

The embryologic origin of ureteral duplication lies in the abnormal development of multiple ureteric buds from the Wolffian duct during the fourth to fifth week of gestation. If these buds persist and develop independently, they give rise to separate ureters, leading to a complete duplex system¹¹. Although unilateral complete duplication is more common, bilateral presentations have also been documented¹³. Urolithiasis

can develop in either moiety of a duplicated system, particularly in the presence of structural anomalies such as ureterocele or pelvi-ureteric junction obstruction¹².

In this case, the stone was located in the proximal ureter of the upper moiety and was complicated by a concurrent ureteric stricture, an unusual presentation that deviates from the Meyer-Weigert law. This embryological principle states that the upper pole ureter tends to have an ectopic insertion and is more susceptible to obstruction, while the lower pole ureter is prone to reflux¹⁴. While lower moiety involvement is more frequently reported, our case highlights an exception, underscoring the variability in clinical presentation.

Imaging studies are pivotal in diagnosing and characterizing duplex systems. While ultrasonography remains a valuable initial tool, it may not fully delineate the ureteral anatomy or associated pathologies. Advanced imaging modalities such as CT urography and MRI provide a more comprehensive view of the collecting system, enabling detailed assessment of anomalies such as ectopic ureters, ureteroceles, and obstructive stones⁴. These tools are crucial in preoperative planning and choosing the appropriate therapeutic strategy.

In a series reviewed by Amis et al., various etiologies of lower pole moiety obstruction were identified, including ureteropelvic junction obstruction, bladder tumors, ectopic ureterocele, and ureteral calculi¹⁵. The current case, however, involved the upper moiety, adding to the spectrum of anatomical and clinical variations observed in duplex systems.

Anatomically, the terminal portions of duplicated ureters may be enveloped in a common adventitial sheath, with the muscular layers of both ureters interweaving particularly in regions where they cross adjacent pelvic structures. In males, this typically occurs near the level of the vas deferens; in females, where the ureters cross the

uterine artery. This close anatomical relationship, along with a reduced ureteral caliber, can complicate endoscopic procedures and increase the likelihood of iatrogenic injury during instrumentation¹⁶. Typically, the ureter draining the lower moiety inserts laterally and superiorly to that of the upper moiety. The lower moiety is more frequently affected by pathological processes, including VUR and obstruction¹⁷. However, our patient demonstrated an uncommon pattern, with the upper moiety being affected by a stone and proximal ureteric stricture. This deviation from the classic Meyer-Weigert rule highlights the importance of individualized diagnostic assessment and tailored management in patients with duplex renal systems.

Conclusion

This article summarizes the importance of the Duplex collecting system sufferer's follow-up as this clinical variety could remain uncomplicated with no consequential morbidity or lead to complication and pose the patient's life at risk of end stage kidney disease. The previously asymptomatic Duplex renal system may develop signs and symptoms later. When identified without symptoms or evidence of renal function compromise, the integrity of the renal moieties may have to be closely monitored for the rest of the life. A thorough evaluation with a high index of suspicion is required in individuals presenting with flank pains to quickly identify Duplex system associated disorders quickly and thus prevent the renal compromise. As Urolithiasis is a common obstacle in developing nations, regular follow-up and care is required in such group of patients.

It thus demonstrates a very rare incidence of Bilateral Complete Duplex systems with ureteric stone and stricture in one moiety system that posed a surgical challenge. Thus double the problems of stones and

stricture in double the systems caused actual trouble and was thus managed successfully.

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Legends Figures

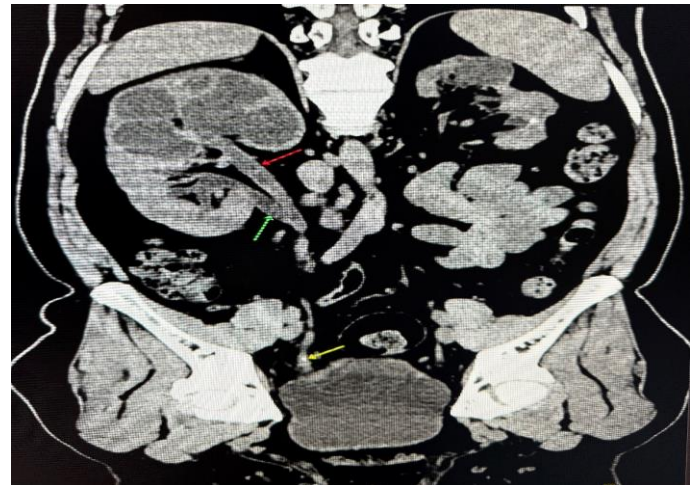


Figure 1A: Sagittal cut of CT KUB Plain showing Right Duplex pelvicalyceal system with Duplex ureter (upper moiety (red arrow), lower moiety (green arrow) with moderate to severe dilatation of calyces and renal pelvis at upper moiety of right kidney. Well-defined ovoid hyperdense calculus of size measuring 3.1x2 mm noted at the distal end of upper moiety ureter causing retrograde hydroureter (yellow arrow).



Figure 1B: Sagittal cut of CT KUB Plain showing Left Duplex pelvicalyceal system with Duplex ureter (upper moiety (red arrow), lower moiety (green arrow) with left renal scarring.

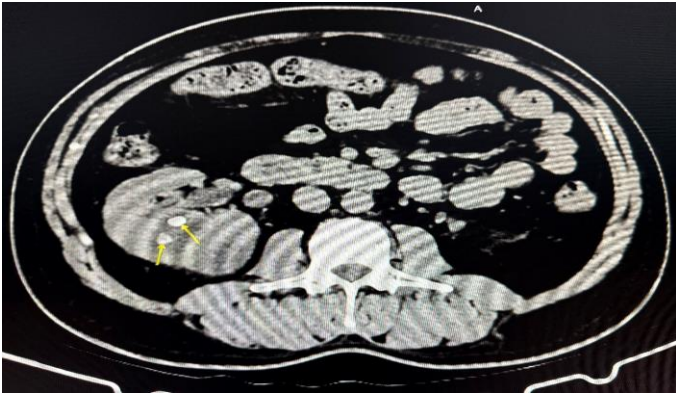


Figure 1C: Axial cut of CT KUB Plain showing moderate to gross right upper moiety hydronephrosis with hyperdense calculi in the upper moiety of right kidney measuring 7.7mm and 6.4mm respectively.



Figure 2: Intraoperative image of Right RGP showing initially unable to negotiate guide wire in right upper moiety ureter. Lower moiety guide wire negotiated successfully.



Figure 3: Right RGP showing stricture in the upper moiety segment at L5 level.



Figure 4: DJ stent negotiated in the right upper moiety ureter with difficulty beyond the stricture segment.

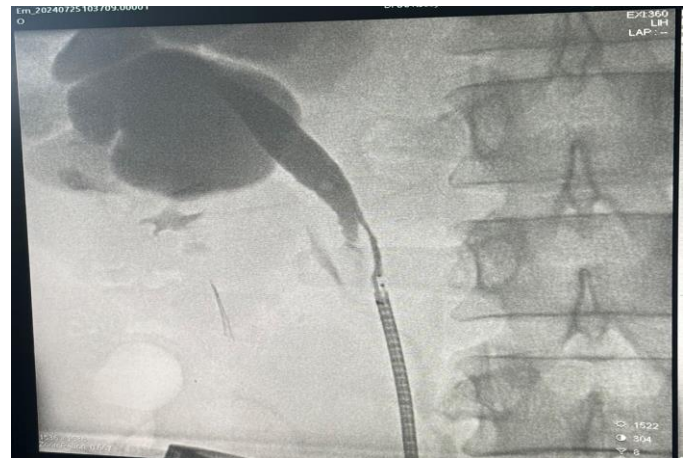


Figure 5: Right upper moiety flexible URS with RGP showing complete narrowing at L3-L4 level with upper moiety gross hydronephrosis.



Figure 6: Flexible URS showing upper moiety stricture.



Figure 7: Intraoperative image of stone retrieved by Right PCNL.

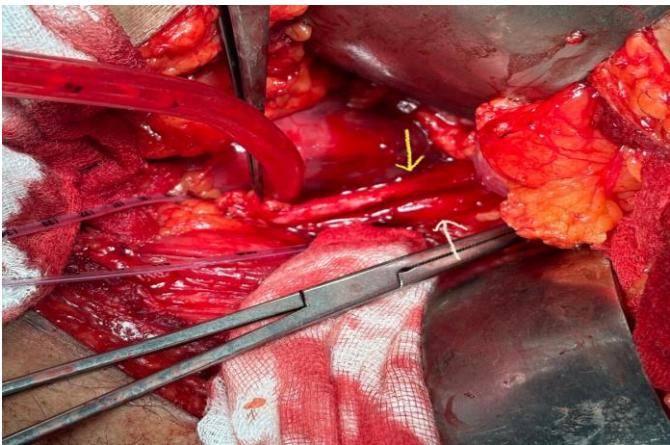


Figure 8: Intraoperative image showing Right Duplex ureter; upper moiety (yellow arrow), lower moiety (white arrow).



Figure 9: Right Uretero-Ureterostomy done of the upper and the lower moiety segments in side to side, tension free, water tight fashion with Vicryl 4-0 over 6Fr/26cm DJ stent.