BILATERAL COMPLETE URETERAL DUPLICATION WITH OBSTRUCTING STONES IN BOTH LEFT URETERS: A CASE REPORT AND A REVIEW OF LITERATURE

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ABSTRACT

Congenital anomalies like duplex renal collecting systems are rare developmental conditions. Among these anomalies, ureteral duplication is rarely observed, with partial ureteral duplication comprising a majority of cases. Bilateral ureteral complete duplication, where fully duplicated ureters drain separately into the bladder via separate orifices, is extremely rare, making it a seldom-documented anomaly in the medical literature. A case of bilateral complete ureteral duplication complicated by calculi in both the upper and lower moieties of the left kidney presented to the urology outpatient department with complaints of left flank pain for one month. His computed tomography-urogram revealed complete duplication of bilateral collecting systems with bilateral double ureters with an 8.2×5.9 mm^2 sized stone in the pelvi-ureteric junction of the left upper moiety and 11.9×5.6 mm^2 sized stone in the proximal ureter of left lower moiety causing hydronephrosis of both upper and lower moiety pelvicalyceal system. Calculi in both the upper and lower moieties were cleared in a single setting using a Ureteroscopic Lithotripsy. Duplex renal systems, while often asymptomatic, are linked to a higher occurrence of treatable conditions like stones and renal reflux diseases. The duplicated structure may lead to stagnant urine, causing obstructions and consequently infections. Stones that obstruct the ureter for a prolonged period can cause renal impairment and require active intervention. Currently, flexible or semirigid ureteroscopy with laser lithotripsy is the standard procedure. Even in this case, both stones were effectively managed in a single session using a standard procedure.

KEYWORDS

Complete ureteral duplication, hydronephrosis, kidney, stones

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INTRODUCTION

A duplex collecting system can be described as a kidney with two pelvicalyceal systems, generally referred to as upper and lower moieties. In complete duplication, two ureters emerge from the pelvis and empty via two separate orifices in the bladder. In contrast, an incomplete duplication or a bifid system is a form of duplication with two pelvicalyceal systems joining at the ureteropelvic junction or before emptying into the bladder.[1] Bilateral duplication of the ureter is rare occurring in 0.13% of individuals while 0.8% of individuals have a bifid ureter.[2] Patients with ureteral duplication are often asymptomatic but may potentially have urinary stones, ureterocele, vesicoureteral reflux (VUR), and obstructive uropathy.[3] We report a case of a 41-year-old male with complete bilateral ureteral duplication complicated by obstructing stones in both left ureters. The case report has been reported in line with the SCARE criteria.[4]

Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

CASE REPORT

A 41-year-old male presented to the Urology Outpatient Department (OPD) of Nepal Medical College Teaching Hospital with complaints of left flank pain for 1 month. Notably, he also had a history of right ureteroscopic lithotripsy (URSL) six years prior to his presentation to the hospital. A CT-urogram revealed an 8.2×5.9 mm calculus in the pelviureteric junction of the left upper moiety causing significant dilatation of the calyx and an 11.9×5.6 mm sized calculus in the proximal part of the left lower moiety ureter resulting in moderate proximal hydronephrosis (Fig. 1). Consequently, a left-sided URSL procedure was planned to address the calculi. During cystoscopy, two separate ureteric orifices were identified on each side of the trigone. Subsequent ureteroscopy also revealed two separate ureters on the left side with each ureter draining independently into the urinary bladder.

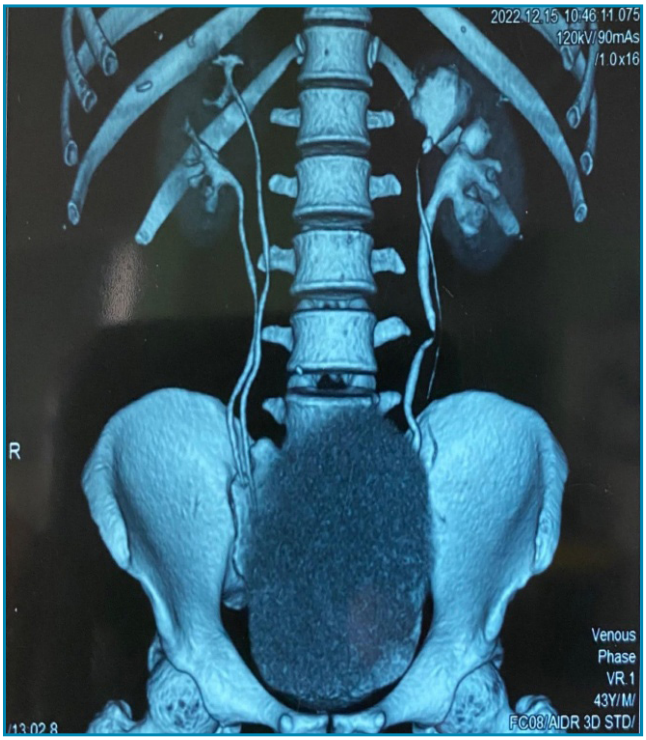

Fig. 1: Preoperative CT-urogram

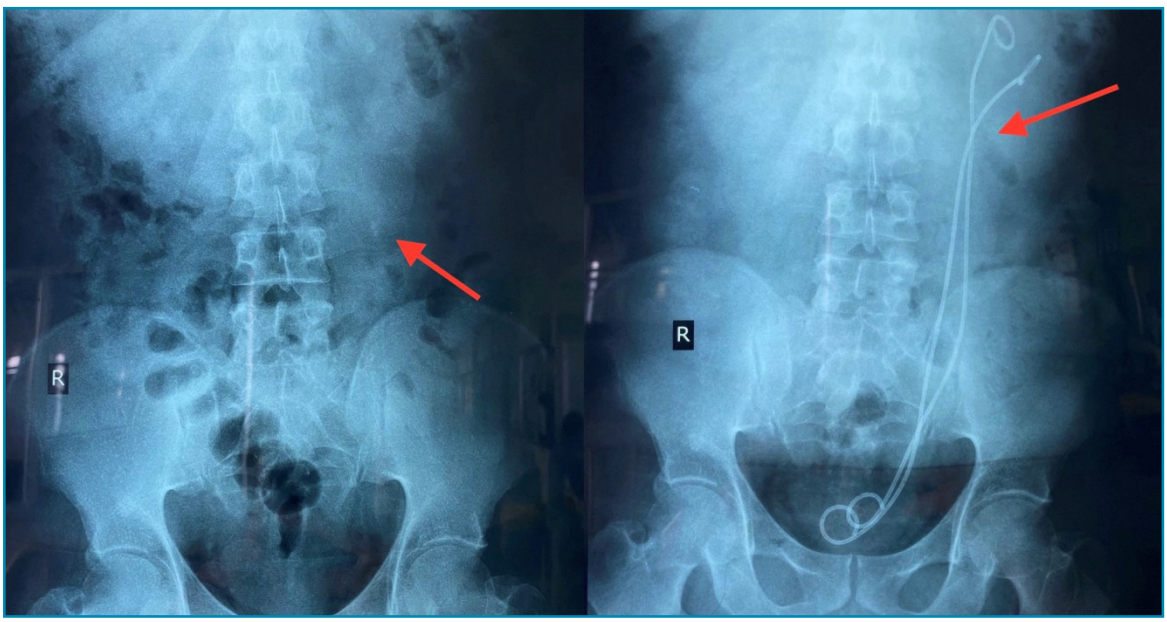

Fig. 2: Pre-operative and post-operative Plain X-ray showing stones and double J stents respectively

A 6.5 Fr semi-rigid ureteroscope was negotiated in the left lower moiety ureter, where the stone was visually confirmed. Laser lithotripsy was performed by breaking the calculus into fragments which were subsequently removed. A 7.5 Fr flexible ureterorenoscope was negotiated into the upper moiety ureter and the stone was completely fragmented with laser. 6 Fr Double J (DJ) stent was inserted under fluoroscopic guidance in both ureters of the left kidney. A postoperative plain radiograph demonstrated the stents to be in a good position (Fig. 2). The postoperative period was uneventful and the patient was discharged on the 3rd postoperative day. As per the established protocol, the standard practice is to remove a DJ stent within a timeframe of two weeks following the procedure. However, considering the patient's unique developmental anomaly, the decision was made to extend the stent removal to six weeks post-procedure. Following the removal of the DJ stent, the patient remained symptom-free during the follow-up visits.

DISCUSSION

The urinary system develops from two sources: metanephric blastema and mesonephric duct. The ureteric bud arises as a diverticulum from the caudal end of the mesonephric duct. This diverticulum elongates and later fuses with the metanephric blastema to form the renal pelvis, further dividing into the major and minor calyces. Abnormalities in the branching pattern of the ureteric bud cause the duplication of the collecting system. In cases of complete duplication, the ureteral bud arises twice, resulting in a double ureter with a double opening into the urinary bladder.[5]

In a study of 5196 excretory urogram performed on both children and adults, duplex systems were present in 1.8% of patients. The condition was bilateral in only 0.3% of patients, while complete duplication was demonstrated in less than one-third of the patients.[6] Therefore, our case complete bilateral ureteral duplication is a rare condition.

Although duplex renal systems are usually symptom-free and often go unreported, they are usually associated with an increased incidence of treatable disorders such as urinary stones, and reflux disease. These issues occur more frequently in individuals with this anatomical variation, likely due to stasis making obstructions and infection rates higher.[7]

An article published in 2015 in the International Journal of Surgery by Aiken *et al*[8] from the University of West Indies claims their case report as the first reported case of bilateral complete ureteral duplication with calculi obstructing both limbs of the left double ureter in the English medical literature. Owing to the narrow caliber of the ureters, gaining access to them was challenging. These authors, thus suggested passive ureteral dilatation through pre-stenting before ureteroscopy for uncomplicated retrograde ureteroscopic access. In our current report, the ureteral lumens were wide enough to accommodate ureteroscopes without the need for ureteral dilatation or pre-stenting.

Studies have confirmed that prolonged obstruction of the ureter by stones can lead to renal impairment, necessitating the active removal of stones. More recently Dino *et al*[9] reported a case of the complete duplex of the left ureter with lower moiety hydronephrosis secondary to ureteral stone in a patient who had to undergo left retroperitoneal exploration with partial nephrectomy. A delayed diagnosis due to low suspicion by primary healthcare providers led to significant kidney damage that could have been avoided with prompt endoscopic treatment.

In a previous case reported at King Abdulaziz University in Saudi Arabia, a patient with bilateral complete duplication of ureters had stones obstructing all four ureters, which were successfully removed through a combination of holmium laser lithotripsy and extracorporeal shock wave lithotripsy (ESWL).[10]

As per the European Association of Urology Guidelines, the initial recommended treatments for ureteral stones are ESWL and URSL.[11] While these two methods remain the most widely used approaches, the optimal treatment of choice depends on various factors, including stone size, composition and location, clinical factors, equipment availability, and surgeon's expertise.[12] For managing ureteral stones sized more than 10 mm, the current standard procedure is flexible or semirigid URSL.[11] In our specific case, both stones were successfully addressed in a single session and the patient was rendered stone-free using this established method.

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