

Laparoscopic management of double moiety: A case report with review of literature

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ABSTRACT

Duplication of the ureter is one of the most common anomalies of the urinary tract. Ureterocele is a developmental anomaly with cystic dilation of the distal aspect of the ureter and is often associated with some urological anomaly, such as a duplicated system or stenotic ureteric orifice. Early detection of these anomalies during the antenatal period has dramatically increased due to advances in imaging technology. However, a few undiagnosed adults also exist. It may remain asymptomatic but may cause repeated urinary tract infections or calculi. This is a case report of a 47-year-old female who had bilateral duplex collecting systems with right ureterocele. The patient had a recurrent urinary tract infection. She underwent cystoscopic deroofting and a laparoscopic right total nephrectomy. Congenital anomalies of the urogenital system should be considered in patients with chronic or recurrent infections. Multimodal imaging techniques such as ultrasonography, computed tomography, or magnetic resonance imaging should be used to confirm the diagnosis, especially before surgical management. The rationale for reporting this case is to highlight this rare condition and the feasibility of its minimally invasive therapy.

Key words: Congenital anomaly, Duplex collecting system, Laparoscopic nephrectomy, Ureterocele, Urinary tract infection

Duplications of the ureter represent one of the most common anomalies of the urinary tract. The duplex collecting system refers to the kidney with two pelvicalyceal systems, which may have either a single or bifid ureter (partial duplication) or a double ureter draining separately into the urinary bladder (complete duplication). It may be associated with other abnormalities such as ureterocele, ectopic ureter, vesicoureteral reflux (VUR), or ureteropelvic junction obstruction (UPJO) [1]. An ureterocele is a congenital disorder that causes a submucosal cystic dilation of the terminal ureter. It has a fibromuscular wall lined with urothelium that swells and causes an abnormality in the flow of urine to the bladder. The appearance is that of a fluid-filled structure within the bladder and is often described as a cyst within a cyst.


CASE REPORT

A 47-year-old female presented with chief complaints of right-sided flank pain with dysuria, burning micturition, and fever for 7 days, for which she was being treated by her family physician with tablet nitrofurantoin 100 mg twice a day. However, she did

not respond and came to the emergency ward with worsening right-sided dull aching continuous flank pain with 3–4 spikes of fever daily associated with anorexia, dysuria, and myalgia. She gave a history of recurrent urinary tract infections.

On examination, she appeared dehydrated and was febrile. Her pulse was 96 beats/min, and her blood pressure was 90/60 mmHg. On abdomen examination, she had right-sided lumbar region tenderness and right renal angle tenderness.

She was admitted to the intensive care unit under the general physician. Her white blood cell count was 18,000/mm³, C-reactive protein was 175.5 mg/dL, and Serum Creatinine was 1.3 mg/dL. The urine was turbid, with 20–24 red blood cells/hpf and 40–45 pus cells/hpf. The urine culture study grew *Escherichia coli*. Bedside ultrasonography evaluation was suggestive of the right hydronephrotic kidney with a dilated ureter filled with debris. The specialist in urology was then called in and he advised to get computed tomography (CT) urography done. After stabilizing the patient, CT urography was done. It was suggestive of a 12×8.4 cm enlarged right kidney with gross hydronephrosis and parenchymal thinning with duplication of the right pelvicalyceal system with dilatation of the upper moiety with internal echoes suggestive of pyonephrosis of the upper moiety with hydroureter

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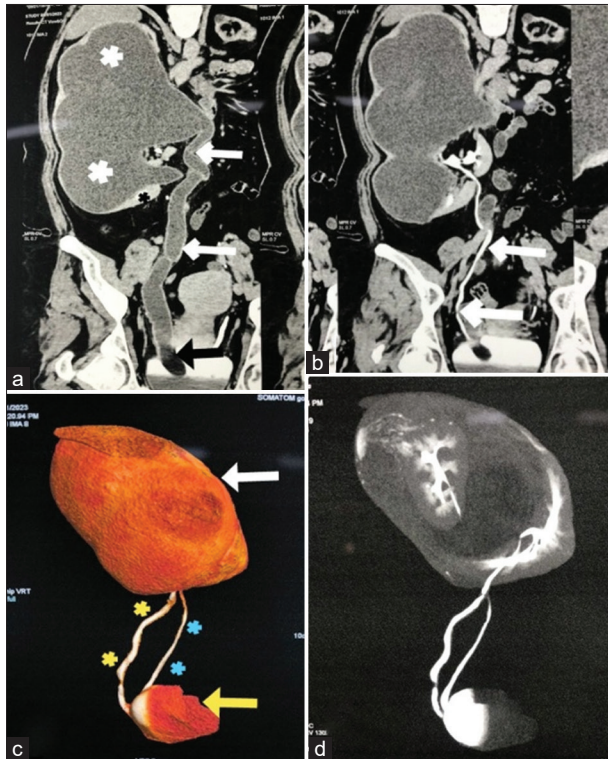


Figure 1: (a) CT urography coronal section shows left kidney with minimally functional lower moiety (black asterisk), non-functional upper moiety (white asterisks), hydro-ureter of upper moiety (white arrows), and ureterocele (black arrow), (b) CT urography coronal section shows normal caliber 2nd right ureter of lower moiety (white arrows), (c and d) CT urography 3D reconstruction showing hydronephrotic upper moiety (white arrow), bladder (yellow arrow), and the 2 ureters: hydroureter (yellow asterisks) and normal ureter (blue asterisks). CT: Computed tomography

and ureterocele at the right ureterovesical junction (Fig. 1a-d). Then, a renal nuclear scan was performed, which was suggestive of <10% of the functional right kidney. Following this, it was decided to take her up for surgical intervention.

A cystoscopic de-roofing of the ureterocele was performed by the specialist endo-urologist (Fig. 2a and b). Subsequently, the patient's position was changed to lateral, and laparoscopic intervention was initiated with the intention to excise only the upper non-functioning moiety. However, during the intraoperative assessment, the remnant right lower moiety appeared to be too thinned out with a very small volume and paper-thin parenchyma. Hence, an “on-table” decision to perform a total nephrectomy was made. The patient and her family were pre-operatively informed and counseled about the possibility. She then underwent a laparoscopic total right nephrectomy (Figs. 2c-f, 3a-f, 4a, and b). The same was performed through 4 trocars (Fig. 4c). The lowermost trocar was widened to retrieve the specimen (Fig. 4d).

On post-operative day 3 (POD), the drain was removed, and she was discharged on POD 5. On her POD 10 outpatient department visit, all her wounds had healed well.

DISCUSSION

The duplex collecting system is one of the most common urological anomalies. The incidence of duplex renal collecting systems is

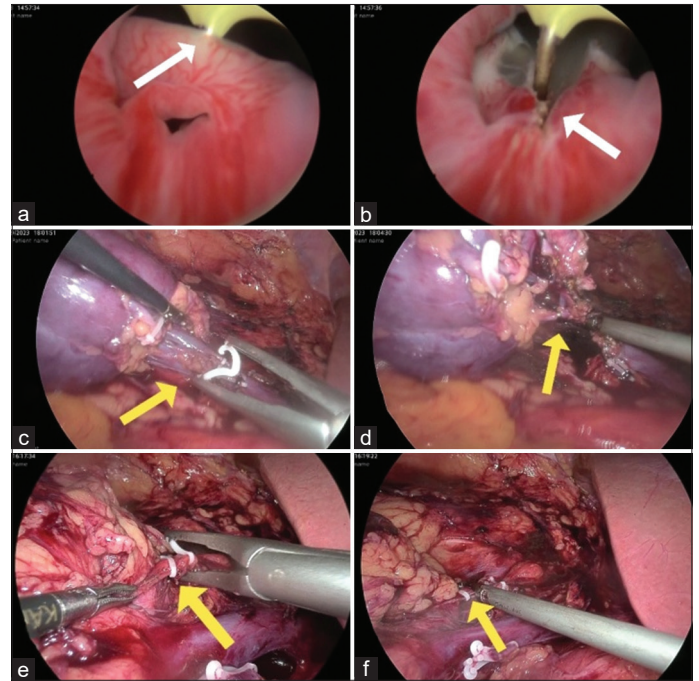


Figure 2: (a and b) Cystoscopic de-roofing of the left ureterocele (white arrows), (c) Hemoclip being applied to renal vein (yellow arrow), (d) Division of renal vein (yellow arrow), (e) Clipping of renal artery (yellow arrow), (f) Division of renal artery (yellow arrow)

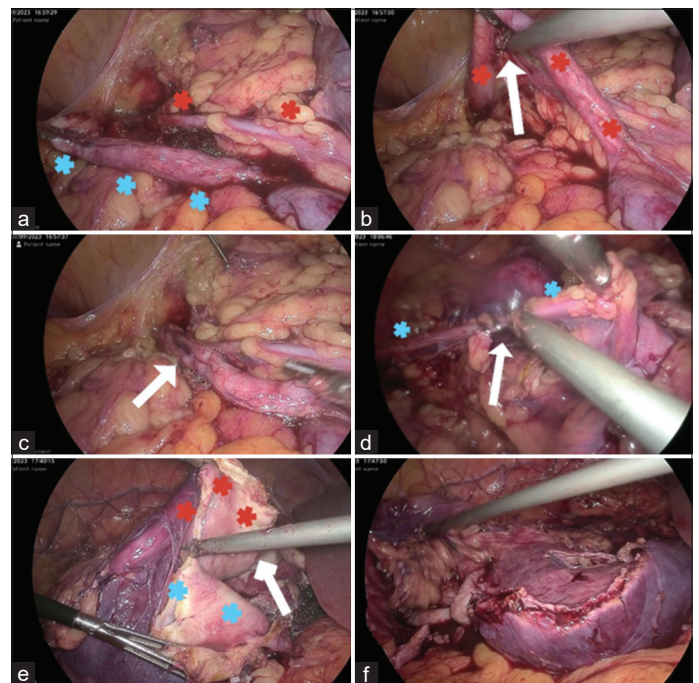


Figure 3: (a) Hydroureter (blue asterisks) of upper moiety and the 2nd normal caliber ureter of the lower moiety (red asterisks), (b) Hydroureter (red asterisks) being dissected (white arrow), (c) Dissection of lower end of hydroureter (white arrow) before clipping and division, (d) Clipping and division of normal caliber 2nd ureter (white arrow), (e) Dissection of upper (red asterisks) and lower (blue asterisks) moieties during nephrectomy, (f) Thinned out lower moiety seen after resection of non-functional upper moiety

in the range of 0.5–3%, with females being more affected than males (2:1) [1]. The kidney is divided into various segments, each supplied by a single “end” arterial branch that arises from one of the main renal arteries. Three groups of primitive vasculature

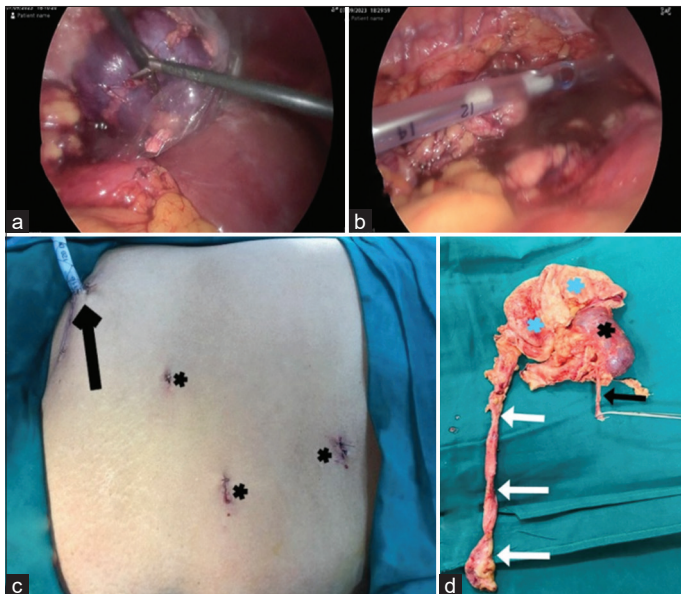


Figure 4: (a) The specimen being retrieved in a plastic bag, (b) Tube drain introduced *in situ*, (c) Trocar sites (black asterisks) and specimen retrieval cum drain site (black arrow), (d) The specimen showing hydroureter (white arrows), normal caliber 2nd ureter (black arrow), upper moiety (blue asterisks), and lower moiety (black asterisk)

derive from the renal arterial tree and coalesce to form the mature vascular pattern for all retroperitoneal structures. The renal parenchyma is divided into five segments based on the vasculature: Apical, upper, middle, lower, and posterior. The lower renal segment, however, is often supplied by an accessory vessel [2]. Two ureteral buds rarely develop independently from a single mesonephric duct during embryonic development, resulting in a duplex kidney with ureteral duplication [3].

An ureterocele is defined as a congenital disorder that occurs due to submucosal cystic dilation of the terminal ureter. Its incidence is 1:4000 people, occurring 4 times more often in women with slightly more predominance on the left side, and 10% of cases are bilateral [4]. Depending on their location, ureteroceles are most commonly referred to as intravesical or extravesical [5]. Intravesical ureterocele is completely confined within the bladder and is associated with good renal function. An intravesical ureterocele causing obstruction can also be called a stenotic ureterocele. Extravesical ureterocele is defined as one involving some portion of the ureterocele in the bladder neck or urethra and is associated with poor renal function [6]. The classic explanation of ureterocele formation is that of Chwalle, who postulated that distal ureteral expansion is produced by the failure of rupture of a distal membrane at the ureteral orifice (Chwalle membrane) [7].

A various range of symptoms might manifest according to the associated distortions and the case severity. The vast majority of cases are diagnosed in early childhood. However, a larger percentage of patients are asymptomatic and diagnosed incidentally. The primary and most common symptoms are those related to VUR and urinary tract infections [8]. One of the major complications associated with duplication is that of an obstruction, which could be calculus (anywhere along the ureter)

or non-calculus obstruction at a confluence of the ureter or bladder insertion [9]. As per the Meyer-Weigert rule, the upper pole is generally seen as ectopic and therefore dysplastic due to obstruction, whereas the lower pole is related to VUR [10]. In our case, the patient had a right complete duplicated kidney with a lower draining ureter and left upper hydronephrotic segment and a right ureterocele causing a dilatation in the proximal part of the right kidney. Amis *et al.* analyzed 11 cases of obstruction involving the lower pole moiety of the kidneys. There were complete duplications in seven cases. The various pathological causes were due to UPJO in 2 cases, bladder tumor in 2 cases, ectopic upper pole ureterocele compressing and obstructing the ureteral orifice to the lower pole in 1 case, and ureteral stones in 2 cases (located at the mid and ureterovesical junction) [11]. Similarly, our patient had a right ureterocele, which caused prolonged obstruction that went unnoticed for an extended period.

The first modality used was ultrasound, which showed our patient's multicystic mid- and upper-pole segments. Later, other imaging modalities used to confirm or rule out conditions must be conducted. To rule out reflux, a voiding cystourethrography can be conducted. Intravenous urography (IVP) and CT urography confirm the duplex system and other obstruction causes, and nuclear imaging is done to realize the split function and level of obstruction [11]. Another option for clarifying the anatomy of duplication anomalies is pre-operative cystoscopy combined with a retrograde ureteropyelogram before surgical correction [12].

Management depends on the cause of obstruction and the cortical loss in the renal moiety [11]. The majority of cases can be managed endoscopically, laparoscopically, and through open surgery in resource-limited facilities [13]. The role of treatment is to relieve the obstruction cause or relieve the pain by removing part of the kidney (partial nephrectomy) [11]. In cases where the upper renal moiety function is poor, upper pole heminephrectomy is a standard surgical treatment [13]. The ureterocele management should be individualized based on clinical presentation, type of ureterocele, patient's age, and other clinical variables that may contribute to the management's best choice [14]. The advantages of endoscopic treatment are simplicity and minimal invasiveness, but there is no consensus on its effectiveness for treating ectopic ureterocele. It has been reported that 50–80% of cases after initial endoscopic treatment need secondary surgery for ectopic ureterocele [15]. If the diagnosis had been made sooner, our patient would have benefited from a simple endoscopic procedure. However, her delayed presentation to appropriate care resulted in this unfortunate complete renal moiety parenchymal loss. A similar result was reported by Anyimba *et al.* [9]. A review of literature on other cases of double moiety with concurrent ipsilateral ureterocele is summarized (Table 1) and reveals that this is only the third such reported case in world literature [16,17]. It is the only such reported case in world literature that was managed in the same sitting by minimal access surgery (cystoscopy and laparoscopy).

Table 1: Review of literature

S. No.	Journal/year of publication	Author/s	Age/sex/presentation	Radiological diagnosis	Management
1	International Journal of Surgery Case Reports/2021	Prahara and Eldo [16]	25 y/F, right flank pain	Right complete duplex collecting system with giant right ureterocele	Cystoscopic deroofting and staged laparoscopic partial nephrectomy
2	BMJ case reports/2017	Ruchir <i>et al.</i> [17]	36 y/F, large abdominal lump	Right complete duplicated collecting system with ureterocele with giant right hydronephrosis	Cystoscopic deroofting and open total nephrectomy

CONCLUSION

As seen in this report, the previously asymptomatic duplex renal system may develop symptoms later in life. 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 their lives. A thorough evaluation with a high index of suspicion is required in individuals presenting with flank pains to quickly identify missed duplex renal systems and duplex system-associated disorders quickly and thus prevent the attendant renal moiety parenchymal loss. Furthermore, as seen here, laparoscopic management of double moiety, where indicated, is feasible in an advanced setup coupled with the concurrent availability of the requisite expertise.

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