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CASE STUDY

UNILATERAL COMPLETE DUPLICATION OF URETER WITH SUCCESSFUL MANAGEMENT

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ABSTRACT

The ureter is a subject to a number of natural variations, and duplex ureter is one of the rare congenital abnormalities affecting the ureter, occurring in around 1% of general population[1]. We here are reporting a case of a 25 year old gentleman, who had come with complaints of pain on the left flank and LUTS (Lower urinary tract symptoms). Investigations revealed that he had ahydronephrotic left kidney, with the upper pole showing pyonephrosis with suboptimal function. This was associated with a unilateral duplex moiety with complete duplication of ureters on the left side and ectopic insertion of the upper moiety ureter into prostatic urethra. The lower moiety collecting system was hydronephrotic but non obstructing and non refluxing. The patient subsequently underwent aleft sided partial upper pole nephrectomy. Most cases of duplex ureters present in childhood and adolescence, and are usually asymptomatic. This case report emphasizes the need to consider the possibility of such anomalies and their varied spectrum of presentation and the need for screening of such anomalies at an earlier stage.

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INTRODUCTION

Ureters are pair of muscular tubes that convey urine from kidneys to the urinary bladder. Embryologically, ureters are developed as an elongation of the diverticulum of the mesonephric duct, known as ureteric bud. An accessory ureteral bud may develop from the mesonephric duct, thereby forming a duplicated ureter. Duplication of the ureter is a congenital anomaly which accounts for 1% of incidence (Husain *et al*., 2013). Duplex ureters tend to have a potential for future complications, such as collecting system obstruction, formation of stones (lithiasis), dilatation (ureterocele) and vesicoureteral reflux (Prakash *et al*., 2011). Though the presence of duplicated ureter may be an asymptomatic phenomenon, its presence cannot be ignored. The early detection of this anomaly helps in avoiding these complications. Since the ureter is closely related to neighbouring vessels and organs, its detailed anatomy is essential in surgical and radiological interventional approaches. Duplication is two to five times more common in females than in males and incomplete duplication is three times more common than complete duplication (George *et al*., 1987). The uniqueness of this case we are reporting here is that tho is a case of a 25 year old gentleman, who had come with complaints of pain abdomen, found to have a completely duplicated right ureteric moiety, and successfully underwent partial nephrectomy for associated right kidney upper pole pyonephrosis.

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Case report

We had a 25 year old gentleman, who had presented with complaints of pain abdomen since the past one year over the left flank, which had increased in intensity since the past fortnight. The pain radiated from loin to groin, and was associated with fever and burning micturition. Patient had no other significant medical or surgical past history. On examination, patient was febrile, with mild tenderness positive over left flank. Routine blood investigations revealed a serum creatinine value of 0.9mg/dL, serum urea value of 31mg/dL, total leucocyte count of 10.49 cells/mm3, and heamoglobin 15.4mg/dl. Urine culture sensitivity showed growth of Klebsiella pneumonia spp, whichshowed sensitivity to the combination of piperacillin-tazobactam. Preliminary investigations on ultrasonography showed left kidney sized 8.9 X 3.7 cms, with evidence of 6.6 and 6.3 cm exophytic cyst in upper pole, with presence of a duplex moiety with papery thin cortex of upper moiety with pyonephrosis with moderate hydronephrosis of lower pole moiety, with lefttortuous hydroureter. Patient underwent further evaluation, with a contrast enhanced Computed Tomography (CECT) of the KUB region. CECTshowed entire left ureter appearing duplicated, with gross dilatation and tortuosity involving the entire left ureteric system. The upper moiety ureter was seen causing gross hydronephrosis of upper pole with papery thin cortex. Moderate hydronephrosis was noted involving mid and lower

pole draining into lower moiety ureter, with cortical thickness of 8mm and 9mm respectively. Upper moiety was seen draining below the base of the bladder into prostatic urethra. There was non-opacification of upper moiety ureter noted even on delayed scans. A percutaneous nephrostomy (PCN) was done to drain the pyonephrosis present in the upper pole of left kidney. A urine culture of the PCN fluid was sent, which showed presence of E.coli, which was sensitive to Norfloxacin and Nitrofurantoin, and the patient was treated accordingly. Once his PCN fluid culture showed no growth, the patient underwent a percutaneous nephrostogram, to check the duplex ureter moiety where a radio opaque iodinated dye, was injected into the left kidney via the PCN tube and subsequent X rays taken to note the excretion of dye into the ureter. The dilated and tortuous left upper moiety ureter was seen, draining into the prostatic urethra, with the lower moiety ureter dilated not visualized in PCN gram suggestive of complete left unilateral duplication of ureter Figure 1,2. A micturating cystourethrogram and a retrograde cystourethrogram were done, and they showed a normal study, with no abnormalities revealed in the urethra, with absence of reflux in any of the left ureters. A further analysis with DTPA scan to assess the renal function and drainage pattern was done. The left kidney showed percentage function of 39.57% and the right kidney showing 60.43%. The GFR of the left kidney was 38.94 ml/min, and the right kidney showing 59.48 ml/min. Further, the lower pole moiety of left kidney revealed adequate transit of tracer in dilated pelvicalyceal system with partial hold up of tracer that clears on furesemide injection, hence showing a non obstructive pattern. The upper moiety revealed a sluggish progressive filling up of tracer, with upper pole showing GFR of 7.77 ml/min as compared to the lower pole showing a GFR of 31.2 ml/min.

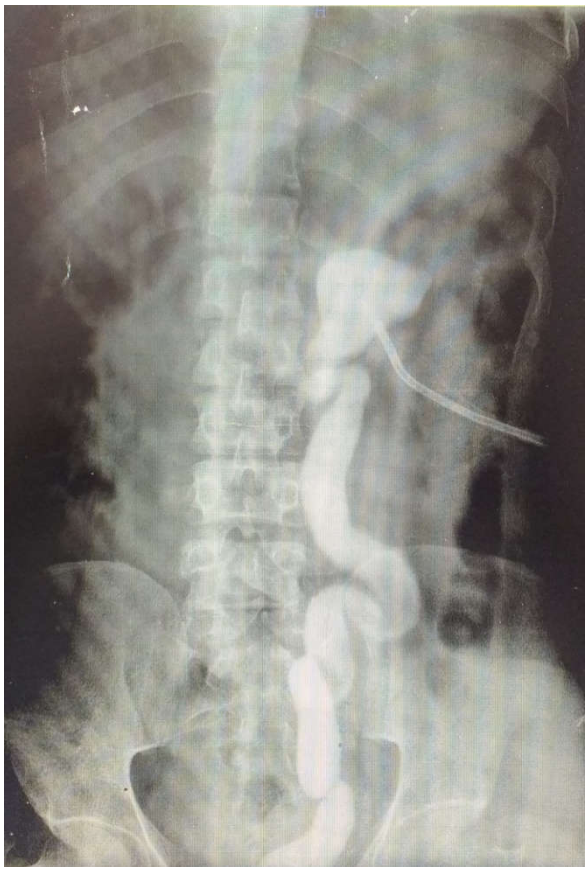

Figure 1. PCN gram showing dilated tortuous upper pole ureter with ectopic insertion beyond pelvic brim

Patient underwent cystoscopic examination with bilateral ureteric retrograde pyelography (RGP) as the percutaneous nephrostogram did not show lower moiety insertion was confirmed to be in the bladder. Subsequently, the patient underwent a laparoscopic partial nephrectomy of the upper pole moiety with ureterotomy. He had an uneventful post operative period and was consequently discharged.

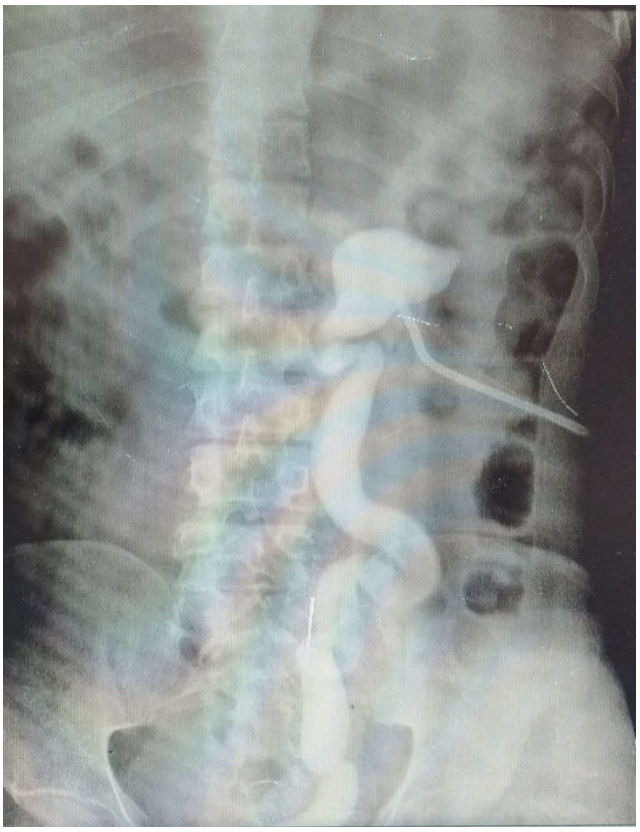

Figure 2. PCN gram showing dilated tortuous upper pole ureter

DISCUSSION

Ureteric anomalies are at important aspect in the realm of urology.They tend to present with varied presentation, from being asymptomatic to presenting with hydronephrosis and pyonephrosis, from being diagnosed prenatally to being diagnosed in healthy adults incidentally, either during routine radiological investigations, or during renal surgeries, anatomical dissection or during autopsy procedures. Embryologically, duplication tends to occurs when two separate ureteric buds arise from a single Wolffian duct and according to the Weiger-Meyer rule (Weigert, 1877; Meyer, 1907), the future lower pole ureter separates from Wolffian duct earlier and thus migrates superiorly and laterally as the urogenital sinus grows.Since the ureter is closely related to neighbouring vessels and organs, its detailed anatomy is essential in surgical and radiological interventional approaches. The blood supply of the duplex ureters at the bladder level, tend to be intertwined within a common ureteric sheath, hence sharing a common adventitial blood supply (Gearhart John *et al*., 2010) Duplication can be variable. At one end of the spectrum, there is merely a duplication of the renal pelvis, draining via a single ureter. At the other extreme, two separate collecting systems drain independently into the bladder or ectopically. Duplex ureters tend to have a potential for future complications, such as collecting system obstruction, formation of stones (lithiasis), dilatation (ureterocele) and vesicoureteral

reflux (Prakash *et al*., 2011). Though the presence of duplicated ureter may be an asymptomatic phenomenon, its presence cannot be ignored. The early detection of this anomaly helps in avoiding these complications. Though usually asymptomatic, Children with duplex kidney may be prone to urinary tract infections because of vesico-ureteric reflux or obstruction. Surgical management is usually required once symptoms occur (Mouriquand *et al*., 2001). The investigation of choice before the advent of computed tomography was the intravenous pyelography study. In this case 3-dimensional CT urography, which has now become the investigation of choice, helps us to clarify ureteric insertion or anomalies. Given the safety of Doppler study and significant differences in flow dynamics of obstructed versus unobstructed ureters, the utility of Doppler ultrasound examination as a useful adjunct to gray-scale ultrasound by improving the accuracy of ultrasound exam in diagnosis of ureteral obstruction (Jandaghi *et al*., 2013). Duplex kidney is an uncommon finding during adult sonographic examinations. Thesensitivity and specificity of sonography in identifying this anomaly are controversial. An experienced hand with good knowledge of the anomaly and its sonographic signs is helpful in more accurately diagnosing this anomaly (Lee *et al*., 2001). MRI can also be considered as an investigative tool in this case, as it can help in define the exact anatomical malformation of the urinary tract, and plan the optimal approach for surgery. Some authors have suggested cystourethroscopy with retrograde catheterization to be carried out in every patient with an ectopic ureter (Husain *et al*., 2013).

The majority of patients with ectopic ureters can be treated by addressing only the upper urinary tract. Whether noted preoperatively or postoperatively, reflux into the ectopic ureter necessitated ureteral stump removal (Plaire *et al*., 1997). Faulty embryogenesis is proposed to explain these findings and the importance of endoscopic positioning of the ureteral orifice is emphasized in determining whether heminephrectomy will be necessary (Duplex Kidneys, 1975). In this patient, there was a complete obstruction of the upper ectopic duplex ureter at the level of joining the prostatic urethra, with the lower ureter opening normally at the vesico-ureteral junction. It is difficult to perform a common sheath re-implantation if the upper pole ureter is situated ectopically (Lawn and Nigam, 2010), and since the upper pole had pyonephrosis, a laparascopic approach aimed at achieving a partial nephrectomy was done and achieved successfully. The upper ureteral stump was left in situ, so as to not cause a de-vascularisation injury to the lower ureter. Laparoscopic retroperitoneal upper-pole heminephrectomy for ectopic ureter is safe, effective and reproducible, being an excellent minimally invasive treatment option and offers the patient the typical postoperative benefits of laparoscopic surgery (Li *et al*., 2011; Wang *et al*., 2003). A vast majority of patients with residual ureteral stumps after heminephrectomy do not require stump resection at long-term follow-up (Caluwé *et al*., 2002). A double ureter has an incidence of 1 % in general population. Of particular note in this case is the presence of minimal complaints in this patient, with complaints of only flank pain, and burning micturition since one year, along with the fact that presentation at the age of 25 years. A double ureter is usually a presentation of the age group of infants, children and adolescents, and hence, the particular importance of this case. This case report thus aims to press upon the importance of keeping the diagnosis of a double ureter, leading to complications associated with it, in any suspected case of flank pain, presumed to be of renal origin.

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