

Case Report

Incidental Finding of Unilateral Complete Duplication of Ureter in a Patient with Large Ureteric Calculus Obstructing Both Left Limbs

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Abstract

Duplication of the ureter is a common congenital malformation, which unfortunately may be accompanied by unpleasant and challenging pathologies. Hereby, a rare case of a patient with obstructive urolithiasis secondary to undiagnosed complete ureteral duplication is presented. A single large calculus in the vesicoureteral junction was obstructing both duplicated ureters. The aim of this article was to discuss both the challenges arising from this clinical entity and the diagnostic approaches. In such complicated cases combined with suspected pyelonephritis or severe hydronephrosis, the option of an urgent lithotripsy should be considered. The obstructed orifices are often inflammatory, hindering their stenting. Asymptomatic and undiagnosed patients with completely duplicated ureters are prone to severe complications. Thus, early screening of these patients is an imperative need for the clinician.

Keywords

calculus, duplication, lithotripsy, ureter

INTRODUCTION

Duplication of the ureter is one of the most common congenital malformations of the urinary tract. A kidney with two pyelocaliceal systems is accompanied by either a single or two partially (or completely) separate ureters inserting into the bladder. [1] Nation et al. reported ureter duplications of any form in 0.9% of the general population in a series of autopsies. The prevalence of this condition is higher for female population and is often bilateral. [2] Its incidence rate ranges from 0.7% to 4% and presents on 0.3% of excretory urograms. [3,4]

In complete duplication, the ureter to the upper segment arises from a cephalad position on the mesonephric duct and consequently, it remains attached to the mesonephric duct longer. As a result, it migrates farther, ending median and inferior to the ureter which drains the lower segment. (Weigert-Meyer law) Thus, the upper segment ureter may become ectopic and obstructed, whereas the lower segment ureter may end laterally and have a short intravesical tunnel leading to vesicoureteral reflux.^[5]

Ureteral duplication is often asymptomatic but may be associated with recurrent urinary tract infections, urolithiasis, and congenital problems such as ectopic and obstructed ureter, often combined with a ureterocele and vesicoureteral reflux. Urolithiasis is precipitated by the relative stasis of urine in such patients but other factors unrelated to the duplication also cumulate to this pathology. [6]

Hereby, we present a rare case of a patient with obstructive ureteric urolithiasis secondary to undiagnosed



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complete ureteral duplication. The large calculus in the vesicoureteral junction was obstructing both duplicated ureters. The aim of this study was to discuss the challenges arising from this clinical entity and the possible diagnostic approaches for those patients.

CASE REPORT

A 44-year-old female patient presented to the Emergency Department with colicky left flank pain, fever, and dysuria. She complained about left flank pain for about a week. Physical examination was significant only for costovertebral angle (CVA) tenderness. Her temperature was 38.7°C and she was slightly tachycardic on her vitals (BP 142/83, HR 105, SpO₂ 97%, RR 20). Blood serum chemistries demonstrated leukocytosis (WBC 14,200), mildly deranged renal function tests, normal serum electrolytes and normal blood sugar level. Her urine tested positive for blood, leucocyte and nitrates indicating urinary tract infection (UTI) and a urine culture was requested. Furthermore, the patient was evaluated by sonography and moderate left hydronephrosis was reported. A plain film of the abdomen revealed a large calculus in the distal left ureter and the patient was admitted to our clinic with a diagnosis of complicated UTI.

While inpatient, a computed tomography (CT) urography was obtained and revealed a complete left duplication of the ureter along with moderate to severe hydronephrosis and the presence of a large calculus in the vesicoureteral junction measuring approximately 2.9×1.8 cm (Figs 1-3). The patient underwent a rigid cystoscopy, which revealed the presence of two separate left orifices and a large stone of the vesicoureteral junction causing obstruction in both orifices. A diffuse inflammation was present in the area of trigone. We managed to insert a safety wire in the upper left orifice up to the

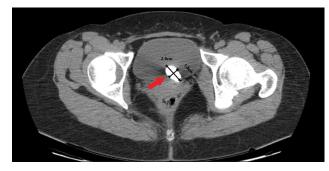


Figure 1. Axial plane of computed tomography (CT) urography depicting a large calculus in the vesicoureteral junction measuring 2.9×1.8 cm.

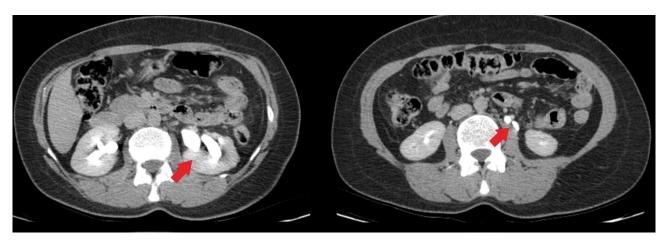


Figure 2. Excretory phase imaging of CT urography demonstrating the presence of complete ureter duplication. Two pyelocaliceal systems are accompanied by completely separate ureters.

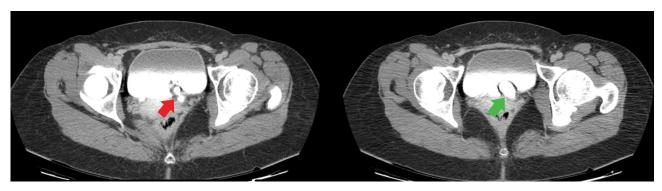


Figure 3. Excretory phase of computed tomography (CT) urography (axial plane), indicating obstruction of both duplicated left ureters by the presence of a large calculus in the vesicoureteral junction.

renal pelvis and a double J ureteral stent (4.8 Fr, 28 cm) was placed. During her treatment in our Clinic, parenteral antibiotics were administered along with adjunctive measures and her fever resolved. Both her renal function tests and her general condition improved, and she was discharged and rescheduled for ureteroscopy (URS).

Thus, the patient was re-admitted two weeks post discharge and after all necessary pre-operative examinations, she underwent a rigid laser lithotripsy under general anesthesia and antibiotic prophylaxis. The large calculus was visually pulverized using our clinic's holmium laser, while the remaining large fragments were extracted with the use of a basket. Finally, the lower left ureter was restented with a double-J stent. (4.8 Fr, 26 cm). Intraoperatively, C-arm radiographics confirmed the proper stenting of both duplicated left ureters (Fig. 4) and a plain KUB film revealed a stone free patient (Fig. 5). Postoperative period was uneventful, and the patient was discharged from the hospital on postoperative day 2, after removing the Foley catheter. Six weeks after surgery, patient underwent a KUB X-ray in order to be evaluated and to confirm that she was stone free and afterwards a cystoscopy was performed for the double-j removal. Both orifices of the duplicated ureters remained inflammatory (Fig. 6).

DISCUSSION

Although some patients with duplicated ureter (complete or incomplete) are asymptomatic, they usually present with persistent or recurrent UTIs. Moreover, an ectopic ureter of the upper pole secondarily to this congenital malformation is not uncommon. The presence of vesicoureteral reflux and ureterocele are reported as well and confirmed by voiding cystourethrogram. Kirby et al., also reported a patient with urinary retention secondary to ureteral calculus in a duplicated system.^[7]

Large ureteral calculi in a completely duplicated ureter are a challenging clinical entity for the clinician but not



Figure 5. Post endoscopic intervention plain X-ray of the kidney, ureter and bladder (KUB). The patient is stone-free.

new to the urologists. Thus, large calculi in the vesicoureteral junction can be obstructing both ureters in completely duplicated ureters. In 1959, Sidoti et al. was the first to report large ureteral calculi in a completely duplicated ureter.^[8] Unfortunately, the problem in the past was that the patient had to undergo a major operation due to lasers unavailability. We searched through the databases of PubMed,

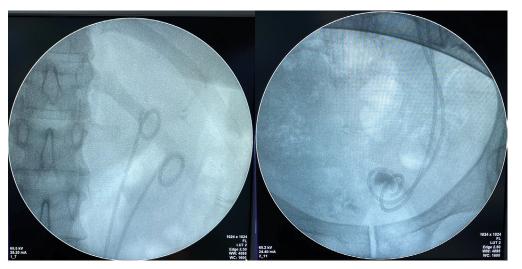
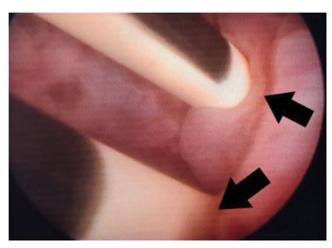


Figure 4. C-arm radiographics of the abdomen depicting the presence of Double-J stents in both limbs of the left duplicated ureter.



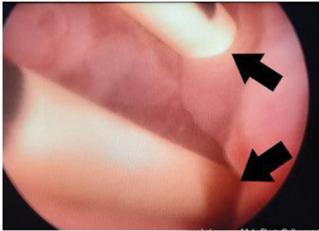


Figure 6. Follow-up post lithotripsy cystoscopy images showing the presence of two separate (stented) ureteric orifices.

Scopus, and Google Scholar and to the best of our knowledge this is the fourth case of patient with complete ure teral duplication concomitant with calculi obstruction of both limbs. $^{[6,9,10]}$

This occurrence may hinder the stenting of the obstructed orifices which are often inflammatory. In such complicated cases combined with suspected pyelonephritis or severe hydronephrosis, the option of an urgent lithotripsy should be considered. Nevertheless, severe sepsis might be an independent prognostic factor of action and therefore should be considered as well. In our case after discussion with the patient, we opted for URS laser lithotripsy as we considered this approach both less invasive and more effective for the patient. Depending on the location and the composition of the stone in obstructed duplicated ureters Bhatia et al., in 1993 highlighted the role of extracorporeal shockwave lithotripsy in duplex system lithiasis.^[11]

In recent years, prenatal ultrasonography has led to the early diagnosis in many asymptomatic neonates. Neglected cases like the one reported above, lead to complicated and often life-threatening pathologies. This highlights the value of prenatal ultrasonography and the need for its catholic application. Asymptomatic and undiagnosed patients with completely duplicated ureters are prone to severe complications like secondary obstructing ureteric or vesical lithiasis and complicated UTIs or chronic urothelial inflammation. Early diagnosis in these patients is an imperative need and the screening protocols should always be applied. Our case demonstrates that in patients with distal ureter lithiasis and concomitant completely duplicated ureters the stenting of either of the two ureters may be tricky and not always feasible. In such cases, alternative options should be considered.

Author contributions

I.T. conceptualized the report and wrote the paper. D.C. collected the data and images. K.G. and D.K. provided insightful comments and expert advice. K.S. helped to write the

manuscript and approved the final variant. All authors have read and approved the submitted version of the manuscript.

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Competing interests

The authors declare that no conflicts of interest exist.

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Consent

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

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Случайная находка одностороннего полного удвоения мочеточника у пациента с большим конкрементом мочеточника, вызывающим обструкцию обоих левых мочеточников

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Резюме

Удвоение мочеточника является частым врождённым пороком развития, который, к сожалению, может сопровождаться неприятными и сложными патологиями. Таким образом, представлен редкий случай пациента с обструктивным уролитиазом, вторичным по отношению к недиагностированному полному удвоению мочеточников. Один большой конкремент в пузырно-мочеточниковом соединении вызвал обструкцию обоих удвоенных мочеточников. Цель этой статьи состояла в том, чтобы обсудить как проблемы, связанные с этой клинической ситуацией, так и диагностические подходы. В таких сложных случаях, сочетающихся с подозрением на пиелонефрит или выраженный гидронефроз, следует рассмотреть вариант экстренной литотрипсии. Закупоренные отверстия часто воспалены, что затрудняет их стентирование. Бессимптомные и недиагностированные пациенты с полностью удвоенными мочеточниками склонны к тяжелым осложнениям. Таким образом, ранний скрининг таких пациентов является настоятельной необходимостью для клинициста.

Ключевые слова

конкремент, дупликация, литотрипсия, мочеточник