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CASE REPORT

Avoidable Pitfalls: Another Encounter of Bilateral Ureter Complete Duplication with Impacted Calculus At Al Habib Hospital/Dubai

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Abstract

The ureter is commonly the subject of congenital anatomical variations involving either complete or incomplete duplication. Complete ureter duplication may not produce symptoms and therefore does not become apparent until later in life. This condition is most encountered incidentally during the extraction of impacted ureteric stones. This type of anomaly is more common in females. However, this condition is not the extremely rare incident as previously thought. Herein, we present our second case of bilateral complete ureter duplication. This aberration was encountered during the extraction of an impacted calculus which was found close to the vesico-ureteric junction of the medial limb of the right complete duplicated ureter. We also present a review of the related English literature. It is essential to emphasize the importance of our basic knowledge relating to most possible renal and ureteric surgical aberrations. A particular consideration prior to any procedure is the meticulous analysis of related radiological images. This clinical practice is a key priority if we are to avoid unpleasant surgical pitfalls. The in-depth diagnosis of an aberration basically remains a radiological entity. This case study provides learning points to complement our daily practice in the management of impacted ureteric calculi.

Keywords Complete ureter duplication · Impacted calculus · Ureteroscopy · Missing of the stone

Abbreviations

ACR	American College of Radiology
AUA	American Urology Association
CT	Scan non-enhanced computerized tomography scan
EAU	European Association of Urology
ESWL	Extracorporeal shock wave lithotripsy
HB	Hemoglobin
KUB	Kidney-ureter-bladder X-ray
RBCs	Red blood cells
URS	Ureteroscopy
US	Ultrasound scan
UTI	Urinary tract infection
VUJ	Vesico-ureteric junction
VUR	Vesico-ureteric reflux
WBCs	White blood cells

1 Introduction

A duplicated ureter occurs in approximately in 0.7% of the population and is more common in females than males, with a ratio of 2:1. The highest prevalence has been reported in White females [1–3]. The ureteral bud branches from the caudal portion of the Wolffian duct during the fourth and fifth week of gestation. Ureteral anomalies usually result from alterations in the bud number, position or even the time of bud development. Genetic mutation and environmental factors are both known to contribute to the formation of these anomalies. Ureter duplication is inherited by an autosomal dominant gene with variable penetration. This duplication may be incomplete, often referred to as a bifid ureter, or complete, in which each limb opens separately to the bladder [1, 4]. In addition to a bifid ureter, this condition may include a trifid or multifid renal pelvis [1]. In cases of complete duplication, the upper renal moiety drains to the first limb of the duplicated ureter, which undergoes ectopic insertion in the bladder, usually medial and inferior to the second limb, which drains the lower pole moiety. This anatomical configuration is commonly known as Weigert–Meyer law, as depicted in Fig. 1 [1]. A violation of Weigert–Meyer law is a rare incident and has been reported

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Fig. 1 The 3D-VRT volume rendering technique and bilateral complete ureter duplication

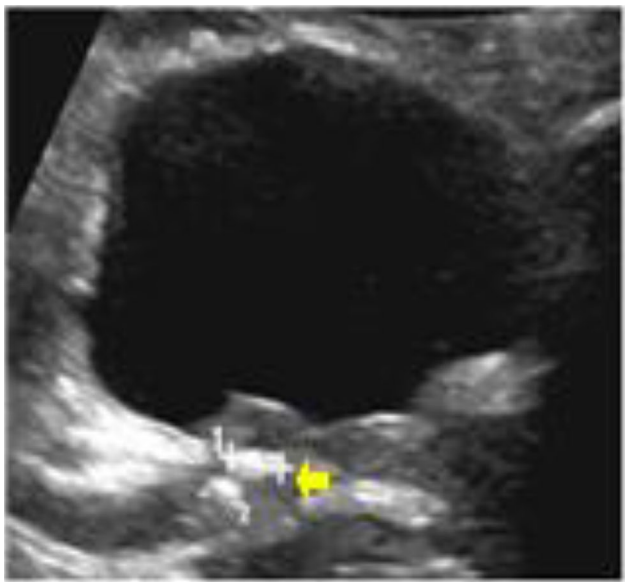

Fig. 2 Real-time ultrasound scan showing a 9-mm stone at the right vesico-ureteric junction

in cases of uncrossed complete ureteral duplication with a dysplastic lower moiety [5]. However, most duplicate aberrations are detected incidentally during pelvic surgery or in research studies during the routine dissection of a cadaver [3, 6]. Furthermore, a few cases have been reported possessing calculi in a single limb of the duplicated ureter [7–9]. In addition, some cases have been reported with an impacted stone in both limbs of one side of the duplicate [10, 11]. Occasional cases have been reported involving the detection of calculi in all limbs of the duplicate [12].

Herein, we present our second case of bilateral complete ureter duplication with an impacted calculus; this anomaly was found incidentally in the medial limb of the right duplicate (Fig. 2). In addition, we present some representative radiological and endoscopic images.

2 Case Presentation

A 46-year-old female was admitted through the emergency room with a complaint of persistent pain in the right lower abdomen and right flank. Previous childbirth deliveries were all normal. She did not have any related significant history and the abdomen was unremarkable. Her blood pressure was 157/72 mmHg. Laboratory results revealed hemoglobin 14.3 g/dl, serum creatinine 47 mg/dl, calcium 2.32 mg/dl and uric acid 5 mg/dl. The urinary white blood cell (WBC) counts were 2–4/hpf (high power field) and red blood cell (RBC) counts were 5–8/hpf; cultures did not result in the growth of any microorganisms. Real time ultrasound scanning revealed a 9-mm calculus at the right vesico-ureteric junction (VUJ) with no ureteric jet, as shown in Fig. 2.

Simultaneous unenhanced computed tomography of the kidneys, ureters and bladder (CT KUB) scanning revealed a 6.5-mm stone that was entrapped within the right VUJ; this was associated with mild right proximal fullness (Fig. 3). In addition, we encountered three tiny non-obstructive stones (2–3 mm in size) in the right superior renal pole (Fig. 4). Multiple tiny gall stones were also noted (Fig. 5). Symptomatic conservative treatment was attempted overnight. The patient was unable to expel the stones spontaneously and the pain remained intolerable. Eventually, right ureteroscopy (URS) was considered. Surprisingly, cystoscopy revealed two ureteric orifices on the right side of the bladder and another two on the left side (Figs. 6, 7). The right medial ureteric orifice of the duplicate was easily negotiated, and its caliber was able to accommodate a rigid 9.5F ureteroscope (Fig. 8). An impacted stone was encountered close to the VUJ of the medial limb of the right duplicate (Fig. 9). Surgery was completed in 1 h and involved laser disintegration of the stone and its tiny particles; these were removed in a piecemeal manner (Figs. 9, 10). Ultimately, double coil stents (24/6) were inserted into the duplicate, which were subsequently removed after 2 weeks (Fig. 11). Chemical analysis of the stone revealed calcium oxalate (75%), calcium phosphate (5%), struvite (10–20%), uric acid (5–10%) and cystine (1%) (Fig. 12).

3 Discussion

Duplicated ureters are usually found during childhood due to presentation with urinary tract infection (UTI) or incontinence. This condition may be associated with ureterocele or

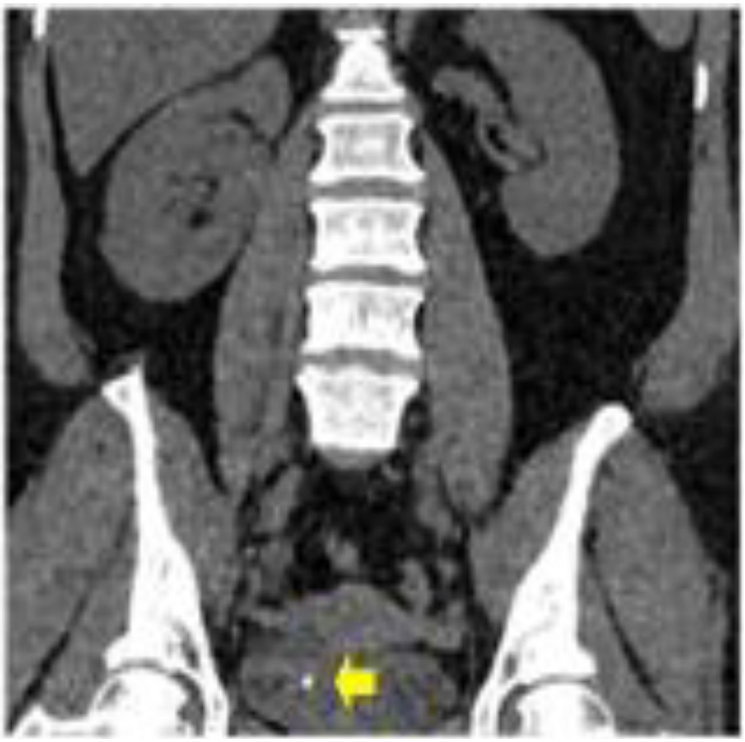

Fig. 3 Coronal CT multiplanar reformation (MPR) reconstruction. Non-enhanced CT abdomen and pelvis coronal section. The right vesico-ureteric junction featured a 6.5-mm stone

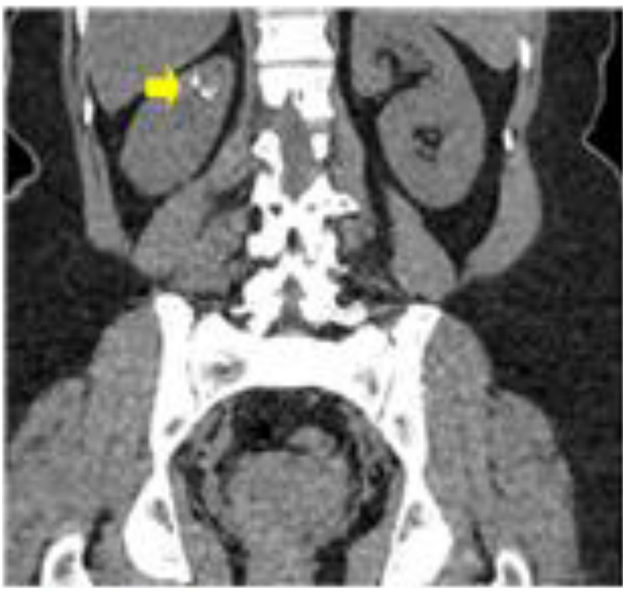

Fig. 4 Right superior pole intrarenal non-obstructive tiny stones (2–3 mm in size)

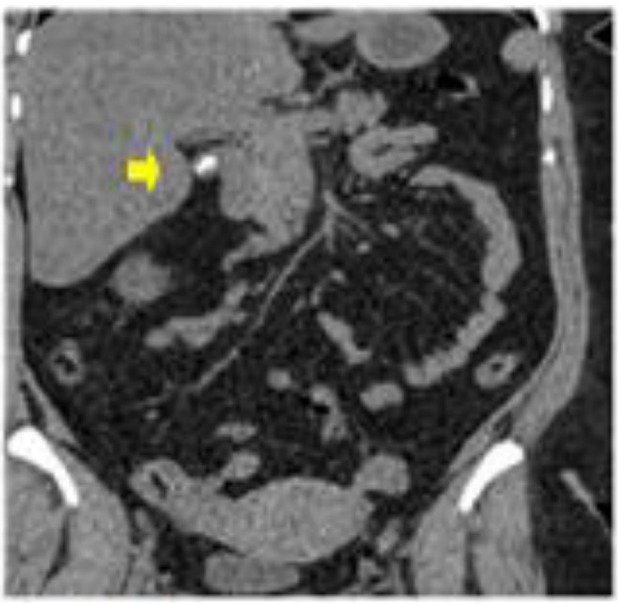

Fig. 5 Coronal CT multiplanar reformation (MPR) showing a stone in the gall bladder

vesicoureteric reflux (VUR). Occasionally, one ureter ends normally in the bladder while the other has an ectopic ending distal to the bladder neck either in the urethra, vulval vestibule or the vagina [13–15].

In adult patients, this may be associated with incontinence, recurrent urinary tract infection and/or stone formation [16–18]. Commonly, the initial diagnosis of an obstructed calculus is elicited in the emergency room by ultrasound scanning of the urinary system or non-enhanced CT scanning, or both. However, this anomaly is mostly encountered by radiologists. In adult patients with

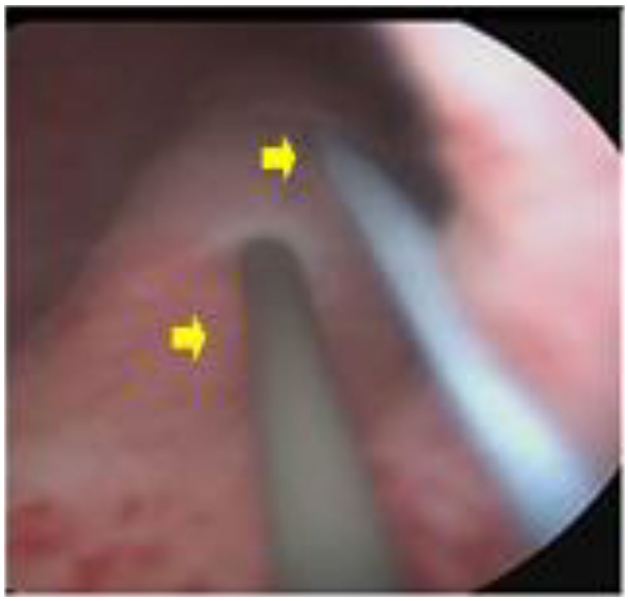

Fig. 6 Cystoscopy view of two separated left ureteric orifices, medial inferior and upper lateral

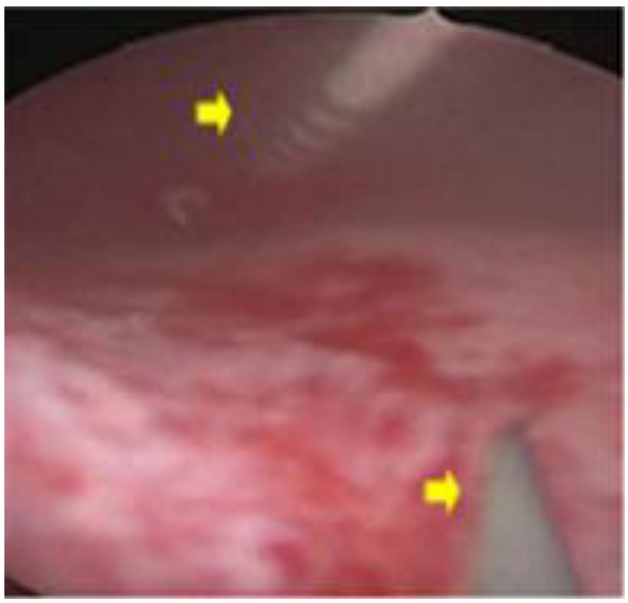

Fig. 7 Cystoscopy view of two separated right ureteric orifices, medial inferior and upper lateral

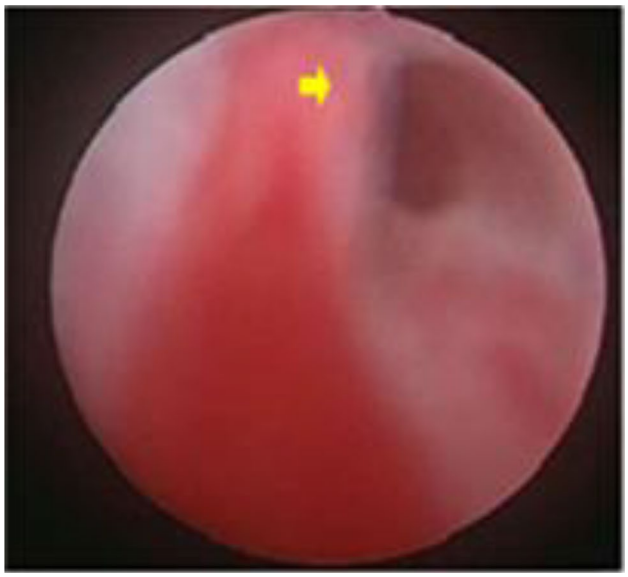

Fig. 8 Cystoscopy view of the right medial and inferior lower ureteric orifices with an adequate caliber

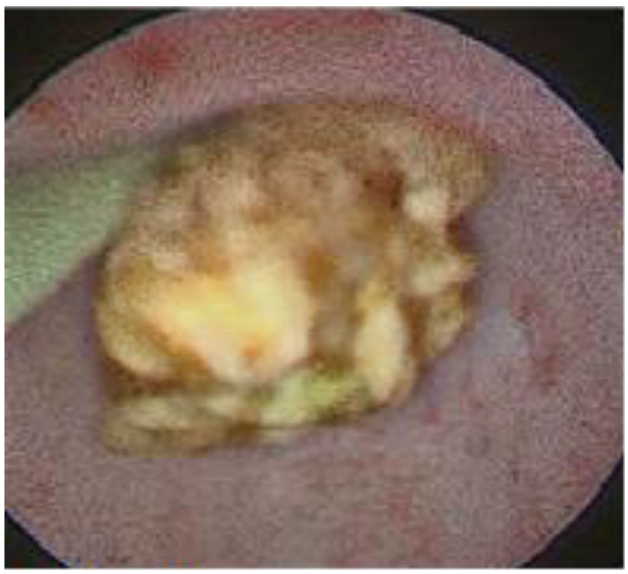

Fig. 9 Impacted stone in the medial limb of the complete right duplicate

an impacted stone, the first line of investigation may differ from one guideline provider to another.

From one perspective, both the American Urology Association (AUA), and the American College of Radiology (ACR) consider non-enhanced CT scans of the abdomen and pelvis as the first line of investigation for all adult patients presenting with symptoms suggestive of obstructing stones. This method has the highest sensitivity (94–97%) and the highest specificity (96–100%). In addition, Hounsfield units can be measured, particularly if extracorporeal shock wave lithotripsy (ESWL) is considered [9, 15]. However, physicians need to consider the associated hazards of radiation and the high costs involved [19, 20].

From another perspective, the European Association of Urology (EAU) consider ultrasound scanning as the first line of investigation. This method has 40% sensitivity and

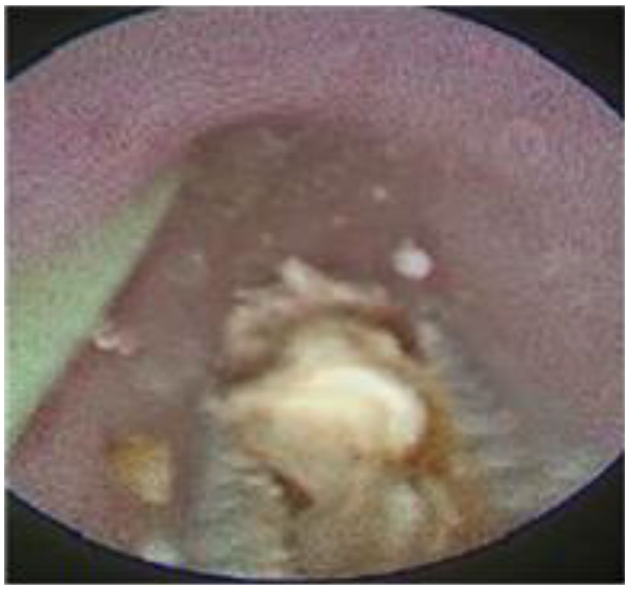

Fig. 10 Disintegrated stone captured by forceps

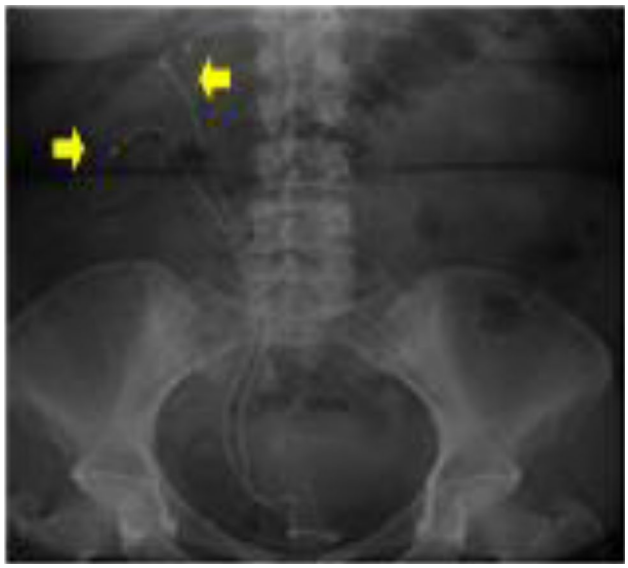

Fig. 11 A double coil stent in position in the right duplicate

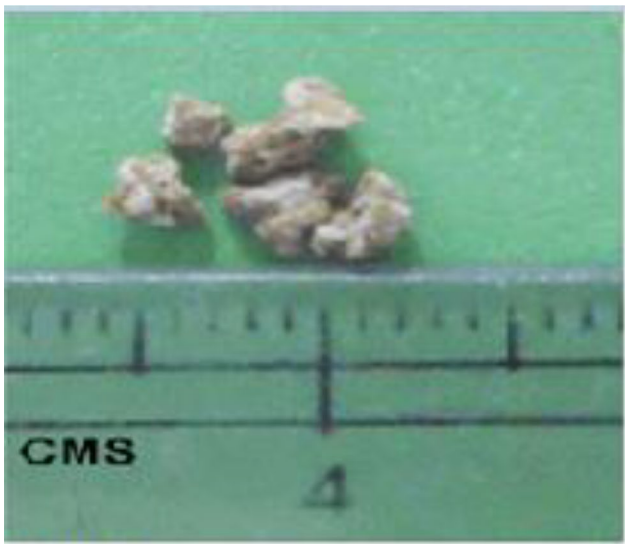

Fig. 12 An extracted stone

84% specificity. A combined study of ultrasound scanning and KUB radiography revealed a wide range of sensitivity (58–100%) and specificity (37–100%) [19, 21]. In pediatric patients and pregnant women, the first line of investigation recommended by all the main guideline providers (the AUA, ACR and EAU) remains the same. All these providers consider ultrasound scanning as the first line of investigation [19–21].

Notably, stone measurement is expected to differ slightly depending on the tool deployed for diagnosis. Currently, ultrasound scanning is less sensitive and specific than CT scanning with regards to the detection and sizing of stones. The sizes of small stones will inevitably be overestimated by ultrasound scanning. Nevertheless, in a previous randomized controlled trial, both ultrasound scanning and non-enhanced CT scanning were found to exhibit equivalent diagnostic accuracies when deployed in the emergency department [19, 22]. However no definitive study has been conducted yet to compare these two modalities for imaging stones in obese patients.

We searched the existing literature for articles describing impacted calculi in a single limb or in both limbs of complete ureteric duplication; some of this literature reported that some of the treated patients subsequently required a definitive surgical intervention [10–12, 17]. This may prolong hospital stay and increase the patient co-morbidity that is usually associated with a substantial clinical and financial burden.

Herein, we present our second identical case of complete ureter duplication with an impacted stone in the medial limb of the right duplicate; this comes shortly after the detection of our first case. The calculus was encountered by both ultrasound scanning and non-enhanced CT scanning, but with expected differences in the measurements. Ultrasound scanning measured the stone at 9 mm while CT scanning measured the stone at 6.5 mm. The duplications were not visualized initially but became evident during cystoscopy, at which point the bilateral duplicate was clearly visible (Figs. 6, 7, 8, 9, 10).

Retrospectively, all images were analyzed meticulously; the anatomical aberrations that were initially missed were

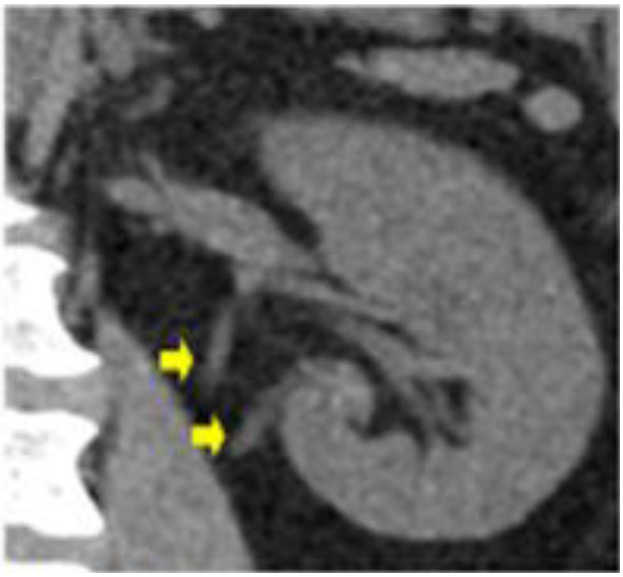

Fig. 13 Unenhanced coronal CT multiplanar reformation (MPR) showing the left kidney with two separated ureters emerging from the upper and lower moiety

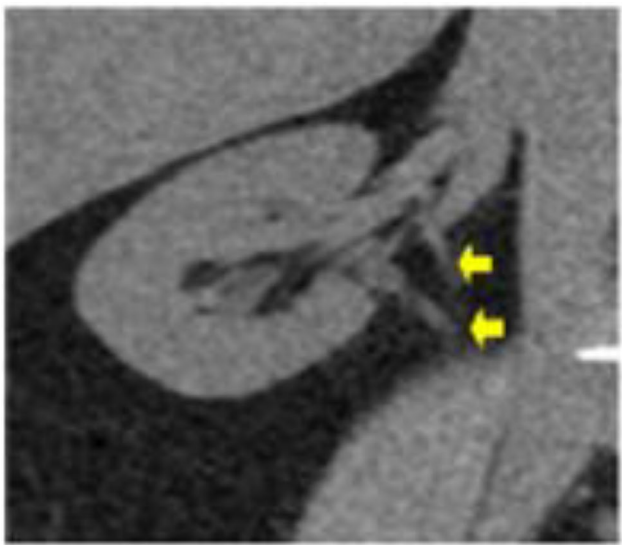

Fig. 14 Coronal CT multiplanar reformation (MPR) showing the right kidney with two separated ureters emerging from the upper and lower moiety

clearly recognized; hence the patient was diagnosed with bilateral complete duplication (Figs. 1, 13, 14).

4 Conclusion

Complete ureter duplication is not the extremely rare incident as was initially considered. Occasionally, a single limb, or more than one limb, of the duplicate is conflicted with an impacted calculus. Occasionally, the diagnosis of such anomalies can be delayed or even missed. As the best standard-of-care, and to avoid clinical pitfalls, it is necessary to diagnose aberrations early, although this remains a radiological challenge.

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Availability of Data and Material The related data and material have been retained and are available from the author.

Declarations

Conflict of interest None of the authors have any conflicts of interest to disclose.

Ethics approval and consent to participate The patient and her family agreed to participate and were aware of our intention to publish.

Consent to publication Written and informed consent was acquired from the patient and her husband.

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