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Bilateral bifid ureter: Case report and clinical discussion

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Abstract
The ureters are conduit for urine as they leave the kidney and enter the urinary bladder. Megaureter, ureterocele, and H shaped ureter are diseases that involve this particular structure. Furthermore, these pathologies are often associated with other conditions, such as horse shoe kidney and ectopic kidney. Bifid ureter is one of the malformations of the kidney collecting system - the minor and major calyces. It can be complete, when the division is total and the supernumerary ureter joins the bladder through another orifice, or incomplete, where the ureters merge together. The incidence of this condition is 0.8% in the overall population, although it is more common in women. It is also more commonly found as a unilateral condition. We report a case of bilateral bifid ureter on a male cadaver, discussing the many forms of variation and their clinical significance.

Keywords: bifid ureter, duplex collecting system, anatomy, embryology, clinical significance

1. Introduction

Essentially, the ureters are tubes that transport urine from the kidneys to the urinary bladder, they begin at a region of the kidney named as renal pelvis, a structured formed by two or three major calyces. The ureter usually has 28 to 34 cm, although the right is 1 cm shorter than the left ureter[1]. Its abdominal part is retroperitoneal, and it is situated medially to the psoas major muscle, more inferiorly, it crosses the common iliac vessels or the origin of the external iliac vessels in order to reach the pelvic region[1].

In the pelvis, the ureters are located in the lateral wall of this area, below the peritoneum, then, it is situated anteriorly to the interior iliac artery, medially to the obturator nerve, the inferior vesicle artery and middle rectal artery. After reaching the inferior border of the greater sciatic notch, the ureter turns to the medial side to enter the lateral angle of the urinary bladder[1].

The right and left ureters have different relations during their trajectory. The right ureter is related to the inferior vena cava, the descending portion of the duodenum, and it is crossed by the right colic and ileocolic vessels, while the left ureter is crossed by the left colic vessels, and passes posteriorly to the sigmoid colon and its mesocolon [1].

Bifid ureter (BU) is a condition in which the ureter is duplicated, and it can be classified into complete (when they join separately at the urinary bladder) or incomplete (when they fuse at some point in their trajectory) [2, 3].

In the literature, the prevalence of duplicated ureter ranges from 0.6 to 0.8%, moreover, it is more common in women [2, 4, 5]. Variations of the ureter are not uncommon, although, according to some authors, they are usually unilateral[1,2,6]. Furthermore, it can be associated with other anomalies, such as horseshoe kidney [7], Goltz's syndrome [6], renal ectopy [8] H shaped ureter [3], ureterocele [9], megaureter [10, 11] and ureteropelvic junction obstruction and dilatation [3, 10]. Vesicoureteral reflux is the most common form BU manifestation [9].

We report a case of bilateral incomplete bifid ureter in a male cadaver from Brazil.

1.1 Case Report

A male cadaver fixed with a 10% formalin solution was dissected for teaching purposes when was accidentally found the presence of a duplex kidney collecting system - a bifid ureter on both sides. Figure 1 shows the right side, while Figure 2 displays the left side.

Both ureters had regular trajectories after the fusion, entering the urinary bladder through one left vesicle orifice and one right vesicle orifice.

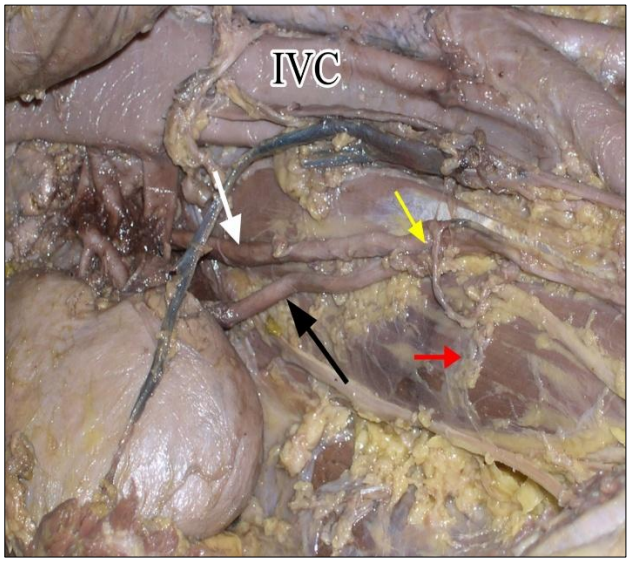

Fig 1: View of the right ureters. The right upper ureter can be seen (white arrow), and the black arrow shows the right lower ureter. Their anastomosis is depicted on the yellow arrow. Psoas major muscle (red arrow) and IVC is the Inferior Vena Cava.

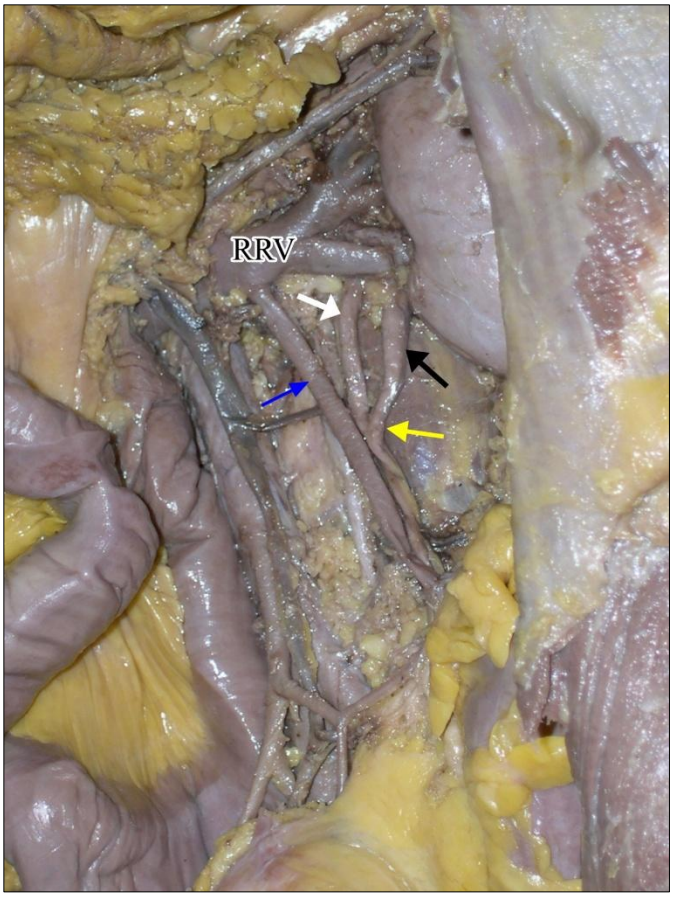

Fig 2: View of the left ureters. The left upper ureter can be seen (white arrow), and the black arrow shows the left lower ureter. Their anastomosis is depicted on the yellow arrow. Psoas major muscle (red arrow) and RRV is the right renal vein. The blue arrow shows the left testicular vein.

2. Discussion

The glans and urogenital ducts develop together as an agglomerate of cells, and most of these structures (pronephros, mesonephros, mesonephric or Wolffian ducts, paramesonephric or Muller ducts, and the metanephros), disappear almost completely before fetal life [1, 5]. The mesonephric duct forms a "diverticulum" in its caudal extremity, which will form the renal calyces and the ureter. Furthermore, the cranial portion of the mesonephric duct - together with the metanephros - will form the kidney, thus, explaining the associated abnormalities that usually accompany the congenital defects of the ureter [1, 5, 6, 12].

Hence, duplication of the ureteric bud from the Wolffian duct will give rise to duplicated ureters. Although, in order to form a complete duplication, it is necessary that the lower end of the ureteral bud to split, and to form an incomplete duplication, this cleavage must occur near the base of the kidney[5].

According to Bergman *et al.* (1988) [13], the most common variation of the ureter is the division of this structure into two, although the ureter can tripled, quadrupled, quintupled or even sextupled, and when this branching happens, they can also be complete (a whole new ureter which joins the bladder at a different side) or incomplete (when the supernumerary ureters fuse at some point of their trajectory).

Another form of ureter variation is the retrocaval position, associated with a malformation of the inferior vena cava[13]. Dinanath *et al.* (2011) described a rare variation of a bilateral complete division of the ureter and the renal pelvis[14]. Chawla *et al.* (2014) reported a variation in which there were incomplete bifid ureters (as they joined before entering the bladder) associated with vascular variations of the renal artery, although the latter was unilateral[15]. Furthermore, they can enter other sites (ectopic ureter), instead of the urinary bladder, like the vagina, seminal vesicle, urethra, prostate, epididymis, and vas deferens [12, 13]. Salakos *et al.* (2009) described a case of double-blind ureteral duplication, a rare condition in which the ureter has no connection to the kidney whatsoever [16].

Béltran Armada *et al.* (2004) described a case of an ectopic kidney (intrathoracic kidney, with abnormal vasculature) associated with duplex collecting system [8].

Anatomy textbooks emphasizes the 3 different areas in which the ureters suffer a constriction: (1) as it exits the renal pelvis (ureteropelvic junction), (2) when it crosses the iliac vessels, (3) when it enters the urinary bladder[1]. We believe that the incomplete BU presented here had a higher probability of constriction and subsequent calculi formation at the site of junction between both ureters, as it had an acute angle. Furthermore, it has been stated that once the ureters merge at different level and join the urinary bladder through one orifice (such as our case presented), an stenosis on the pyeloureteral transition or retrograde peristalsis could cause the YOYO-phenomenon, where urine goes from one ureter to another, unable to move forward to the bladder, although this phenomenon is more common on blind end ureters [5, 17, 18].

Incomplete BU may remain asymptomatic, although it can produce symptoms of uretero-ureteric reflux, frequent urinary tract infections, urinary lithiasis, ureteric stenosis [6], and renal colic [2]. According to García Rodríguez *et al.* (2008) and Lee *et al.* (2015), BU is more seen in patients with frequent urinary infections than in the normal population [2, 19]. Knowledge regarding the presence of such anomaly is important due to the fact that the division can be complete, and we believe that increases the chances of an ectopic ureter, as Mascatello *et al.* (1977) stated that they are commonly associated [20]. Dorko *et al.* (2015) classifies the BU as proximal (ureter fissus proximalis) or distal (ureter fissus distalis), our variation is of proximal nature [5].

Women are more susceptible to this condition and associated pathologies, and urinary incontinence is usually one of the main symptoms [20]. Furthermore, the BU may also be a surprise during surgery, as it has been shown in female patients with vesicovaginal fistula that underwent through surgical repair, and should be considered as a danger during the procedure and should be included in differential diagnosis of vesicovaginal fistula [21].

Cystoscopy, ureteroscopy, computed tomography, magnetic resonance imaging, ultrasonography, and renal scintigraphy are useful to determine the presence of these anomalous ureters [16, 18]. Symptomatic patients should be treated accordingly with the intensity of the symptoms, and no intervention should be performed in asymptomatic patients [18].

3. Conclusions

Bifid ureters are a common condition, although it usually manifests on one side, and with a higher frequency in women. We believe that the YOYO-phenomenon may be present in both incomplete bifid ureter and blind end supernumerary ureter, due to an obstruction of the main ureter. It is vital to pay

attention to the symptoms (although it can be asymptomatic) and to diagnose other associated malformations. The surgeon should be aware of these variations in order to reduce possible iatrogenic injuries and to prevent misdiagnosis of vesicovaginal fistula.

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