Unusual Pattern of Violation of Weigert Meyer Rule with Ureterocele in an Adult: A Case Report and Literature Review with Review of Associated Embryology

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Abstract

Background: *Duplication of renal system is one of the most common congenital anomalies of the urinary tract. The relationship between the drainage of the two moieties into the urinary bladder is described by the Weigert-Meyer rule which states that the moiety opening into the bladder medially and caudally drains the upper pole while the moiety opening laterally and cranially drains the lower pole of the kidney.*

Case presentation: *We present a case of violation of the Weigert Meyer rule in a 48 year old male who presented primarily in adulthood. On imaging studies, the patient was discovered to have a duplex left renal collecting system with the both moieties draining ectopically into the the bladder and with the lower pole moiety draining caudally and anteriory to the upper pole moiety. We present this case as it describes a rarer variant of a common anomaly as well as for its importance to radiologists and surgeons.*

Conclusions: *Radiologists and urologists should bear in mind the possibility of these violations when dealing with patients with a duplex system. Furthermore, patients with congenital urinary tract anomalies may present primarily in adulthood and have an uneventful childhood.*

Keywords: *CAKUT, Ureterocele, Weigert-Meyer rule, Stephens ectopic pathway*

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Background

Congenital abnormalities of the kidney and urinary tract (CAKUT) are the most common anomalies detected in neonates, with an incidence of nearly 3 to 6 per 1000 live births (1). Some of the common anomalies include horseshoe kidneys, duplicated systems, ureteropelvic junction obstruction, etc. The genetic basis for most of these anomalies involves the same molecular pathway which plays a key role in the development of the renal system (2). Duplication of the renal system, albeit a relatively rare occurrence, is one of the most common renal anomalies, seen in about 0.2-2% of the population. It is more common in females, and unilateral involvement is more commonly seen than bilateral involvement (1, 3). Duplication may be complete or incomplete. In an incomplete duplex system, the draining systems of both the renal poles drain into the bladder through the same ureteral opening while in complete duplex systems the drainage is via separate ureteral openings (1). In a complete duplex system, the relationship between the drainage of the two moieties into the bladder is described by the Weigert-Meyer rule (4, 5). Urinary tract anomalies are commonly diagnosed in children who usually present with obstructive symptoms, reflux, or urinary tract infections associated with complications of these anomalies (4 - 7). In this article we describe the case of an adult patient with an uneventful childhood who presented with an unusual pattern of deviation from the Weigert Meyer rule.

Case presentation

A 48 year old male presented in 2023 with episodic lower abdominal and pelvic pain with burning micturition for 1 year. Physical examination was unremarkable. The patient had no significant prior clinical history or family history. Urine microscopy showed microscopic hematuria. Routine investigations including renal function tests were within the normal limit.

The patient underwent ultrasonography (Figure 1A-C and Figure 2A-C) which revealed focal ecta-

sia of the calyces of the upper pole of left kidney. The right kidney was normal in morphology. Two echogenic structures showing posterior acoustic shadowing were visualised within the lumen of the urinary bladder on either side, suggestive of calculi. The calculus visualized on the left side was surrounded by a thin rim of echogenic soft tissue and showed no displacement on changing the position of the patient. The calculus on the right side showed displacement to the dependent position on change in patient decubitus.

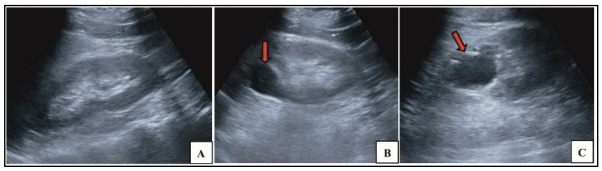

Figure 1(A-C): Ultrasonographic images of bilateral kidneys. Longitudinal ultrasonographic images of the right (A) and left (B and C) kidneys reveal normal appearing right kidney (A) and focal ectasis of the upper polar calyces (red arrows) of the left kidney (B and C)

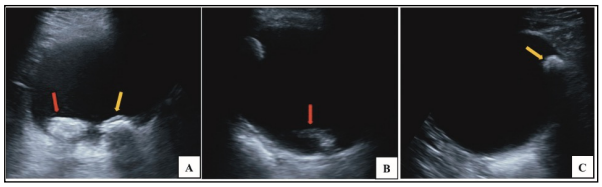

Figure 2(A-C): Ultrasonographic images of the urinary bladder. Transverse ultrasonographic images of the urinary bladder in supine position (A), and sagittal images on left (B) and right (B) lateral decubitus positions show two echogenic structures, one each on the right (red arrows) and left (yellow arrows) sides, within the lumen of urinary bladder showing posterior acoustic shadowing, representative of calculi. A thin echogenic rim of soft tissue is seen around the calculus on the left (yellow arrows). The calculus on the left side shows no change in position with the change in patient decubitus, while the calculus on the right (red arrows) shows displacement to the dependent position.

For better assessment and to see the excretory function of the kidneys, further investigation with a CT urography (Figure 3A-F) was performed. The scan and the volume rendered images (Figure 4A-C) derived from it revealed complete duplication of the collecting system of the left kidney with the upper and lower pole moieties showing normal contrast uptake and excretion. The upper pole moiety showed a focal dilation of the intravesical portion, suggestive of ureterocele, with a calculus noted within the same. Few other smaller calculi were also seen in the distal aspect of left upper pole ureter just proximal to the vesicoureteric junction, with resultant upstream hydro-uretero-nephrosis of the entire left upper pole collecting system. The lower pole moiety, which was expected to be orthotopic, drained slightly inferior and anterior to the upper pole moiety, thus deviating from the Weigert-Meyer rule. An intravesical calculus was also visualised. Right kidney was normal in morphology, contrast enhancement and excretion.

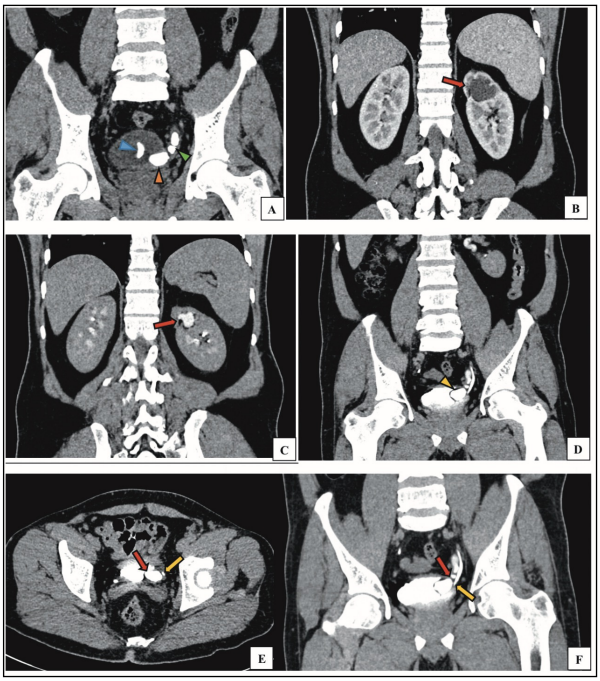

Figure 3(A-F): CT urography images. Coronal reformatted non contrast (A), corticomedullary phase (B), and excretory phase images (C, D and F), and axial excretory phase (E) CT urography images show normal contrast uptake (B) and excretion (C) in bilateral kidney. The left kidney shows a complete duplex system with hydronephrosis of left upper pole moiety (red arrows). The left upper pole moiety shows a ureterocele with a calculus noted within it. Few smaller calculi are also seen in the distal aspect of left upper pole ureter (green arrow head in A). Delayed (E and F) images show the anamolous relation of the upper (red arrows) and lower (yellow arrows) pole moieties with the lower pole moiety seen opening anterior (E) and inferior (F) to the upper pole moiety.

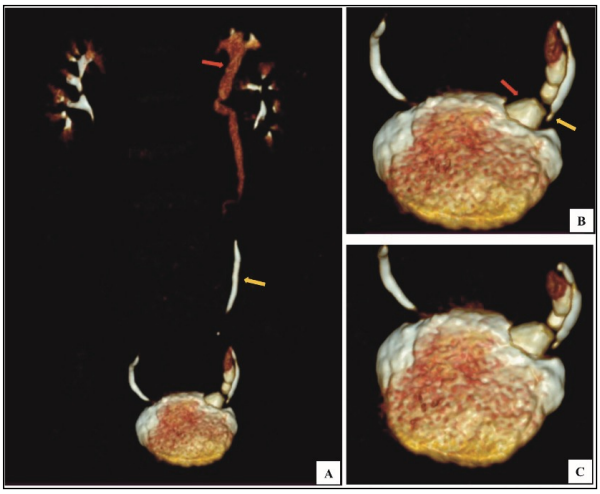

Figure 4(A-C): Post processed 3D volume rendered CT images. 3D VRT images derived from CT urography shows complete duplication of the collecting system on the left. The upper pole moiety (red arrows) shows calculi in the distal ureter and vesicoureteric junction with upstream hydroureteronephrosis. Inferior moiety (yellow arrows) appears unremarkable. The orthotopic inferior moiety (yellow arrow) is seen draining inferior and anterior to opening of the ectopic moiety.

Limited sections of T2 weighted coronal MRI (Fig. 5A-C) was performed for better anatomical delineation, which revealed a duplicated collecting system on the left with a ureterocele of the left upper pole moiety. Few T2W hypointense filling defects were seen within the ureterocele and the

distal aspect of left upper pole ureter with resultant upstream hydro-uretero-nephrosis of the upper pole moiety. However, the lower moiety appeared unremarkable. An unusual finding that was observed was the drainage pattern of upper and lower moieties; the ureters draining both the upper pole moiety and the lower pole moiety had an ectopic drainage into the bladder at a location more inferior to the normally expected vesico-ureteric junction. Furthermore, the lower pole moiety drained into the bladder slightly caudal and anterior to the upper pole moiety, thereby violating the Weigert Meyer rule.

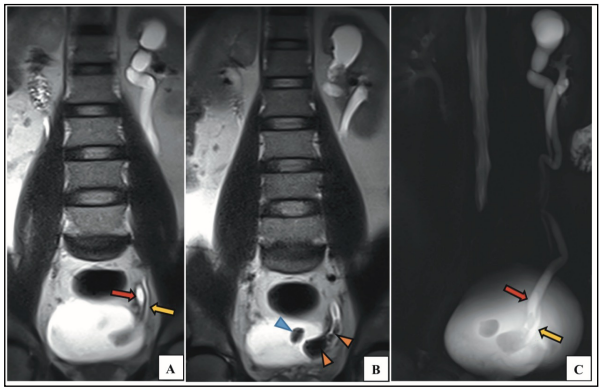

Figure 5(A-C): MR urography images. Coronal T2W (A and B) and oblique thick slab T2W (C) MR images show complete duplication of the left collecting system with a calculi seen as T2W filling defects (orange arrow heads) within the ureterocele and in the distal ureter of the upper pole moiety causing hydroureteronephrosis. An intravesical calculus (blue arrow head) is also seen. The lower pole moiety (yellow arrows) is seen to drain into the bladder inferior and anterior to the upper pole moiety (red arrows) clearly represented in the thick slab images.

The patient was not in favour of undergoing invasive management and was managed conservatively for pain and infection. Unfortunately the patient was lost to follow up following resolution of the infection.

The embryonic development of the urinary tract begins when the nephric duct induces changes in the adjacent mesoderm to sequentially form the pronephros, mesonephros, and ultimately the metanephros. The pronephros is a rudimentary structure and regresses soon after its development. The mesonephros gives rise to the mesonephric duct (Wolffian duct), which forms the bladder and parts of the genital system in males. The metanephros, formed around 5-6 weeks of gestation, is the precursor of renal development. The ureteric bud arises from the mesonephric duct and grows to join with the metanephric mesenchyme. The former gives rise to the collecting system of the kidneys, while the latter forms the renal parenchyma (6).

In an incomplete duplex system, the draining systems of both, the upper and lower renal poles drain into the bladder through the same ureteral opening. It occurs when the ureteric bud arising from the mesonephric duct branches prematurely before reaching the metanephric mesenchyme. The point of branching gives rise to a variety of phenotypic appearances of an incompletely duplicated system, which may range from a bifid pelvis to a bifid ureter. In a complete duplication, the two ureteric moieties course separately and drain into the bladder through separate orifices (6). This occurs when two separate ureteric buds arise from the mesonephric duct and join with the metanephros (1, 4, 7).

In a complete duplex system, the relationship between the drainage of the two moieties into the bladder is described by the Weigert-Meyer rule, which states, ‘In a complete ureteral duplication, the ureter whose orifice is more medial and caudal reaches the upper moiety and the other ureter whose orifice is more lateral and cephalad reaches the lower renal moiety’. The upper pole moiety which drains into the bladder caudally is said to be ectopic, while the cephalad draining lower pole moiety is considered to be orthotopic (4, 5, 8).

The basis for this lies in the embryologic development of a duplex system. Embryologically, the lower pole moiety is considered to be akin to the normal ureter of a normal single system kidney, and therefore it drains in its usual orthotopic location. However, the upper pole moiety is said to be similar to an accessory ureter, and follows the pathway of the Wolffian duct to drain into any other mesonephric duct derivative, such as the trigone of the bladder, urethra, genital ducts in males and vagina in females (4).

The complications associated with a duplicated system can be separately categorised into those involving the upper pole moiety (ectopic drainage) and those involving the lower pole moiety (orthotopic drainage) (4, 9). The complications associated with the upper pole moiety are more common, and are majorly due to its ectopic insertion. The common upper pole moiety complications include ureterocele, obstruction, and incontinence, while the lower pole is usually afflicted by reflux and its sequelae (4 - 7).

Duplicated systems, in most cases, show an obstructed upper pole system and a refluxing lower pole collecting system. Furthermore, it presents during the childhood as recurrent urinary tract infections and other related complications while initial presentation in adults is rare (8, 11).

However, in our case the primary detection of the anomaly occurred in the middle age of the patient. Moreover, although the upper pole moiety did show a ureterocele and an obstructed system, the lower pole moiety did not show any signs of VUR or its sequelae. The patient also did not have any history of recurrent urinary tract infections during childhood.

Majority of the duplicated systems obey the above described Weigert-Meyer rule. However, a handful occurrences of violation of the Weigert Meyer rule have been reported in literature, with most of these deviations being reported in children (10 - 17). The data regarding prevalence of violation of the stated rule are lacking.

One of the first violations of the rule was reported by Weigert himself in 1878, in which an upper pole moiety was seen draining into the bladder cranially to the lower pole ureter (12).
Stephens in 1958 mentioned 7 cases of violation of the Weigert-Meyer rule wherein the lower pole moiety was seen draining caudally to the upper pole moiety. He also postulated that the ectopic opening were found along a specific pathway and named it the 'ectopic pathway' (13).
Ahmed in 1981 reported a case of violation of the stated rule in which the lower pole moiety was seen draining caudally and was associated with a ureterocele, while the upper pole moiety appeared to be attached to a non-functioning ectopic kidney (14).

Brown et al reported a case in 1988 where the lower pole moiety was seen ectopically draining into the ipsilateral epididymis and presenting as epididymitis on the background of a urinary tract infection (12). Slaughehoupt et al reported a similar case in 1997 wherein the ectopic ureter arose from the lower pole of the kidney and was seen draining into the ipsilateral vas deferens (15).
A case of ectopic lower pole ureter with associated cystic dysplasia was reported by Jain et al in 2008 (16).
As depicted in the cases reported by Brown et al and Slaughenhoupt et al where the drainage of the ectopic ureter was noted into a Wolffian duct structure, Mishra et al reported another similar case in 2017 wherein an ectopic ureter arising from the lower pole of the left kidney was seen to drain into the scrotum (10). They were the first to depict the violation of Weigert Meyer rule via a CT reconstruction.

A case of ectopically draining lower pole ureter with associated megaureter, similar to that of Jain et al, was reported by Darr et al in 2020 (11).

Stormont et al reported a peculiar violation of Weigert-Meyer rule in which there was involution of the lower pole moiety (17). Rathbun et al (2020) reported a case of uncrossed ureteral duplication in an infant. with an adynamic obstruction of the lower pole segment, presenting with hydronephrosis in utero (18). Another case of ectopically draining lower pole moiety with stenosis of its vesicular opening and associated megaureter was reported by Kagantsov et al (2022) in a 5 month old infant (19).

In our case, there was a clear diversion from the Weigert Meyer rule, with both the moieties draining into the bladder ectopically; inferior and medial to the expected site of opening. Further, the lower pole moiety was seen draining slightly anteroinferiorly and lateral to the upper pole moiety.
Although many theories have been postulated explaining the mechanism behind the violation of Weiger-Meyer rule, the embryological and molecular basis of these violations still remains unclear.

Conclusion

Thus, while most cases of renal duplication follow the Weigert Meyer rule, this rule is not an absolute law and various phenotypic violations and deviations have been documented. Radiologists and treating urologists should bear in mind the possibility of a violation while dealing with cases of duplicated renal collecting systems. Further, it is possible for patients with duplicated systems to present primarily during adulthood while having an uneventful childhood.

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Declarations

Consent for publication: The author clarifies that written informed consent was obtained and the anonymity of the patient was ensured. This study submitted to Swiss J. Rad. Nucl. Med. has been conducted in accordance with the Declaration of Helsinki and according to requirements of all applicable local and international standards. All authors contributed to the conception and design of the manuscript, participated in drafting and revising the content critically for important intellectual input, and approved the final version for publication. Each author agrees to be accountable for all aspects of the work, ensuring its accuracy and integrity.
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