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Co-existing pelvi-ureteric junction obstruction and retrocaval ureter in a 7-year-old Nigerian girl: a case report

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Abstract

Pelvi-ureteric junction obstruction is a common cause of upper urinary tract obstruction in children and most often due to congenital anomaly of pelvi-ureteric junction. It is the most common cause of significant hydronephrosis in children. It is associated with many congenital urologic anomalies. Retrocaval ureter is a congenital anomaly of the ureter resulting in varying degree of proximal ureteral obstruction. Though retrocaval ureter is associated with many congenital anomalies, it is infrequently reported in association with pelvi-ureteric junction obstruction. There is no data report of co-existing pelvi-ureteric junction obstruction and retrocaval ureter to the best of our knowledge. Clinical presentation of pelvi-ureteric junction obstruction occurs commonly as flank pain in older children and adults. We present our experience in the diagnosis and management of a 7-year-old Nigerian girl with co-existing right pelvi-ureteric junction obstruction and retrocaval ureter in a Nigerian tertiary hospital. A 7-year-old girl who presented with 2-year history of colicky right flank pain. She had been to several hospitals with no definitive diagnosis or effective treatment. Computerized Tomography Urogram (CTU) showed right moderate hydronephrosis with non-visualization of right ureter which precluded preoperative diagnosis of retrocaval ureter. At surgery through an open flank extraperitoneal approach, a right hydronephrosis, stenotic right pelvi-ureteric junction and normal right retrocaval ureter were found. Excision of stenotic segment, retrieval of retrocaval ureter and Anderson-Hynes pyeloplasty was done over 3Fr DJ stent. Postoperative period was not adversely eventful. Pelvi-ureteric junction obstruction can co-exist with retrocaval ureter. If both are responsible for ureteral obstruction, it may pose challenges to reconstruction. It is therefore important for urologists to be aware of possible associations and treatment challenges that may result.

Key words: pelvi-ureteric junction obstruction, retrocaval ureter, congenital.

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Introduction

Pelvi-Ureteric Junction Obstruction (PUJO) refers to an impediment to the normal antegrade flow of urine from renal pelvis to the ureter which results in elevated intra-pelvic pressure and urinary stasis that contributes to progressive renal damage and function deterioration.[1] It is the most common cause of functionally significant hydronephrosis in children. It accounts for 80% of causes of antenatally detected hydronephrosis.[1,2] Most cases of PUJO are congenital and diagnosed prenatally in advanced countries.[1] Proposed pathogenetic bases for congenital PUJO include incomplete canalization of proximal ureter, premature arrest of muscular development affecting proximal ureter, deficiency of interstitial cells of Cajal, proximal ureteral valves or polyps and extrinsic compression by aberrant lower renal pole vessles.[1] Being mainly congenital, there are a number of associated congenital urologic anomalies. These include horseshoe kidney, multicystic dysplastic kidney, duplex collecting systems, vesico-ureteral reflux, prostatic utricles, hypospadias and undescended testes.[3] Retrocaval Ureter (RCU) is a congenital anomaly in which the proximal ureter takes a route posterior to the Inferior Vena Cava (IVC) before insertion into the renal pelvis resulting in varying degree of ureteric obstruction.[4] This anomaly results from the persistence of right sub-cardinal vein ventral to the ureter in which the ureter is made to course behind and medial to IVC over the third lumber part of the ureter located posterior to the IVC.[4] Its prevalence is 0.06-0.17 % globally and it is more common in males.[4,5] It is more common on the right side.[5] RCU commonly occurs in isolation, existing in association with other anomalies in 21% of cases.[6,7] Associated anomalies with RCU that have been reported include solitary kidney, horseshoe kidney, congenital absence of bilateral vas deferens, branchial arch syndrome, retroperitoneal fibrosis, renal cell carcinoma.[6,7] Although PUJO may be asymptomatic, flank or loin pain is the most common presenting symptom in older children and adults.[1] Patients may also present with symptoms of complications of PUJO such as recurrent UTI and non-functioning kidney. Abdominopelvic ultrasound scan, Computerized Tomography Urogram (CTU), magnetic resonance imaging and nuclear scan are currently less invasive modalities of diagnosing PUJO and RCU.[1,8] Pyeloplasty remains the gold standard of treatment of PUJO with good outcome in >90% of cases.[1] Though many cases of PUJO with associated anomalies have been reported, no case of RCU co-existing with PUJO has been reported to the best of our knowledge. We therefore report a case of co-existing PUJO and RCU in a 7-year-old Nigerian girl.

Case Report

A 7-year-old girl was referred from a private hospital on account of recurrent right flank pain of two-year duration. The pain was described by the mother as intermittent, necessitating frequent presentations at several hospitals and with an adverse impact on the child's education. She was first reviewed at the urology clinic in our center with an abdominopelvic ultrasound scan report of moderate right hydronephrosis. Physical examination was not remarkable. CTU was done in our facility which showed hydronephrotic right kidney with abrupt cessation of contrast at right pelvi-ureteric junction. Right ureter was not opacified by the contrast on the delayed film (Figure 1). A retrograde ureteropyelogram was not done in the index patient. Thus, right ureter could not be visualized on the imaging precluding preoperative diagnosis of associated right RCU. Her hemogram, renal function test, urine analysis and other investigations were essentially normal. She was subsequently scheduled for open right pyeloplasty through flank extraperitoneal approach. Intraoperative findings included dilated right renal pelvis, stenotic pelvi-ureteric junction, retrocaval ureter segment with normal caliber (Figures 2 and 3). Excision of stenotic pelvi-ureteric junction was done. RCU was meticulously retrieved and positioned anteriolateral to IVC. Patency of right ureter was ascertained by passing lubricated size 5Fr feeding tube down into the bladder. Anderson-Hynes pyeloplasty was done over a size 3Fr DJ stent using PDS 4/0 suture. Retroperitoneal drain was left *in situ*. Post-operative period was not adversely eventful. Patient was discharged home at 5th post-operative day after removal of retroperitoneal drain. DJ stent was removed at 6th week post-operative period. She is currently on follow-up at the urology outpatient clinic.

Discussion

Pelvi-ureteric junction obstruction is the most common cause of significant upper tract obstruction in children diagnosed and managed by urologists. There are many associated congenital anomalies with PUJO with which urologists are familiar. However, co-existing PUJO and RCU are rare and there are no data report of it to the best of our knowledge. This may be due to the rarity of RCU when compared with PUJO.[4] Also, it may also be due to under-reporting of cases of RCU as the majority of patients with this anomaly are asymptomatic.[4] In addition, non-opacification of ipsilateral ureter in a significant PUJO might cause missed diagnosis of co-existing RCU, contributing to paucity of data on co-existing PUJO and RCU as in this index patient. Part of RCU may also be compressed by IVC, kinked or adynamic resulting in ureteral obstruction. Stenosis of pelvi-ureteric junction is a common underlying pathology of PUJO as found in the index patient.

Patients with PUJO can be diagnosed prenatally and anytime in post-natal period. The peak age of presentation is 20-40 years.[1] The majority of older children and adults with PUJO present with flank pain as in our index patient.[1] CTU plays crucial role in the diagnosis of PUJO and RCU. In PUJO, the urogram shows classical abrupt cessation of contrast at pelvi-ureteric junction as in the index patient, as shown in Figure 1. However, fishhook or S-shaped deformity of the proximal ureter on urogram clinches the diagnosis of RCU which was not observed in our patient due to non-opacification of right ureter on urogram. Treatment of PUJO may be non-operative or operative depending on the symptomatology, severity of hydronephrosis, residual renal function or presence of complications. Recurrent flank pain (as in the index patient), severe hydronephrosis, compromised renal function and recurrent UTI are indications for operative care. Pyeloplasty is the gold standard in the management of PUJO and was offered to this patient. Intraoperative findings are shown in Figures 2 and 3.

Conclusions

Pelvi-ureteric junction obstruction can co-exist with retrocaval ureter. If both are responsible for urinary tract obstruction, this may pose grave challenges associated with complex reconstruction. It is therefore important for urologists to be aware of this possible association and treatment challenges that may result.

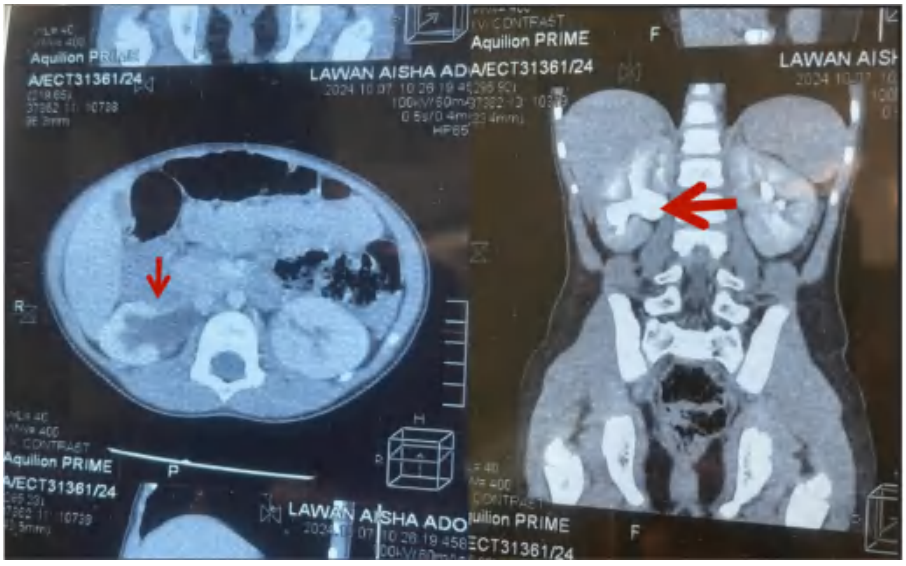

Figure 1. Computed Tomography Urogram (CTU) showing right hydronephrosis with abrupt cessation of contrast at right pelvi-ureteric junction (thick red arrow), hydronephrotic right kidney (narrow red arrow).

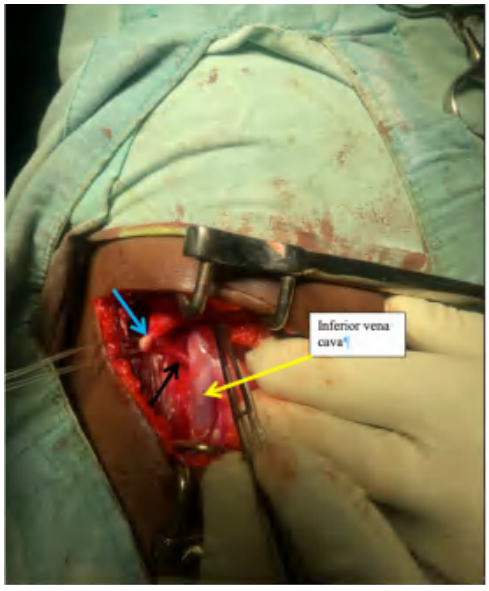

Figure 2. Intraoperative photograph showing right pelvi-ureteric junction (blue arrow) and retrocaval ureter (black arrow).

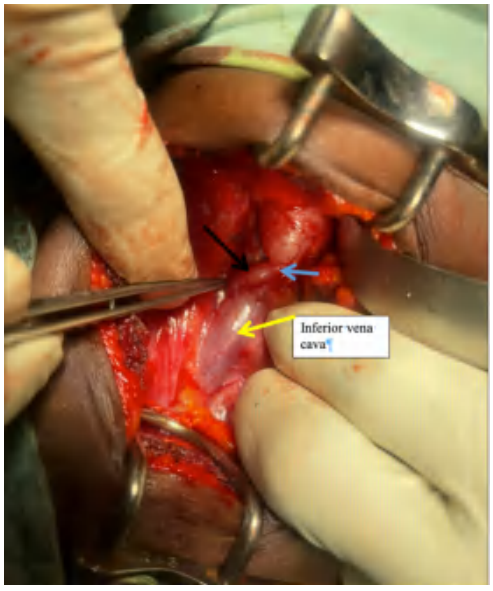

Figure 3. Intraoperative photograph showing right pelvi-ureteric junction (blue arrow) obstruction and co-existing retrocaval ureter (black arrow).

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