VOLUME - 14, ISSUE - 05, MAY - 2025 • PRINT ISSN No. 2277 - 8160 • DOI : 10.36106/gjra

Original Research Paper

Urology

CURIOUS CASE OF HYDROURETERONEPHROSIS: URETERS ON A DETOUR

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ABSTRACT Bilateral Hydroureteronephrosis secondary to Bilateral Ectopic Ureters is a rare presentation in the reported cases of duplex kidneys and ectopic ureters. Duplex collecting system is a common urologic malformation with a wide range of clinical symptoms. It is also associated with variety of urologic abnormalities such as an ectopic ureter, ureterocele, vesicoureteral reflux and ureteropelvic junction obstruction. This report presents a case of a 14-year-old girl who had a bilateral duplex collecting system that was revealed accidentally by a bilateral hydroureteronephrosis. The duplication was complete on both sides. Initially right nephrostomy was done to relieve the hydronephrosis and was followed by bilateral ureteric reimplantation by Modified Politano-Leadbetter procedure along with double DJ stenting.

KEYWORDS :

INTRODUCTION

A duplex kidney is a common congenital anomaly of the kidneys with two pelvicalyceal collecting systems. It may have either single or bifid ureter (partial duplication) or double ureter draining separately into the urinary bladder (complete duplication)[1]. A duplicated ureter is commonly found in association with other congenital anomalies and defects[1]. Unilateral duplication is six times more common than bilateral duplication and it has more female preponderance[2]. Duplication of collecting system occurs due to incomplete fusion of the renal moieties. The normal bud inserts in the correct site of trigone of bladder and the lower bud draining the lower pole of kidney inserts in the bladder with laterally placed ureteric orifice and is therefore more associated with vesico-ureteric reflux (VUR)[2]. Usually, duplication is associated with other congenital anomalies and may cause other complications like ureterocele, urolithiasis and vesico-ureteral reflux[3]. Ectopic ureters are almost associated with ureteric duplication and 10% bilateral with female preponderance[2]. In females, ectopic ureter may either open into urethra below sphincter or into vagina and in males the opening of ureter is above external urethral sphincter. Hence females may experience incontinence [2].

As time progresses ectopic ureters may cause VUR and subsequently hydroureteronephrosis which may need nephrectomy or ureteric reimplantation. Here we have a case of bilateral complete ectopic ureters with duplex collecting system which were found during the investigation of bilateral hydroureteronephrosis.

Case Report

A 14-year-old female was hospitalized due to burning micturition and right loin pain for the last 6 months. She complained of dribbling of urine and retention of urine associated with feeling of urgency. All the symptoms were insidious in onset. She has no history of fever, discharge or haematuria.

On presentation the patient's vitals were normal. Physical examination revealed tenderness over right loin region. The opening of urethra is normal and there were no other examination findings. Her labs on the day of admission are as follows.

Table1. Initial Laboratory Analysis of the Patient

Hemoglobin	8.6 gm/dl
TLC	10.04 per mcL
Platelets	404 per mcL
Bleeding Time	1 min 30 sec
Clotting Time	3 min 15 sec
Serum Creatinine	2.2 mg/dl
Serum Sodium	144 mmol/L
Serum Potassium	4 mEq/L

Increased serum creatinine suggests acute kidney injury. Viral markers were non-reactive and chest x-ray and ECG were normal. 2D echo revealed 58% ejection fraction. Urine analysis and urine culture showed no significant findings ruling out any infective etiology.

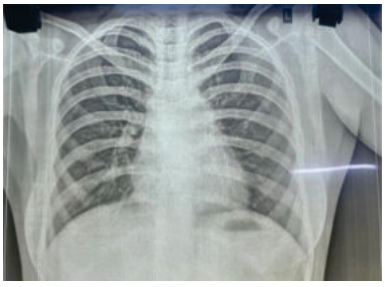

Fig 1. Chest X ray PA view.

Ultrasound abdomen showed severe bilateral hydro-ureteronephrosis of the pelvicalyceal system with cortical thinning and dilatation of the ureters with its terminal end bulging into the bladder lumen. It also gave rise to the suspicion of ectopic ureters. To get a good grasp of the situation magnetic resonance urogram was done which gave

an impression of bladder outlet obstruction. CT urography was advised to gain further insight.

CT KUB Plain revealed a mildly dilated left pelvicalyceal system and sub centimetric calculi in the right upper and lower calyx. It also revealed a 7.4mm calculus in the left vesico-ureteric junction causing proximal ureteric dilatation. Final impression was left mild hydroureteronephrosis secondary to VUJ calculus and right renal calculi.

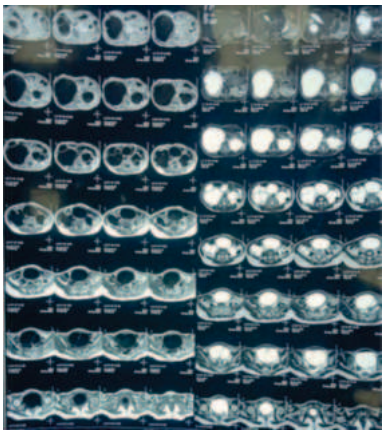

Fig 2. CT KUB Plain and Contrast

CT KUB with urography revealed bilateral gross hydro-ureteronephrosis with parenchymal thinning. One hour delayed phase images show excretion of contrast into the urinary bladder.

Fig 3. CT KUB Contrast

Technetium scan showed left hydroureteric kidney showing reduced cortical function and delayed drainage, right enlarged gross hydroureteronephrotic kidney with severely reduced cortical function and delayed drainage due to VUJ obstruction.

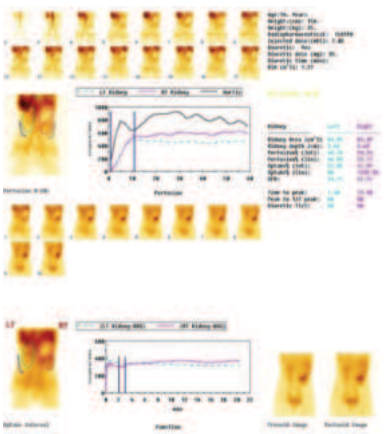

Fig 4. DTP scan

Surgical Management

Initially a right percutaneous nephrostomy was done to relive the pressure on kidney by draining some urine. This would also give some time to plan the intended surgical management. Diagnostic cystoscopy was done to get a view on the insertion sites of ureters.

Based on the diagnostic findings, the decision was made to perform the Modified Politano- Leadbetter procedure to surgically correct the bilateral ectopic ureters. This procedure is approached by the Pfannensteil incision. The bladder is mobilized and the ureter is identified and transected. Free end of the proximal ureter is spatulated, the bladder is opened on the anterior surface and the ureter is tunneled into the bladder wall in the fashion of Politano- Leadbetter. Left ureteric reimplantation was done and on the right side a common sheath was formed for the two ureters. The ureters were implanted using an anti-reflux technique, followed by placement of DJ stents on both sides. Post operatively x-ray KUB was performed to confirm the positioning of DJ stents. Penrose drain was inserted and supra pubic catheterization was done with 14Fr Foley's to decrease the burden on bladder. 2 units of PRBC was transfused post op instead of pre op as hemoglobin levels were 8.6 gm/dl pre op. Ureteric Tissue samples were sent for HPE examination which revealed histological features of right and left ureteric duplication with squamous metaplasia and obliterated ectopic ureteric cord.

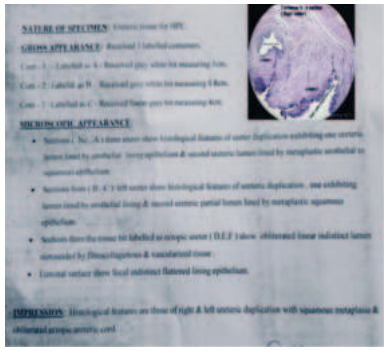

Fig 5. Histopathological Report

Drain was removed on POD-5. She was advised to follow-up after 3 weeks. After 3 weeks the patient underwent another set of blood tests where all of them were normal, serum creatinine was decreased to 1.2 mg/dl. Follow up ultrasound showed mild bilateral hydroureteronephrosis. DJ stent removal was done one month after the surgery. The patient was kept on follow up for every 6 months and she is asymptomatic at present.

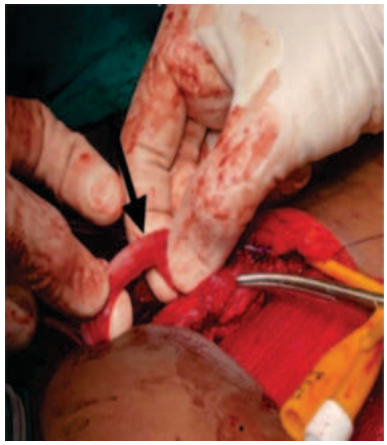

Fig 6. Grossly Dilated Right Ureter

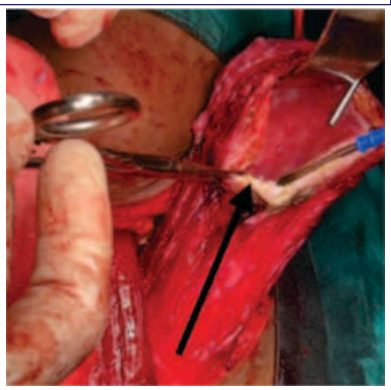

Fig 7. Opened Bladder

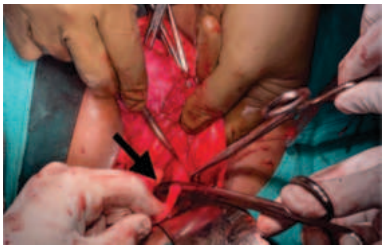

Fig 8. Ureter Divided at Distal end

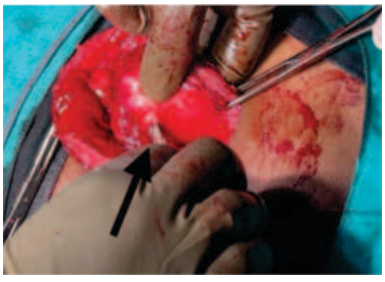

Fig 9. Creation of Dubmucous Tunnel

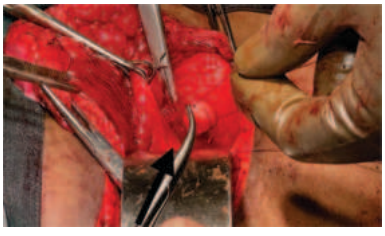

Fig 10. Trans Vesical Ureteric Reimplantation – Leadbetter Politano Technique (Non refluxing)

DISCUSSION

Intermediate mesoderm forms the genitourinary system from the mesonephric tubules. They join to form the Wolffian duct or the mesonephric duct. The ureteric bud arises from the mesonephric duct in 5th week of intra uterine life. The ureteric bud and the metanephric tissue form the renal pelvis, calyces and the collecting system. Sometimes the ureteric bud and metanephric tissue divide before penetrating and give rise to bifid ureters which may drain into bladder through single opening or dual openings [4]. Most commonly the ectopic ureter follows the Weigert-Maeyer law. In cases of duplex kidney and complete ureteral duplication, the upper renal and lower renal moieties are drained by separate ureters, each having its own ureteral orifice in the bladder. Upper renal moiety ureter has ectopic insertion medial and inferior to the lower renal moiety ureter, and commonly ends in a ureterocoele whereas lower renal moiety ureter has orthotopic insertion lateral and superior to the upper renal moiety ureter and vesicoureteral reflux can commonly occur in the normal ureter [5]. If the ectopic ureter doesn't follow this path and if it's drained medially and superiorly to the normal ureteric orifice, it is termed as Stephen's Ectopic Pathway.

Many studies revealed variable incidence of duplex kidney with incomplete or complete duplication. The study done by Prakash et al.[6] and Dahnert[7] by analysing urograms found that partial duplication of ureter to complete duplication is in the ratio 3:1. A study done by Whitaker and Danks[8] found that unilateral duplication to bilateral duplication is in the ratio 6:1. Siomou et al[9] found that duplex kidney is more common in girls as compared to boys. And in another study done by Rege VM et al.[9] concluded that bifid ureter was often presented on the right side.

The two most important clinical consequences of a duplex kidney are vesicoureteral reflex and ureterovesicular junction obstruction. Previous reports on this condition in children detail the many possible anatomic variations of a duplicated collecting system, differing mostly in where the redundant ureter inserts. This case was unusual as the patient was found to have bilateral ectopic ureters, both causing hydro-ureteronephrosis. Diagnosis by radio imaging plays an important role in identifying duplex kidneys. Dual collecting systems are better identified in magnetic resonance imaging rather than intra venous pyelography. Most of the cases become symptomatic in childhood[10], as reflected in the present case.

Surgical treatment is the preferred course of action in cases which are similar to the present case. The main dilemma is the should augmentation of bladder neck be performed to improve the continence of the patient. In a study done by Kesavan et al.,[11] the neck of the bladder and the trigone were maldeveloped in most of the bilateral and unilateral ectopic ureters. The incontinence maybe due to insufficient development of trigone and bladder neck as suggested by Heuser et al.[12]

Various other procedures such as using pubo vaginal sling and artificial sphincter to increase the bladder neck resistance were proposed. But a study done by Podesta[13], reported that a bladder with bilateral ectopic reimplantation can achieve normal function with continence. Hence only ureteral reimplantation was considered in this case and the patient didn't complain of any incontinence in the regular follow up.

CONCLUSION

Even though duplex kidney is a commonly found congenital malformation, a case with bilateral ectopic ureters is uncommon. And the symptoms it caused make the case more interesting to deal with. Numerous anatomical variations of the ectopic ureters must be reported and studied to understand the symptoms with which the patient is presented and to choose the surgical technique required.

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