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Research Paper

A Rare and Unique Case of Bilateral Double Collecting System with Congenital Ureteric Anomalies Presenting as Abdominal Distension in a Two-Year Old Baby Girl.

Medical Science

KEYWORDS : Malrotated duplex kidney, Congenital Giant Megaureter, Right upper moiety dilatation,Abdominal distension.

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ABSTRACT

Development disorders of kidneys and urinary tract constitute a spectrum of malformations ranging from the complete absence of kidney tissue to minor structural abnormalities. We report a unique case of malrotated duplex kidney with complete duplication of ureter with upper moiety grossly dilated tortuous ureter suggestive of Congenital Giant Megaureter (CGM) on Right side and duplex kidney on left side with partial duplication of ureter in a two year old female child which presented as abdominal distension since birth.

Such anomaly with the normal function and drainage in both the kidneys has not been reported so far, in this age group.

Introduction :

The incidence of renal and urinary tract malformation identified in fetal ultrasound structures is 0.3 to 1.6 per 1000 infants (both live born an still born)[1]. Abnormalities of the lower urinary tract can be identified in approximately 50% of affected patients. These anomalies include vesicoureteral reflux (25%), ureteropelvic junction obstruction (11%), and ureterovesical junction obstruction (11%) [2]. Renal malformations, other than mild antenatal pelviectasis occur in association with non-renal malformations in about 30% cases[1]. Most incidence rates are not derived from population-based studies during pregnancy; rather, they are derived from autopsy series (or) studies in selected live-born infants. Duplication of renal collecting system, both complete and partial, is the most common congenital anomaly of urinary tract [3]. According to autopsy studied, the incidence is estimated to be 0.8% to 5% [4].

Similar rate was reported in a study of 13,705 fetuses for which antenatal ultrasound examinations were performed in a tertiary center in Turkey [5]. However. a study in which 132,686 Taiwanese school children (6 to 15 years of age) were screened revealed a lower incidence (1 per 5000 children)[6].

In this case we report bilateral double collecting system with duplication of ureters. Right kidney: malrotated duplex with complete duplication of ureter with upper moiety grossly dilated tortuous ureters - Congenital Giant Megaureter (an extremely rare subgroup of megaureter) (CGM) and Left kidney: duplex with partial duplication of ureter in a 2 year old baby girl presenting as abdominal distension

Case Report:

A 2 year old baby girl, weighing 10kg presented with progressive distension of abdomen (FIG - 1). No abnormality was detected on general physical examination. Abdominal examination, on inspection shows generalized distension in right lumbar and iliac region. On palpation, liver was normal, no organomegaly noted. Bimanual palpation of kidneys revealed ill-defined renal angles on both sides. On auscultation bowel sounds heard normally. The urinalysis, blood urea and serum creatinine were found normal. Ultrasonography was advised which revealed bilateral duplex kidneys; Right Kidney: Duplex kidney with complete duplication of ureter with upper moiety dilated and Left Kidney: Duplex kidney ? Partial duplication of ureter. Further Intravenous pyelography(IVP) and Contrast-enhanced computed tomography(CECT) scan of abdomen and pelvis was done which showed – Bilateral double collecting system with duplication of ureters. Right kidney: Malrotated duplex with complete duplication of ureters with upper moiety grossly dilated tortuous ureters suggestive of Congenital Giant Megaureter(CGM); Left kidney : Duplex with partial duplication of ureter. Micturating Cystourethrogram(MCU) showed normal bladder and urethra – No finding of vesicoureteric reflux(VUR) noted. Nuclear Scan DTPA Renogram done at Nizam's Institute of Medical Sciences (NIMS) reveals normal drainage and normal functions of both the kidneys.

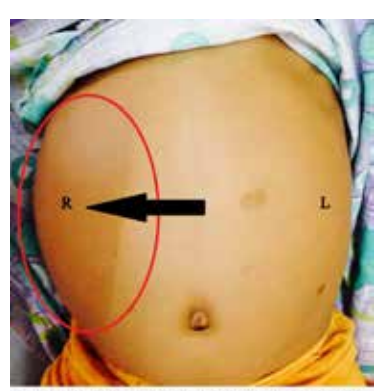

Image showing : Distension of abdomen on right side

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FIG – 1 : SHOWING ABDOMINAL DISTENSION ON RIGHT SIDE

Materials and Methods:

Investigations:

Complete Blood Picture:

Hemoglobin: 10.0gm/dl (100g/L), WBC Count: 10,800/cu.mm, Neutrophils: 59%, Lymphocytes: 37%, Eosinophils: 02%, Monocytes: 02%; Platelets: Adequate, Red Cell Morphology suggestive of Hypochromic, normocytic, microcytosis and anisocytosis.

Biochemical tests:

Blood urea: 22mg/dl, Serum Creatinine: 0.6mg/dl, Alkaline Phosphatase: 105U/L.

Urine Analysis:

Colour : pale yellow, Transperancy: Hazy, Reaction: Acidic, Albumin & sugar: Nil, Pus cells: 2-3, Red blood cells: nil, Epithelial cells: 2-3.

Ultrasonography in this case:

Ultrasonography gives excellent information on anatomical aspects. It is especially suited for children since it is painless, requires no sedation (or) radio contrast administration and can be repeated safely. In this case, ultrasonography reveals duplex kidney on both right and left side. On right side: duplex kidney with complete duplication of ureter with upper moiety grossly dilated(FIG - 2). On left side: Duplex kidney partial duplication of ureter(FIG - 3).

FIG – 2 ULTRASONOGRAPHY SHOWING RIGHT UPPER MOIETY DILATATION AND RIGHT LOWER MOIETY, ALSO SHOWING RIGHT DUPLEX KIDNEY.

FIG – 3 ULTRASONOGRAPHY SHOWING LEFT DUPLEX KIDNEY

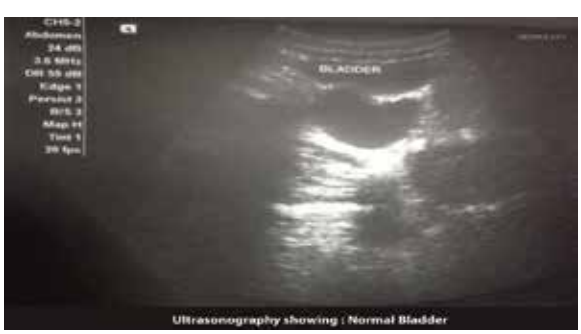

FIG – 4 ULTRASONOGRAPHY SHOWING NORMAL URINARY BLADDER

Intravenous Pyelography:

Intravenous Pyelography(IVP) is useful for detailed evaluation of structural anomalies such as duplex kidneys and horseshoe kidney, and for ureteric calculi. IVP requires bowel preparation and an ionic contrast (urograffin, 3-4mL/kg) was administered. Films were taken at 1-5 minutes, 10-15 minutes and a late pelvic film for bladder was taken. IVP in this case shown duplex kidneys on both the sides, with right side showing complete duplication of ureter with upper moiety grossly dilated tortuous ureter which is suggestive of Congenital Gaint Megaureter(CGM) and left side showing partial duplication of ureter(FIG – 5 & 6).

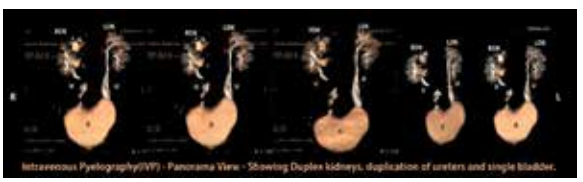

FIG – 5 PANORAMA VIEW

FIG – 6 PANORAMA VIEW

FIG – 5 & 6 INTRAVENOUS PYELOGRAPHY SHOWING BILATERAL DUPLEX KIDNEYS, WITH RIGHT SIDE SHOWING COMPLETE DUPLICATION OF URETER WITH UPPER MOIETY GROSSLY DILATED TORTUOUS URETER AND LEFT SIDE SHOWING PARTIAL DUPLICATION OF URETER.

Contrast-Enhanced Computed Tomography Scan – WHOLE ABDOMEN:

CECT Scan was taken outside the institute where the findings showed:

LIVER: Normal in size, shape and attenuation. No intra hepatic duct dilatation. No focal Lesions.

Common Bile Duct and Portal Vein: Normal.

GALL BLADDER: Normal in contours. Wall thickness appears normal. No calculi.

SPLEEN: Normal in size, shape and attenuation. No focal lesions noted.

PANCREAS: Appears normal in size, shape and attenuation. No ductal dilatation. No calcifications/calculi. Peripancreatic fat planes appear normal.

SUPRARENAL REGIONS: Clear appear normal.

BOTH KIDNEYS – Bilateral double collecting system with duplications of ureters.

Right malrotated duplex kidney and complete duplication of ureter with upper moiety grossly dilated tortuous ureter suggestive of Congenital Giant Megaureter.

Left duplex kidney with partial duplication of ureter. No evidence of left hydrouretronephrosis.

URINARY BLADDER: Normal in contours. No filling defects noted. No calculi. Wall thickness appears normal.

RECTUM: Appears normal. Peri-rectal and peri-vesical fat planes

appear normal.

AORTA and Inferior Vena Cava: Normal, no paraaortic/paracaval adenopathy.

No free fluid in the peritoneal cavity.

Opacified stomach and bowel loops appear normal.

Impressions of CECT Scan were : Right malrotated duplex kidney and complete duplication of ureter with upper moiety grossly dilated tortuous ureter S/O Congenital Giant Megaureter.

Left duplex kidney with partial duplication of ureter(FIG – 7,8 and 9).

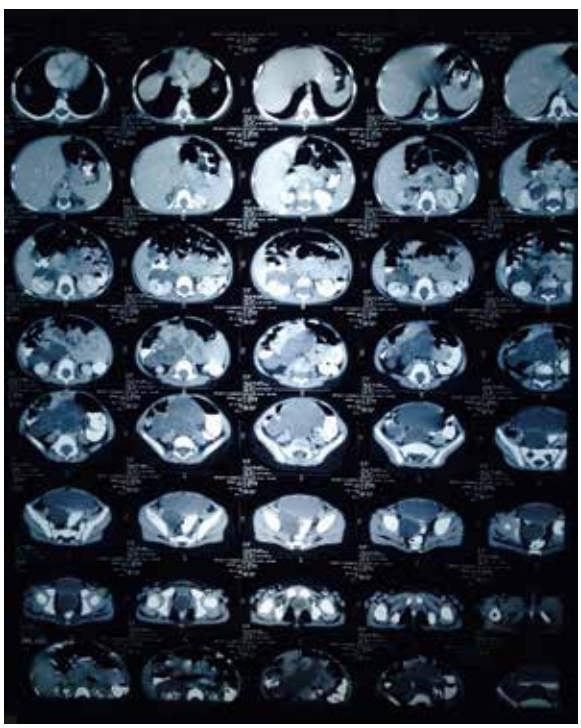

FIG – 7 CECT SCAN – WHOLE ABDOMEN

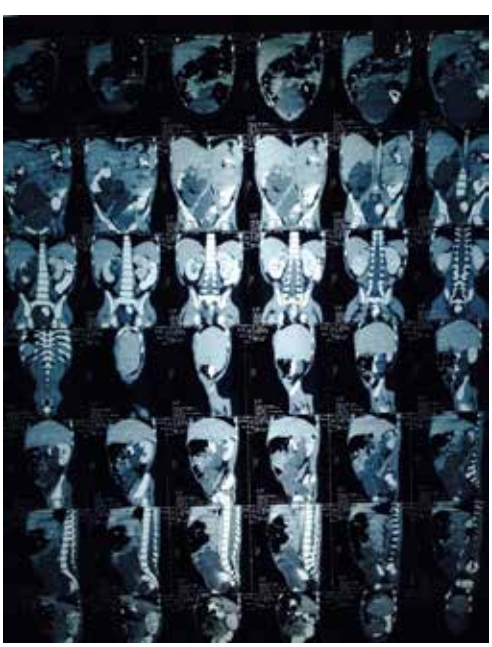

FIG – 8 CECT SCAN – WHOLE ABDOMEN

FIG – 9 CECT SCAN – PANORAMA VIEW

DTPA Renogram:

Renography/renal perfusion study monitors the arrival, uptake and elimination of a radiopharmaceutical by the kidney. Agents used include 99 mTc labeled diethylene triamine-penta-acetic acid (DTPA), an agent excreted purely by glomerular filtration.

In this case, 99 mTc labeled diethylene-triamine-penta-acetic acid (DTPA) was used, with diuretic. PROTOCOL : F0 lasix, and 3m Ci99 mTc- DTPA injected I.V. and posterior dynamic views taken.

A diuretic renogram helps differentiate obstructive dilatation of upper urinary tract from non-obstructive hydronephrosis; in the latter, the radionuclide clears promptly following the injection of frusemide. DTPA scan finding showed Normal drainage and normal function of both the kidneys (FIG - 10, 11 and 12).

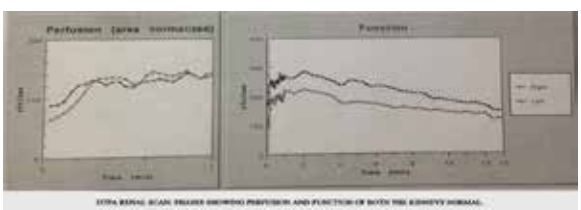

FIG – 10 DTPA RENAL SCAN SHOWING PHASES OF PERFUSION AND FUNCTION OF BOTH THE KIDNEYS NORMAL

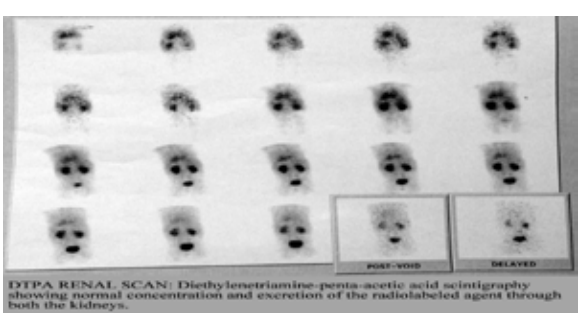

FIG – 11 DTPA RENAL SCAN SHOWING NORMAL CONCENTRATION AND EXCRETION OF THE RADIOLABELED ISOTOPE THROUGH BOTH THE KIDNEYS

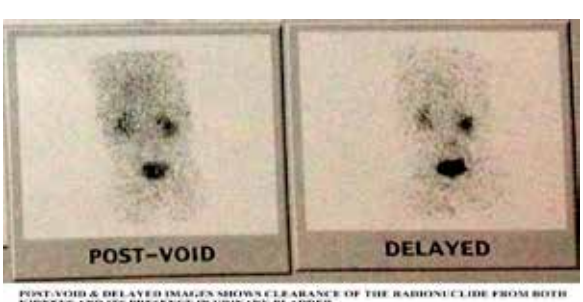

FIG – 12 DTPA RENAL SCAN SHOWING CLEARANCE OF THE RADIONUCLIDE FROM BOTH THE KIDNEYS AND ITS PRESENCE IN URINARY BLADDER.

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History of DTPA Renogram Scan:
DTPA Renogram Scan (1986) was developed at University of Utah by Dr. Alan R. Fritzberg, Dr. Sudhakar Kasina & Dr. Dennis Eshima[15].Drug underwent clinical trials in 1987[16] and passed phase III testing in 1988[17].

Micturating Cystourethrogram:
The micturating cystourethrogram(MCU) is useful for diagnosis and grading of vesicoureteric reflux(VUR) and detection of bladder and urethral abnormalities. Following urinary catheterization, radiocontrast agent is introduced into the bladder; films are taken while the child is voiding. Strict aseptic precautions are required.

In this case MCUG showed normal bladder and urethra.

Abbreviations:
CGM(Congenital Gaint Megaureter), CAKUT(Congenital Anomalies of Kidney and Urinary Tract), CECT(Contrast-Enhanced Computed Tomography), DTPA(Diethylene-Triamine-Penta-Acetic acid), MCU(Micturating Cystourethrogram).

Discussion:
Double Collecting System :
Complete (or) partial duplication of the renal collecting system is the most common congenital anomaly of urinary tract[3]. A double collecting system is thought to result from duplication of the ureteric bud, whereby the superior bud is associated with the upper renal pole and the inferior bud with the lower renal pole. In complete duplication, the kidney has two separate pelvicalcyceal systems and two ureters. The ureter from the lower collecting system usually enters the bladder in the trigone, whereas the ureter from the upper collecting system can have a normal insertion in the trigone (or) be inserted ecopicalcy in the bladder (or) elsewhere. In boys, insertion occurs in posterior urethra ejaculatory ducts (or) epididymix; whereas in girls, I ntothe vagina (or) uterus Ectopic insertion of the uretr can result in obstruction or vesicoureteral reflux. Depending on the location of the ectopic insertion, incontinence also may be present. Partial duplication is more common than complete duplication.

In this case we report Bilateral double collecting system with duplication of ureters.

On Right side, there is malrotated duplex kidney with complete duplication of ureter with upper moiet grossly dilated tortuous ureter – s/o CGM and on left side, we report duplex kidney with partial duplication of ureter.

Duplication of the ureter :
Nation (1944) found some form of duplication of ureter in 0.9% of a series of autopsies[9]. The condition occurs more in females than in males and is often bilateral. The male of inheritance is autosomal dominant, although the gene is of incomplete penetrance (At well et al, 1974)[7].

The incomplete (4) type of duplication is caused by branching of the ureteral bud before it reaches the metanephric blastema. In most cases, this anomly is associated with no clinical abnormality. However, disorders of peristalsis may occur near the point of union (O'Reilly et al, 1984)[10]. In such cases one segment may be abstructed (or) dilated owing to ureteroureteralreflux. A ureteropyelostomy is effective treatment in most cases (Sole, Randall and Arkell, 1987)[11].

In complete duplication of ureter, the presence of 2 ureteral buds leads to formation of 2 separate ureters and 2 separate renal pelves. Because the ureter to the upper segment arises from a cephalad position on the mesonephric duct, it remains attached to the mesonephric duct, it remains attached to mesonephric duct longer and consequently migrates farther, ending medical and inferior to the ureter draining the lower segment (Weigert-Myer law)[12]. Thus, the ureter draining upper segment may, migrate too far caudally and become ectopic and obstructed, while the ureterdraining the lower segment may, end laterally and have a short intravesical tunnel, which leads to vesicoureteral reflux (Kaplan, Nasrallah, and King,1978; Tanagho, 1976)[8]. The same general principle is noted in rare ureteral triplication (Zaontz & Maizel's, 1975)[12].

Here in this case (2015) we report you both complete and partial duplication of ureters.

There is complete duplication of ureter with upper moiety grossly dilated tortuous ureter-CGM on right side and also partial duplication of ureter on left side shown on Intravenous pyelography (IVP).

Taking advise DTPA scan was done at Department of Nuclear Medicine, Nizam's Institute of Medical Sciences (NIMS) where the scan revealed normal drainage and normal function by both the kidneys.

Upper Urinary Tract Duplication:
Duplex collecting systems are the most common anomaly of the upper urinary tract, with a reported incidence of 0.8%. A duplex kidney is one that has two separate pelvicaliceal systems. If two separate ureteric buds originate from the mesonephric duct, two separate interactions will develop between the ureter and the metanephricblastema, resulting in the formation of a duplex system. This duplex system includes two separate renal units and collecting systems, ureters, and ureteral orifices. Bifid pelvis, bifid ureters, or double ureters can be formed depending on the level at which the ureters join. Duplication anomalies are usually asymptomatic; there- fore they often remain undetected. However, a proportion of duplex kidneys may be associated with VUR or obstruction. Fetal urinary tract dilatations are related to complicated renal duplication in 4.7% of cases.

The final position of the ureteral orifices in the bladder follows somewhat counterintuitive so-called Weigert-Meyer rule: the lower orifice belongs to the upper pole orifice, and the higher orifice to the lower pole. To achieve these positions, the two ureters and orifices rotate 180 degrees clockwise in their longitudinal axes. The orifice draining the lower moiety is commonly refluxive, whereas the ureter draining the upper moiety may end in an obstructive ureterocele that inserts into the bladder in a more distal position than normal.

The routine postnatal radiologic workup of abnormal duplex kidneys is based on ultrasonography and voiding cys- tourethrography. Voiding cystourethrography is performed to detect VUR and to evaluate the ureterocele. Isotope studies are required to determine the function of the dilated renal moiety.

Megaureter:
Megaureters are usually discovered through screening ultrasonography of the kidneys and bladder because of a prenatal diagnosis of hydronephrosis or postnatal UTI, hematuria, or abdominal pain. A careful history, physical examination, and VCUG identify causes of secondary megaureters and refluxing megaureters as well as the prune-belly syndrome. Primary obstructed megaureters and nonobstructed megaureters probably represent varying severities of the same anomaly

The primary obstructed nonrefluxing megaureter results from abnormal development of the distal ureter, with collagenous tissue replacing the muscle layer. There is disruption of normal ureteral peristalsis, and the proximal ureter widens. Usu-

ally there is not a true stricture. On IVP, the distal ureter is more dilated in its distal segment and tapers abruptly at or above the junction of the bladder. The lesion may be unilateral or bilateral. Dilatation of the upper collecting system and calyceal blunting are suggestive of obstruction. Megaureter predisposes to UTI, urinary stones, and flank pain because of urinary stasis. In most cases, diuretic renography and sequential sonographic studies can reliably differentiate obstructed from nonobstructed megaureters. Generally, hydronephrosis lessens and most megaureters diminish in size over time. Truly obstructed megaureters require surgical treatment, with excision of the narrowed segment, ureteral tapering, and reimplantation of the ureter. The results of surgical reconstruction are usually good, but the prognosis depends on preexisting renal function and whether complications develop.

If differential renal function is normal (>45%) and the child is asymptomatic, it seems safe to follow the patient with serial ultrasonography and diuretic renography to monitor renal function and drainage. These children should receive prophylactic antimicrobial therapy while there is urinary stasis in the upper ureter and kidney. If renal function deteriorates, upper urinary tract drainage slows, or UTI occurs, ureteral reimplantation is recommended. Overall, 20–30% of children with a nonrefluxing megaureter undergo ureteral reimplantation.

Clinical Management of CAKUT:
CAKUT play a causative role in 30% to 50% of cases of end-stage renal disease in children[13], it is important to diagnose and initiate therapy to minimize renal damage, prevent or delay the onset of end-stage renal disease, and provide supportive care to avoid complications of end-stage renal disease.

Overall approach to Management of CAKUT:
In newborns with bilateral renal malformation, a solitary malformated kidney, (or) a history of oligohydramnios, abdominal ultrasonography is recommended within the first 24hours after birth, because an intervention such as decompression of the bladder with a transurethral catheter may be required .

Newborns with unilateral involvement do not need immediate attention. In these infants, renal ultrasonography is generally performed between 48 hours in one week after birth. USG before 48-hour of age may not reveal collecting system dilatation because a newborn is in a relatively volume-contracted state during that time[14]. The serum creatinine concentration ca be used to estimate the extent of renal impairment and should be assessed when renal disease is bilaterally or a solitary kidney is present. The serum creatinine concentration at birth is similar to that in mother(usually <1.0mg/dL[88umol/L]). Thus, serum creatinine concentration should be measured after the first 24hours after birth. It declines to normal values(0.3 to 0.5mg/dL[27 to 44umol/L]) within approximately 1 week in full term infants and 2 to 3 weeks in preterm infants.

Management in this case:
Abdominal and pelvic USG is indicated to determine the presence of a collecting system. Voiding Cystourethrography should be undertaken, particularly if hydronephrosis is present, in view of the risk of VUR and urinary tract obstruction. If VUR is detected, then prophylactic antibiotics should be consider, especially in patients who have a history of UTIs.

Further evaluation is based on the results of renal USG and Voiding Cystourethrography and on the serum creatinine concentration. No further evaluation is required in the patient with normal appearing contralateral kidney and no evidence of hydronephrosis.

For further review DTPA renogram studies are done for detecting any obstruction.

In this case, serum creatinine values were in range; no evidence of UTIs, VUR, DTPA and MCUG normal.

So, presently no surgical intervention is required, however a follow-up of yearly is necessary.

Conclusions:
The incidence of Congenital Giant Megaureter is a rare variation. The present case report is a rare combined variation of Congenital Giant Megaureter and Bilateral Double Collecting System.

Right Kidney : Malrotated Duplex with complete duplication of ureter with upper moiety grossly dilated tortuous ureters - Suggestive of Congenital Giant Megaureter and

Left Kidney : Duplex with ? Partial duplication of ureter shown on CECT scan and IVP. DTPA Renogram of both kidneys showed normal drainage and normal function, MCUG scan showed no evidence of vesicoureteric reflux (VUR).

As there is normal function and drainage of both the kidneys no intervention is required in this case as for now. However, a follow-up is necessary.

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