

Case Series

Crossed fused renal ectopia: a report of twelve cases at a tertiary health centre and literature review

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ABSTRACT

Crossed fused renal ectopia (CFRE), second most common renal fusion anomaly of the kidney. These patients were mostly asymptomatic and present with complaints due to associated conditions. The management of these conditions was complicated not only by the approach to be picked, but also by the investigation to be performed. The following study was done at a tertiary care centre, with the duration of this study being from January 2019 to April 2021. A total of twelve patients with CFRE required surgical intervention for their symptoms during this period. As a routine history, routine investigations and ultrasound (US) of the abdomen and pelvis was followed for all the patients. Other imaging modalities were opted on case to case basis. Out of n=12 patients of CFRE with fusion who underwent surgical intervention, majority were male patients (n=10). The most common symptom was abdominal pain (n=9). The most common crossed renal ectopia was the left to right fusion (n=7), as compared to right to left CRE (n=5). The most common fusion anomaly was L-shaped (n=8). The endourological procedures (n=6), laparoscopic procedures (n=2) while open procedures (n=4). It is important for a urologist to bear in mind the various presentations of this less known anomaly with diverse presentations. We have highlighted the investigations used at our center which would help plan further treatment and surgical approach in such patient in view of complex renal as well as vascular anatomy.

Keywords: Fusion anomalies, Renal anomaly, Renal calculus, Ectopic kidney

INTRODUCTION

CFRE is a condition in which one kidney crosses the midline and is displaced to the opposite side. It is second most common renal fusion anomaly after horseshoe kidney with incidence of 1/7500 on autopsy findings and 1/1000 in live births. It is of two types: crossed-unfused renal ectopia (10%) and crossed-fused renal ectopia (90%). These patients are mostly asymptomatic and present with complaints due to associated conditions such as renal stones, ureteropelvic junction obstruction (UPJO), renal tumors and cystic disease of the kidneys.¹⁻⁵ The management includes choosing appropriate imaging

for the diagnosis of the type of fusion anomaly, the urinary tract abnormality and associated anomalies other than the urinary tract. We presented twelve different cases of CRE with fusion, which required endourological, laparoscopic or open surgical intervention for their respective conditions.

CASE SERIES

The following study was done at a tertiary care centre, with the duration of this study being from January 2019 to April 2021. A total of twelve patients with CFRE required surgical intervention for their symptoms during

this period. A routine institutional protocol including assimilation of history with clinical examination, complete blood count, renal function tests, urine routine analysis and microscopy, urine culture and sensitivity and US of the abdomen and pelvis was followed for all the patients. Other imaging modalities such non-contrast CT (NCCT), contrast enhanced computed tomography (CECT), micturating cystourethrogram (MCU), magnetic resonance imaging (MRI), and intravenous urogram (IVU) was done depending on the type of urinary tract abnormalities and patient related factors. NCCT was performed for stone disease, CECT was performed for better anatomical and vasculature delineation before the procedure and diagnosis of uretero-pelvic junction obstruction. MCU was done in cases where vesicoureteric reflux was suspected in view of hydronephrosis on ultrasound. In a pregnant lady, MRI was done to visualize the anomaly. Interventional procedures included endourological procedures (n=5), laparoscopic procedures (n=2) and open procedures (n=4). The patients were followed up duly, for a minimum of three months after the treatment.

Out of n=12 patients of crossed renal ectopia (CRE) with fusion who underwent surgical intervention, majority were male patients (n=10). The youngest patient was 3 months old while the eldest one was 71 years in age. In terms of clinical presentation. The most common symptom was abdominal pain (n=9), recurrent UTI (n=2) and one patient was followed up case of antenatal hydronephrosis. One patient had impaired renal function with sepsis, one presented with fever and chills along with abdominal pain while one female patient was pregnant with 30 weeks of gestation. All patients had USG abdomen and pelvis as the initial imaging, followed by NCCT (n=6), CECT (n=4), MCU (n=2), MRI (n=1) and IVU (n=1). The most common crossed renal ectopia was the left to right fusion (n=7), as compared to right to left CRE (n=5).

The most common fusion anomaly was L-shaped (n=8), disc shaped (n=2), cake shaped (n=1) and superior ectopia (n=1). The endourological procedures (n=6) done were PCNL (n=3), DJ stenting (n=2), ureteroscopic lithotripsy (URSL) (n=1). Laparoscopic procedures (n=2) included lap-assisted PCNL (n=1), cyst de-roofing (n=1) while open procedures (n=4) included nephroureterectomy (n=1), Boari flap (n=1), Cohen cross trigonal ureteric reimplantation (n=1) and Anderson Hynes dismembered pyeloplasty (n=1). The details regarding the same are mentioned in Table 1.

Case 1

31-year-old male with uncontrolled type 1 diabetes on insulin therapy presented with complaints of high grade fever with chills, vomiting and generalized abdominal pain. No history of high risk behavior or lower urinary tract symptoms (LUTS) was noted. On physical examination patient was febrile with low blood pressure

recordings and left renal angle tenderness was present. Laboratory investigations revealed raised total leucocyte count and deranged renal function. Patient was started on empirical intravenous antibiotics after sending a sample for urine culture. NCCT was done which suggested left sided CFRE with bilateral perinephric fat stranding but no evidence of obstructive pathology. The urine culture report revealed *E. coli* (>10⁵), sensitive to cefoperazone. Bilateral double J stenting was done as the patient's general condition further deteriorated on conservative management. Post operatively patient's condition improved and was discharged after 5 days with oral antibiotics for two weeks. On follow-up the ultrasound was normal and both stents were removed.

Case 2

30 year old male patient with nil co-morbidities presented with severe right sided colicky pain associated with two episodes of vomiting since one day. Physical examination was normal but laboratory investigations revealed deranged renal function (serum creatinine 2.1 mg/dl). The patient was subjected to a NCCT which revealed L-type left sided CFRE with tiny vesico-ureteric junction (VUJ) calculus of the size 4 mm in the right ureter. Patient underwent ureteroscopic lithotripsy (URSL) under regional anesthesia with 6/7.5 FR semi-rigid ureteroscope. A calculus was noted at the right VUJ and the stone was fragmented with a pneumatic lithotripter. All fragments were retrieved and 6 Fr/26 cms double J stent was placed in the right ureter. Patient was discharged the next day and stent was removed after 2 weeks.

Case 3

71 year old male with diabetes and hypertension presented with left sided lower back ache since three months which was insidious in onset and gradually progressive in nature over the last one month. His physical examination was unremarkable and orthopaedic causes were ruled out. The patient underwent US of abdomen which showed absent right kidney in the right renal fossa with mild hydronephrosis of the left kidney with calculi.

His renal function parameters were deranged so NCCT was done which showed features suggestive of L type or tandem type of cross fused ectopic kidney on the left side with multiple calculi in the left renal pelvis with mild ectasia of the upper pole calyces and also multiple calculi in upper and lower pole moieties of ectopic right kidney with mild dilatation of renal pelvis.

Patient was advised to undergo standard bilateral prone percutaneous nephrolithotomy (PCNL). Retrograde pyelogram (RGP) was done and 5 Fr ureteral catheters were placed in both the ureters. Under fluoroscopic guidance, left superior calyceal puncture was made and dilated upto 24 Fr. 20.8 Fr mini nephroscope and swiss

pneumatic lithotripter were used to fragment the stones (Figure 1 A and B). Multiple stones were noted with largest 2 cm in renal pelvis. Similarly superior calyceal

approach was taken on the right kidney, dilated upto 24 Fr and 1.6 cm calculus was fragmented. Double J stents were placed in both renal units.

Table 1: Diverse presentations of crossed fused ectopia with management options used at our centre.

S. No	Age (in years)/sex	Presenting complaints	Imaging	CFRE/fusion type/diagnosis	Intervention
Endourological procedures					
1.	31/male	Fever with chills, vomiting and abdominal pain	USG NCCT-KUB	Left Sided CFRE; L-shaped; acute pyelonephritis with AKI	Bilateral double J stenting with IV antibiotics as per
2.	30/male	Right sided colicky abdominal pain, vomiting	USG NCCT-KUB	Left sided CFRE; L-shaped; right VUJ calculus	Right URSL With stenting
3.	71/male	Low back ache with dull aching pain abdomen	USG NCCT-KUB	Left sided CFRE; L-shaped; bilateral renal calculus	Bilateral PCNL with double J stenting
4.	47/male	Dull aching pain abdomen since 1 month	USG NCCT-KUB	Left sided CFRE; L-shaped; bilateral renal calculus	Bilateral PCNL with double J stenting tab. febuxostat 40 mg/day for 2 weeks
5.	34/female	Left loin pain Primigravida 30wks gestation	USG MRI	Right to left; disc/doughnut right hydronephrosis	Left USG guided double 'J' stenting
6.	38/male	Left loin pain	USG NCCT	Left to right; L type; left renal calculi	Left mini PCNL with double J stenting
Laparoscopic procedures					
7.	46/male	Fullness in the right upper quadrant of the abdomen with dull aching type of pain since one month.	USG abdomen CECT-Abdomen and pelvis	Right Sided CFRE; L-shaped; renal cyst	Open cyst deroofing
8.	50/female	Right loin pain	USG CECT	Left to right; L-shaped; right hydronephrosis with 2cm pelvic calculi	Laparoscopy assisted percutaneous nephrolithotomy
Open procedures					
9.	3/male	Antenatally detected hydronephrosis	USG IVU MCU	Left to right; superior; left atretic Ureter right grade III VUR hydrocephalus	Left nephroureterectomy
10.	31/male	Recurrent urinary tract infection	USG CECT MCU	Right to left; L-shaped; bilateral grade IV VUR	Cohen cross trigonal reimplantation
11.	17/male	Recurrent urinary tract infection	USG CECT RGP	Right to left cake/lump bilateral hydronephrosis; left orifice hypoplastic	Bridged Boari flap (short flap)
12.	40/male	lower abdominal pain, impaired renal function, sepsis	USG NCCT RGP	Right to left; disc/doughnut; bilateral pelviureteric junction obstruction	(B/L) Anderson Hynes dismembered pyeloplasty

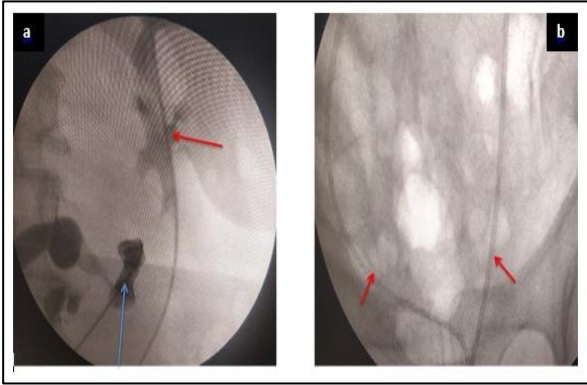


Figure 1: (A) RGP and ureteral catheter insertion of bilateral kidneys (blue arrow depicting the ectopic kidney) (red arrow showing normal left kidney); (B) fluoroscopic image: normal position of bilateral ureteric orifices with ureteral catheters *in situ* (marked by 2 red arrows).

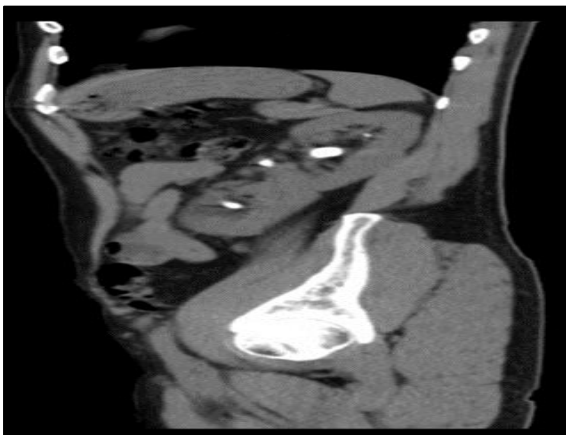


Figure 2: Sagittal section showing inferior type of left sided CFRE with calculi in both kidneys indicated by arrows.

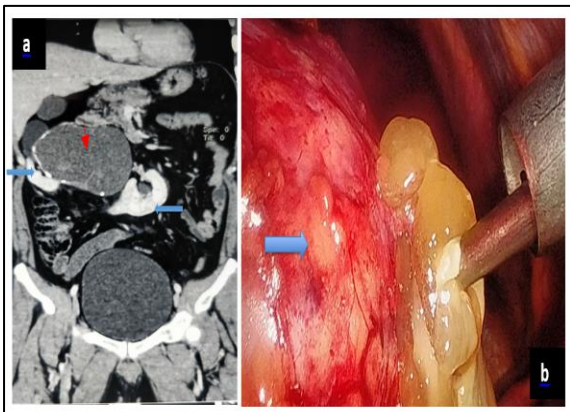


Figure 3: (A) Coronal section showing L-type left sided CFRE with renal cyst; crossed fused kidneys (blue arrows); renal pelvic cyst (red arrow); (B) intra-operative picture showing renal pelvic cyst (blue arrow).

The operative time was 82 minutes. Post-operative period was uneventful and patient was discharged on day 3. X-ray KUB was advised after 2 weeks on follow up which revealed no significant residual fragment and so both stents were removed. At 6 months of follow-up, the patient was asymptomatic with no recurrence detected on ultrasound.

Case 4

A 47 year old gentleman with no comorbidities was referred with diagnosis of left sided CFRE with calculi in both the kidneys. He presented with complaints of dull aching abdominal pain. His physical examination was unremarkable but laboratory investigations showed deranged renal function.

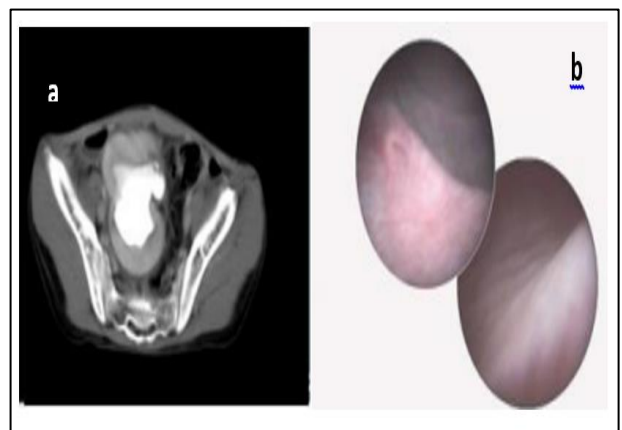


Figure 4: (A) CECT KUB revealed a right to left cake shaped fusion; (B) cystoscopy images showing left hypoplastic ureteric orifice.

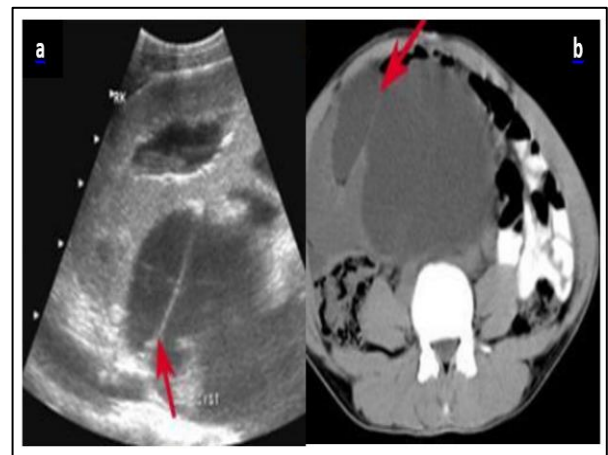


Figure 5: (A and B) USG and CECT showed right to left disc shaped fusion, (R) hydronephrotic kidney fused with (L) grossly hydronephrotic unit.

To plan further management, patient underwent a NCCT which showed left sided crossed-fused renal ectopia with multiple calculi in both kidneys (Figure 2). The laboratory work up showed hyperuricemia,

hyperuricosuria with normal complete blood picture and renal function. Patient was advised for standard simultaneous bilateral prone PCNL. The procedure done was similar to the previous case with operative time of 88 minutes. Post-operative period was uneventful and patient was discharged on day 2. Patient was started on therapy with febuxostat for hyperuricemia. After 3 months of follow-up, the patient was asymptomatic with no recurrence detected by abdominal USG with normal uric acid levels in the urine and blood.

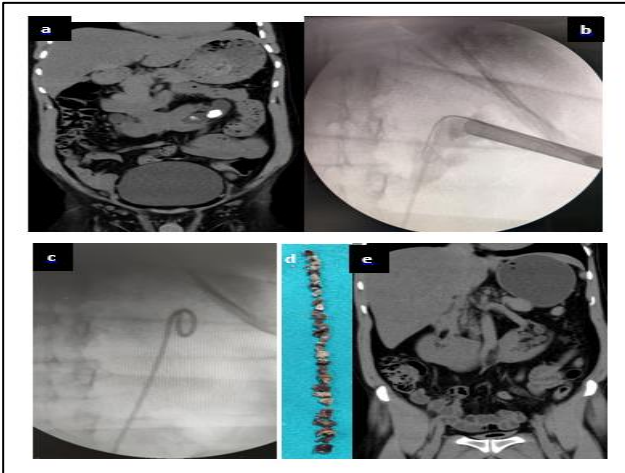


Figure 6 (A and B): NCCT KUB showing multiple left renal calculi; Mini PCNL done with superior calyceal approach; (C-E) complete stone clearance confirmed on fluoroscopy and NCCT KUB.

Case 5

A 46 year old gentleman diagnosed with right sided CFRE with a large renal cyst in the right renal pelvis region measuring 12×11×11 cm on CECT (Figure 3). Patient was referred to our center for further management. Patient had history of dull aching pain in the right sided upper abdomen since three months which had increased over the last month. On examination, fullness was noted in the right upper quadrant of the abdomen and a cystic swelling was palpable. Rest of the examination was normal. Patient was advised for laparoscopic de-roofing of the cyst. Cystoscopic examination was normal. Bilateral RGP was done which showed communication of the cyst with the right pelvicalyceal system and 5-Fr ureteral catheters were placed in both the ureters. De-roofing of cyst was done and all the mucoid-jelly like content was aspirated. Cyst wall and part of the mucoid material was sent for histopathological examination (HPE). Post-operative period was uneventful and patient was discharged on day 5. Cytological examination did not show any atypical cells. Histopathology of the cyst wall showed fibrous tissue with inflammatory changes. On follow up at 1 and 3 months, the patient was asymptomatic with no recurrence detected by abdominal US.

Case 6

A 3 month male child had presented with an antenatal sonogram revealing bilateral hydronephrosis. USG revealed an empty left (L) renal fossa with right (R) hydroureteronephrosis (HUN). IVU showed right (R) low placed malrotated kidney, left (L) hydronephrotic unit with atretic ureter. MCU showed right grade III reflux. On cystoscopy, right gaping orifice and left orifice hypoplasticity was seen. It was a case of left to right CRE with superior fusion anomaly, where the (L) kidney was placed across the pelvic brim and fused with the (R) kidney at the upper pole. He underwent extirpation of the left kidney and ureter. The right grade III VUR was managed conservatively in view of the young age and probability of resolution of reflux as the child attains growth.

Case 7

A 31 year male with a history of recurrent urinary tract infections (UTI) was being evaluated by department of medicine. The USG demonstrated empty right renal fossa with fused kidneys in the left lumbar region with moderate hydronephrosis. IVU/CECT showed bilateral moderate hydroureteronephrosis with 'L' shaped fusion on the left side. He was transferred to the department of Urology and MCU was done which revealed bilateral grade IV reflux. The anomaly here was a right to left L-shaped crossed fusion ectopia. He underwent Cohen cross trigonal ureteric reimplantation.

Case 8

A 17 year male, with recurrent UTI was admitted for evaluation. USG reported fused kidneys in the pelvis with gross hydronephrosis. CECT kidney ureter bladder (KUB) revealed cake shaped fusion anomaly with atretic left ureter. On cystoscopy, left ureteric orifice was hypoplastic and retrograde pyelogram (RGP)/CECT KUB showed (R) solitary ureter crossing to the left side. It was a case of right to left Cake shaped fusion anomaly with solitary crossed ureter. He was treated with modified pyelovesicosotomy with a short bridge of Boari flap.

Case 9

A 40 year man was admitted with dull aching lower abdominal pain, fever and lower abdominal mass. He had impaired renal function with a creatinine of 4.6 mg/dl and total count of 14,500 cells/ μ l. There were no associated anomalies. On USG and NCCT, he was found to have (R) mildly hydronephrotic kidney fused with (L) grossly hydronephrotic unit. He was diagnosed with right to left Disc shaped crossed fusion ectopia with bilateral hydronephrosis (Figure 5 A and B). He underwent bilateral nephrostomy, the tubes were inserted by the radiologist under ultrasound guidance and he was started on intravenous antibiotics. The creatinine improved from

4.6 mg/dl to 1.1 mg/dl over a week with nephrostomy drainage and antibiotics. The renogram was not performed due to financial constraints. Nephrostogram showed bilateral hydronephrosis (left more than right) and bilateral retrograde pyelogram showed positive jet sign with features of bilateral PUJ obstruction. He underwent bilateral pyeloplasty under general anaesthesia. The approach was through the lower midline incision. Intraoperatively, the right kidney was fused to the left unit as a disc shaped fusion with both the pelvis facing anteriorly and hydronephrosis, there was a crossing vessel on the right unit. Hence bilateral pyeloplasty were performed. The decision for right pyeloplasty was taken due to the presence of a crossing vessel, hydronephrosis on administration of diuretic showing tense pelvis and difficulty in access to the unit at a later date for a repeat surgery due to complex anatomy.

Case 10

A lady in her late 50s presented with right lower abdominal pain. USG showed right hydronephrosis with absent left kidney. CECT KUB reported a left to right L-shaped crossed fusion ectopia with a right renal pelvic calculi measuring 2 cm with (R) unit hydronephrosis. The stone was removed by laparoscopy assisted percutaneous nephrolithotomy (PCNL).

Case 11

A 34 year primigravida at 30 weeks period of gestation presented with left loin pain. On USG abdomen, absent right kidney in the renal fossa with left kidney showing gross hydronephrosis and a gravid uterus with single live fetus of 30 weeks of gestation with no anomalies was visualized. Her serum creatinine measured 1.2 mg/dl and urea 36 mg/dl. She had proteinuria of 2 grams/day. MRI showed right to left CRE with disc shaped fusion and gross hydronephrosis of the right renal unit with a gravid uterus and a single live foetus. On cystoscopy there was a right hemitrigone with hypoplastic left ureteric orifice. Under local anaesthesia with USG guidance, right double 'J' (DJ) stenting was done. Her renal parameters improved and she delivered a 2.6 kilogram healthy baby at 37 weeks of gestation. She has been planned for right Anderson Hynes dismembered pyeloplasty.

Case 12

A 38 year old gentleman with no comorbidities was referred with diagnosis of left sided CFRE with calculi in left lower calyx and uretero-pelvic junction. He presented with complaints of dull aching abdominal pain. His physical examination was unremarkable and laboratory investigations were within normal limits. To plan further management, patient underwent a NCCT showed left sided crossed-fused renal ectopia with multiple calculi in left kidney. Left sided mini PCNL was performed with superior calyceal puncture and complete stone clearance

was confirmed on fluoroscopy. The total operative time was 58 minutes. Post operative period was uneventful and patient was discharged on day 2 (Figure 6 A-E).

DISCUSSION

Types of CFRE and fusion anomalies

CFRE is second only to horseshoe kidney in terms of fusion anomaly with incidence of 1:1000 in live births and 1:7500 on autopsy studies. In spite of both kidneys being on the same side, the ureter of the ectopic kidney descends and crosses the midline and enters the urinary bladder at its normal position. The ectopic kidney is mostly located inferior to the normal kidney with fusion. CFRE is more common in males and left to right shift with an empty left renal fossa. The postulated pathogenesis states that at four to eight weeks of gestation, there is aberrant migration and crossing of the metanephric blastema and the ureteral bud to the opposite side, thereby both the kidneys lie on the same side, but the ureteric bud which arises from the metanephric duct arises normally. In literature, six types have been described as inferior ectopia (type 1; most common), superior ectopia (type 2), sigmoid or S-shaped (type 3), lump kidney, also called a pancake kidney (type 4), L-shaped (type 5) and disk shape (type 6). A much rarer type of crossed-fused renal ectopia which is the "Y" type ectopia was described where the ureters of the ectopic and normal kidneys are fused and draining into the bladder as a common channel.⁶⁻¹⁰ In our case series, 10 patients were males and 2 were females but contradictory to the current literature, we had seven cases (58.3%) of left sided CFRE with L-shaped fusion or the tandem fusion being the most common anomaly seen in 8 cases (66.6 %).

Imaging modalities for diagnosis

Various imaging modalities have been described for the diagnosis of this condition. US can reveal the absent kidney but CECT scan can better identify renal vessels which will help in achieving a safe access during surgery, identification of any anomalies present in the collecting system and the ureters and also determine the exact anatomy and relations of the ectopic kidney before an intervention if required. Doppler US can also be used to delineate abnormal vasculature.¹¹ Most cases of crossed renal ectopia are asymptomatic and diagnosed incidentally while being evaluated for associated conditions such as renal and ureteric stones, pyelonephritis, vesico-ureteric reflux (VUR), cysts, UPJO or malignancy with renal cell carcinoma being the most common. Such cases may present with vague abdominal or flank pain, dysuria, haematuria and very rarely a palpable mass. Ectopic ureteric orifices are seen in less than 3% cases.¹²⁻¹⁵

Endourological procedures for upper tract stone management in CFRE

Less than 50 cases have been reported on CFRE with stones and none with surgical intervention for a large renal cyst. There is no clear consensus or guidelines on management of the same. The treatment options for these renal stones included conservative therapy, extracorporeal shock wave lithotripsy (ESWL), PCNL, retrograde intrarenal surgery (RIRS) and laparoscopic pyelolithotomy or open surgery. The choice of treatment of renal calculi in such cases depend on multiple factors such as stone size and vascular anatomy. PCNL is a good option in case of large stone burden but it is associated risks of bleeding and injury to the gastrointestinal tract due to the abnormal vascular anatomy of the ectopic kidney.^{16,18} In our case series, two patients underwent standard simultaneous bilateral prone standard PCNL with double J stent placement after complete stone clearance for bilateral renal calculi. Mini PCNL was performed for one patient with left side renal calculus while one patient underwent URSL for distal ureteric calculus. Double J stenting for bilateral acute pyelonephritis and urosepsis was performed for one patient while unilateral DJ stenting was performed for the pregnant female with VUJ calculus.

CFRE and associated conditions

A high incidence of other urological abnormalities has been associated with renal ectopy. Vesicoureteral reflux (VUR) is the most common associated abnormality followed by other conditions such as aberrations such as renal multicystic dysplasia, hypospadias, cryptorchidism and hydronephrosis. Mullerian duct anomalies have also been reported in females with CFRE.¹⁹ The incidence of PUJO in horseshoe and ectopic kidneys ranges from 25% to 33% and from 22% to 37%, respectively. No data are available in the literature regarding the incidence of PUJO in fusion anomalies other than horseshoe kidneys.²⁰ In our series we encountered one patient with right to left CFRE with disc/doughnut fusion anomaly with bilateral UPJO. Open bilateral Modified Anderson Hyne's pyeloplasty was performed.

Guarino et al demonstrated that VUR occurred in 20% of crossed renal ectopy, 30% of simple renal ectopy, and 70% of bilateral simple renal ectopy cases.¹⁹ In our series, the reported patient was an adult with bilateral grade 4 VUR presenting with recurrent UTI. We predicted little chance of spontaneous resolution because of the complicated VUR associated with crossed fused kidney. Cohen's cross trigonal ureteric re-implantation was performed as ureteral length was adequate. Other options as per literature are Politano-Leadbetter technique and Paquin technique. The former is a better option for short ureters and can be performed under direct vision, thus reducing the risk of peritoneal injury. One patient was diagnosed to have right sided grade 3 VUR with left non-functioning kidney with atretic ureter who underwent left

open nephrouretrectomy. Very limited literature is available on the associated conditions like unilateral hypoplastic/atretic ureter with CFRE. Open short bridge Boari flap was performed for one such patient with right to left cake/ lump fusion anomaly with bilateral mild hydronephrosis and left atretic ureter with hypoplastic ureteric orifice.

CONCLUSION

It is important for a practicing urologist to bear in mind the various presentations of this less known anomaly of renal fusion. We have highlighted the role of different imaging modalities for diagnosis of various associated conditions with cross-fused renal ectopia This is important to plan further treatment and surgical approach in such patient in view of complex renal as well as vascular anatomy. There are no specific guidelines available for the management of various manifestations of crossed fused kidney ectopia and so each case should be individualized as per the surgeon expertise and patient preference.

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