

Crossed Fused Renal Ectopia with Separate Collecting Systems and Functional Obstruction Treated with a Novel Common Sheath Pyeloplasty: A Case Report

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ABSTRACT

Background: Cross-fused renal ectopia (CFRE) is a rare congenital anomaly involving abnormal renal fusion and positioning, with an estimated occurrence of 1 in 1000 live births. Most patients remain asymptomatic, but complications such as hydronephrosis, urinary tract infection, or pelvi-ureteric junction obstruction (PUJO) can develop. Functional obstruction without mechanical narrowing is exceptionally uncommon. We report a case of a 2-year-old boy with CFRE and functional PUJO who was managed with a novel dismembered common sheath pyeloplasty, with a satisfactory outcome. **Case Presentation:** We report a 2-year-old male who presented with recurrent fever, progressive abdominal swelling, and abdominal pain since birth. Antenatal scans revealed an ectopic kidney. Examination showed a central cystic abdominal mass and a non-palpable right testis. Ultrasonography demonstrated bilateral ectopic pelvic kidneys with marked right hydronephrosis. Computerised Tomographic (CT) urography revealed cross-fused renal ectopia draining into a massively dilated renal pelvis with the "fountain sign." Laboratory investigations were normal. The patient underwent a novel common sheath dismembered pyeloplasty and right orchidopexy. The patient had an uneventful recovery. The drain and catheter were removed on postoperative days 8 and 10, respectively. He was subsequently discharged on postoperative day 12 and remains asymptomatic on follow-up. **Conclusion:** Cross-fused renal ectopia can rarely present with hydronephrosis due to functional obstruction. Advanced imaging is essential for accurate diagnosis. Dismembered common sheath pyeloplasty remains a successful reconstructive option of the anomalous systems of cross renal ectopia, representing a novel modification of the procedure described by Anderson and Hynes.

Keywords: Cross-fused renal ectopia, Separate system, Functional obstruction, Hydronephrosis, Pyeloplasty

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INTRODUCTION

Renal fusion anomalies represent a small portion (<10%) of congenital renal malformations, with cross-fused renal ectopia (CFRE) being the second most common form, after horseshoe kidney. [1,2] Among the four categories of cross renal ectopia (CRE), CFRE is the most frequently observed (90%). The other categories, in order of increasing rarity, include CRE without fusion, solitary CRE, and unfused bilateral CRE [3]. The incidence of CFRE occurs in approximately 1 in 1000 live births, with a male-to-female ratio of 2:1 and a left-to-right predominance [3,4]. Dominicus Panarolus first described the anomaly in 1654 [1]. It is a developmental defect that results from the abnormal ascent of the kidneys, with fusion of both kidneys within the pelvis. The two main theories suggest either an abnormal positioning of the umbilical arteries, impeding renal ascent, or the presence of two ureteric buds leading to malrotation and fusion [3,5].

Cross-fused renal ectopia involves one kidney traversing the midline to fuse with the orthotopic kidney. At the same time, its ureter crosses backward to drain into the bladder via its original opening into the trigone. [6] The crossed fused renal ectopia with a single ureter is a scarce variant that has been infrequently reported. [7,8] Most cases are asymptomatic and found incidentally during imaging. Still, some patients present with abdominal swelling, pain, recurrent urinary tract infections, or pelvi-ureteric junction obstruction (PUJO). [8,9] Intervention is necessary in symptomatic cases, and treatment should be tailored to the specific complication. [4] We report a rare case of left-to-right cross-fused renal ectopia with separate collecting systems, functional PUJO, and a single right ureter, treated with a novel common sheath dismembered pyeloplasty.

CASE REPORT

A 2-year-old male presented with recurrent fever, progressive abdominal swelling, and abdominal pain since birth. Pregnancy and delivery were uneventful,

but antenatal ultrasound revealed an ectopic kidney. He was the fourth child in a monogamous family with no family history of similar illness. General physical examination was grossly normal. There was gross asymmetrical abdominal distension, with a ballotable non-tender right flank mass (Figure 2A). The phallus was uncircumcised with a normal meatus. The left testis was normal, but the right was non-palpable. Haemogram, urine culture, serum electrolytes, urea, creatinine, serology, and clotting profile were all normal. Haemoglobin genotype was AA. Abdominal ultrasound revealed a right ectopic (pelvic) kidney with (SFU) grade 4 hydronephrosis. The left renal fossa was empty and non-palpable testis was not visualised. The CT urography revealed a fused, malrotated pelvic kidney at L4-S1 with a separate collecting system draining into a massively dilated renal pelvis (13.6×12×10.1 cm) with delayed contrast excretion and the characteristic "fountain sign" (Figure 1 A&B). The fused ectopic kidney was drained by a single ureter to the right side of the urinary bladder (Figure 1C).

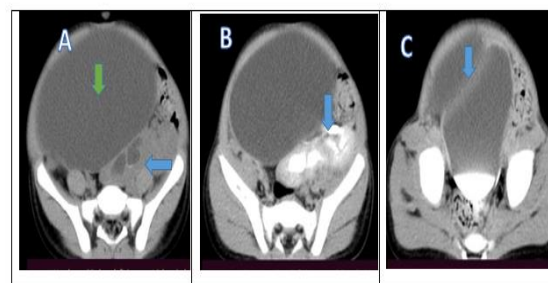


Figure 1 (CT Urogram): A (non-contrast); hydronephrotic fused pelvic kidneys (blue arrow) with a huge common renal pelvis (green arrow). **B (contrast);** Independent collecting systems of the right CFRE draining into the huge renal pelvis demonstrating the 'fountain' sign (blue arrow). **C (contrast);** shows compression of the right superior part of the urinary bladder by huge renal pelvis (severe hydronephrosis) (blue arrow)

A diagnosis of left to right cross-fused renal ectopia with pelvi-ureteric junction obstruction and right undescended (non-palpable) testis was made. He

underwent a dismembered common sheath pyeloplasty via Gibson's incision and right orchidopexy by the subdartous pouch method. Intraoperative findings included left-to-right cross-fused renal ectopia, right and left collecting systems draining separately into a large pelvis, and a single patent ureter draining into the right side of the urinary bladder (Figures 2B-E). The PUJ was found to be patent, with no form of mechanical narrowing observed. There was a right inguinal testis (Figure 2F). The procedure was novel and a modification of the technique described by Anderson and Hynes, as in our case, the 2 systems, representing each of the fused kidneys, were connected together and to the common renal pelvis drained by a single patent right ureter. The huge renal pelvis was resized, and the proximal 5 cm of the ureter including the PUJ was excised to cater for the redundancy. The anastomosis of the two collecting systems was performed side-to-side, followed by end-to-end anastomosis of the renal pelvis to the ureter. The completed reconstruction is shown in Figure 2F. Two improvised internalized stents via each kidney (fenestrated size 5 NG-tubes passed percutaneously) and a retroperitoneal drain were placed. The drain dislodged spontaneously on day 8, and the stents, along with the urethral catheter, was removed on day 10. The child was discharged home on day 12 after an uneventful recovery and postoperative period. Histopathological finding of the PUJ segment include bands of extensive fibrocollagenous tissues with few bundles of muscles. The patient has been on regular follow-up without complaint.

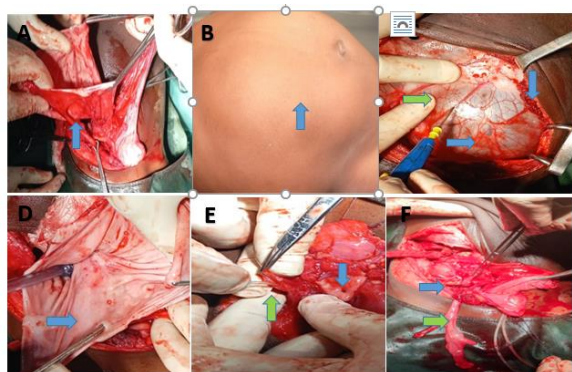


Figure 2 (Intraoperative): (A) Pre-operative photograph showing grossly distended abdomen with ballotable right flank mass (blue arrow); (B) Huge renal hydronephrotic renal pelvis (blue arrow); (C) Openings of the 2 independent systems (blue arrows) of CFRE and the pelvis ureteric junction (green arrow); (D) Single ureter (blue arrow) draining the huge renal pelvis into the urinary bladder; (E) The 2 independent collecting systems after spatulation (green and blue) of the cross fused ectopia before common sheath pyeloplasty into the single ureter; (F) Showed the completed common sheath dismembered pyeloplasty (blue arrow) and right undescended testis (green arrow).

DISCUSSION

Crossed fused renal ectopia is an uncommon birth defect in which the two kidneys are joined, resulting in a single renal mass often with two collecting systems on one side of the abdomen. However, the ureter of the 'translocated' kidney crosses the midline again to insert into the bladder through its original ureteric orifice. [6] During fetal development, between the 4th and 8th weeks, the kidneys normally ascend to the level of the L2 vertebra in the retroperitoneal space. Crossed fused renal ectopia results from abnormal renal ascent combined with fusion of both kidneys at the pelvis. Two main theories have been proposed to explain the pathogenesis of CFRE. Left-to-right fusion occurs three times more often than right-to-left, and in most cases, two separate ureters descend to the bladder, although variants with a single ureter have also been reported. [7,8,10,11] Our patient had the more common left-to-right fusion, which explained the right-sided abdominal swelling observed at presentation. In 1957, McDonald and McClellan classified crossed fused renal ectopia into six subtypes (I-VI) based on the orientation and position of the renal fusion and ureteral insertion into the bladder. [2] Type III, known as the lump kidney, involves broad fusion of both kidneys with the ureter of the ectopic kidney crossing the midline. The current case conforms to this Type III classification and features a single ureter drainage system. Habtemichael et al. [12] reported the pancake (Type

III) form of CFRE with a single ureter in a 17-year-old male in 2021.

Most cases of crossed renal ectopias are asymptomatic and often discovered incidentally. [9] However, symptomatic presentations frequently occur with accompanying urinary tract complications such as hydronephrosis, ureteropelvic junction obstruction, vesicoureteral reflux, recurrent infections, and urolithiasis. These conditions can cause symptoms like flank pain, abdominal mass, recurrent fever, hematuria, among others. [6] Our patient presented late at the age of 2 years with progressive abdominal swelling and recurrent infection due to a functional PUJO. This contrasts with findings from a previous study by Loganathan and Bal. [4] where most of their 36 patients presented within 1 year of age because of recurrent infection, abdominal pain from pelvi-ureteric or vesicoureteric junction obstruction, and reflux. However, other authors such as Kaur et al. [7], Dahal et al. [8] and Naseri [13] have reported the conservative management of much older patients at ages 22, 46, and 6 years with CFRE and a single ureter, presenting with lithuria and reflux, respectively. The late presentation (at 2 years) of our patient, despite being symptomatic for several months, may be related to limited access to specialized urological care, the necessity of out-of-pocket payment despite financial constraints, and the absence of comprehensive health insurance coverage in our society.

Other system anomalies that may be associated with CFRE include genital (penile duplication, undescended testis, vaginal agenesis), GIT (imperforate anus, gut malrotation), cardiovascular (septal defects, aortic coarctation), tethered cord, and limb abnormalities, among others. [3,14] Besides the rare duplex-drainage system and a single ureter, our patient also presented with an undescended testis. Similarly, Drevna et al. [15] and Yadavalli et al. [16] have reported instances of CFRE simultaneously associated with ipsilateral cryptorchidism in a 30-year-old and a 10-year-old male, respectively. Imaging plays a vital role in diagnosis, monitoring,

preoperative planning, and follow-up. Ultrasonography remains an essential initial modality for detecting the location of fused ectopic kidneys and the presence of hydronephrosis. [3] Advanced three-dimensional imaging such as CT and MR urography are crucial for defining urinary tract anatomy, vascular supply, presence of obstruction, and associated anomalies. [9,17] In our case, CT urography was used to establish the diagnosis, as it demonstrated the radiological features of the left-to-right cross-fused renal ectopia, including the classic “fountain sign.” [17] While asymptomatic cases are observed by periodic monitoring, symptomatic cases require intervention tailored to the presenting complication. Functional obstruction of an ectopic kidney is an uncommon but recognized phenomenon that can result from an abnormal calyceal orientation or impaired peristalsis. The finding of independent duplex systems with functional obstruction in our patient, evidenced by a massively dilated renal pelvis without intrinsic PUJ narrowing or external compression, is extremely rare. [18] This warranted a novel procedure that incorporated the two collecting systems into a single channel before anastomosis with the dilated pelvis. Although most procedures previously performed for CFRE were due to stone, a few, such as those by Goyal et al. [20], have involved pyeloplasty. [14,21] Anderson-Hynes dismembered pyeloplasty is the surgical intervention of choice for PUJO in an anomalous kidney, but this procedure was originally described in a single collecting system kidney. [21,22] Our novel technique combines the separate systems into a single channel through side-to-side anastomosis, forming a neo-renal pelvis before connecting it to the single ureter as part of a common sheath pyeloplasty. This modification of the original Anderson-Hynes dismembered pyeloplasty, which we found to be effective and rewarding, is presented here.

CONCLUSION

Cross-fused renal ectopia with a duplex collecting system is a rare anomaly that can present with

hydronephrosis caused by functional pelvi-ureteric junction obstruction. Diagnosis can be difficult and often requires advanced imaging. Modified Anderson–Hynes dismembered common sheath pyeloplasty, even in cases with complex separate anatomy, remains an effective surgical option with excellent outcomes.

Study Limitations: Radionuclide study was not done as it was not available in our facility, and the parents could not afford it, where it was available.

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Informed Consent: Patient's parent has consented to this publication

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