INTRODUCTION

A horseshoe kidney (HSK) is the most common congenital kidney anomaly with an incidence of 1 in 500 [1]. An HSK is a subtype of the fused kidney, e.g., L-shaped kidney, sigmoid kidney, or pancake kidney. These kidney anomalies show fusion between the right and left kidneys. Crossed fused renal ectopia (CFRE) is a congenital ectopia of one kidney to the contralateral side with fusion to the kidney on the same side. The origin and course of the renal vessels are variable depending on the shape and size of the CFRE as well as the HSK. Knowledge of the renal vessels surrounding the kidney anomalies helps us to better understand possible complications of the surgeries in the region they are located.

CASE REPORT

During routine dissection of the abdominal cavity, an artery was found arising from the right common iliac artery in a male, Caucasian fresh-frozen cadaver whose age at death was 85 years. The artery was traced distally and found to be the renal artery, which traveled to a kidney displaced in the pelvis. Further dissection of this kidney revealed that this was the right side of the fused kidney. This fused kidney was slightly

Summary: Herein we present a case of a horseshoe kidney with crossed fused renal ectopia. Both of these pathologies are congenital anomalies; however, to date, there are few cases that present with both. In this case, discovered during routine dissection, the fused kidney was mostly left-sided and very low in the pelvis. No renal artery arose from the right wall of the abdominal aorta, and the right renal vein drained into the lower part of the inferior vena cava (IVC) where the right and left common iliac veins joined. It is essential for clinicians and surgeons to understand these types of congenital anomalies, as they could impact patient care.

Key words Ectopic kidney, inferior vena cava, horseshoe kidney, cross fused renal ectopia, renal artery, cadaver, anatomy
curved with deviation to the left side, which is defined as CFRE (Figure 1). The CFRE was positioned between the third lumbar vertebra and upper part of the sacrum level so that the right kidney was in the pelvis. The CFRE was then resected from the abdominal cavity with related structures to be fixed in the formalin (Figure 2). The right kidney was located in the midline and anterior to the right and left common iliac arteries. Both hila faced anteriorly, and the left renal pelvis was slightly enlarged. The right renal hilum received two right renal arteries, which arose from the anterior aspect of the abdominal aorta 15 mm inferior to the inferior mesenteric artery and the anterior wall of the right common iliac artery, respectively. The left renal hilum was supplied by only one renal artery that arose from the lateral wall of the abdominal aorta. No renal artery was branched off the right wall of the abdominal aorta. Two renal veins drained from the right renal hilum. The one drained into the lower part of the IVC where the right and left common iliac veins joined and the other drained into the left wall of the IVC 25 mm inferior to the left renal vein. The left renal vein received the left testicular vein, ran in front of the abdominal aorta below the superior mesenteric artery and drained into the inferior vena cava (IVC). No renal vein drained into the right wall of the IVC.

**Fig. 1.** Ex vivo image of the pelvic crossed fused renal ectopic kidney reported herein. AA, abdominal aorta; CIA, common iliac artery; IMA, inferior mesenteric artery; IVC, inferior vena cava; RA, renal artery; RV, renal vein; TV, testicular vein; U, ureter.

**Fig. 2.** Anterior view of the embalmed crossed fused renal ectopia and adjacent structures with formalin. before (a) and after (b) the abdominal aorta and arteries retracted to the left. AA, abdominal aorta; CIA, common iliac artery; CIV, common iliac vein; IVC, inferior vena cava; RA, renal artery; RV, renal vein; SMA, superior mesenteric artery; TA, testicular artery; TV, testicular vein; U, ureter.
DISCUSSION

CFRE is a relatively uncommon congenital kidney anomaly with an incidence at autopsy of 1 in 2000 [1,2]. According to Liu et al., CFRE occurs up to two times more often in men than in women, and more often on the right side [3]. In the present case, the fused kidney was mostly located on the left side, and the right kidney was on the midline. To our knowledge this is a unique finding. Also, no renal artery arose from the right wall of the abdominal aorta, which is why we diagnosed this kidney as CFRE. CFRE is the second most common fusion anomaly of the kidney. There are six unique types of CFRE that have been identified to date: inferior crossed fused ectopia, sigmoid or S-shaped kidney, lump kidney, disc kidney, L-shaped kidney, and superior crossed fused ectopia [3]. The present case was the inferior crossed fused ectopia. Surgeons should be aware of this type of kidney anomaly as it may affect the position and course of the ureter, renal vessels, and kidneys themselves [4,5]. These anomalies can be diagnosed using computed tomography and magnetic resonance images, but may also could be overlooked if knowledge of variants is lacking. Patients presenting with CFRE are often asymptomatic, but they sometimes report the following varied symptoms: flank pain, urolithiasis, abdominal pain, blood in the urine, no creation of urine, frequent urinary tract infections, renal failure, fever, hydronephrosis, and high blood pressure [3]. Also, a CFRE could be misdiagnosed as an abdominal mass [6].

Embryologically, the kidney begins developing in the pelvis during the fourth week of gestation, followed by ascension at week seven—with internal rotation over the iliac crest—and at week nine it obtains its final position [7]. In patients with CFRE, there is a lack of fusion and lack of complete rotation. It is important to note that CFRE may lead to fistula formation between the collecting ducts and the lymphatics and can present as milky-colored urine [3].

The blood supply to the kidneys appears to be abnormal in all known case reports. Of the 22 case reports that Hertz reviewed in 1977, each had a unique blood supply, and no consistent arterial pattern could be discerned [8]. From his studies, Hertz noted that renal arteries were branching from the abdominal aorta as high as the T12 level and as low as the L3-4 level [8]. He also discovered in some cases that blood supply to the kidney could also be coming from the iliac arteries. Hertz showed that even the arteries supplying the non-ectopic kidney arose from abnormal sites [8]. Malek et al. showed that 90% of pediatric patients with CFRE are highly likely to have other anomalies in organ systems outside of the genitourinary system (18/20 patients) [9]. However, in adults anomalies outside of the genitourinary system were approximately 23% [10].

Most case reports of CFRE were found incidentally. Treatment is only indicated when symptoms in the upper urinary tract are significant—most frequently these include nephrolithiasis and vesicoureteral reflux [11]. If an ectopic kidney is found to be non-functioning, then surgical excision may be necessary [12]. Surgery may also be necessary due to the potential of an entrapped ureter, for which the kidney’s isthmus requires division [12]. In cases that require surgical management, ureteric reimplantation can be performed using the cross trigonal Cohen’s technique [12]. However, to date, there are no specific guidelines for the management of CFRE. It is important for clinicians to know that the fused kidneys do not need to be separated unless symptoms are present [11]. CFRE can occasionally be palpated as abnormal abdominal masses [7]. Surgical procedures for kidney anomaly such as CFRE may lead to complications because of the complex vascular system [2]. Additionally, there have been case reports where complete nephrectomy led to surgical complications [9].

CONCLUSION

An HSK with a CFRE is a rare anomaly, which can be clinically significant. As clinicians, it is essential to understand variations of the kidneys and their symptomologies [13-18]. Additionally, such variations may be relevant to surgeons when planning lateral surgeries such as the extreme lateral interbody fusion of the lumbar spine due to variations in the vasculature of a CFRE. Appropriate sonography and comprehensive imaging should be completed on patients who have CFRE anomalies to determine if any interventions or modifications of surgical procedures need to take place.

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REFERENCES

1. Baggenstoss AH. Congenital anomalies of the kidney. Med Clin North Am 1951; 1:987-1004.
2. Iwanaga J, Watanabe K, Saga T, Tahara N, Tabira Y et al. Anatomical and Radiological Analyses of L-shaped Kidney with Vascular Anomalies. Kurume Med J 2018; 64(1.2):21-24.
3. Liu DY, Wang HF, Xia WM, He HC, and Shen ZJ. Right-Crossed, Fused Renal Ectopia L-Shaped Kidney Type with Urinary Chyluria. Urol Int 2015; 95(2):243-245.
4. Iwanaga J, Yilmaz E, Tawfik T, Abdul-Jabbar A, Vetter M et al. Anatomical Study of the Extreme Lateral Transpsoas Lumbar Interbody Fusion with Application to Minimizing Injury to the Kidney. Cureus 2018; 10(1):e2123.
5. Canova G, Masini R, Santoro E, Bartolomeo S, Martini C et al. Surgical treatment of abdominal aortic aneurysm in association with horseshoe kidney. Three case reports and a review of technique. Tex Heart Inst J 1998; 25(3):206-210.
6. Bergman RA, Tubbs RS, Shoja MM, and Loukas M. Bergman’s comprehensive encyclopedia of human anatomie variation. Hoboken, New Jersey: John Wiley & Sons, Inc.; 2016, 1432 pages.
7. Natsis K, Piagkou M, Skotsimara A, Protogerou V, Tsitouridis I et al. Horseshoe kidney: a review of anatomy and pathology. Surg Radiol Anat 2014; 36(6):517-526.
8. Hertz M, Rubinstein ZJ, Shahin N, and Melzer M. Crossed renal ectopia: clinical and radiological findings in 22 cases. Clin Radiol 1977; 28(3):339-344.
9. Malek RS, Kelalis PP, and Burke EC. Ectopic kidney in children and frequency of association with other malformations. Mayo Clin Proc 1971; 46(7):461-467.
10. Gledhry B, Petersen J, Hofmann KJ, Schenk C, Herwig R et al. Kidney fusion anomalies revisited: clinical and radiological analysis of 209 cases of crossed fused ectopia and horseshoe kidney. BJU Int 2009; 103(2):224-235.
11. Bhatt K and Herts BR. Crossed fused renal ectopia. J Urol 2014; 191(2):475-476.
12. Solanki S, Bhatnagar V, Gupta AK, and Kumar R. Crossed fused renal ectopia: Challenges in diagnosis and management. J Indian Assoc Pediatr Surg 2013; 18(1):7-10.
13. Granger A, Zurada A, Zurada-Zielinska A, Gielecki J, and Loukas M. Anatomy of turner syndrome. Clin Anat 2016; 29(5):638-642.
14. Subramaniam H, Taghavi K, and Mirjalili SA. A reappraisal of adult thoracic and abdominal surface anatomy via CT scan in Chinese population. Clin Anat 2016; 29(2):165-174.
15. Shen XH, Su BY, Liu JJ, Zhang GM, Xue HD et al. A reappraisal of adult thoracic and abdominal surface anatomy via CT scan in Chinese population. Clin Anat 2016; 29(2):165-174.
16. Bouzada J, Vázquez T, Duran M, Delmas V, Larkin T et al. New insights into the morphogenesis of the gubernaculum testis and the inguinal canal. Clin Anat 2017; 30(5):599-607.
17. Bilal M, Voin V, Topale N, Iwanaga J, Loukas M et al. The Clinical anatomy of the physical examination of the abdomen: A comprehensive review. Clin Anat 2017; 30(3):352-356.
18. Pak N, Patel SG, Hashemi Taheri AP, Hashemi F, Eftekhari Vaghefi R et al. A reappraisal of adult thoracic and abdominal surface anatomy in Iranians in vivo using computed tomography. Clin Anat 2016; 29(2):191-196.