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Case Report

Crossed fused ectopic kidney in a patient with irritable bowel syndrome with diarrhea: a case report

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ABSTRACT

Irritable bowel syndrome (IBS) is the most commonly diagnosed gastrointestinal condition that reduces patients' quality of life. It has multifactorial etiology. Nowadays it is thought that more than one etiologic factor may contribute to heterogeneous symptoms of IBS. Crossed fused ectopic kidney is a condition where both kidneys are situated on one side and fused and drain bilaterally into the urinary bladder. Most often, it is discovered by chance when evaluating another illness. A drum-shaped bulge was discovered in the descending colon of an 18-year-old girl during the examination of a protracted period of abnormal bowel habits in the form of diarrhea, with normal overlying mucosa. On additional examination, an abdominal CT scan revealed that she had a fused left renal ectopia and an empty right renal fossa. In most of the crossed fused renal ectopia (CFRE) cases patient remains asymptomatic; in a few cases it is associated with renal agenesis, vascular malformation, incontinence, a palpable abdominal mass, urinary tract infection, high incidence of stone formation, and genital anomalies. In this case report, we emphasize on crossed fused kidney causes an external compression to the intestinal wall which may create increased visceral hypersensitivity resulting in irritable bowel syndrome with diarrhea (IBS-D) type symptoms.

Keywords: IBS, CFRE, IBS-D

INTRODUCTION

Irritable bowel syndrome (IBS) is an important disease entity in gastroenterology due to its high prevalence, substantial morbidity, and enormous cost.[1] 7-16% of adults have symptoms compatible with IBS in the USA.[2] Altered motility, visceral hypersensitivity, abnormal gas handling, low-grade inflammation, immune activation, abnormal 5HT metabolism, and psychological and genetic factor-all may contribute to IBS as single or in combination. Among all identifiable congenital anomalies in adulthood 10-12% account for congenital renal anomalies that include abnormal number, positional anomalies, shape and fusion, and urinary tract anomalies.[3] Renal ectopia means a kidney is placed outside the normal renal fossa. Crossed ectopic kidney presents when the kidney is located across the midline with its contralateral kidney; it may be fused or unfused.

CASE REPORT

An 18 year-old normotensive, nondiabetic female presented to us with the complaint of altered bowel habit in the form of diarrhea for the last 2 years without any history of the passage of blood or mucous. There was no history of abdominal pain, urinary problem or weight loss. Family history was unremarkable. Physical examinations revealed nothing significant. Routine laboratory studies including renal function tests did not document anything. For evaluation of her bowel complaint a colonoscopy was done which revealed a

drum-shaped swelling in the descending colon with overlying normal mucosa. The vascular pattern was normal. Findings were consistent with external colonic compression. For further evaluation abdominal USG was done which showed that the right renal bed was empty and the right kidney was fused with the lower pole of the left kidney. Impression was crossed fused ectopic kidney. To confirm the diagnosis a CT scan of the whole abdomen with contrast was done that revealed the right renal bed was empty and the right kidney was fused with lower pole of the left kidney. The right kidney was malrotated with its hilum directed anterolaterally. The inferior part of the right kidney was anterior to the abdominal aorta and common iliac arteries. The left ureter was inserted into the left side and the right ureter was inserted into the right side of urinary bladder. Both kidneys showed normal excretion of contrast media. Both ureters were not dilated. After IV contrast no abnormal enhancement was noted. There were no other congenital anomalies seen in the CT scan. Impression was crossed fused ectopic kidney. Patient developed an IBS-D type symptom due to external compression to colonic wall from crossed fused ectopic kidneys. She was managed accordingly and planned for regular follow-ups.

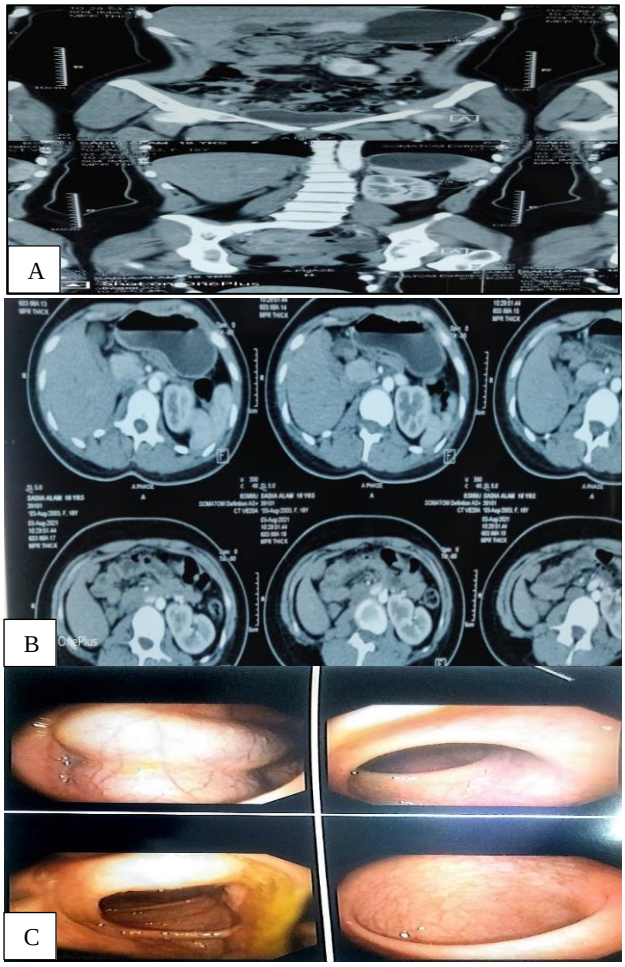

Figure 1: CECT of crossed fused ectopic kidney (A,B), and colonoscopy of a drum-shaped swelling in descending colon with overlying normal mucosa (C).

DISCUSSION

Irritable bowel syndrome (IBS) is characterized by the presence of abdominal pain associated with disturbed defecation.[4] The prevalence of IBS varies anywhere from 1-45% worldwide.[2] People under 50 years have a higher prevalence of IBS in the community.[2] It is more common in women. Researchers are trying to explore the underlying organic pathophysiology rather than labeling it as a functional disease. In recent research, there has been a growing scientific interest in studying the role of visceral hypersensitivity which refers to an increased intestinal perception, whereby otherwise physiologic stimuli are perceived as discomfort and pain.[5] It is associated with the upregulation and sensitization of transient receptor potential cation channels (TRPV) in the peripheral sensory neuron.[6] Several pieces of research have demonstrated that increased TRPV1 nerve fibers are observed in IBS and may contribute to the development of visceral hypersensitivity and pain in IBS -D patients.[7,8] The working hypotheses include altered expression of microRNA in gastrointestinal tissue, increased intestinal permeability that causes a visceral nociceptive drive, and nociceptive input from the colon that causes hypersensitivity. Crossed fused renal ectopia secures the second most common fusion abnormalities after horseshoe kidney among urinary tract fusion anomalies in which one kidney crosses the midline and fuses with the other kidney during development. During the 4^{th}-8^{th} weeks of gestational life, abnormal renal ascent with fusion kidneys in the pelvis causes crossed fused renal ectopia (CFRE). CFRE is divided into four main categories: 1. Crossed renal ectopia with fusion; 2. Crossed renal ectopia without fusion; 3. Unilateral crossed renal ectopia (with unilateral renal agenesis); and 4. Bilateral crossed renal ectopia (without fusion).[9] Around 90% of crossed renal ectopic cases, the kidneys are partially or completely fused, that's why it is named after CFRE. It is more common in men; the male-female ratio is 3:2.[10] Left-to-right ectopia is three times more common than right-to-left ectopia.[11] There are six anatomical variations of CFRE eg: inferior CFRE, sigmoid kidney, lump kidney, L- shaped kidney, disc kidney, and superior CFRE.[12] The variants are more often inferior to CFRE. Here, the upper pole of the crossed ectopic kidney in an inferior position is fused to the lower pole of the kidney in a superior position. The patient in our situation has CFRE of the inferior form. In most cases, it is asymptomatic and found during radiological evaluation for other diseases. USG and contrast CT scans are preferred modalities for the diagnosis of CFRE. In USG, there may be a characteristic "notch" between two fused kidneys.[13] Often hydronephrosis, recurrent UTI, and stone formation complicate CFRE. There are some case reports of renal cell carcinoma and Wilms tumor-associated CFRE.[14,15] In our patient CFRE creates external compression to the intestinal wall and may cause increased visceral hypersensitivity to result in IBS- D type symptoms which were managed accordingly and planned for further follow-up. To our best knowledge, it

is the first case report that demonstrates CFRE was found during the evaluation of altered bowel habits for a prolonged period.

CONCLUSION

Mostly it is an incidental finding detected during evaluation of other conditions unless it is complicated by other anomaly or complicated by obstruction or infection. This should be treated according to clinical presentation and anatomical abnormality.

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