Case Report

An Interesting Case Of A Horseshoe Kidney With Unilateral Single Ureter And Associated Anorectal Malformation

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Abstract

Horseshoe kidney (HSK) has an incidence of about 1 in 400 or 0.25% of the general population and is seen more commonly in males as compared to women in a ratio of 2:1. Usually, Horseshoe kidney would have two normal ureters each draining separately into the bladder. We present a rare and interesting case of Horseshoe kidney with unilateral single ureter and associated anorectal malformation.

Keywords: Anorectal malformation; Horseshoe kidney; Unilateral single ureter.

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Introduction

Horseshoe kidney (HSK) has an incidence of about 1 in 400 or 0.25% of the general population and is seen more commonly in males as compared with women in a ratio of 2:1.[1] It is associated with genitourinary anomalies with duplication of ureters seen in 10% of HSKs.[1] The association of HSK with single-system ectopic ureter is an extremely rare finding with only one similar case reported in literature. Most cases describe HSK with bilateral single ectopic ureters draining separately into the bladder. This report aims to highlight the radiological and clinical findings in a case of HSK with unilateral single ureter with associated anorectal malformation.

Case report

An 18-year-old female was referred to the Urology outpatient department from the paediatric surgery department in view of incidental radiological finding of horseshoe kidney with right sided hydronephrosis. The patient was asymptomatic on presentation.

A detailed history revealed the following information- Female child was delivered at full term by lower segment caesarean section in view of breech presentation. She passed meconium and urine normally after birth. At one month of age, she was started on top feeds after which she developed constipation that required intermittent rectal evacuation and enemas. On further evaluation she was diagnosed to have anteposed anus for which she underwent anterior sagittal anorectoplasty (ASARP) at the age of four months. She was then asymptomatic until the age of 11 years when she presented to a tertiary care hospital with faecal incontinence and abdominal distention associated with non-bilious vomiting. She underwent an

exploratory laparotomy with ascending colon colostomy with operative notes mentioning the presence of colonic duplication cyst. She presented one year after surgery with complaints of obstipation and underwent excision of total colonic duplication cyst with colo-colonic anastomosis and covering ileostomy, ileostomy closure was done three months later. The patient was on regular follow up annually in paediatric surgery department; routine ultrasound revealed presence of horseshoe kidney and right sided hydronephrosis. The horseshoe kidney was diagnosed already but hydronephrosis was a new finding.

On further history taking, there was no history of recurrent urinary tract infections in the past. Patient attained menarche at the age of 12 years and had age adequate intelligence and development of secondary sexual characters. On clinical examination, patient was afebrile and haemodynamically stable. Per abdomen examination revealed previous abdominal surgery scars. Perineal examination revealed normally placed urethral meatus and vaginal orifice. Scar seen near perianal area was seen suggestive of surgery done in the first year of life.

A review ultrasound of Kidney, Ureter and Bladder (KUB) revealed Right Kidney with normal dimensions of 10.1 X 4.2cm, right sided mild hydronephrosis (AP diameter= 22.8mm) associated with mild entire hydroureter (Pelvic ureter = 9.4mm). However, terminal most ureter appeared normal in calibre. The Left Kidney measured 7.5 x 3.5 cm, MPPT- 12mm, normal in shape, echotexture and cortico-medullary junction associated with Mild hydronephrosis (AP diameter= 19.4mm). Lower poles of both the kidneys were fused in the midline. Urinary Bladder was found to be distended with smooth & regular outline. There was significant postvoid urine volume of 130cc with a prevoid bladder of 300 ml capacity on ultrasoumd.

Laboratory investigations revealed normal serum creatinine values and Urine routine and microscopic examination.
Micturating Cysto-urethrogram was done which was found to be normal (**Figure 1**).

Patient was further evaluated by CT urography which revealed normal dimensions of Right kidney: 9.8 x 4.8 cm with Right sided mild hydronephrosis and Left kidney with normal dimensions of 8.1 x 4.6 cm. Both kidneys were normal in enhancement pattern. The lower poles of the bilateral kidneys were fused (glandular fusion) at the level of L4 vertebra suggestive of horseshoe kidney. The isthmus/connecting part measured 15 mm in diameter at L4 vertebra. Single ureter was seen arising at the level of isthmus forming common pelvi-ureteric junction and then ureter was seen inserting into right postero -lateral wall of urinary bladder (**Figure 2**).

Hydroureter was seen with maximum diameter of ureter being 13 mm. Urinary bladder was distended with normal wall thickness of 10 mm. Right common iliac artery was seen passing posterior to the connecting part. Left common iliac artery was seen passing anterior to the connecting part and taking 360-degree rotation while giving rise to internal iliac artery.

Patient is planned to be kept on six-monthly follow up with clinical examination, Ultrasound examination of KUB with measurement of post void residual urine volume and serum creatine and urine routine examination.

Discussion

A number of anomalies associated with HSK have been reported in the past. HSK with bilateral single-system ectopic ureters was reported by Khong et al[2] as a very rare entity with only five cases reported in literature. Christoffersen and Iversen stated that combination of HSKs with bilateral ureteral duplication is a very rare entity with only two reported cases.[3] Sharma et al described bilateral duplex collecting system in a HSK and found only three reported cases.[4] Tyagi et al reported HSK with complete unilateral duplication of ureter and pelvicalyceal system and found it to be the first of its kind reported in literature.[5] We reviewed literature by searching in PubMed database and Google Scholar using 'horse shoe kidney with unilateral single ectopic ureter' as keywords and found one case reported by Dr. Samarth Agarwal.

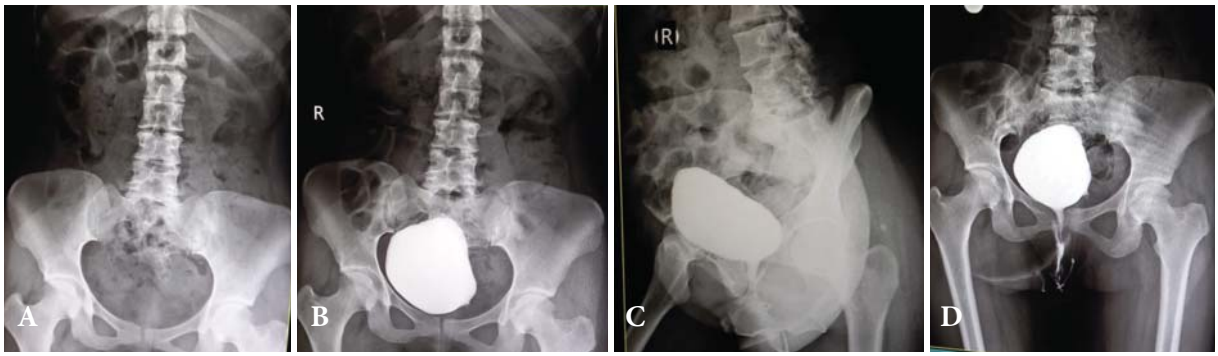

Figure 1. Scout Film- XRAY KUB- Normal scan, no bony deformity, no evidence of calculus (A). Serial images of micturating Cystourethrogram show normal Bladder contour and capacity, No evidence of vesicoureteric reflux, Normal urethra (B,C,D)

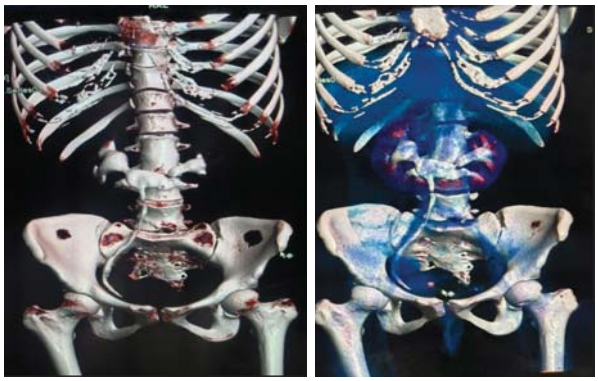

Figure 2. CT scan reconstructed image showing Horseshoe shaped kidney with a single unilateral right sided ureter.

Our case presented with a unique association of Horseshoe kidney with Anorectal malformation of anteposed anus and cystic variant of colonic duplication.

Clinicians should be cautious while approaching a case of HSK. A thorough clinical and radiological assessment should be undertaken as HSK presents a diagnostic and interventional challenge owing to a number of associated anomalies. Also, any surgical intervention in HSK will be real challenging to urologist because of its abnormal morphology and unpredictable blood supply. Along with it, this patient has already underwent laparotomy multiple times.

Conclusion

For a urologist, horseshoe kidney presents unique challenges in diagnosis and technical challenges in management. A detailed evaluation using imaging techniques especially CT scan with reconstruction images and a thorough discussion with the reporting radiologist is essential to understand the unique anatomy of such cases which will guide in deciding the treatment plan for these patients.

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