CASE REPORT

Horseshoe kidney with unilateral single ectopic ureter

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SUMMARY

The horseshoe kidney (HSK) is not an uncommon entity with an incidence of about 1 in 400 or 0.25% of the general population. It is also more commonly found in men as compared with women in a ratio of 2:1. An increased association of genitourinary anomalies have been found with HSK. Duplication of ureters occurs in 10% of HSKs. However, the association of HSK with single-system ectopic ureter is extremely rare. To our knowledge, no case with similar presentation has been reported in literature. Most cases of HSK with bilateral single ectopic ureters have been described. The aim of this report is to highlight the radiological and surgical findings in a case of HSK with unilateral single ectopic ureter.

BACKGROUND

Horseshoe kidney (HSK) is a commonly found anomaly during urological procedures and has varied clinical presentations. One must keep in mind different associations of this anomaly for better treatment outcome.

CASE PRESENTATION

A 5-year-old girl with complaints of urinary incontinence with intermittent normal voiding since birth presented to us.

The antenatal history was humdrum with uncomplicated normal vaginal delivery at 38 weeks of gestation. Baseline renal function tests were normal with a creatinine of 0.5 mg/dL and Hb of 11.5, and urine routine microscopy showed pus cells 5–10/hpf.

Clinical and abdominal examination was unremarkable. On physical examination of genitalia, she was found to have three separate openings (figure 1).

On further calibration with infant feeding tube, the child was found to have a single urethral meatus, a single vaginal opening and an ectopic right ureter opening just to the right of the vagina (figure 2).

Abdominal ultrasound of patient was normal with insignificant post-void residual urine.

Micturating cystography (MCU) did not demonstrate any vesicoureteric reflux on either ureter even on post-void films (figure 3).

On cystourethroscopy (figure 4), the patient was found to have a single non-patulous left ureteric orifice with clear urine efflux. It was located at the centre of a deformed trigone with absent right ureteric orifice.

On further evaluation with CT–intravenous pyelogram (IVP), the HSK was well seen with lower

Figure 1 Three separate perineal openings on physical examination.

Figure 2 External urethral meatus calibrated with infant feeding tube.

poles fused in the midline (figures 5 and 6). There was good excretion of contrast from both fused moiety. Right ureter was seen mildly dilated and coursing posterior to the bladder neck and ending at the level of urethra. Contrast was also seen in vagina. Left ureter was normal in course and calibre (figures 7 and 8).

DIFFERENTIAL DIAGNOSIS

Ectopic ureter
 Ureterocolocele
 Neurogenic bladder

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Figure 3 Micturating cystography (MCU) demonstrating no reflux.

Figure 4 Left ureteric orifice located at the centre of deformed trigone on cystoscopy.

TREATMENT

Right ureteric reimplantation (extravesical Lisch Gregor's technique) was performed after 6 weeks (figure 9).

OUTCOME AND FOLLOW-UP

An ultrasound and MCU performed at 6 months did not reveal any hydronephrosis or vesicoureteric reflux. The child was continent immediately following surgery and Foley catheter removal and remained continent in follow-up as well.

Figure 5 Horseshoe kidney fused in the midline.

Figure 6 Horseshoe kidney fused in the midline.

After 1 year of follow-up, there was not a single episode of urinary tract infection and the child has remained well since then.

DISCUSSION

Ectopic ureter or ureteral ectopia is a condition when the ureter does not terminate in the urinary bladder but enters a different site. This site is usually urethra in men and urethra/vagina in women.¹

Eighty per cent of all ectopic ureters is associated with duplicated system especially in women. Single-system ectopic ureters are more troublesome than duplicated system as they are more prone to renal dysplasia, urinary tract infection and urinary incontinence.²

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* as a very rare entity with only five cases reported in literature.³

Christoffersen and Iversen stated that combination of HSKs with bilateral ureteral duplication is a very rare entity with only

Figure 7 CT urogram showing dilated right ureter.

Figure 8 Contrast in right ureter is seen to extend below the bladder and enter the vagina.

Figure 9 (A and B) Right ureter seen extending below the bladder. Ureter spatulated with infant feeding tube. (C and D) Bladder mucosa identified and spatulated. Extravesical ureteric reimplantation done.

two reported cases.⁴ Sharma *et al* described bilateral duplex collecting system in a HSK and found only three reported cases.⁵

Tyagi *et al* reported HSK with complete unilateral duplication of ureter and pelvicalyceal system and found it to be first of its kind reported in literature.⁶

We reviewed literature by searching in PubMed database and Google Scholar using 'horse shoe kidney with unilateral single ectopic ureter' as keywords and found no such case reported since then.

Learning points

- ▶ Horseshoe kidney (HSK) is not an uncommon entity.
- ▶ Clinicians should be cautious while approaching a case of HSK.
- ▶ Ectopic ureter should be considered as an important differential diagnosis of any child with urinary incontinence.
- ▶ A thorough clinical and radiological assessment should be undertaken as HSK presents a diagnostic and interventional challenge owing to a number of associated anomalies.

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