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Congenital Uretero-Vaginal Fistula - A Rare Case of Single System Ectopic Ureter Associated with Renal Dysplasia

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Abstract

Urinary incontinence resulting from functional disturbances is common in childhood, however diagnosis of urinary incontinence due to organic causes such as ectopic ureter and genital fistulas is important because urinary continence in females has a negative impact on the quality of life and correct diagnosis carries a potential of absolute treatment by surgical corrections. We present a case of a 9-year-old female with complaints of involuntary leakage of urine from the vagina despite normal bladder filling and emptying. A diagnosis of ectopic ureter with uretero-vaginal fistula was made. Radiological evaluation is important in cases of suspected ectopic ureter and best results can be achieved by using multi-disciplinary approach.

Keywords: Urinary incontinence, ectopic ureter, congenital, uretero-vaginal fistula.

Introduction

Diagnosis of urinary incontinence secondary to organic causes such as ectopic ureter and uro-genital fistulas is extremely important as it is curable and best outcomes can be obtained by surgical approach [1], [3]. Ectopic ureter is defined when the ureter opens into any region other than its usual opening into the trigone of urinary bladder [1], [4]. The prevalence of EU is uncertain because many are asymptomatic and the diagnosis is usually overlooked [1], [5], [7]. Female-to-male ratio is 2–6:1 [1], [5]. When an ectopic ureter drains only one kidney, it is referred as a single system ectopic ureter. Approximately 8% of ectopic ureters in females are associated with duplex kidney. However, single system ectopic ureter with dysplastic kidney is rare, especially in females [3], [7], [15]. The surgical approach for the treatment of ectopic ureter depends on the extent of the

renal function, accompanying anomalies and the site of opening of the ectopic ureter.

We describe case report of a 9-year-old female patient with a history of urinary dribbling studied retrospectively after receiving permission from concerned authorities (Head of the Department of Radio-diagnosis and Hospital Director) of MGM Hospital, Navi Mumbai.

Case Report

A 9-year-old female patient came with urinary incontinence and continuous dribbling of urine from the vaginal orifice.

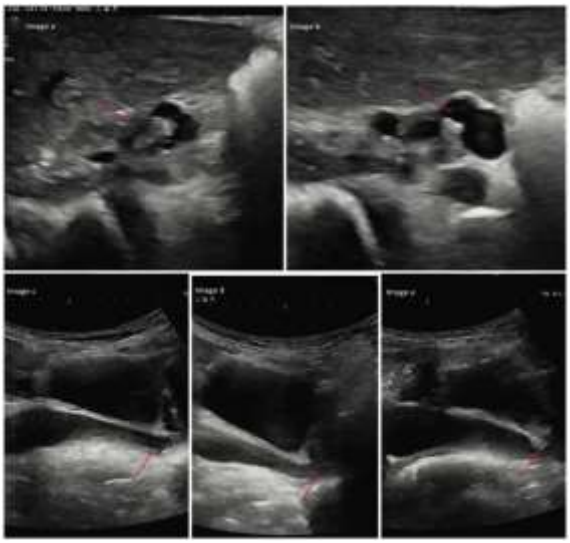
On Ultrasound examination: the right kidney was small in size measuring 3.9 x 1.9 cm with dilated renal pelvis. There was dilatation of proximal and distal segments of right ureter with distal segment showing cystic dilatation and tortuosity in the course. The right ureter did not appear to insert into the urinary bladder with absent right sided urinary jet. The dilated lower end of the ureter was traced distal to the bladder neck and appeared to be inserting into the vaginal vault.

On cystoscopy; left ureteric opening was normal and right ureteric opening was absent with no evidence of any other opening in bladder neck or urethra.

On vaginoscopy; there was evidence of a tubular structure entering into the anterior vaginal wall with peristalsis at 10 O'clock position.

99mTc DTPA Renogram done for kidney function showed non-functioning right kidney with normal function and excretion of left kidney.

Figure 1:



Small sized right kidney with dilated renal pelvis, proximal hydro ureter and few cysts (Image a and b).

Dilated lower end of ureter with its tip traced distal to the urinary bladder in the lower pelvic region (Image c, d and e).

Figure 2:

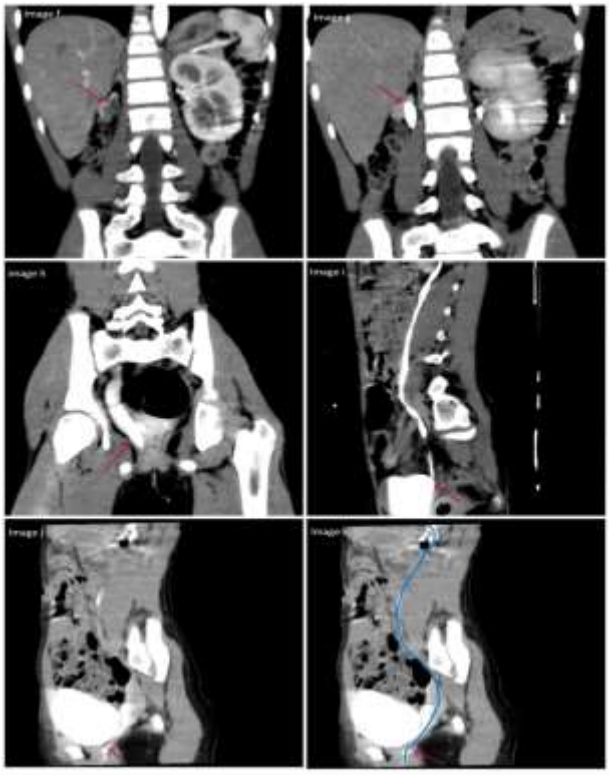


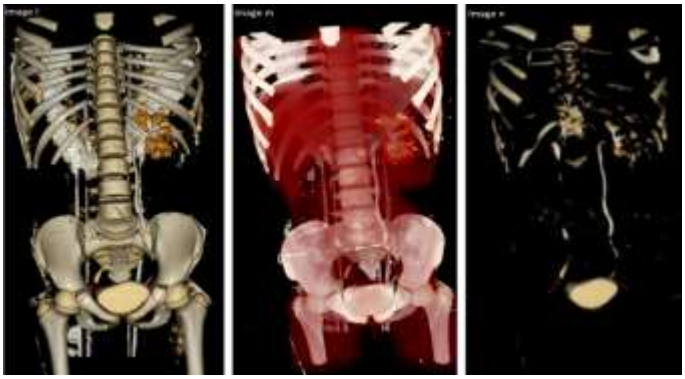
Image f showing small in size right kidney with significant cortical thinning.

Images g and h showing dilated proximal and distal ureter and there was delayed excretion of contrast into the right pelvi-calyceal system on 25 minutes delayed images.

Image i showing normal course of left ureter with insertion into the urinary bladder.

Images j and k showing dilated distal ureter with pooling of contrast within, and it is seen traversing posterior to the urinary bladder not inserting into the bladder. It shows ectopic insertion into the vaginal vestibule.

Figure 3:



3D reconstructed images showing normal insertion of left ureter into the urinary bladder with non-insertion of right ureter into the urinary bladder and absent right vesico-ureteric junction.

Discussion

The prevalence of ectopic ureter is unknown because most patients are asymptomatic and the diagnosis is often delayed. Single system ectopic ureter is difficult to diagnose especially when it is associated with small, dysplastic and poor functioning kidney which may be difficult to visualize on the conventional imaging [1], [3], [11].

Embryologically, ureteric bud arises from mesonephric duct at approximately fifth week of gestation. Ectopic ureter occurs when the ureteric bud arises abnormally high than the usual position and delayed or non-separation of the ureteric bud from the mesonephric duct. The ureteric bud remains attached to the mesonephric duct for long and migrates more caudally to get inserted distal to the urinary bladder or the genital tract. When the ureteric bud gets incorporated into the adjacent structures of paramesonephric duct origin, this leads to opening of ectopic ureter into the female genital tract [1], [5], [6]. In females, the common sites of insertion are urinary bladder neck, urethra, vestibule between the urethra and the introitus, vagina, cervix or uterus. In males, common sites of insertion are lower urinary bladder, posterior urethra, seminal vesicle, ductus deferens, ejaculatory duct or rectum [10], [12].

Clinically, it presents with urinary incontinence, continuous dribbling of urine despite normal bladder filling and emptying and recurrent urinary tract infections, often associated with abnormal urinary odour. They often suffer from wet and erythematous perineal rash due to continuous dribbling [1], [2], [4], [13].

Treatment of ectopic ureter is surgical and it not only targets the treatment of incontinence, but also to prevent the renal damage due to recurrent urinary tract infections. Plan of treatment depends on the degree of renal function, location of the ureteric orifice and the associated anomalies [1], [4], [7]. If it is associated with normal kidney, treatment is targeted to preserve the normal renal function [9]. In cases of intramural ectopic ureter in which ureter is attached to the serosal surface of bladder but it doesn't terminate into the trigone, instead it tunnels through the trigone in the submucosa and terminates distal to the bladder neck. In such cases ureteric re-implantation is done by neo-ureterostomy and urethral-trigonal reconstruction. In cases of extramural opening, neo-uretero-cystostomy is done re-implantation of ureteric orifice closer to the urethral sphincter which restores normal urinary outflow control [1], [8], [9], [14]. If the ectopic ureter is associated with dysplastic or non-

functioning kidney, target of treatment is alleviation of symptoms as soon as possible and can be planned for nephro-ureterectomy [1], [2], [7].

Conclusion

Urinary incontinence due to functional cause is common and the treatment is ineffective but the diagnosis of urinary incontinence secondary to the organic cause is extremely important as it is potentially curable and can be corrected surgically. Congenital single system ectopic ureter associated with dysplastic kidney is a rare condition. Urinary incontinence that persists even after the toilet training should be assessed thoroughly and managed by a multi-disciplinary approach especially in females so that appropriate treatment can be done at early age not affecting the psycho-social development as the urinary incontinence creates a profound negative impact on a woman's quality of life. In cases of continuous urinary dribbling and incontinence despite having normal voiding patterns, possibility of uretero-vaginal fistula should be considered irrespective of the age. Along with the diagnosis of ectopic ureter, additional investigations to look for renal functions should also be done as the treatment options depend on degree of renal function and the location of ectopic ureter. Incontinence can be treated by nephro-ureterectomy or ureteric reimplantation.

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