Case Report

Complete Left Ureteric Duplication with Left Ectopic Ureter Presenting as Ureterovaginal Fistula in a Nigerian Girl; A Case Report

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Abstract

Ureteric duplication is associated with ectopic ureter and manifests in many ways. It is commonly seen in the female gender. Most patients with this anomaly present with recurrent urinary tract infections, total urinary incontinence and enuresis while others may remain asymptomatic. We present a 26-year-old nulliparous Nigerian lady with complete left duplicated ureters who was initially leaking urine intermittently from a spot on the vestibulum but developed continuous leakage of urine per vaginam following a surgical attempt in another hospital. She had exploratory laparotomy at our hospital and re-implantation of the left duplicated ureters in the bladder. A piece of gauze from previous surgery was found to have eroded into the proximal third of the rectum. This was removed and rectal repair done. Duplicated ureters should be suspected in female patients presenting with urinary incontinence and their care should be left to experienced urologists to prevent complications.

INTRODUCTION

Duplication of the ureters is a common congenital malformation of the urinary system [1,2]. Naton (1944) found some form of duplication of the ureters in 0.9% of a series of autopsies [1].

Ureteric duplication may be incomplete when the two ureters fuse to become one before reaching the urinary bladder or complete in which the two ureters run separate courses. In complete duplex ureters, the upper pole ureter may be ectopic, when its ureteral orifice is caudal to the normal insertion on the trigone thereby presenting as urinary incontinence. Some patients have normal pattern of voiding together with continuous incontinence (continent incontinence) [1-4].

CASE PRESENTATION

E.C. was a 26-year-old single, nulliparous Nigerian lady who presented to our department with 4 month history of continuous leakage of urine per vaginum associated with normal voiding pattern in between (continent incontinence).

She presented at the referring hospital with leakage of urine from an ectopic site on the vestibule which she noticed when she was 24 years old. At that hospital, she was treated for urinary tract infection with oral antibiotics twice before she had two unsuccessful abdominal operations aimed at correcting the condition. The diagnosis made by the referring doctor and the type of operation performed were not stated in the referral letter given to the patient, so we did not have those information. She then started leaking urine from the vagina after the second surgery and no longer leaked from the previous site on the vestibulum.

At presentation, she was a young woman with normal external urethral meatus. There was a depressed spot on the labial minora located on the left side of the meatus and there was continuous egress of urine from the introitus.

Speculum examination showed pooling of urine in the posterior fornix with an area of in duration on the left superolateral wall of the vagina.

A diagnosis of suspected left ureterovaginal fistula involving the ectopic ureter of a duplicated system was made.

Cystoscopy showed normal urinary bladder mucosa with two ureteric orifices located at normal sites on the either side of the trigone and were both jetting out urine.

Intravenous Urography showed prompt excretion of contrast from upper and lower poles of both kidneys. There was a left duplicated system and the ureter draining the lower pole was dilated up to its renal pelvis while the upper pole ureter seemed normal in caliber (Figure 1). The hydronephrosis of the left lower pole ureter was probably caused by vesicoureteric reflux that usually accompanies lower pole ureter because of its short intravesical portion. This was taken into account when performing the anti-refluxing ureteric re-implantation.

Additional information was obtained from Abdominal CT Urography which showed unilateral left duplicated ureters with the upper pole ureter coursing farther down beyond the urinary bladder. The right ureter and pelvicalyceal system were normal.

Complete blood count and serum electrolyte, urea and creatinine were normal.

She underwent exploratory laparotomy using a midline infraumbilical incision through the previous scar. The right ureteric orifice and the left lower pole ureteric orifice were seen when the bladder was opened and were easily catheterized with size 5FR feeding tube.

The left duplicated ureters enclosed in a common sheath was mobilized on the medial side to free them from the adherions of previous surgery (Figure 2). One of the ureters inserted into the bladder while the other one, the upper pole ureter coursed further down posterior to the bladder and was traced to the posterior fornix of the vagina.

A piece of gauze swab from the previous surgery at the referring hospital was found adherent to the left posterior surface of the urinary bladder and eroded into the rectum (Figure 3). Surrounding adhesions had covered the gauze and the bowel. The gauze was removed, bowel edges were freshened and closed primarily in two layers with vicryl 2/0 sutures. Both ureters were divided as a unit close to the bladder and a common sheath ureteric re-implantation was done by Politano-Leadbetter technique.

Her post-operative period was satisfactory. She remained dry with no gastrointestinal or urologic sequelae and was followed up for up to 6 months before she defaulted from follow up. All the investigations requested to monitor her progress post operatively were not done before she defaulted. (Abdominal/pelvic USS, IVU or CT Urography.

DISCUSSION

Like the case being reported, majority of patients with complete ureteric duplication are females [4-8]. Most cases are unilateral but could also be bilateral [5].

Patients with ectopic ureter in a duplicated system usually present with recurrent urinary tract infections, urinary incontinence or enuresis [3,4,9]. Some are without urinary incontinence despite ectopic ureteric orifices [10,11]. Presentation is usually in childhood but patients who became symptomatic in adulthood have been reported [10,12].

In males, urinary incontinence does not occur as the ectopic ureteric orifice always opens proximal to the external sphincter [1,2]. Normal urination together with continuous incontinence (continence incontinent) are features of infrasphincteric ureteral openings [1,2,13].

Ectopic sites of ureteral openings could be at the urethral margins, urethropelvic septum [4] or the vestibulum [7,10,11]. The ectopic ureters opened into either the uterus or vagina in some reported cases [7,12,14] and have also been found to insert
at the bladder neck or prostatic urethra [7,11,15,16]. All the four ureters in bilateral duplex ureters reported by Morhason-Bello et al opened into the urinary bladder [5].

In the management of these patients, one of the challenges faced is identification of the exact location of ectopic ureteric orifice. This could delay the diagnosis as most diagnostic tools like abdominal and pelvic ultrasound scan, cystoscopic evaluation and intravenous urography only give incomplete details. So, in order to gather sufficient information about the state of the pelviccalycal systems, the duplicated ureters and identification of ectopic orifice, Multi-detector Computed Tomography (MDCT) Urography with non-ionic iodinated contrast and Magnetic resonance Urography are very essential [12,15].

Treatment depends on the extent of renal damage caused by ureteral obstruction, vesico-ureteric reflux (VUR) and urinary tract infections. Renal Scintigraphy is important to ascertain the residual function in the kidney and to decide the type of surgery to do.

Though the current approach in the management of ectopic ureter is to remove the anomalous kidney together with the ectopic ureter especially if the ureter opens into regions outside the urinary tracts, using a nephron-sparing surgery [9,17], but uroterovesical re-implantation could also be an option if the renal segments have good function. Several cases of such ectopic ureters with good split functions of the affected kidneys have been reported and managed by ureteric re-implantation [7,11,12].

In instances where the ectopic ureters open into other places but within the urinary systems, the functions of the renal segments are often preserved and ureteric re-implantation is usually performed [5,7,15,16].

If the kidney is hypofunctional or a segment is dysplastic, a left heminephrectomy or heminephroureterectomy would be necessary [7,8,9,10,17,18].

The ectopic ureter opened into the vagina in our case and she had left common sheath ureteroneocystostomy because the calyceal systems of both upper and lower poles of the left kidney demonstrated good contrast excretion on CT Urography.

El Ghoneimi et al studied 31 ectopic ureters with complete ureteric duplication. The locations of ectopic orifices were identified in 25 of them. 17 children were treated with upper pole nephroureterectomy as they had non-functioning renal segments while the remaining 14 who had good renal functions had uroterovesical re-implantation in 12 and ureteropyelotomy in 2. In 7 cases, the ectopic orifices opened into the vagina like the case being reported, 2 had good renal function and had ureteric reimplantation while the remaining 5 had partial nephrectomy due to non-functioning kidney [7].

The finding of a piece of gauze which had eroded into the proximal rectum made this case an interesting one. This showed one of the challenges faced when managing patients in our environment. Most cases would have been dabbled into by non-specialists before presentation. Similar scenarios have been reported in other surgical specialties [19].

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REFERENCES


