

Case Report

Bilateral Single Ectopic Ureters Draining Into a Grossly Dilated Vagina in an Adolescent Female

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A 16-year-old female presented with dribbling of urine along with voluntary voiding since birth. Renal imaging revealed hydroureteronephrosis on the right side; the uterus and ovary were normal. A radionuclide scan showed a left nonfunctional kidney. On cystovaginoscopy, the urethra was shown to be normal and the urinary bladder was tubular with small capacity and an absent trigone. Although the vagina was capacious, no ureteric orifices were found. Computed tomography corroborated the diagnosis of bilateral, single ectopic ureters draining into a grossly dilated vagina. This case is unique because it is a bilateral single-system ureteral ectopia in a completely differentiated female genital tract that presented late in adolescence. To the best of our knowledge, this is the second such ureteral abnormality reported in the literature so far. The patient underwent ileocystoplasty with right ureteric reimplantation and nephroureterectomy for the left nonfunctional kidney, which histopathology showed to be tuberculosis. The patient is continent with cystometric capacity of more than 300 mL.

Keywords: Abnormalities; Ureter

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INTRODUCTION

Ectopic ureters are common in females, 80% of which are duplicated and present at birth or in childhood. The urethra and vaginal vestibule are the most common sites of drainage in females. Bilateral single-system ectopic ureters are rare. Furthermore, their drainage in a completely developed Mullerian system has been reported only once before, although there are reports of bilateral single-system ureteral ectopia into the urogenital sinus. We report one such case of bilateral, single ectopic ureters draining into a grossly dilated vagina that presented late in an adolescent female.

CASE REPORT

A 16-year-old female presented with a complaint of involuntary leakage of urine since birth. The patient re-

ported being able to void normally apart from the involuntary leak. The previous leak of a few drops had increased in amount over the past 2 months, thus prompting medical attention. She could manage with 2 to 3 pads during the day, with 10 to 12 daytime voids in amounts ranging from approximately 90 to 240 mL. Her nighttime incontinence was severe; she consistently woke up wetting her bed every night.

On examination, the external urethral meatus was normal as was the external genitalia, and the vaginal introitus was moist. Renal parameters and the results of urine analysis were normal. Ultrasonography revealed right kidney hydroureteronephrosis (HDUN) changes and an atrophic left kidney. The intravenous urogram showed left kidney calcification with nonvisualisation of the same. The right kidney was hydronephrotic with a dilated ureter and dilated vagina wrongly interpreted as a normal urinary bladder (Fig. 1A, B). A diethylenetriaminepentaacetic acid re-



FIG. 1. (A) Intravenous urogram (IVU) showing right hydroureteronephrosis with contrast filling the capacious vagina. (B) IVU: full apparent urinary bladder (vagina).

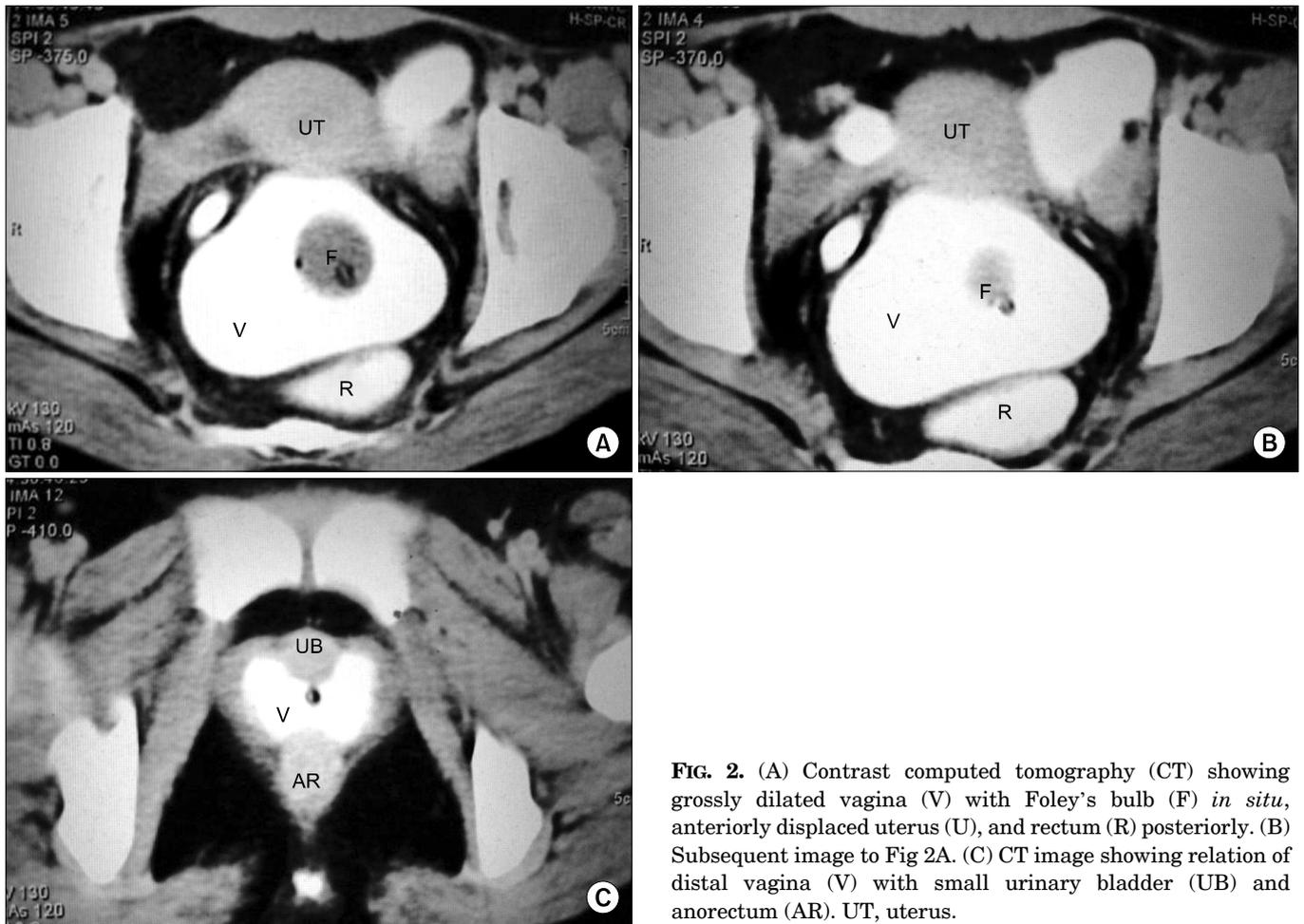


FIG. 2. (A) Contrast computed tomography (CT) showing grossly dilated vagina (V) with Foley's bulb (F) *in situ*, anteriorly displaced uterus (U), and rectum (R) posteriorly. (B) Subsequent image to Fig 2A. (C) CT image showing relation of distal vagina (V) with small urinary bladder (UB) and anorectum (AR). UT, uterus.

nal scan showed a nonfunctioning left kidney, delayed excretion phase of the right kidney, and a glomerular filtration rate of 65.71 mL/min. On cystovaginoscopy, the ex-

ternal urethral meatus and urethra appeared normal, with a small urinary bladder with approximately 20-mL capacity and an absent trigone. Urine could be seen coming out

through the vaginal introitus. The vagina was highly capacious but ureteric orifices could not be identified. Computed tomography revealed calcification in the left kidney and left ureter, a nonexcreting left kidney, HDUN changes on the right side with delayed excretion, and a small urinary bladder separate from the capacious vagina (Fig. 2A-C). The accumulating contrast was well delineated.

On exploration, the urinary bladder was represented by a tubular structure engulfed by the capacious vagina. The right ureter was moderately dilated, and the left ureter was minimally dilated and calcified. Both ureters terminated on the anterolateral wall of the vagina (Fig. 3). Ileocystoplasty with right ureteral reimplantation was done. The bowel contributed 90% of the reconstructed bladder wall, because the native bladder was atrophic. Left-sided nephroureterectomy was done. The histopathological examina-

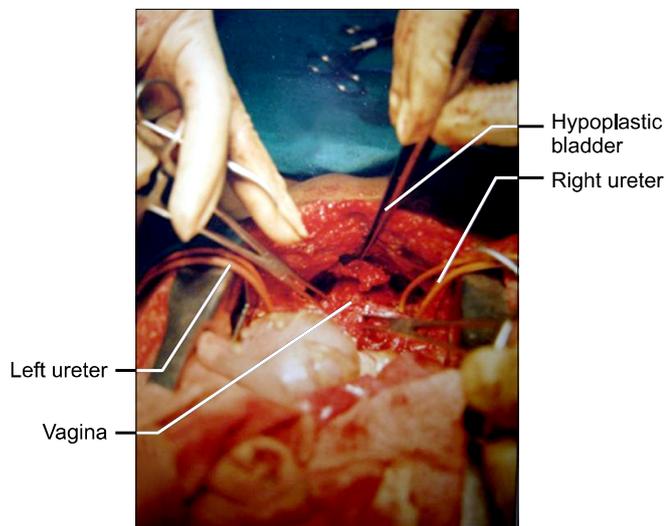


FIG. 3. Intraoperative picture.

tion revealed tuberculosis, for which antitubercular therapy was given. There was no evidence of tuberculosis in the excised urinary bladder specimen. The postoperative period was uneventful. The patient voided well with straining and is fully continent. Clean intermittent self-catheterization fourth hourly was advised for the initial few months, which the patient was later weaned from because she maintained good voiding.

DISCUSSION

In our review of the available literature for the past 50 years, we found only one such published case report of bilateral, single vaginal ectopic ureters by Chatterjee and Chatterjee [1] in 2011. Sheldon and Welch [2] and more recently Singh et al. [3] reported bilateral, single ectopic ureters draining into the urogenital sinus. The ureters are derived from the mesonephric duct outpouching known as the ureteric bud. The ureters find their way into the definitive bladder as the distal mesonephric duct is absorbed into the developing bladder to form the trigone, bladder neck, and proximal urethra. Ectopic ureters draining into the female genital tract are thought to be via embryonic remnants of the mesonephric duct (Gartner's duct), which breaks open into the Mullerian system. This elusive embryonic event is unexplained and rare, more so in bilateral, single ectopic ureters. Occurrence of a simultaneous urogenital abnormality is expected here, such as agenesis of urinary bladder or persistent urogenital sinus. Bilateral single-system vaginal ectopia in a completely differentiated urogenital tract should arguably be very rare. This case represents one such unique occurrence.

Bilateral ectopic ureters in females usually present early with constant dribbling of urine. In this case, however, the presentation was during adolescence and with 150 to 200 mL voided apparently normally. This is explained by the

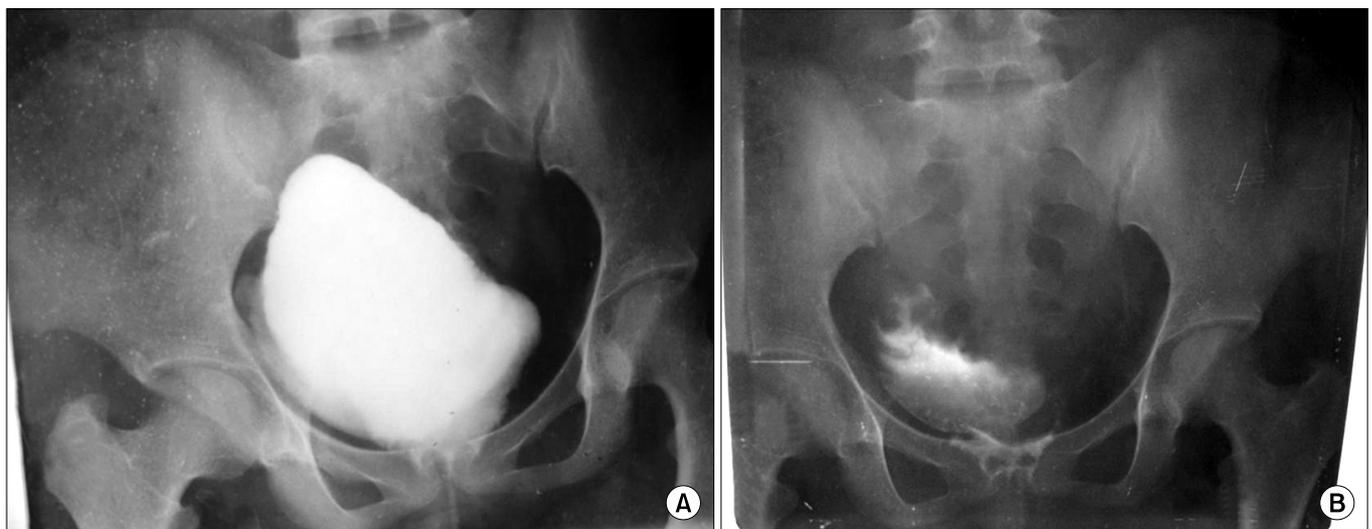


FIG. 4. (A) Micturating cystourethrogram (MCU) showing reconstructed bladder postileocystoplasty. (B) MCU: postvoid film with adequate emptying.

fact that patient had, over the years, learned to hold the urine in her vagina by contracting the pelvic floor musculature. Computed tomography was crucial for arriving at a diagnosis in this case, as was reported by other authors in similar cases, although magnetic resonance urogram can be further precise [4]. There was a therapeutic dilemma regarding reconstruction of the urinary tract. An ileocystoplasty with right ureteric reimplantation was done anticipating future need of a continence procedure or undiversion to nonorthotopic diversion [5]. Because apparently good urethral mucosal coaptation was seen on cystoscopy, orthotopic diversion was preferred. The issue of continence is also discussed by Podesta et al. [6] in their article, in which they inferred that the bladder in bilateral, single ectopic ureters may retain a good continence mechanism with adequate augmentation. Hence, on the basis of this evidence, no bladder neck continence procedures were attempted in our case. Fortunately, the patient was continent with cystometric capacity of 300 mL at 3 months. She was weaned off clean intermittent self-catheterization by 6 months and at present voids satisfactorily by abdominal straining (Fig. 4A, B).

CONFLICTS OF INTEREST

The authors have nothing to disclose.

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