CASE REPORT

Normal functioning single system ectopic ureter draining into a Gartner's cyst: laparoscopic management

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SUMMARY

Association of single system ectopic ureter with normal functioning bilateral kidney and absence of congenital anomalies is very rarely reported in the literature. We are reporting for the first time a case of this type of anomaly in which uretral ectopia was draining into a Gartner's cyst and was managed by laparoscopy. A 16 year girl presented with normal voiding with continuous dribbling since birth. Voiding cystourethrogram, intravenous urogram, cystovaginoscopy and retrograde contrast study confirmed the diagnosis. Ultrasound of the whole abdomen and physical examination ruled out any associated congenital anomalies. Transperitoneal laparoscopic ureteric reimplantation was performed and distal stump was ligated. On follow-up at 3 months she was completely dry; her vaginocopy showed disappearance of cystic bulge and her voiding cystourethrogram showed normal study without any reflux. When single system ectopic ureter opens into small-to-moderate size wide opened mouth vaginal Gartner's cyst, laparoscopic ureteric reimplantation and ligation of distal stump is an appropriate procedure.

BACKGROUND

Most of the time the ectopic ureter arises from the upper moiety of a duplex kidney.¹ The single system ectopic ureter is a rare condition which is usually associated with non-functioning dysplastic kidney.²⁻³ Unlike duplicated system ectopic ureter/ureterocele, a single system ectopic ureter or ureterocele is very rare in women and has a strong risk of additional congenital cardiac and urogenital anomalies⁴⁻⁶ and usually managed by nephroureterectomy of ipsilateral dysplastic kidney.¹ To the best of our knowledge we are reporting for the first time a case of single system ectopic ureter draining into vaginal Gartner's cyst (GC) with normal functioning bilateral kidney and without any associated congenital anomalies, which was successfully managed by laparoscopic ureteric reimplantation.

CASE PRESENTATION

A 16 year girl presented with continuous dribbling of urine along with normal voiding since birth. There was no history of any other lower urinary tract symptoms, stress incontinence, urinary tract infection or fever. Physical examination showed leakage of urine per vaginum. Routine urine and blood investigations were within normal limits. Ultrasonography (USG) revealed bilateral normal-sized kidney with normal cortical echo texture and

corticomedullary differentiation and mild dilation of left ureter. Intravenous urography (IVU) showed normal excretion of contrast through both kidneys, both pelvicalyceal systems and ureters were visible and showed bilateral single system, left lower ureter was dilated (7 mm) and pooling of contrast into vagina (figure 1A). Voiding cystourethrogram (VCUG) was normal. On cystourethroscopy right hemi-trigone was seen with normal right ureteric orifice and urethra. Left-sided trigone and ureteric orifice were absent (figure 1B). Vaginoscopy showed left ureteral opening on a cystic bulge in left vaginal wall (figure 1C). After ingestion of phenazopyridine (400 mg, by mouth), efflux of yellow-coloured urine was seen through this opening (figure 1D), which was suggestive of ectopic ureter opening into vaginal GC. On negotiation with 0.038" hydrophilic guide wire, wire was coiled into cavity in the left vaginal wall (figure 2A). Retrograde contrast study (with urograffin) through this opening showed dilated and tortuous left lower ureter terminating as a cystic cavity in left vaginal wall, which confirmed the diagnosis (figure 2B, C). The patient was managed by laparoscopic re-implantation of left ureter.

Operative technique

After general anaesthesia, the patient was placed in 20° supine Trendelenburg position. A 16 Fr Foley's catheter was introduced into the bladder per-urethral and clamped. A four-port technique was used. The first port was an infraumbilical 10 mm camera port. A 10 mm working port was placed on left lateral edge of rectus in midclavicular line, and the other, a 5 mm port, was placed on right lateral edge of rectus in midclavicular line, each 1.5 cm below the level of umbilicus. The fourth port, a 5 mm port, was placed 2 cm above the right midinguinal point (figure 3A). The procedure was started by incising the peritoneum just above the bifurcation of common iliac vessel. The left ovary and fallopian tube were mobilised, and an infundibulopelvic ligament was transected after clipping. The ureter was dissected and followed up to its insertion into vagina (figure 3B). The ureter was clipped near its insertion into vagina and transected. The bladder was retracted anteriorly by stay suture and site for ureteric reimplantation was marked on the left posterior bladder wall. Here a 4 cm longitudinal incision on the bladder was made. The detrusor was incised in full thickness while keeping the mucosa intact. On both sides of the incision lateral detrusor flaps were developed (figure 3E).

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Figure 1 (A) Intravenous urography showing bilateral normal excretory kidney, dilated left lower ureter and pooling of contrast in vagina. (B) Cystoscopy showing right hemitrigone with normal right ureteric orifice; left hemitrigone and ureteric orifice absent. (C) Vaginoscopy showing cystic bulge of Gartner's cyst in upper part of left vaginal wall. (D) Vaginoscopy showing efflux of yellow-coloured urine through opening in left vaginal wall.

The ureter was partially divided transversely (figure 3C) and then ureteral spatulation of 1.5 cm was performed using scissors via right lower 5 mm port and holding the ureter with the left hand (figure 3D). The bladder mucosa was incised, and it was anastomosed to the spatulated ureter using 4-0 polyglycolic continuous sutures starting from the apex of ureteral spatulation and continuing on either side (figure 3F). A 5 Fr JJ stent was kept across the anastomosis (figure 3F). The bladder muscle flaps were then approximated using 4-0 polyglycolic interrupted stitches (figure 3G). The distal stump was ligated using non-absorbable suture.

The abdominal tube drain was kept near the anastomosis via right lateral 10-mm port. The operative time was 120 min. Perioperative periods remained uneventful. The abdominal drain was removed on the second postoperative day, and next day the patient was discharged.

OUTCOME AND FOLLOW-UP

At 3 weeks postoperatively, the stent was removed. At 3 months follow-up, the patient was asymptomatic; vaginoscopy showed disappearance of the cystic bulge and VCUG showed no reflux.

Figure 2 (A) Fluoroscopic view showing coiled guide wire in cavity of left vagina. (B) Retrograde contrast study showing pooling of contrast in a cavity in left vaginal wall. (C) Retrograde contrast study showing dilated and tortuous left lower ureter terminating as a cystic cavity in left vaginal wall.

Figure 3 (A) Location of port placement. (B) Left ureter dissected up to its insertion into vagina. (C) Partial transversely divided ureter. (D) Spatulated ureter (E) Transacted and spatulated left ureter along with JJ stent in situ, full thickness detrusor flap and intact mucosa in posterior bladder wall. (F) Anastomosis of bladder mucosa with spatulated ureter by 4/0 polyglycolic suture. (G) Final anastomosis.

DISCUSSION

Ectopic ureter is defined as a ureteric orifice outside the posterolateral extremity of the bladder trigone.⁷ This anomaly comprises a variety of ureteral insertion shifting from nearly normal to extravesical location. In men the extravesical ectopic orifice is always suprasphincteric and enters the prostatic urethra, seminal vesical, ejaculatory duct or vas deferens.⁴ In women the ectopic orifice may be either suprasphincteric or infrasphincteric, with most common insertion being into the urethra or vestibule and rarely it may insert into fallopian tube, cervix or vagina.⁸⁻⁹ Rupture of the Gartner's duct into adjacent organ has been postulated to explain insertion in the above locations.¹⁰

The basic defects in the ureteral ectopia is that, the ureteric bud arise more cephalic than normal position from the mesonephric duct. Rather than being normally incorporated into bladder, it moves caudally with mesonephric duct and inserts ectopically either in or out of the bladder.¹¹ When the ureteral bud originate from an even higher point along the mesonephric duct, incorporation to the point of separation of the ureteral orifice from the Wolffian duct becomes impossible and in women this results in persistence of caudal portion of Wolffian duct (GC) with ectopic ureter draining into it.¹²

The single ectopic ureter is often associated with a degree of renal dysplasia.¹³ In these instances, the ureteral bud, because of its abnormal cephalic position on the mesonephric duct, may fail to meet the mesonephric blastema properly and normal nephrogenesis is not induced.¹⁴⁻¹⁵ Fernandes and Gonzalez¹⁶ showed that most of the reported cases of non-duplicated ectopic ureterocele (34 of 36) in English literature were associated with ipsilateral non-functioning kidney. Johnson and Perlmutter⁶ reported a very high incidence of associated congenital anomalies in single system ectopic ureterocele. In the case of our patient the ipsilateral kidney was adequately functioning and any associated congenital anomalies were absent.

Approximately one-half of the girls with ectopic ureter suffer from continuous dribbling and incontinence despite a normal voiding pattern.¹⁷ GC can be found in the broad ligament, vagina and the vulva with variable clinical presentations like vaginal discharge, recurrent urinary tract infection, incontinence and dyspareunia in large cyst¹⁸ while small GC are usually asymptomatic.¹⁹

An ectopic ureter draining into a GC can be confused with an ectopic ureterocele²⁰⁻²² and can be differentiated from ureterocele by its lack of communication with the bladder, bladder neck or urethra; it does not obviously change in shape and size when intravesical pressure increases owing to bladder filling and voiding.²² The cyst is mostly located on the vaginal wall and may be connected to the vaginal cavity.²²

Classical radiological methods such as USG and IVU are usually performed to determine abnormal duplex kidneys and ectopic ureter.²³ VCUg is particularly important in providing further information about missing ureters. Although, 84% of ectopic ureters are diagnosed by intravenous pyelogram (IVP) during childhood²⁴ the upper moiety in some cases may not be detected by IVP because of the absence of upper pole calyx or because of poorly functioning very thin dysplastic and/or pyelonephritic parenchyma; hence further imaging modalities like CT/MRI²⁵ or dimercaptosuccinic acid (DMSA) scan may be required.

In the case of our patient VCUg was normal, and in IVU both kidneys and ureters were very well-delineated, hence we did not perform further costly investigations like MRI or DMSA scan because of the economic condition of the patient.

Endoscopy of the vagina and urinary bladder are advocated to detect ectopic ureter termination.²⁶ In cystoscopy we found absent left hemitrigone and left ureteric orifice so a vaginoscopy was performed, which showed ectopic orifice and bulging in left lateral wall with efflux of urine through it. However, MRI is the modality of choice for diagnosing this condition but GC can also be confirmed by direct cyst puncture and injection of contrast medium. It may show a communication with the genital tract.²¹ We also confirmed our diagnosis by negotiation of hydrophilic guide wire and retrograde contrast study through vaginal opening (figure 2A–C).

Kidney function status is the most important parameter while selecting the treatment option. Because of poorly functioning dysplastic kidney, most of the time nephroureterectomy is the procedure of choice.²⁷ In patients with significant renal function, either open or laparoscopic ureteral reimplantation is treatment of choice for a single system. For the single system ectopic ureter in women, laparoscopic ureteric reimplantation has been recently reported with good results.²⁸

But when the ectopic ureter opens into the GC the scenario may be different from the ectopic ureter. If the distal residual cavity of the large GC does not collapse it may be problematic owing to persistent discharge and in case of stenotic orifice there is risk of blind loop and abscess formation.

To the best of our knowledge we are reporting for the first time the laparoscopic ureteric reimplantation for single system ectopic ureter draining into GC. We performed laparoscopic reimplantation and suture ligation of the distal stump. Although clip ligation may be enough, there may be a risk of clip erosion in long run when we clip so close to vagina/bladder. Our patient was a big girl, so there was adequate space in pelvis and we did not encounter any problem in suture ligation, but in case of small children suturing may be difficult so a clip ligation would be better option.

Asymptomatic GC can be observed and surgery is usually indicated for symptomatic ones.²⁹ Surgical excision is frequently associated with bleeding owing to the natural vascularity of vagina and other postoperative complications like pain, haemorrhage and dyspareunia.³⁰ The large GC can be treated by various ways like aspiration and 5% tetracycline sclerotherapy³⁰ de-roofing or marsupialisation³¹ with reasonable results.

In our patient the orifice in vagina was large enough and visible (figure 1D), as we did retrograde study through it using 5 Fr catheter it dilated well with nice drainage, we thought not to incise and give it trial. It did work! and the patient did not develop any problem in follow-up. At 3-month follow-up the patient was completely dry and we performed vaginotomy which showed complete disappearance of the cystic bulge. So laparoscopic ureteric reimplantation and ligation of distal stump is a safe, feasible and adequate minimal invasive procedure for single system ectopic ureter draining into small-to-moderate size wide open mouth GC; however, in conditions of large GCs or stenotic mouth additional procedure like vaginal deroofing, marsupialisation or incision of cyst may be required.

Learning points

- ▶ Continuous urinary incontinence with normal voiding since birth in a female child or young adolescent should raise the suspicion of ectopic ureter and this should be considered the first differential diagnosis.
- ▶ Most common sites of ectopic ureter in women are the urethra and the vestibule but rarely may involve the vagina and the cervix.
- ▶ Ectopic ureter opening into vaginal Gartner's cyst (GC) can be seen as a bulge in lateral wall and the opening may be difficult to visualise; however, oral phenazopyridine may help in detecting the opening.
- ▶ Single system ectopic ureter is usually associated with ipsilateral renal dysplasia and congenital anomalies but may rarely happen with bilateral normal kidney and without any congenital anomalies. Treatment depends on ipsilateral renal function. In dysplastic kidney nephroureterectomy is the treatment of choice while in normal functioning kidney ureteric reimplantation should be performed. Laparoscopic ureteric reimplantation ensures faster recovery with good results. When opens into small to moderate size wide open mouth GC, conservative management after ligation of distal stump is sufficient but in conditions of large cystic mass or stenotic mouth deroofing/marsupialisation may be an option.

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