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Case Report

Vesicouterine fistula: Youssef's syndrome

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Introduction

Vesicouterine fistula is an uncommon pathological communication developing between the uterus or cervix and the urinary bladder most commonly after an iatrogenic injury during Caesarean section (83%–93% of cases)^{1–3} especially emergency Caesarean section. Youssef's syndrome classically presents as a triad of cyclical hematuria, amenorrhea and urinary continence, that is, there is no vaginal leakage of urine.³

Case report

A 28 year old female patient, para 1 presented with cyclical hematuria (menouria) and secondary amenorrhea following an emergency Caesarean section done for non progress of labor in

the second stage of labor 4 years earlier. There was no urinary incontinence, although the patient complained of occasional “wetting” of the vagina. She had hematuria after the Caesarean section that was treated with catheterization for two weeks. She later developed amenorrhea and cyclical menouria. The diagnosis was made four years after the delivery when she presented to the gynecologist with the above complaints. Physical examination done per abdomen and per vaginam was unremarkable except for a well-healed Pfannenstiel incision. There was no urine leak from the anterior vaginal wall or cervix during the speculum examination. The biochemical, hemato-logical and hormonal investigations were normal.

Ultrasonography of the abdomen and pelvis revealed no abnormalities. Based on a strong clinical suspicion of an iatrogenically created VUF, a magnetic resonance imaging (MRI) of the pelvis was done that revealed a well-defined, fluid-filled fistulous communication between the urinary bladder and the uterus (Fig. 1).

The cystoscopy and hysteroscopy revealed an opening approximately 2.0 cm in diameter in the supratrigonal region (Fig. 2) with irregular congested margins; a guide wire passed through this opening was hysteroscopically visualized in the supraisthmic region anteriorly. The uterine endometrium was thinned out.

She underwent a transabdominal, transvesical VUF repair. An initial cystoscopy was done and both the ureters were canalized with a 6F ureteric catheter. A 2.5 cm × 2.5 cm-sized fistula was identified in the supratrigonal region. A cystostomy was done. A racquet-shaped incision was made around the fistulous connection and was deepened up to the uterus. The urinary bladder was bivalved. The fistulous tract was excised and the opening of the uterus closed with No. 1

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Fig. 1 – Sagittal and axial turbo spin-echo T2-weighted MR images show an abnormal fluid filled communication (arrow) between the endometrial cavity and the urinary bladder.

polyglactin suture transversely. The urinary bladder was closed in two layers with No. 2 polyglactin and No. 3 polyglactin. The vesico-uterine peritoneum was closed to act as a transposition between the bladder and the repaired uterus. The abdomen was closed over a suprapubic catheter and a perurethral catheter was applied. She was given GnRH (Gosarelin Acetate) analogs. Her post-op recovery was uneventful and the suprapubic catheter (SPC) followed by the perurethral catheter (PUC) was removed after 3 weeks.

An MRI was repeated 4 weeks after the surgery to reassess the VUF. The MRI revealed a well-defined, hypointense scar on T2-weighted images (Fig. 3) in the region of the previously visualized fistulous tract. Presently the patient has resumed regular menstrual cycles and has no urinary complaints.

Discussion

Iatrogenic injury to the lower urinary tract is an uncommon (0.1%–0.3%) complication associated with Caesarean delivery.^{1–4} However, the causes of peripartum bladder and uterine injury resulting in fistula formation are nearly always iatrogenic. Risk factors include delivery in the late first or second stages of labor, wherein injury may arise because of difficulty or inadequate reflection of the bladder from the lower uterine

segment. Excessive intraoperative bleeding may also cause injury from attempts to achieve hemostasis, and may involve the distal ureter. Other risk factors include severe dystocia, forceps delivery, placenta percreta, manual removal of the placenta, uterine rupture and previous Caesarean section.^{3–7} An unrecognized bladder injury might persist leading to a VUF. The causes of a vesicovaginal fistula (VVF) are obstructed labor and hysterectomy, the former being the leading cause in developing countries with poor health care infrastructures and the latter in developed countries.

Youssef first described “Menouria” resulting from VUF in 1957.⁵ A classification of VUF based on the routes of menstrual flow has been proposed by Józwick and Józwick⁶ that divides VUF into three types. Type I, characterized by the triad of amenorrhea, menouria and complete continence of urine has been known as Youssef’s syndrome. Type II is associated with dual menstrual flow via both the bladder and vagina. Type III is associated with normal vaginal menses and lack of menouria.⁶ Our patient qualified into type I VUF; the presence of a supraisthmus fistula helped in maintaining continence, leading to a delay in patient presenting to the gynecologist and its subsequent diagnosis.

Hystero-graphy and cystoscopy remain the “gold standard” in the diagnosis. The use of intravenous urogram and cystometrogram for diagnosis have shown variable results in literature and are fraught with the risks associated with radiation and contrast media administration, and hence were not used for diagnosis in this case. Transabdominal ultrasound evaluation of urogenital fistulae, supported poorly by the limited data available has shown variable sensitivity in the diagnosis of genitourinary fistulae, with some case reports reporting positive identification of fistula in as low as 29% of cases.⁸ The ultrasound done in this case was essentially normal. The available data on MRI diagnosis of VUF is very limited.⁹ In a study with a small number of patients by Abou-El-Ghar et al,⁹ 100% sensitivity was achieved in the diagnosis of VUF. A few other authors have also reported accurate detection of VUF on MRI.¹⁰ The fistulous tract was exquisitely depicted by MRI in our case.

Conservative management with continuous bladder drainage for 4–8 weeks along with antibiotics and anticholinergics is recommended in cases where a small fistula is detected in the early or immediate postpartum or postoperative phase. The success rate of conservative management is less than 5%.⁷ Transvesical fulguration with continuous catheterization and hormonal treatment can be

Fig. 2 – Cystoscopic view of the fistula.

Fig. 3 – Sagittal and axial turbo spin-echo fat suppressed T2-weighted MR images shows post-op status with obliteration of communication between the endometrial cavity and the urinary bladder by hypointense scar tissue (arrow).

successful for the treatment of VUF with a small, well epithelialized orifice, though the results in the presence of extensive scarring are poor and recurrence is high.¹¹ Open surgical management with transabdominal correction management has good results^{7,11,12} as it involves excision of the tract. Transpositioning a graft gives better results. In our case, we interposed the vesical peritoneal fold.

The patient made a full clinical and anatomic recovery, confirmed by the post-op MRI and has no ongoing irritable bladder symptoms or incontinence. Clinical assessment and interval MRI have been suggested for follow-up. Careful monitoring will be required in subsequent pregnancies as there is a small but potential risk of scar dehiscence and/or recurrent fistula formation.

Youssef's syndrome, an uncommon if not rare entity has been presented here reiterating the fact that meticulous practice of obstetric and surgical principles during Caesarean section and other obstetric interventions can prevent formation of these fistulae.

Conflicts of interest

All authors have none to declare.

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