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Correspondence

An unusual case of post-cesarean vesicouterine fistula (Youssef's syndrome)

Dear editor,

A vesicouterine fistula (VUF) is a medical condition characterized by an abnormal communication between the anterior wall of the uterus and the posterior wall of the bladder.[1] VUFs are categorized as one of the least common types of urogenital fistulas (UGFs), affecting only 1%~4% of individuals.[2] When a woman exhibits symptoms of cyclic hematuria, medical practitioners should be particularly attentive as this may be indicative of a VUF.

VUF was once believed to be a complication of assisted delivery methods such as vacuum and forceps techniques.[3] However, today, VUFs are predominantly caused by cesarean deliveries and specific gynecological procedures, accounting for 83%~93% of cases.[4] The main symptoms of VUF include cyclic hematuria, amenorrhea, and urinary incontinence. Youssef's syndrome classically presents as cyclical hematuria, amenorrhea, and urinary continence,[5] which was first described in the literature in 1938, and later named after Dr. Abdel Fattah Youssef in Egypt in 1957.[6] Although conservative management has been reported, surgical intervention is typically required to treat the condition. This article presents a single case of VUF that occurred after a second cesarean delivery, causing secondary amenorrhea and cyclic hematuria without urinary incontinence. The discussion focuses on reviewing and updating this rare condition, as reported in the literature.

A 27-year-old woman, who had undergone two cesarean deliveries and had a six-month history of amenorrhea and cyclic hematuria, was referred to the gynecology outpatient clinic. The patient did not report any incontinence. She had undergone a cesarean section one year prior to her current visit, during which she had severe adhesions of the uterus and bladder that were successfully repaired. The patient had a urethral catheter for two weeks after the procedure, and no voiding difficulties were reported upon its removal. However, six months later, the patient began experiencing episodes of cyclic hematuria without any menstrual blood flow. Between hematuria attacks, urine analyses were normal, and urine cultures were sterile. To determine the location of the fistula, a urogynecological examination using Computed Tomography (CT), along with cystoscopy and hysteroscopy, were performed.

During the cystoscopy, a fistula measuring 1/5 cm was discovered, located on the supraregional portion of the posterior bladder wall, with uneven and congested edges (Fig. 1). Additionally, a Methylen Blue injection was seen running through the supra isthmic area during hysteroscopy, and the uterine endometria was thinned anteriorly. After explaining the clinical condition to the patient and her family, laparotomy was chosen as the preferred approach. The patient opted for an extraperitoneal approach via a Pfannenstiel (PF) incision to reach the bladder, as she wished to preserve her fertility.

During surgery, the suprapubic region was explored, revealing significant adhesions between the superior and posterior regions of the bladder and uterus. Although there was no apparent bladder rupture, the necrotic fistula tract was excised following the bladder's removal. After discovering the fistula focus between the bladder and uterus, the uterine mucosa and bladder were sutured in two layers using interrupted 3-0 polyglycolic acid sutures. The suture lines showed no signs of leakage, and sterile methylene blue solution was used to control any potential leaks. The bladder was drained with the assistance of urethral and suprapubic catheters. To prevent recurrence, the omentum was inserted between the bladder and uterus. The catheters were removed three weeks later without any complications. A CT urography was performed four weeks after the procedure to evaluate the VUF. The patient was discharged on the fifth postoperative day and remained symptom-free after 40 days and six months of observation.

VUF is the rarest form of genitourinary fistulas, and it is mostly associated with cesarean section. It can occur immediately after the procedure, during the puerperium period, or after multiple surgeries.[4] The first case was reported in 1908.[7] During a cesarean section, direct trauma, inadequate downward mobilization, or improper suturing can cause bladder damage.[8] Patients typically report urine incontinence, which can be intermittent or persistent and may be associated with hematuria. VUF that develop gradually may become infected, devascularized, clamped, or develop hematomas in the bladder. After a cesarean section, the bladder becomes firmly attached to the uterus, and during subsequent vaginal delivery, it is subjected to extreme pressure, which can result in the formation of a VUF to thin the lower segment during labor. Studies have shown that multiple cesarean sections can damage the bladder's vascular network, leading to gradual devitalization and scarring.[9]

Based on the clinical presentation, Jówik proposed a classification system for VUF in 2000, which divides them into three categories[10] (Fig. 2). Type I presents with symptoms of amenorrhea, menouria and complete continence of urine, which has been known as Youssef's syndrome. While Type II presents dual menstrual flow via both the bladder and vagina. Type III presents with normal vaginal menses, without menouria.[10] In our case, the clinical presentation aligns with Type I of this classification system.

VUF can also arise as a result of dilatation and curettage after hysteroscopy, although this is a less common cause. Risk factors for VUFs include the inability to fully separate the bladder from the uterine segment, excessive intraoperative bleeding, use of forceps and vacuum, placenta previa totalis, abnormal placental insertion (such as acreta, inreata, and percolate), uterine rupture, prior cesarean section, and a history of multiple abortions. Additionally, VUFs can be caused by endometriosis, inflammatory bowel conditions, migration of intrauterine

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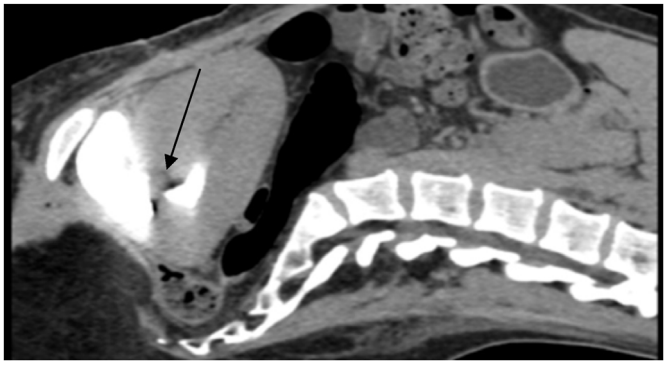

Fig. 1. CT of cystoscopic view of the fistula (arrow indicates the fistula).

devices, bladder infection, and congenital defects, although these are less common causes.[4] In our case, both the cesarean delivery bladder and uterine adhesion may have contributed to the development of the VUF.

The location of the fistula tract in a vesicovaginal fistula has an impact on the likelihood of persistent incontinence. Depending on how a healthy cervix functions as a sphincter, menouria memoria can occur without urine incontinence. In our case, there was no urinary inconsistency. Diagnosing a VUF can be more challenging when urine incontinence is not present. There have been reports of delayed presentation of symptoms. For instance, Ugurlucan et al.[11] reported a case of a 55-year-old woman who presented with urine incontinence 30 years after having had a cesarean section. They suggested that a VUF should be considered as a differential diagnosis for urinary incontinence. However, accidental cases and congenital development defects have also been documented in the literature.[12]

Various diagnostic techniques, including cystoscopy, cystography, intravenous pyelography, saline infusion sonohysterography, hysterosalpingography, helical computed tomography, and MRI, can be used to diagnose VUF. Radiographic screening techniques can aid in surgical planning.[13] Cystoscopy is a critical tool for detecting the presence of a fistula, identifying its location, and understanding its interaction with the trigone, although it may not accurately pinpoint the fistula tract's upper pole. In our case, a cystoscopy, CT scan, and hysteroscopy were utilized to determine the diagnosis and plan the surgical treatment.

Various treatments are suggested for VUF, including surgical resection through open abdominal surgery, laparoscopic correction, vaginal correction, and robotic surgery. Surgical treatment may be performed through various routes, including vaginal, transvesicaltransversal, transperitoneal, laparoscopic, and robotic.[14] The selection of the surgical approach depends on the location of the fistula and the surgeon's expertise in non-conventional routes. In this study, the surgical treatment was performed through open laparotomy. Laparoscopic and robotic techniques have success rates comparable to open surgery in treating VUF, but they offer greater postoperative patient comfort and earlier discharge. Thus, minimally invasive procedures are superior to open surgery in terms of patient comfort and recovery time.

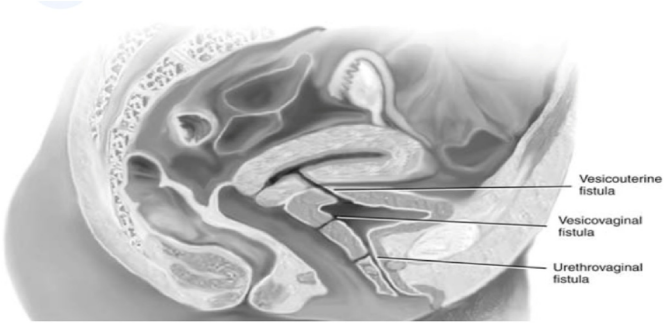

Fig. 2. Vesicouterine fistula classification [10].

Ethical approval

This study was approved by the Ethics Committee of Urmia University of Medical Sciences with code: IR.UMSU.HIMAM.REC.1401.090.

Consent to participate from patients

The patients provided written informed consent before reporting the disease.

Author contributions

All authors contributed equally to this paper.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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