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## Post caesarean vesicouterine fistula: A rare entity

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### Abstract

**Introduction:** Vesicouterine fistulas (VUF) are an uncommon and pathological connection between the uterus and the bladder. Although rare, they are usually related to caesarean section.

**Case Report:** A 28 year old female, P<sub>2+1</sub>, consulted in gynecology OPD for a 5 years history of episodes of painless cyclical hematuria (menouria) every month that started immediately after her last caesarean section. Her past obstetric history was significant for one previous caesarean section. After admission and confirming the diagnosis of vesicouterine fistula by cystoscopy and CT scan pelvis, surgery was done. The surgical repair included a fistulectomy with primary uterine and bladder repair followed by omental flap interposition. The recovery was uneventful with no recurrence of symptoms.

**Conclusion:** This case highlights the fact that although vesicouterine fistulas are uncommon, but the diagnosis should always be on top of mind in patients presenting with menouria and/or urinary incontinence associated with a history of caesarean section.

**Keywords:** Vesicouterine fistula, Youssef's syndrome

### 1. Introduction

Vesicouterine fistulas (VUF) are the least common type of urogenital fistulas, comprising 1 - 4% of all urogenital fistulae<sup>[1]</sup>, and are defined as an uncommon communication between the urinary bladder and uterus<sup>[2]</sup>. It is mostly noticed after obstetrical intervention. In the past it was a rare complication of prolonged vaginal birth or with the use of forceps. Nowadays, with the wider indications of caesarean section, the most common is the post-caesarean section vesicouterine fistula. It comprises 83-93% of all cases<sup>[3]</sup>. Most result from emergency caesarean sections, or from previous caesarean sections, even after a long period. It may have an early or delayed presentation. Patients usually experience urinary incontinence (intermittent or continuous) which may be associated with hematuria<sup>[1]</sup>. The diagnosis is usually made with cystoscopy, cystography and/or computed tomography (CT) scan<sup>[4]</sup>. It is usually recommended to delay surgical repair<sup>[5, 6]</sup>. However, recent cases demonstrate successful fistula closure with conservative management<sup>[4, 7]</sup>.

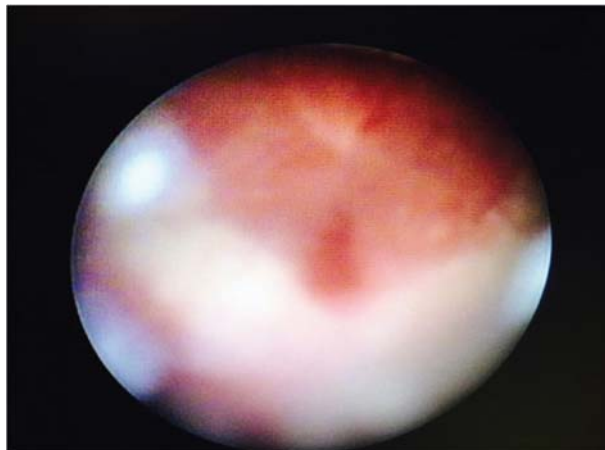
### 2. Case Report

A 28 year old female, P<sub>2 L 2</sub>, consulted in gynecology OPD for a 5 years history of painless cyclical hematuria (menouria) that started immediately after her last caesarean section, done 5 years back and amenorrhoea for 4.5 years. She had hematuria in the immediate postoperative period that was not taken care of. She was in lactational amenorrhoea for the next 5 months and thereafter she resumed her menses in the form of painless cyclical hematuria and no vaginal bleeding. There was no associated urinary incontinence. She had repeat LSCS for previous one LSCS (8 years back) and fetal distress. On examination, general examination was normal. Systemic examination was within normal limits. External genitalia was normal. Speculum examination showed normal vaginal wall and cervix with no urinary leakage. Per vaginum examination – Uterus was anteverted, parous size, bilateral fornices free and non tender. After initial workup, transvaginal ultrasound was done and noted was focal defect in posterior urinary bladder wall and a fluid filled tract (width 4.4 mm), extending to the endometrial cavity at the site of LSCS scar (Figure 1). Further, CECT abdomen and pelvis was done and the findings were bulky and elongated uterus with LSCS scar and focal soft tissue thickening at the level of scar at the interface of uterus and posterior urinary bladder wall with focal small outpouching in posterior bladder wall.

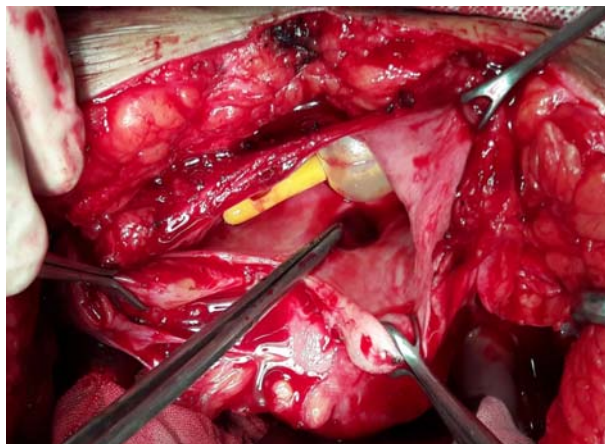
The findings were highly suggestive of Youssef's syndrome. Next, cystoscopy was done and showed a fistulous opening on posterior urinary bladder wall above trigone (Figure 2). After discussion and obtaining informed written consent from the patient, surgical intervention was planned. The surgical repair included a fistulectomy with repair of defect in uterus and bladder followed by omental flap interposition between the uterine repair and the bladder closure (Figure 3). The postoperative period was uneventful with no recurrence of symptoms.



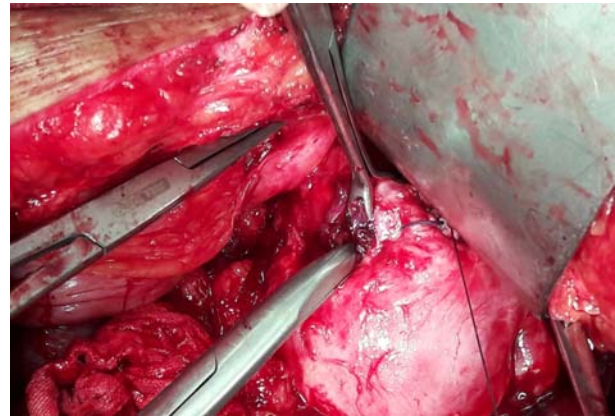
**Fig 1:** TVS showing a fistulous tract between uterus and urinary bladder wall



**Fig 2:** Cystoscopic view of fistula site on posterior urinary bladder wall



a) Fistula opening in urinary bladder



b) Fistula opening in anterior surface of uterus

**Fig 3:** Perioperative view

### 3. Discussion

Vesicouterine fistulas represent 1% to 4% of all urogenital fistulas, with a peak incidence in young women between 25 and 33 years old [5, 8, 9]. The exact epidemiology is not well known. The rise in prevalence may be explained by the increase in number of cesarean section and vaginal delivery following previous cesarean section.

The main causes of urogenital fistulas can be classified as obstetrical, surgical, radiation necrosis or related to a malignancy. Currently, the main cause of VUF is an iatrogenic injury during cesarean section which accounts for 83–88% of cases [5, 8]. These injuries occur more often after repeated caesarean sections [8, 9]. Some risk factors have been advocated for the development of VUF, such as an inadequate reflection of the bladder from lower uterine segment, accidental placement of sutures through the bladder wall, excessive intraoperative bleeding, severe dystocia, forceps delivery, vaccum delivery, manual removal of the placenta, placenta percreta with bladder invasion, uterine/bladder rupture after obstructed labour, and dilatation and curettage (criminal abortion). Other less frequent causes are intrauterine device migration, bladder tuberculosis and congenital lesions [2, 10]. It has also been documented after Shirodkar and Mc Donald cerclage as well. Repeated caesarean sections may result in progressive devitalization and scarring of the uterus and bladder base by damaging their vascular network, thus predisposing to fistula formation [4, 7].

The symptoms of vesicouterine fistula are dependent upon the level of fistula. This can be explained by the sphincteric mechanism of the uterine isthmus and the different pressure gradients [11]. During the menstrual cycle a change is seen in the shape and diameter of the isthmus lumen. The menstrual blood accumulates in the uterine cavity and when the pressure rises above 25–30 mmHg, the sphincter of the isthmus relaxes and a bloody discharge occurs. If the fistula is above the level of the isthmus, the blood present in the uterine cavity flows freely into the bladder causing menouria. There is no accumulation of blood in the uterine cavity, so the cavity does not distend and the pressure does not increase, thus the sphincter of the isthmus fails to relax, producing the classical combination of symptoms of menouria, amenorrhoea with a patent cervical canal and absence of urinary leakage, known as “Youssef syndrome”, which comprises > 90% of all cases, as in this case, the patient showed the classical features of Youssef's syndrome.

This syndrome was reported in 1957<sup>[11]</sup>. However, if the fistula is located below the level of isthmus, the menstrual blood, not only flows into the bladder but also through the cervix into the vagina, thus, having normal menses along with menouria. Conversely, when submitted to high pressure in the bladder, urine leaks through the fistula into the uterine cervix and vagina.

Patients with VUF can have early or delayed clinical presentations. Usually, immediate presentation occurs when there is direct injury to the bladder during surgery. Patients can have early hematuria and/or urinary leakage as in this case, voiding difficulty, low-grade pyrexia, urinary sepsis or be completely asymptomatic<sup>[10]</sup>. Delayed presentation can occur when there is an infection or a progressive devitalization of the posterior wall of the bladder<sup>[5, 9]</sup>. Patients with delayed presentation often have symptoms of urinary leakage from the vagina if the cervix is incompetent, cyclic hematuria (menouria) and amenorrhea<sup>[5, 9]</sup>.

Accurate and early diagnosis of vesicouterine fistula can be difficult, since there are many different clinical pictures. There are multiple means of investigation for VUF and several examinations may be required to confirm the diagnosis. When there is macroscopic hematuria and/or urinary leakage early in the postoperative period of cesarean section or other gynecological procedures, early exclusion of VUF should be done<sup>[1]</sup>. The mainstays of diagnosis are cystoscopy and urinary tract imaging<sup>[1]</sup>. A cystoscopy may be necessary to detect a fistula tract and to evaluate location, size and proximity to ureteral orifices<sup>[1]</sup>. The diagnosis of VUF can be made indirectly by injection of methylene blue in the bladder. A VUF is present if dye comes out of the cervix. Radiological evaluation remains the gold standard evaluation for diagnosis of urogenital fistulas. The integrity of the upper urinary tract should also be evaluated with an intravenous pyelography, a retrograde pyelography or a pyridium test<sup>[12]</sup>. A cystography may demonstrate a tract or retrograde filling of the uterine cavity. During hysterography, if a VUF is present, the radio-opaque solution fills the bladder. Additional diagnostic procedures include CECT, MRI and TVS with or without Doppler (abnormal fluid in the uterine cavity)<sup>[2, 10]</sup>.

Treatment options for VUF include conservative, medical or surgical treatment<sup>[5, 9, 13, 14]</sup>. Some studies advocate conservative treatment for early diagnosed small fistula with bladder catheterization alone with antibiotics for 3 weeks<sup>[1]</sup>. Others proposed induced amenorrhea by oral contraceptive pills or by GnRH analog because epithelial and stromal cells that contain sex hormone receptors (as in the endometrium) had been observed within the fistula,<sup>[1]</sup> thus, allowing the fistulous tract to heal and close by reducing tract menstrual flow. This may be useful especially in Youssef syndrome. This management technique may be less successful in women with a mature tract (6 weeks or longer). The ideal length of treatment is unknown, but most reports treated the patient for 6 months.<sup>[1]</sup> There are also some reports describing the endoscopic fulguration of the fistula.<sup>[12]</sup> Spontaneous closure of the fistula in up to 4% of cases may be expected with the involution of the uterus.

Surgery is the definitive treatment and is the treatment of choice in most cases, especially for large vesicouterine fistulas which invariably necessitate surgical closure<sup>[5, 12]</sup>. Hysterectomy is not always necessary but is the preferred one if uterus preservation is not desired. If uterus

preservation is desired, uterus sparing surgery could be done as in our case. The first effort at surgical closure of the fistula is the most likely to succeed. Surgical repair of VUF are performed by different approaches, which include vaginal, transvesical, transperitoneal and laparoscopic or robotic procedures<sup>[15]</sup>. A bladder injury diagnosed during the Cesarean section should be repaired preferably with tissue interposition during the same surgery to prevent fistula development and the need for repeat surgeries in an already fibrous environment. When there is urinary leakage and pain related to VUF, early diagnosis and treatment are warranted to avoid social and familial embarrassment.<sup>16</sup> It is usually recommended to delay surgery up to 3 months after the causative surgery to allow spontaneous closure of the fistula with the involution of the uterus and to have less inflammation making the surgery easier to perform with a lower risk of complication<sup>[5, 6]</sup>. However, if it is suspected that the fistula is large or the patient has pronounced abdominal pain then immediate repair may be preferred to expedite recovery and minimize impact on quality of life during a period of expectant management.

#### 4. Conclusion

The important learning point from this case was that presence of painless menouria with amenorrhoea must raise suspicion of vesicouterine fistula for early diagnosis and treatment to improve patients quality of life. Vesicouterine fistulas are uncommon, yet they are becoming more prevalent due to changes in modern obstetrical care. They should always be on top of mind in patients with a history of cesarean section or who have experienced a gynecological procedure associated with signs of hematuria and/or urinary leakage. As the old proverb says, "Prevention is always better than cure", meticulous care of bladder during caesarean section even in the presence of adhered bladder may prevent its formation.

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