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Post-cesarean Vesicouterine Fistulae: A Report on a Case and an Update of the Literature

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ABSTRACT

Vesicouterine fistula (VUF) is an abnormal communication between the posterior wall of the bladder and the anterior wall of the uterus. Although it represents only 1 to 4% of all urogenital fistulae, its prevalence is increasing all over the world because of the large indications of cesarean section [1-3]. VUF usually presents with urine leak, amenorrhoea, and cyclic hematuria. It has a considerable marital and social impact and may cause serious urogenital infections. Herein we report on one more case of VUF following cesarean section with review and update of the recent literature regarding the aetiological, diagnostic, and therapeutic aspects of this entity. Some suggestions are mentioned concerning surgical repair and how to minimize their occurrence.

INTRODUCTION

Vesicouterine fistula (VUF) is an abnormal communication between the posterior wall of the bladder and the anterior wall of the uterus. Although it represents only 1 to 4% of all urogenital fistulae, its prevalence is increasing all over the world because of the large indications of cesarean section (CS) [1-3].

VUF usually presents with urine leaks, amenorrhoea, and cyclic hematuria. It has a considerable marital and social impact and may cause serious urogenital infections.

Herein we report on 1 more case of VUF following cesarean section with a review and update of the recent literature regarding the aetiological, diagnostic, and therapeutic aspects of this entity. Some suggestions are mentioned concerning surgical repair and how to minimize occurrence.

CASE REPORT

A 28-year-old uniparous woman presented with involuntary continuous urinary leakage by vagina, secondary amenorrhea, and cyclic hematuria occurring at intervals of 26 to 28 days, lasting 3 to 4 days, and causing serious psychological distress. She had undergone a cesarean section 2 years previous. She begun complaining of urinary leakage exactly 15 days after surgery. During this period, she developed several urinary tract infections.

Upon physical examination the abdomen and the vagina were apparently normal; the uterus had normal size with no tenderness, vaginal bleeding, or discharge. Both the external urethral meatus and the urethra were normal. There was no

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case report

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Figure 1. **Hysterosalpingography**: Early filling of the bladder cavity with contrast material confirming the communication between bladder and uterine cavity.
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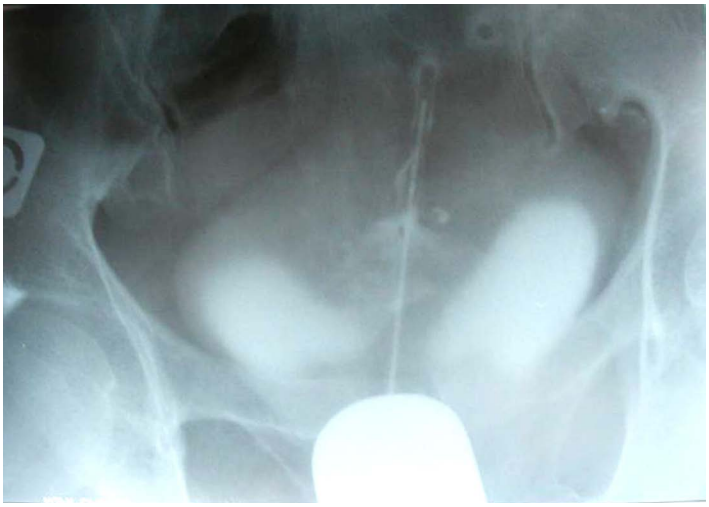

Figure 2. **Preoperative view**: The fistulous opening was catheterized with a Foley catheter.
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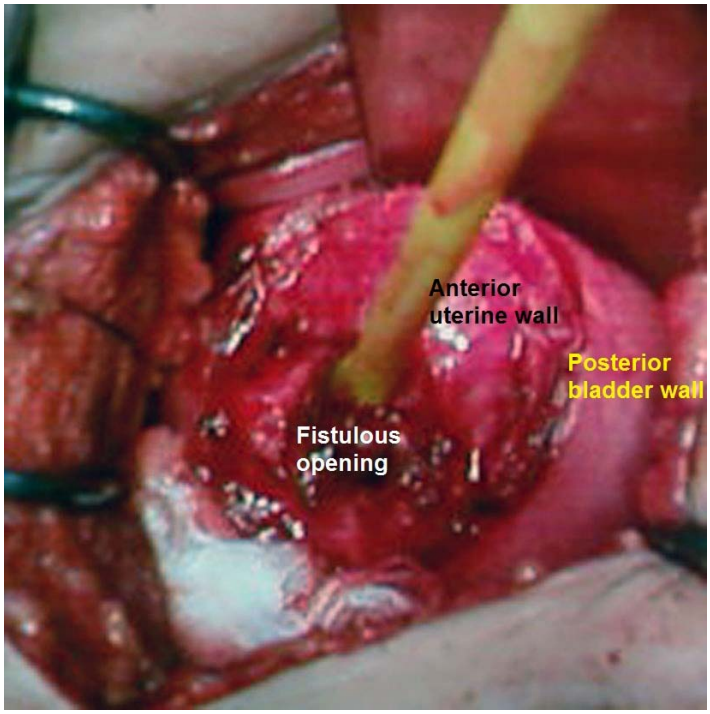

urine leakage from the anterior vaginal wall or cervix during the speculum examination.

Standard laboratory data, urinalysis, and urine culture (out of the time of hematuria) showed no abnormalities. Ultrasonography of the urinary tract showed posterior bladder-wall thickening only.

The intravenous urography was unremarkable. A hysterosalpingography demonstrated contrast with the bladder and suggested a VUF (Figure 1). After instilling into the bladder a solution of methylene blue, the speculum examination showed leakage of the dye from the external orifice of the cervix.

Cystoscopy revealed a fistulous opening in the supratrigonal area above the interureteric ridge. It was a large defect, which was 10 mm in diameter and surrounded by hyperaemic, exophytic, and edematous mucosa. The 2 ureteric orifices were normal.

The diagnosis of VUF following cesarean section was carried out. We decided to close the fistulous tract surgically through a suprapubic transvesical approach.

The bladder was densely adherent to the uterus. The fistulous opening was catheterized with a 20 F Foley catheter (Figure 2). The fistulous track was sharply dissected and included surrounding cuffs of scar tissue from both the bladder and the wall of the uterus. The defect of the uterine wall was closed by 2 layers of interrupted sutures of 2-0 Vycril (Figure 3). The bladder was closed by 2 layers of interrupted sutures of 2-0 Vycril. Bladder resistance was checked by filling the bladder with 200 ml of sterile saline solution. The abdomen was then closed in the standard fashion. The bladder was drained by a 22 F Foley catheter.

The postoperative course was uneventful, and the patient was discharged on the second postoperative day with the Foley catheter. Histology of the specimen revealed inflammatory changes with no evidence of malignancy. A cystogram was done on the 21st postoperative day, which did not reveal any extravazation confirming a complete closure of the fistulous tract, and hence the Foley catheter was removed. A cystoscopy done at that time also showed a well-healed scar.

Figure 3. **Preoperative view**: Suture of the defect of the uterine wall (green arrow).
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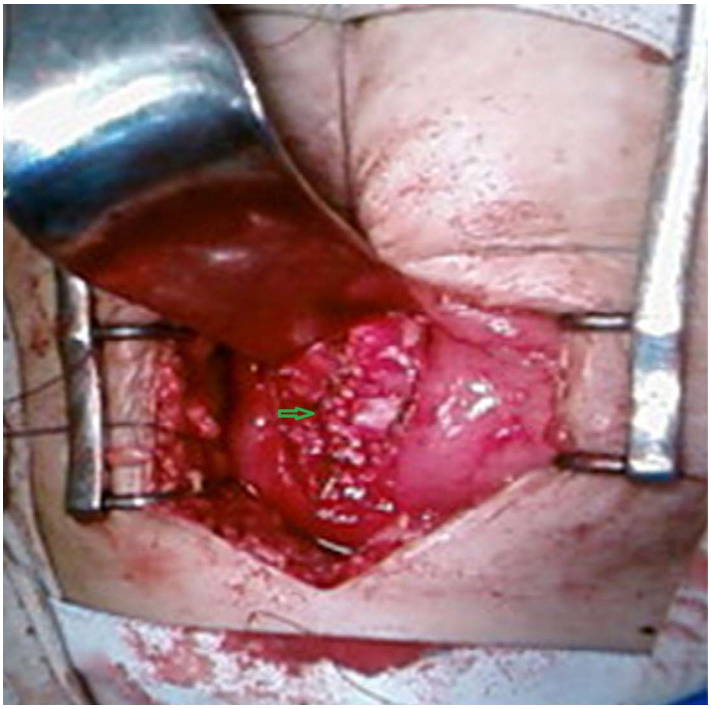

The patient started menstruating 3 months after surgery. Actually, within a 24-month follow-up, she reported no difficulty in urination, complete urinary continence, normal menses, and no hematuria, but she continued consulting with her husband for secondary infertility.

DISCUSSION

Genitourinary fistulae (GUF) are common in developing countries because of the higher incidence of obstetric and iatrogenic gynecological complications. Fistulae are still far from being rare in the Maghreb countries, such as Algeria. They are a sensitive parameter of the social, economic, and medical conditions in these populations.

VUF is one of the least common types of GUF, accounting for only 1 to 4% of all cases [3]. It is a rare complication most commonly associated with cesarean section [4]. Its incidence has been increasing, secondary to a corresponding increase in the use of low segment cesarean section (LSCS) [5]. Fistulae are encountered either after the first CS or after a repeat one [5].

VUF may be due to endometriosis, contraceptive devices, malignant tumors, inflammations, rupture of the uterus and bladder after obstructed long labor, or radiation therapy and iatrogenic trauma of intermittent bladder self-catheterization [6,7]. They also occur following high vaginal forceps-aided delivery, external cephalic version, curettage or manual removal of the placenta [8], placenta percreta, myomectomy [5], uterine artery embolization, tuberculosis of the genital tract, and brachytherapy for carcinoma of the cervix [1,5,8,9].

Most VUFs occur when the bladder is pushed down vigorously with the finger, resulting in trauma to the bladder that may end a late VUF formation. Also, it could result from aberrant suturing of the lower segment and inadvertent inclusion of the bladder wall [1,5,10,11].

After a cesarean section, the bladder gets so adherent to the uterus that it is subjected to severe tension during a vaginal delivery, which may result in VUF formation for thinning of the lower segment during labor [2,8]. Delayed VUF formation may result in infection, ischemia, clamping, or hematoma of the urinary bladder [1].

The fistulous communication is usually reported, as in our patient, between the posterior supratrigonal part of the bladder, the anterior lower segment of the uterus, or more rarely in the cervix [12]. For the fistula opening above the internal cervical opening (os), the menstrual blood might be forced to flow into the bladder because of the functional valve made by the isthmic sphincter, which also gets sufficient pressure to prevent urinary leakage from the vagina [12]. In addition, the menstrual blood may be forced to flow into the bladder when the internal uterine orifice is occluded by granulations on the posterior uterine wall or by cervix fibrosis, which may also prevent urine flow into the vagina [13]. When the fistula is located below the isthmus, the menstrual blood accumulates normally in the uterine cavity, and the menstrual blood passes as it should through the cervix into the vagina and not through the fistula into the bladder.

The isthmic sphincter may be unable to prevent urinary flow from the cervix during micturition because of the increased intravesical pressure, which gets higher than the intrauterine one [11,14]. Urinary incontinence also occurs if the level of the VUF is at or below the internal os or if the os is incompetent [10]. Total vaginal urinary leakage, without any relation to the voiding or filling phase, occurs when the fistula opens ahead of the internal cervical os [11].

Spontaneous disappearance of the menstrual blood flow into the bladder will occur with the onset of menopause [9].
VUFs impair fertility, which can be partially restored after closure of the fistulous track [15].

VUF may only manifest with amenorrhoea, cyclic hematuria, urinary vaginal leakage, or all. Urinary incontinence may be total, intermittent, continuous, or transient. Menouria with apparent amenorrhea may present late, usually a few weeks postpartum or when menses recover. According to the routes of the menstrual flow, fistulae may present with menouria (type I), with dual flow via both the bladder and the vagina (type II), and with normal vaginal menses (type III) [16]. Our patient had a type II VUF. The symptoms can appear early after surgery, as in our case, or months or even years later [15].

Generally, one can easily make the correct diagnosis of VUF by relying on the patient's clinical history. Amenorrhea, cyclic hematuria without urinary incontinence in combination with a history of LSCS, has been described as pathognomonic of VUF [10]. In our case, scant urine leaks occurred even in the presence of competent os with fistula communicating with the uterus above the isthmus. A pelvic examination in the early postoperative period shows only an enlarged, subinvoluted uterus. A speculum examination may show urinary leakage from the external os of the cervix [12]. A nonspecific clinical presentation and negative findings on examination lead to a considerable delay in diagnosis [14].

Usually in UVF, methylene blue instilled into the bladder is detected in the vagina or if after instilled into the uterine cavity it appears promptly in the urine. In addition, methylene blue instilled through catheterization of a visible lesion in the bladder wall can confirm the fistula. This test, however, does not show directly the fistulous tract and its specific location. Moreover, this test can be negative in patients with a long and tortuous tract [17].

Definitive diagnosis can be achieved by cystography and hysterosalpingography [10], computed tomography (CT) and magnetic resonance imaging (MRI) may be indicated in difficult cases [8,9,18].

Although VUFs are difficult to diagnose sonographically, Park et al. reported that sonography can demonstrate the fistulous tract as double echogenic lines between the endometrium of the anterior wall of the uterine body and the mucosal layer of the posterior wall of the bladder.

Transvaginal sonography revealed an abnormal hypoechoic line connecting the endometrial cavity with the bladder wall that appeared encased in the uterus and was focally disrupted [14]. However, sonography has inherent difficulty in differentiating the VUF tract from different patterns of an uncomplicated cesarean scar [14].

Cystography usually shows an abnormal communication between the uterus and the posterior wall of the bladder [18]. Sometimes it can be falsely normal with no filling of the uterine cavity [9].

The final diagnostic clue will be given by the hysterosalpingography. It shows the fistulous track as an abnormal communication between the uterus and the bladder. In Tancer's review of published reports, he found that hysterography was the most reliable diagnostic technique [10].

When a low VUF is present with communication below the isthmus, helical CT after IV contrast injection is a good method to show the fistula [19]. When a high VUF is suspected, helical CT with sagittal reformation gives more information about the precise topography of the fistulous tract [15]. The MRI provides a better definition against the surrounding tissues [20]. A high-resolution, T2-weighted MRI clearly demonstrates the hypointense fistula tract and hyperintense endometrial cavity due to the stagnation of urine. The MRI can denote the exact position of the fistula and surrounding anatomy [5,20]. The injection of a contrast medium in the uterus or the bladder is necessary to show the fistula and to visualize the fistulous tract. It is very useful in the diagnosis of VUF with atypical clinical presentation, obviating the need of invasive conventional radiographic contrast examination.

Cystoscopy, even when repeated, can fail to confirm the fistula [17]. On the other hand, when the fistulous tract is wide, it can be diagnosed easily. It shows on the posterior wall of the bladder a retrotrigonal, irregular hole or depression that may be surrounded by edema and inflammation of the urothelial mucosa [9,13,15,18,20].

A complete urological workup by radiological and endoscopic investigations is important prior to surgery to assess the relationship of the fistulous tract with the sphincter and ureters.

A differential diagnosis must always be made between VUF and vesical endometriosis, which may also appear following a lower segment cesarean section and may cause cyclic hematuria. In these cases, cystoscopy and vesical biopsy may help establish the correct diagnosis. Other forms of urinary incontinence after delivery should be excluded such as vesicovaginal as well as the

ureterovaginal fistulae [2,10].

Treatment options for VUF following cesarean section range from conservative treatment to surgical repair [2]. Rarely, patients refuse any kind of treatment because of the benignity of symptoms and prognosis of the disease.

When detected soon after surgery or delivery, conservative management may be tried since there is a good chance for spontaneous closure of the fistulous track [1,2,15]. This is done with continuous bladder drainage for 4 to 8 weeks [21]. In larger fistulae, significant reduction in size may occur. This treatment includes nutritional improvement, anemia correction, and the treatment of any urinary infections [12].

A recent review of world data showed a high efficacy of hormonal manipulation by the induction of amenorrhea in the treatment of VUFs. In fact, VUFs were first hormonally regulated due to the fistulous canal being lined by endometrium [22]. Hormonal management should be tried first [2]. A combination of levonorgestrel and ethinyl estradiol daily for at least 6 months or analogues of LH-RH are suggested [23]. The effectiveness of hormonal treatment varies. A small fistula may be totally closed, but a large fistula invariably necessitates surgical closure [12].

Endoscopic treatment, which is indicated for small fistulae, consists of destroying the epithelium covering the fistulous track. The fistula opening is cannulated with a ureteral catheter. The fistulous tract is thoroughly fulgurated with a 6 F fulgurating electrode. Foley catheter drainage is left indwelling for 6 weeks [12].

Despite these different therapeutic options, surgery is still the mainstay and definitive treatment of VUFs [1], and 95% of patients will need it. The timing of surgery is important [8]. Injuries to the bladder discovered while performing cesarean sections or a laparotomy for a ruptured uterus should be immediately repaired [1,15]. When discovered later, fistulae should not be repaired before all sloughing, edema, and inflammatory reactions have subsided. Generally, surgical repair is performed 3 to 4 months after a cesarean section because uterine involution has taken place at that time, and the reaction to initial surgery should be minimal [1].

Surgical repairs of VUFs are performed by different approaches, which include mainly extraperitoneal transvesical and transperitoneal routes [24]. The extraperitoneal transvesical approach has the advantage to lead easily and directly to the orifice of the fistula in the bladder [1,11]. The bladder is opened and the hole of the fistula is located. The ureters are catheterized to prevent their damage, and a probe is introduced into the fistula. The bladder is sharply dissected away from the anterior wall of the uterus and incised down to the fistula, which is resected [12].

In the abdominal transperitoneal approach, the fibrous tissue obliterating the vesicouterine fossa is sharply excised and the bladder completely freed. After opening the bladder posteriorly, utereteric catheters may be inserted bilaterally. The fistula is dissected and excised to include the surrounding cuffs [12].

In both approaches, the bladder and the uterus are closed separately in 2 to 3 layers with absorbable, interrupted, and tension-free sutures of Vycil (3-0 or 2-0). The omentum may be mobilized and interposed between the suture lines as a protective barrier, obliterating dead spaces [20,24]. A myouterine flap can be utilized to reinforce the repair if the omentum is of insufficient length or absent [25]. The bladder is drained by a transurethral or suprapubic catheter for at least 2 weeks. Urine cultures before surgery and appropriate antibiotic therapy before and after operation are mandatory.

There is no specific indication for hysterectomy [2]. It could be indicated in selected cases such as multiparous women nearing menopause or a patient who has hereditary or uterine diseases requiring radical surgery.

Recently, numerous reports of laparoscopic [21,26] and robot-assisted surgical repair of these fistulae have shown that these techniques can be used with efficacy and safety with added advantages for less pain, lower blood loss, shorter hospital stays, reduced morbidity, better cosmesis, and quicker recovery than open surgery [26-28]. However, the steep learning curve and the high cost of robotic surgery are limiting factors.

Effective and successful treatment of VUFs is followed by the disappearance of vaginal leakage and/or menouria with the recovery of normal menses [8,15,18,20,26]. The pregnancy rate after VUF repair is reported at 31% with a 25% rate of term deliveries [29].

The best treatment is preventive by the improvement of obstetric care and the quality and technique of surgery [30]. VUF may be prevented by emptying the bladder as well as by carefully dissecting the lower uterine segment. Intraoperative diagnosis is the gold standard in detecting VUFs for allowing immediate repair. Porcaro et al. propose intraoperative sonography for suspecting bladder injury while dissecting the uterine lower

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segment, and for the Foley transurethral catheter-producing bloody urine [12]. It is advisable that after VUF repair, delivery should be performed by repeating a cesarean section due to the risk of fistula recurrence [2,12].

The hope is that with better hospital care, improvement in the quality of medical practices, and the improvement of socioeconomic status in the Maghreb countries, the incidence of GUF will be reduced

CONCLUSION

VUF is a rare entity, mostly iatrogenic after cesarean section. Diagnosis is evoked clinically and confirmed by hysterography. Treatment is often surgical with an excellent cure rate.

The best treatment is prevention based on faster access to care and improving the quality of surgery.

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