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Vesicouterine Fistula Following Vaginal Birth After Cesarean in a Woman With Grand Multiparity: A Case Report and Review of the Literature

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ABSTRACT

Vaginal birth after cesarean (VBAC) is rare in multiparous women in the developed world. A vesicouterine fistula (VUF) is a rare complication of VBAC. The present case is the first known report of a patient with grand multiparity who presented with a VUF after her fourth VBAC. A tentative diagnosis was made following her cystogram on postpartum day 1; the definitive diagnosis was reached with cystoscopy on postpartum day 4. Successful uterine-sparing repair of the fistula was obtained via laparotomy with fistula closure and an omental flap. A review of potential diagnostic and management methods is presented.

INTRODUCTION

The safety of vaginal birth after cesarean section (VBAC) in women with a prior vaginal delivery is well established [1]. Although grand multiparity (≥ 5 deliveries) associated with VBAC is rare, particularly in developed countries, it does occur [2]. Dyack et al [3] examined VBAC in women with grand multiparity and found an increased risk of serious complications, including 4 out of 45 cases (9%) with uterine scar dehiscence or uterine rupture. It is thought that with scar dehiscence or rupture, the shearing force may be transmitted to the bladder and cause the subsequent development of a fistula [4].

A vesicouterine fistula (VUF) remains an uncommon type of urogenital fistula, but its prevalence has been rising [5]. These types of fistulas can occur spontaneously after vaginal delivery following a previous cesarean section. Few cases have been described after VBAC, and none of these reported cases were grand multiparous [6-8]. Theoretically, multiparous women are at risk because the combination of grand multiparity and a prior uterine scar creates 2 risk factors: uterine rupture and VUF formation. We describe for the first time a patient with grand multiparity presenting with a VUF after her fourth VBAC, thereby illustrating the need for continued awareness of this potential complication.

CASE REPORT

A 45-year-old woman (gravida 7, para 5) presented for induction of labor. Gestation was at 38 weeks and 6 days, secondary to advanced maternal age. Her obstetric history was significant for a low-transverse cesarean section performed 15 years earlier because of breech presentation, followed by 4 successful vaginal births.

KEYWORDS: Vesicouterine fistula; VBAC; Vaginal birth after cesarean.

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Abbreviations and Acronyms

CT = computed tomography
IVP = intravenous pyelogram
MRI = magnetic resonance imaging
VBAC = vaginal birth after cesarean section
VUF = vesicouterine fistula

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case report

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Delivery

The patient received dinoprostone followed by oxytocin (Pitocin; JHP Pharmaceuticals LLC, Rochester, MI, USA). An amniotomy was performed and an epidural was placed for analgesia. She progressed to complete dilatation. As she started pushing, recurrent moderate to severe variable fetal heart decelerations were noted. Her expulsive efforts were diminished due to lack of sensation, and the decision was made to perform an operative delivery. A vacuum extractor was placed with the fetal vertex at the plus 4 station. There was one pop-off with the first pull and the fetal vertex delivered with the second pull. The anterior shoulder delivered, followed by a gush of blood. The patient delivered a female infant weighing 3153 grams. A first-degree perineal laceration was repaired with a 3-0 polyglactin 910 suture.

Identification of the VUF

On the evening of postpartum day 0, the patient began to complain of "gushing" drainage from her vagina that increased when she squatted to void. This was followed by what appeared to be blood-tinged urine that saturated a Peri-Pad (Versalon; Covidien, Mansfield, MA, USA) every 1-2 hours. A transurethral catheter was placed, which decreased the amount of vaginal drainage (although her urine remained lightly blood-tinged). The following morning, urology was consulted for her persistent hematuria.

A cystogram performed on postpartum day 1 demonstrated immediate extravasation of contrast into a collection that was superior to the dome of the bladder (possibly within the endometrial canal) and into the vagina (Figure 1). The patient was conservatively managed with a transurethral catheter and discharged home on postpartum day 2 with the catheter in place and instructions to return as an outpatient for follow-up examination. On postpartum day 4, an outpatient cystoscopy was performed. The test identified an opening within the posterior bladder wall. This was followed by an operating-room cystoscopy on postpartum day 9, where indigo-stained saline placed within the bladder was identified as leaking from the cervix, confirming a VUF.

Management of the VUF

Conservative management was attempted with the catheter in place for 6 weeks. The fistula did not resolve and, because the patient desired to preserve her fertility, a primary surgical fistula repair with an omental flap interposition was performed via laparotomy. During surgery, the posterior bladder wall was dissected from the anterior lower uterine segment. A defect approximately 1.5 cm in size was discovered at the junction of the lower uterine segment and cervix. It was closed in a running-locked fashion with 2-0 polyglactin 910 suture. The bladder defect was then closed in 2 layers: the mucosal layer was closed with 2-0 polyglactin 910, and the detrusor layer and serosa were closed in a similar fashion. The bladder was then filled and the closure appeared to be watertight. Two

Figure 1. Cystogram Performed on Postpartum Day 1, Demonstrating Immediate Extravasation of Contrast.

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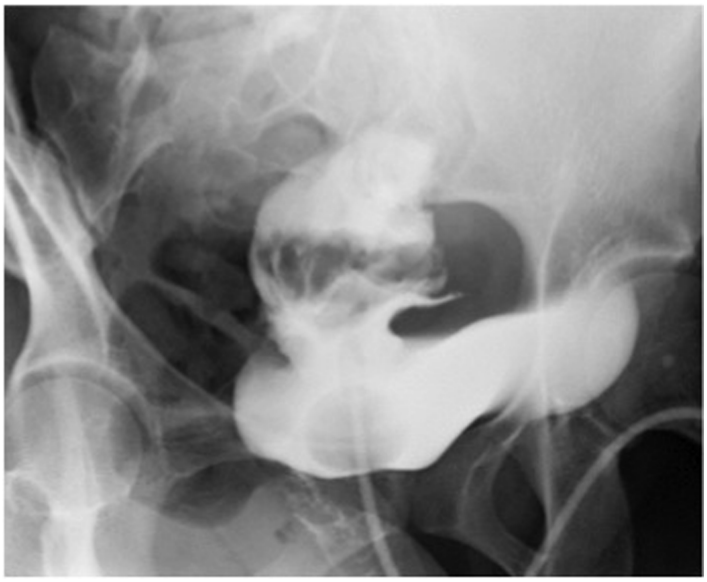

Figure 2. Cystogram Performed 4.5 Weeks After Surgery Showing an Intact Bladder.

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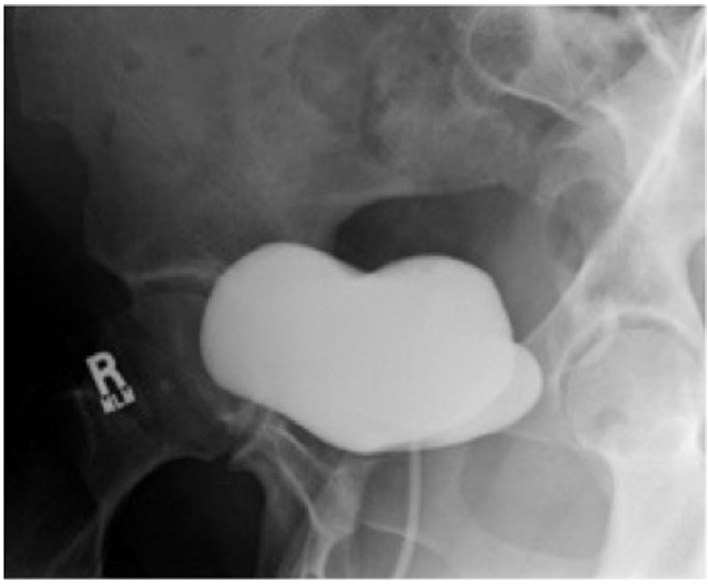

sutures of 2-0 polyglactin 910 were then placed at the lowest point of the uterine closure. The omentum was mobilized, brought down, and secured between the bladder and uterus. Cystoscopy was performed and the bladder defect appeared closed. A transurethral catheter was replaced at the conclusion of the surgery.

The patient had an uneventful postoperative period and was discharged home on postoperative day 2 with no complications. She was given a 5-day course of ciprofloxacin 500 mg orally, twice daily. A repeat cystogram (Figure 2) performed 4.5 weeks after the surgery showed a normal, intact bladder. The catheter was removed on postoperative day 31.

DISCUSSION

Vesicouterine fistulas are among the most rare types of urogenital fistulas [5]. With a VUF, the most common abnormal passage is one connecting the posterior supratrigonal part of the bladder to the anterior lower segment of the uterus [9]. The present patient developed a VUF despite having had multiple successful vaginal births after cesarean. The VUF likely resulted from shearing forces on the uterine scar (and subsequently, the bladder) that occurred with delivery of the infant's anterior shoulder. A tentative diagnosis was made following the patient's cystogram on postpartum day 1, with the definitive diagnosis reached with cystoscopy on postpartum day 4. Although conservative management was attempted, surgery was required to repair the defect that developed during her vaginal delivery. Surgical treatment is contingent on the fertility desires of the patient. In this case, uterine preservation was accomplished with a primary surgical fistula repair with an omental flap interposition via laparotomy.

Presentation of a VUF

Cases of VUF have been documented after cesarean section, VBAC, brachytherapy for cervical cancer, uterine artery embolization for symptomatic uterine fibroids, infections of the genital tract, pelvic surgery, traumatic bladder catheterization, and use of intrauterine contraceptive devices [6, 9-12]. A review of the literature reveals that VUF as an obstetric complication occurs most often in women with a history of cesarean delivery; from 1947-1986,Tancer [5] reported that 54 of 74 VUFs followed cesarean section. Youssef [13] described the classic presentation of a VUF, which includes cyclic hematuria (termed *menouria*), apparent amenorrhea, and urinary continence (because the cervix acts as a sphincter, stopping the flow of urine at the level of the internal os). In the case of a patent cervix, as is the circumstance after vaginal delivery, a patient may present with constant urine leakage [14]. Parulekar [15] termed the immediate postpartum transvesical passage of puerperal lochia in those patients with a VUF as *lochiauria*. Our patient initially presented on postpartum day 0 with vaginal leakage of urine and lochiauria.

Diagnosis of a VUF

High clinical suspicion can lead to an early diagnosis. Urogenital fistulas can be detected by various diagnostic tests including the methylene blue dye test, cystography, hysterography, transvaginal ultrasound, sonohysterography, intravenous pyelogram (IVP), computed tomography (CT), magnetic resonance imaging (MRI), and cystoscopy. With the methylene blue dye test, a transurethral catheter is used to instill methylene blue into the bladder. Using a speculum for direct visualization, dye seen leaking from the cervix confirms the fistula, but does not directly show the fistulous tract or its location [9,16]. Cystography and hysterography are typically successful in detecting the presence of a fistula with extravasation of contrast medium into the uterus or bladder, respectively; however, the tract is often difficult to localize. For this reason, Smayra et al [16] suggested using a helical CT with sagittal reformation after hysterography to obtain precise location and details of the fistulous tract.

Huang et al [17] described the transvaginal ultrasonographic features of a VUF after performing the scan 1-week postpartum in a patient with a suspected VUF. They found a mixed echogenic area in the lower segment of the uterus with loss of continuity of the anterior wall. Four weeks postpartum, another ultrasound was performed and the mixed echogenic area had become anechoic with a hyperechogenic focus, which was determined to be air bubbles within the fistulous tract. Ramamurthy et al [18] supported the use of sonohysterography, using saline infusion into the uterine cavity and a transvaginal ultrasound to detect the VUF. They visualized the saline jetting into the bladder from the uterus through the fistula. This simple, inexpensive procedure (which is similar to transvaginal ultrasound) was well tolerated and effective in locating and diagnosing the fistula with no reported side effects. In addition to sonography, use of Doppler ultrasound may be helpful in detecting the abnormal flow of fluids between the bladder and uterus through the fistula [9].

Intravenous pyelogram, also referred to excretory urography, will show a normal upper urinary tract and be helpful in excluding ureteral involvement [16]. Similarly, CT and MRI may be helpful in excluding involvement of other adjacent organs. Some authors argue that CT and MRI are more precise in visualizing the extent and location of the fistulous tract. Murphy et al [19] described the use of T2-weighted MRI to diagnose a VUF, stating that the multiple contiguous sections

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provide a three-dimensional picture of the fistula. Although this is a noninvasive test with clear advantages of providing detailed images that demonstrate the full extent of the fistula, it still may be considered an expensive diagnostic test for a VUF.

Attempts to directly visualize the fistula with cystoscopy may show a defect, depression, or opening in the posterior bladder wall usually above the trigone with surrounding edematous mucosa; however, findings may be negative [9]. A fistulogram can be performed if a lesion is visualized in the bladder during cystoscopy, with passage of a ureteral catheter into the tract and injection of contrast material [9].

Each of the diagnostic modalities offers advantages and disadvantages. The methylene blue dye test, cystography, and hysterography are noninvasive and inexpensive tests that are useful for initial evaluation and diagnosis; however, they are not adequate for determining the location and extent of the fistula. For this reason, additional inexpensive and simple studies such as transvaginal ultrasound and sonohysterography have been shown to be effective in localizing and diagnosing a VUF. CT scan, MRI, and IVP can be used to rule out involvement of the upper urinary tract and adjacent organs. Additionally, preoperative imaging and endoscopic evaluation are needed to determine the relationship of the fistulous tract with the trigone and ureters, the degree of damage, and the position of the fistula; evaluation is also needed to determine local tissue viability and suitability for planned management and surgical repair [9].

Management of a VUF

There are many management approaches to the treatment of a VUF. Spontaneous closure with continuous bladder drainage occurs in approximately 5% of VUFs [20]. In the postpartum period, continuous bladder drainage allows complete uterine involution and reduction in inflammation [9]. Novi et al [21] placed an indwelling catheter for 5 weeks immediately postpartum for a patient with a VUF. A subsequent CT scan after removal of the catheter was negative for a VUF, and the patient remained asymptomatic at 6 months postpartum.

Other conservative measures include hormonal therapy (eg, medroxyprogesterone, combined oral contraceptives, leuprorelin acetate), which has been used to induce amenorrhea by hormonal manipulation and been found to be an effective, noninvasive, and conservative method of VUF management [20-24]. It is believed that lowering menstrual flow across the fistula allows for healing. Eogan and McKenna [22] used continuous combination oral contraception for 6 months in a patient with a VUF, and subsequent menstruation was normal with no further menouria after completion of treatment. Ravi et al [23] used continuous bladder drainage with a transurethral catheter and endometrial suppression with medroxyprogesterone 20 mg daily by mouth for 2 months to prevent epithelialization of the fistulous tract with endometrium. The patient was also given antibiotics (trimethoprim-sulfasoxazole) while the catheter remained indwelling. Four weeks after initiating this therapy, a repeat cystography showed complete resolution of the fistula. Similarly, Jozwik and Jozwik [20] reported treating a patient with 500 mg of medroxyprogesterone twice a week for 3 months for VUF management. Follow-up examination 2.5 years later showed that the patient remained asymptomatic. In a review of spontaneous closure of VUFs, Jozwik and Jozwik [20] reported that 8 out of 9 patients (88.9%) experienced spontaneous healing with hormonal manipulation. Yokoyama et al [24] used subcutaneous monthly injections of leuprorelin acetate (an LHRH analog) for 6 months to treat a patient with VUF; treatment resulted in amenorrhea. Two months following cessation of her treatment, the patient had a normal menstrual cycle with no menouria, and at 15 months follow-up she was still asymptomatic.

Molina et al [25] recommended fulguration of the fistula tract and destruction of the epithelium of the fistula, followed by prolonged transurethral catheter drainage for 6 weeks, as a simple, effective, and low-risk option for management. However, there has been documented recurrence after cystoscopic fulguration [26].

Although conservative and noninvasive methods have been proven effective in management of VUF, surgery remains the mainstay of treatment. Because access to the fistula is not possible through a vaginal approach, surgical correction is done through a transabdominal approach. Selection of the type of surgical procedure performed is based on a woman's desire for future fertility. A total abdominal hysterectomy with excision of the fistula and primary closure of the posterior bladder can effectively repair a VUF if the woman no longer desires to bear children. If the woman desires future fertility, uterine preservation is possible. Placement of an omental flap between the bladder and uterus or vaginal cuff is done to reduce the risk of fistula and adhesion formation. Kumar et al [27] first described this technique of abdominal repair with mobilization and interposition of omentum between the bladder and uterus. Most other post-VBAC repairs utilize this method with good success.

With the advancement of minimally invasive surgical procedures, laparoscopic and robotic repairs of VUFs have been successfully performed. When compared with previous methods, they

have resulted in decreased pain, shorter hospitalization stay, quicker recovery time, and better cosmesis [28,29]. Following sharp dissection, the fistulous opening is sutured closed and omentum is interposed between the uterus and bladder for adequate healing, followed by 3 weeks of Foley catheter drainage [28,29].

In the past, surgical repair has been delayed until 3 months postpartum, allowing the uterus to involute and return to normal size and inflammation to decrease [9]. A proposed approach is to correct VUF earlier, to prevent social and emotional stress to women with menouria and incontinence [8].

Prognosis

Pregnancy rates following VUF repair, recently reported as 37.5%, are encouraging [30]. In the literature reviewed, all deliveries following VUF repair were done via cesarean section. At our own institution, another patient has had a successful pregnancy following VUF repair. She underwent a 3-layer closure of the bladder, uterine defect repair, and placement of a piece of thick AlloDerm Regenerative Tissue Matrix (LifeCell Corporation, Branchburg, NJ, USA) in place of omentum. In this patient, omentum could not be mobilized because she had a prior gastric bypass and ventral hernia repair. She conceived approximately 10 weeks after repair and delivered at term via classical cesarean section.

In summary, although a VUF is a rare type of genitourinary fistula, there are many risk factors for fistula formation including an obstetric complication following low transverse cesarean sections, VBAC, and operative deliveries. Initial diagnosis of the fistula can be made by simple, noninvasive tests including the methylene blue dye test, cystograms, and hysterograms. Additional inexpensive workup to locate the fistula includes transvaginal ultrasound, sonohysterography, and direct visualization with cystoscopy. At our institution, cystogram is often the initial test of choice and our recommendation for diagnosis. Although conservative management has proven to be effective, surgical therapy remains the definite option. Minimally invasive approaches, including laparoscopic and robotic repairs of VUF, should become a standard of care. Although more surgeons are leaning toward minimally invasive approaches, the recommendation for surgical management is dependent on surgeon experience and comfort level. Future studies may compare the various treatment options and their effectiveness in management of VUF based on fistula size, location, and time to presentation.

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