NYU Langone Health

Department of Urology
Case of the Month

Iatrogenic Vesicouterine Fistula Presenting with Intermittent Gross Hematuria and Secondary Infertility

CASE PRESENTATION

A 38-year-old female, gravida 2 para 2, was referred with intermittent gross hematuria for 6 years. She began noting blood in the urine following her second cesarean delivery. The hematuria was cyclical, coinciding with her menses, and lasted 3 to 5 days each month. She denied associated dysuria, urgency, or frequency. She reported occasional watery discharge, particularly when her bladder was full or when walking but denied overt urinary incontinence. She denied any significant urologic or gynecologic history and reported 2 prior emergent cesarean deliveries performed for arrest of descent. She was otherwise healthy, without major medical comorbidities. Her only medication was a daily prenatal vitamin. She was a lifelong nonsmoker with no risk factors for urologic malignancy.

The patient had been attempting pregnancy unsuccessfully for 5 years and had undergone an extensive infertility workup including 2 hysterosalpingograms that demonstrated a proximal tubal occlusion at the right uterine cornua, a patent left fallopian tube, and a normal uterine cavity. Her husband's semen analysis was within normal limits. She had failed a trial of ovulation induction with clomiphene citrate. In 2021, the patient underwent a saline infusion sonogram at an outside provider's office that revealed a possible cesarean scar defect (Figure 1) and was referred to NYU Langone Urology for further evaluation and management.

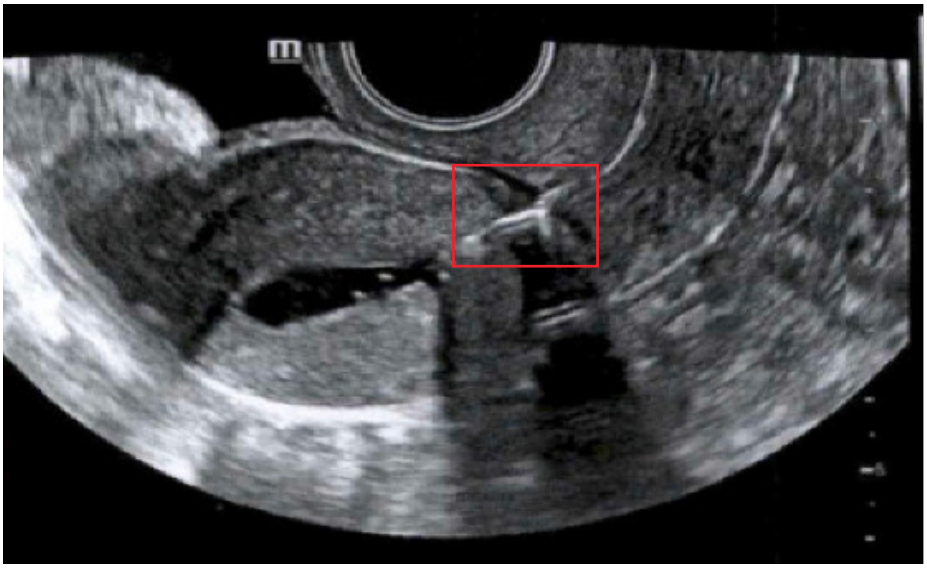

Figure 1. Saline infusion sonohysterogram depicting a possible defect in the anterior lower segment of the uterine wall, possibly the fistulous tract (red box).With permission from C. Brandon and L. Stewart, 2021.

EVALUATION

The patient's pelvic exam was within normal limits, without abnormal discharge or pooling of fluid in the vagina. The vaginal epithelium was intact. The cervix was normal in appearance, without efflux of fluid.

The patient underwent diagnostic cystoscopy, which was notable for a 1 x 1 cm midline defect roughly 2 cm cephalad to the trigone and remote from the ureteral orifices (Figure 2). Bilateral ureteral efflux was visualized. The flexible cystoscope passed easily through the defect, allowing visualization of the uterine fundus and the bilateral tubal ostia, confirming a diagnosis of vesicouterine fistula (VUF).

The patient was offered surgical repair using either an open or a robot-assisted laparoscopic approach. The patient opted for a robotic repair with interposition of an omental J-flap.

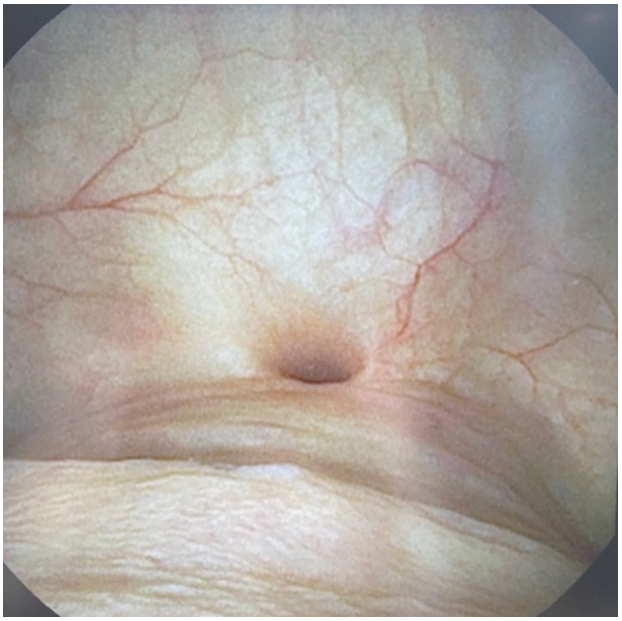

Figure 2. Preoperative cystoscopic evaluation of the bladder revealing a 1 x 1 cm midline supratrigonal defect. With permission from C. Brandon and L. Stewart, 2021.

MANAGEMENT

The patient was taken to the operating room, where she was placed in the dorsal lithotomy position. Cefazolin 1 gm IV was administered for intraoperative antibiosis. The patient was prepped and draped in normal sterile fashion. A cystoscopy was performed with a 30-degree lens, and a supratrigonal 1 x 1 cm midline defect was again identified. A Sensor guidewire was inserted into the defect and pulled out through the cervix with a Kelly clamp. This was done to allow easy identification of the fistulous tract throughout the case. A urethral catheter was inserted and immediately clamped to allow backfilling of the bladder for the initial dissection.

Following peritoneal access and port placement, the omental J-flap was initiated laparoscopically using a LigaSure bipolar cautery device. After adequate mobilization, the distal aspect of the J-flap was tagged with a suture which was brought up through the left lower quadrant abdominal wall with an Endo Close device so that the flap would remain easily accessible once the patient was in steep Trendelenburg position.

The patient was placed in steep Trendelenburg and the robot was docked. Dense adhesions between the uterus and the anterior abdominal wall were lysed with electrocautery and sharp dissection. Lysis of adhesions allowed identification of the vesicouterine plane. This plane was developed until the location of the fistulous tract was identified with both a spill of urine from the bladder and the visualization of the previously placed guide wire. Wide mobilization of the vesicouterine plane with margins of at least 2cm lateral and distal to the tract was carried out to insure a tension-free repair of the tract. At this point, the guide wire was removed. The epithelialized portion of the uterine defect was excised and the hysterotomy was closed in 2 layers with 2-0 V-Loc suture.

Attention was turned to the cystotomy. Direct visualization of the ureters was possible, which confirmed the bilateral efflux and that the cystotomy was at least 2 cm above the trigone. Minimal tissue from the epithelialized edges of the fistulous tract was excised with care not to unnecessarily widen the cystotomy. To minimize tension on the repair, the bladder was mobilized from the anterior abdominal wall by entering the retropubic space and dissecting to the level of the pubic symphysis. The first layer of the cystotomy repair was full thickness bladder wall reapproximated with interrupted 4-0 Vicryl sutures. The bladder was backfilled with 300 mL sterile saline mixed with methylene blue and the repair was found to be watertight. The second layer of the repair was an imbricating layer of bladder muscularis accomplished with running 2-0 Vicryl suture. A Foley catheter was placed to gravity.

The omental J-flap was liberated and secured with 4 interrupted sutures of 2-0 Vicryl between the uterine and bladder closures to prevent overlapping sutures and provide well-vascularized tissue to the site. The robot was undocked. The patient was taken out of the Trendelenburg position and the flap was ensured to be tension-free.

The patient was discharged home on day of surgery with a catheter in place for 10 days. She underwent a voiding cystourethrogram that demonstrated no extravasation from the bladder wall and complete bladder emptying. She was initiated on combined oral contraceptive pills for 3 months to prevent pregnancy during healing of the hysterotomy. Six weeks postoperatively, she reported normal menstruation without any hematuria and no further watery discharge. An office cystoscopy was performed that demonstrated a completely healed urothelial lining (Figure 3).

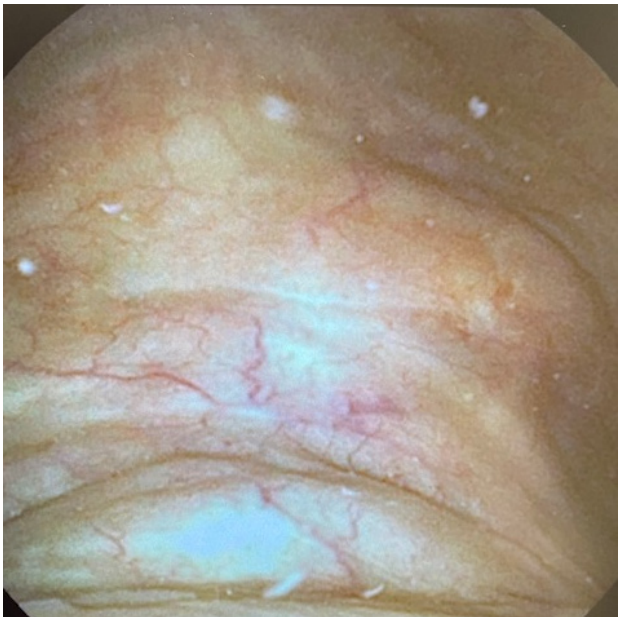

Figure 3. Postoperative cystoscopic evaluation revealed a healed, intact urothelium without defect. With permission from C. Brandon and L. Stewart, 2021.

COMMENT

Genitourinary (GU) fistulae are abnormal connections between a reproductive organ and a urinary tract organ, such as the bladder and the uterus (e.g., a VUF.) The overwhelming majority of GU fistulae are found in women in developing countries and are sequelae of obstructed labor and delayed access to medical care.[1] An estimated 130,000 new GU fistulae occur annually, and more than 2 million women are currently living with GU fistulae.[2,3] Obstructed labor leads to prolonged compression of the pelvic soft tissue between the fetal head and the bony pelvis, resulting in ischemia and eventual necrosis.[4] Tissue breakdown and subsequent scarring and fibrosis ensue. The level of injury reflects the level at which the fetal descent arrests in the birth canal. Often, the fetal descent arrests relatively distally, involving the trigone of the bladder, the bladder neck, or the urethra and resulting in a large vesicovaginal or urethrovaginal fistula surrounded by poorly vascularized tissue. This leaves patients with socially stigmatizing and debilitating urinary incontinence (or fecal incontinence in the case of rectovaginal fistulae).

In the developed world, access to cesarean delivery in the event of obstructed labor has nearly eradicated obstetric fistulae. The true incidence of GU fistulae in this setting is unknown, but rates are estimated at 0.13% to 2%.[5] Most GU fistulae are secondary to iatrogenic injury (90%) at the time of pelvic surgery, with 75% of injuries occurring at the time of a hysterectomy.[6] Although rates differ by study, it has been suggested that bladder injury occurs in 1% to 5% of hysterectomies, while ureteral injury is slightly less common, at 0.2% to 2.5%. The risk of injury increases with distortion of anatomy, intraoperative hemorrhage, significant adhesive disease, large masses, malignancy, and history of radiation. The route of surgery may also affect risk of urologic injury and subsequent GU fistula formation. Only 9.6% of GU fistulae reportedly occur following a recognized injury, highlighting the importance of prompt identification and repair of urinary tract injury.[7]

The most common GU fistula is a vesicovaginal fistula (VVF). In industrialized nations, this occurs mostly after hysterectomy at the level of the vaginal cuff. Typically, bladder defects are supratrigonal and midline or medial to the ureteral orifices. These fistulae often result from inadvertent incorporation of the bladder into the vaginal cuff closure or from delayed thermal injury at the time of colpotomy. Radiation is another important cause of VVFs, with studies indicating that up to 2% of women irradiated for cervical cancer will develop a VVF.[8] Finally, nonabsorbable mesh, particularly transvaginal mesh kits, have been associated with VVFs, with an estimated incidence of 9%.[9] The classic presenting symptoms of a VVF include continuous urinary incontinence regardless of position, as well as possible dysuria, recurrent urinary infections, and hematuria. These symptoms typically will present within 10 days of a surgery or may coincide with removal of a Foley catheter.[10] The diagnosis can be made with a thorough pelvic exam, looking for vaginal pooling. The bladder can be backfilled with methylene blue saline to observe any blue pooling in the vaginal vault. A cystoscopy should be performed to evaluate the location of the defect in relation to the ureteral orifices as well as to evaluate the defect's size and complexity. Upper tract imaging of the ureters should always be performed—by computed tomography with a delayed excretory phase, intravenous pyelogram, or retrograde pyelogram—to evaluate for concomitant ureteral injury, which is estimated to occur up to 25% of the time.[2] Repair of VVFs can be performed vaginally or abdominally and can employ minimally invasive techniques including laparoscopic and robot-assisted laparoscopic closures. The decision to pursue one route over another is multifactorial, including the location and size of the fistula, vaginal access, the proximity to the ureters and need for concomitant procedures, and the surgeon's comfort. Ultimately, all repairs must adhere to the principles of fistula repair, which are listed in Table 1.

Table 1. Basic Principles of Fistula Repair

Principle	Comment
Evaluate for concomitant injury	• Consider upper tract imaging to evaluate ureters and other necessary investigations to confirm extent of surgical repair
Rule out malignancy	• In patients with history of malignancy, radiation, or any suspicious lesion, biopsy of tract or lesion should be performed
Position for adequate exposure	• Consider split-leg position to allow access to vagina and transurethral route to bladder
Mobilize margins for tension-free closure	• Ensure adequate separation of bladder from uterus with adequate margins for tension-free closure of each layer
Avoid overlapping sutures	• Consider perpendicular suture lines on uterus and bladder • Consider use of interposing vascularized tissue flap or graft
Ensure watertight closure	• Absorbable suture is preferable • Confirm integrity of closure with retrograde fill • Reinforce areas of leak with suture or tissue glue
Ensure continuous bladder drainage	• Adequate bladder drainage (i.e., transurethral and/or suprapubic catheter) • Consider confirming integrity of bladder closure with postoperative cystogram prior to removal of catheter

A VUF, which is an abnormal connection between the bladder and the uterus, is one of the rarest GU fistulae, accounting for approximately 1% to 9% of all GU fistulae. VUFs are most commonly associated with cesarean delivery.[11] Vaginal birth after cesarean delivery (particularly if complicated by uterine rupture), manual extraction of the placenta, induced abortion, misplaced intrauterine device (IUD), brachytherapy, dilation and curettage, pelvic tuberculosis, and congenital anomalies are other known causes. Women with VUFs often present with a constellation of symptoms referred to as Youssef's syndrome or Youssef's triad and characterized by cyclical hematuria (menouria), amenorrhea (absence of vaginal bleeding), and watery vaginal discharge. First trimester losses and absence of urinary incontinence have also been described as part of Youssef's syndrome. The timing of presentation depends on the etiology of the VUF, with direct injury to the uterus and/or the bladder resulting in a sooner presentation than VUF secondary to obstructed labor resulting in necrosis and tissue breakdown.

Diagnosis should start with a high index of clinical suspicion, followed by a detailed examination. One can look for expulsion of urine from the cervical os. This can be achieved again with backfilling of the bladder and observing blue-stained saline spilling from the cervix. A cystoscopy can evaluate the bladder defect, often found to be supratrigonal, and delineate the relationship of the defect to the ureters. The cystoscope can often pass safely into the uterine cavity, allowing visualization of bilateral tubal ostia. Similarly, saline infusion sonograms and hysterograms may be able to identify a uterine wall defect and extravasation into the bladder, but this identification may be more operator-dependent and still requires a high level of suspicion.

Management of VUFs is typically surgical given that only about 5% of VUFs have been successfully managed with continuous bladder drainage.[12] Case reports have indicated that in patients with amenorrhea, hormone therapy for at least 6 months may have a role in fistula closure.[13] However, successful closure is less likely in mature fistulous tracts. Cystoscopic fulguration of the tract has been described for small fistulae, but such procedures are rarely successful.[14]

With respect to surgical repair of VUFs, timing following the inciting event is important and yet controversial, as with all fistula repairs. Some advocate for early closure to reduce psychosocial burden on the patient, while others advocate for a minimum wait of 3 months to allow inflammation to settle. Should the VUF follow a cesarean delivery, as is often the case, some authors advocate for a 3- to 4-month wait to allow the uterus to fully involute prior to repair.[12] The most common surgical approach is abdominal, now increasingly being replaced by minimally invasive techniques. A transperitoneal approach enables complete mobilization of the bladder and the uterus from one another, which is crucial for a tension-free fistula repair (Table 1). In one study of 17 patients with a VUF undergoing a transperitoneal abdominal approach, the success rate was 100%.[12] However, follow-up time was not specified. In a series of 3 patients undergoing robot-assisted laparoscopic repair of a VUF, all were symptom-free at follow-up of at least 3 months.[15] It is known that VUFs are associated with infertility. One case series noted a pregnancy rate of 31.25% and a term birth rate of 25% following closure of a VUF.[16] It is recommended that patients give birth via scheduled cesarean delivery given the risk of uterine rupture, a risk extrapolated from the myomectomy population. Patients should also be counseled regarding the risk of recurrence; prior case series have reported this risk to be as high as 20% over a 25-year period.[17]

CONCLUSION

VUFs are rare entities that require a high level of suspicion for diagnosis. The constellation of symptoms, referred to as Youssef's syndrome, is unique and includes menouria, watery discharge, and amenorrhea. Ultimately, these symptoms in the setting of prior uterine surgery should prompt an evaluation for a VUF. Surgical management is the most widely reported intervention, with an 80% to 100% success rate. With the increasing rate of cesarean deliveries in the developed world, the incidence of VUFs may rise, and it is imperative that we remain open-minded in the evaluation of GU symptoms.

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