Severe hemorrhage due to acquired uterine arteriovenous malformation/fistula following first-trimester aspiration abortion: A case report☆

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ARTICLE INFO

ABSTRACT

Uterine arteriovenous malformation/arteriovenous fistula is a rare, but potentially life-threatening, cause of severe hemorrhage. A case of uterine arteriovenous malformation/fistula causing severe hemorrhage following a first-trimester aspiration abortion procedure in a patient with a history of prior cesarean sections is presented. In this case, the patient was promptly diagnosed and effectively treated with uterine artery embolization. Consideration of uterine arteriovenous malformation/fistula in the differential diagnosis of severe hemorrhage following first-trimester aspiration abortion, especially in women with risk factors, can lead to timely recognition and appropriate treatment.

1. Introduction

An acquired uterine arteriovenous malformation/arteriovenous fistula (AVM/AVF) is an abnormal vascular communication that can cause abnormal uterine bleeding. The aberrant connection lacks an intervening capillary network and results in a direct, high-pressure, abnormal connection between uterine arteries and veins [1]. Menorrhagia can occur when the aberrant connection(s) are disrupted either through endometrial sloughing or external factors. Causes of acquired uterine AVM/AVF include trauma, prior instrumentation or surgery, and pre-existing uterine pathology such as infection, gestational trophoblastic disease, and malignancy [1]. The incidence of uterine AVM/AVF is unknown, but fewer than 100 cases were reported between 1926 and 2005 [2]. However, the incidence likely exceeds previous estimates based on a series from 2006 reporting a uterine AVM/AVF rate of 4.5% in 464 women aged 18 to 41 who had a pelvic ultrasound for vaginal bleeding [3].

First-trimester aspiration abortion is an exceedingly safe procedure [4]. Hemorrhage during an aspiration abortion usually results from atony and/or retained products of conception and occurs in approximately 0–4.7% of office-based procedures, with a rate of 1% or less in most studies [5]. Uterine AVM/AVF, on the other hand, is a rare cause of hemorrhage following first-trimester abortion. Few cases of AVM/AVF following aspiration abortion have been published [3,6–8]. A case of severe hemorrhage following a first-trimester abortion procedure is discussed here to highlight the diagnosis and management of this potentially life-threatening event.

2. Case Presentation

A 29-year-old woman, gravida 6, para 4, with a history of four prior cesarean sections presented to an ambulatory reproductive health center requesting an aspiration abortion at 12 weeks and 1 day of gestation dated by last menstrual period consistent with an unremarkable ultrasound at 10 weeks and 1 day of gestation. She was asymptomatic and had no prior history of irregular or heavy bleeding. She had had an

aspiration abortion in the past without any adverse event and reported receiving a blood transfusion following her first cesarean section. Her BMI was 26.9, hemoglobin 11.8 g/dl, and she was Rh positive. She received misoprostol 400 μg buccal for cervical preparation 2.5 h prior to the procedure, doxycycline 200 mg for antibiotic prophylaxis, and ibuprofen 800 mg for analgesia. She also received local cervical anesthesia, fentanyl 50 μg IV, and midazolam 1 mg IV. The obstetrician-gynecologist dilated the cervix using Pratt dilators and evacuated the uterus using a 13 mm rigid plastic cannula attached to electric suction followed by gentle sharp curettage. Post-aspiration, gross tissue examination revealed placental tissue and all fetal parts.

Shortly after the procedure, the patient started bleeding heavily. Bimanual examination revealed a flaccid uterus consistent with uterine atony. The patient was treated with vigorous uterine massage, methylergonovine 0.2 mg IM, and misoprostol 1000 μg rectally. The patient's bleeding notably decreased and her vital signs remained stable. Ultrasound revealed a heterogeneous mass in the uterus consistent with a large blood clot, and the physician evacuated it using brief suction under ultrasound guidance. Heavy bleeding resumed immediately following this maneuver, prompting initiation of hospital transfer. The patient received a second IV line for fluid administration, an additional dose of methylergonovine 0.2 mg IM and oxytocin 20 units IV, and was transferred via ambulance to a local tertiary hospital.

On arrival to the hospital, the patient continued to have moderate bleeding. The patient's vital signs were as follows: blood pressure 89 mmHg/48 mmHg, heart rate 90 beats per minute, respiratory rate 13 breaths per minute, oxygen saturation 99%. Her hemoglobin and hematocrit were stable at 10.5 g/dl and 31.6%, respectively. She received IV fluid and hydromorphone hydrochloride for pain. She underwent a formal ultrasound by a maternal fetal medicine specialist. Real-time grayscale ultrasound imaging of the uterus demonstrated several elongated tortuous cystic spaces containing demonstrable swirling of blood at the site of the cesarean section scar. The uterus was distended with 7.58 cm clot, concerning for an expanding hematometra (Fig. 1). Color and spectral Doppler imaging was highly suggestive of AVM/AVF. The maternal fetal medicine specialist reported the ultrasound findings directly to the care team. The care team discussed management options with the patient, including exploratory laparotomy with possible hysterectomy, and uterine artery embolization. The patient desired future pregnancies and opted for uterine artery embolization, prompting consultation with interventional radiology.

Angiography confirmed an AVM/AVF along the lower anterior uterine segment with early venous shunting into a dilated vein. Selective angiograms demons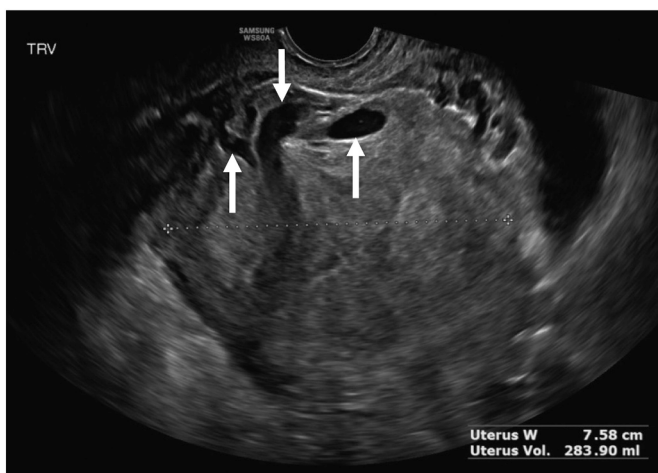trated arterial contribution from both uterine arteries, as well as branches of the cervicovaginal, cystic, and pudendal arteries (Figs. 2A-D). The interventional radiologist performed selective bilateral embolization using liquid embolic n-butyl-2-cyanoacrylate (n-BCA) glue, which polymerizes rapidly, allowing for a more selective embolization and minimizing the risk of systemic/non-target embolization. The patient had an uneventful post-operative course, was discharged home and recovered well, resuming her normal menses.

3. Discussion

Acquired uterine AVMs/AVFs typically present in women of childbearing age (mean age in the 30s) with a history of at least one prior pregnancy. Patients commonly present with intermittent and often heavy bleeding, but can be asymptomatic until undergoing an inciting event such as uterine surgery. Hormones associated with pregnancy may cause existing AVMs/AVFs to proliferate, worsening the symptoms [1,7]. In this case, the patient had a history of four prior cesarean sections and one prior aspiration abortion, but was asymptomatic prior to her presentation.

The differential diagnosis for hemorrhage during first-trimester abortion includes retained products of conception, uterine atony, uterine perforation, cervical laceration, and, rarely, uterine AVM/AVF, cesarean scar pregnancy and abnormal placentation. Retained products of conception and atony causing hematometra usually require re-aspiration to remove blood clots and tissue. In the case of a suspected uterine AVM/AVF, however, additional suction curettage can further
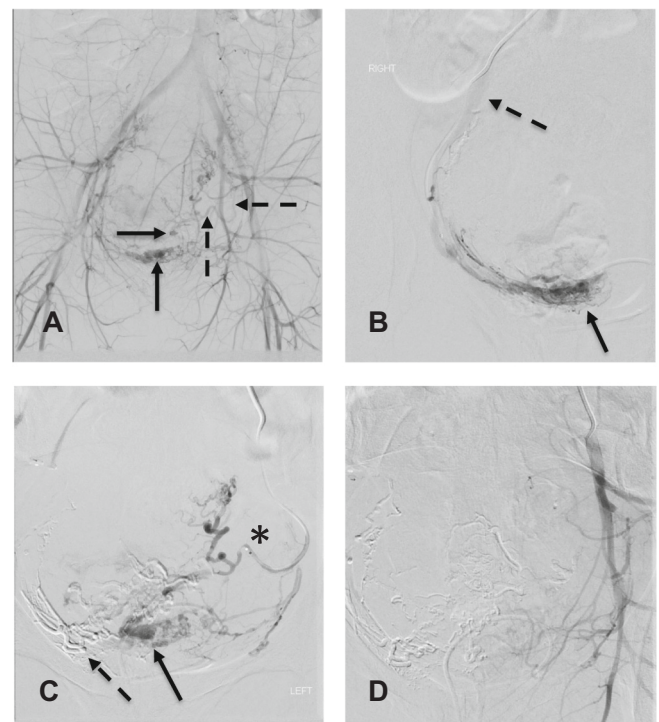

Fig. 1. Transverse transvaginal grayscale ultrasound demonstrates enlarged anechoic tubular structures (arrows) confirmed to be large draining veins on conventional angiography.

Fig. 2. A. Aortogram demonstrates the left uterine artery (dashed arrow) and early filling of large, patulous venous structures (solid arrows). The right uterine artery is obscured by overlapping vessels. **B.** Selective angiogram via a right uterine artery branch shows dilated, early filling venous structures (solid arrow) draining into the right internal iliac vein (dashed arrow). **C.** Selective angiogram from the left uterine artery (asterisk denotes microcatheter tip). There is persistent filling of the AVM (solid arrow) with contribution from the uterine fundal branches and cervicovaginal branches. Glue casts (dashed arrow) from preceding right uterine artery embolization are present. **D.** Completion angiogram via the left internal iliac artery confirms complete embolization of the AVM and normal enhancement of adjacent structures, including the bladder and vagina.

disrupt the AVM/AVF and worsen bleeding, as occurred in this case [9].

Transvaginal ultrasound with color and spectral Doppler imaging is considered the initial imaging study of choice for the diagnosis of uterine AVM/AVF. On grayscale ultrasound, the nonspecific finding of a tubular, hypoechogenic structure in the myometrium raises suspicion for the diagnosis. Color and spectral Doppler imaging showing a hypervascular area with turbulent flow and multiple tortuous feeding vessels with high velocity and low resistance in the myometrium of the uterus is highly suggestive for uterine AVM/AVF [2,7,10]. While not performed in this case, magnetic resonance imaging (MRI) can be useful when ultrasound is insufficient [1].

Since the introduction of uterine artery embolization in the 1980s [9], transcatheter uterine artery embolization has become the treatment of choice for patients wanting to preserve fertility or preferring a less invasive approach. Hysterectomy is reserved for patients who do not desire fertility preservation, have limited access to interventional radiology, or are not hemodynamically stable enough to undergo embolization. Advantages of uterine artery embolization include decreased blood loss, shorter hospital stay, and faster recovery [1]. In this case, the patient was hemodynamically stable and transferred expeditiously to a hospital with access to transcatheter embolization. Angiography identified the arterial supply to the uterine AVM/AVF from both sides requiring bilateral embolization in this case.

The choice of embolic agents rests on the preference of the interventional radiologist and availability. There is no consensus on the type of embolic material to use, and the use of liquid embolics, particles, coils, and ethanol has been described [6,11]. Particle embolics could enter the systemic circulation due to the lack of a normal capillary bed, while coils could block more proximal vessels, leaving the distal vascular nidus perfused. For these reasons, the interventional radiologist in these cases prefers to use the liquid embolics, n-BCA, Onyx™ (ethylene vinyl-alcohol copolymer) or ethanol, as first-line agents. The efficacy of the different agents is unknown due to the lack of controlled trials comparing outcomes [1].

Reported success rates for transcatheter uterine artery embolization range from 61% to 82%, with 5–24% of patients undergoing repeat embolization due to continued symptoms [1,3,11,12]. After repeat embolization, reported overall success rates increase, ranging from 69% to 95% [1,3,11,12]. In addition to repeat embolization, other options for treatment include medical therapies and minimally invasive surgery [1].

Reported side-effects and adverse events associated with transcatheter embolization are minimal. The most common side-effects are pelvic pain or cramping. Post-embolization syndrome, including low-grade fever, nausea/vomiting and tachycardia, can occur and is usually self-limited [13]. Although successful pregnancies have been reported post-embolization, the effect of embolization on fertility and placental abnormalities in future pregnancies is not known [7,8,14].

4. Conclusion

The success of this case highlights the importance of rapid recognition and diagnosis of uterine AVM/AVF, as well as the appropriate use of uterine artery embolization for its treatment. Repeated uterine suction curettage can inadvertently worsen the bleeding in these cases. As the incidence of uterine AVM/AVF may be higher than previously thought, providers should have a high index of suspicion for the diagnosis in surgical abortion cases complicated by hemorrhage that fails to resolve with additional suction curettage and uterotonic administration.

Contributors

Kathryn E. Sharpless drafted and reviewed the manuscript.
India I. Pappas drafted and reviewed the manuscript, and was involved the patient's care.
Ethan M. Dobrow drafted and reviewed the manuscript, and was involved in the patient's care.

Matthew Moccia reviewed and revised the manuscript.
Alison Bates reviewed and revised the manuscript, and was involved in the patient's care.
Michael G. Pinette reviewed and revised the manuscript and was involved in the patient's care.
Maureen Paul drafted and reviewed the manuscript, and was involved in the patient's care.

Patient consent

Written consent for publication of the report and accompanying images was obtained from the patient.

Provenance and peer review

This article was not commissioned and was peer reviewed.

Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

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