

Postpartum hemorrhage caused by uterine artery pseudoaneurysm: A case report

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ABSTRACT

Background: Uterine artery pseudoaneurysms (UAPs) are a rare life-threatening complication presenting as vaginal bleeding. Transvaginal ultrasound doppler scans can diagnose UAPs in the immediate and later postpartum period. This case report highlights UAP management using minimally invasive interventions for fertility preservation.

Case: A 21-year-old woman presented on post-operative day 10 following a primary cesarean section with heavy vaginal bleeding and a UAP was confirmed on doppler sonography. A multidisciplinary approach determined the optimal management taking the patient's fertility into consideration. Initially, the UAP was injected directly with thrombin under ultrasound guidance. However, due to a subsequent hemorrhage, a uterine artery embolization was performed.

Conclusion: Recognition of UAP is critical in the management of postpartum vaginal bleeding. Patient goals should be balanced with the severity of UAPs to determine optimal management.

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1. Introduction

Uterine artery pseudoaneurysm (UAP) is a rare cause of postpartum vaginal bleeding, with presentations ranging in severity. A pseudoaneurysm may be caused by damage to the arterial wall, resulting in a locally contained hematoma that has the color doppler ultrasound appearance of turbulent blood flow and a vascular neck that typically does not close spontaneously [1,2]. A pseudoaneurysm does not include any layer of the vessel wall. Instead, blood is contained by a partition developed by the products of the clotting cascade. This barrier formed from fibrin/platelet crosslinks is weaker than that of a true vascular aneurysm [2]. The prevalence of UAP is estimated to be present in 2–3/1000 deliveries and includes asymptomatic patients [3]. Due to its rarity, UAPs are often unrecognized at initial presentation. It should be suspected in all patients with delayed postpartum hemorrhage, defined as significant heavy vaginal more than 24 h after delivery and up to 6–12 weeks postpartum, especially when accompanied by abdominal pain [3,4].

Cesarean sections are associated with an increased risk of developing UAPs compared with vaginal deliveries [5]. Most often UAPs are found extending from the angle of the uterine incision and may be related to inadequate wound closure. UAP may also result from local trauma, neoplasm,

or infection leading to vascular injury resulting in blood entering the periarterial space within the myometrium. Other conditions associated with delayed postpartum hemorrhage include retained products of conception, involution/partial involution of the placental bed, endometritis and ruptured arteriovenous malformations (AVM), all of which need to be ruled out in making a definitive diagnosis [5].

Both UAP and AVM are vasculopathic etiologies for delayed postpartum hemorrhage with similar presentation and can be distinguished using transvaginal color doppler sonography. UAPs on color doppler studies show the classic “yin yang” sign (Fig. 1A), whereas AVMs display a mosaic pattern with high flow and low resistance [1]. CT angiography is typically reserved for confirmation and treatment of these vasculopathies with uterine artery embolization.

Bleeding from UAP is unpredictable and can occur spontaneously days to weeks after initial development. Most women with UAP present with vaginal hemorrhage and abdominal pain. Emergency management involves bilateral uterine artery embolization (UAE) to avoid hysterectomy [3]. However, UAEs may negatively impact future fertility if not judiciously applied due to infarction of surrounding normal myometrium. Few case reports have been published on thrombin injection into a UAP, which might be a safer, less destructive alternative to microsphere injection [5]. Advancements in interventional radiology combined with the development of new embolization techniques provide additional options for clinicians to consider in the management of patients who desire future fertility [6]. We present a case of pseudoaneurysm managed by direct ultrasound-guided thrombin injection and uterine artery embolization.

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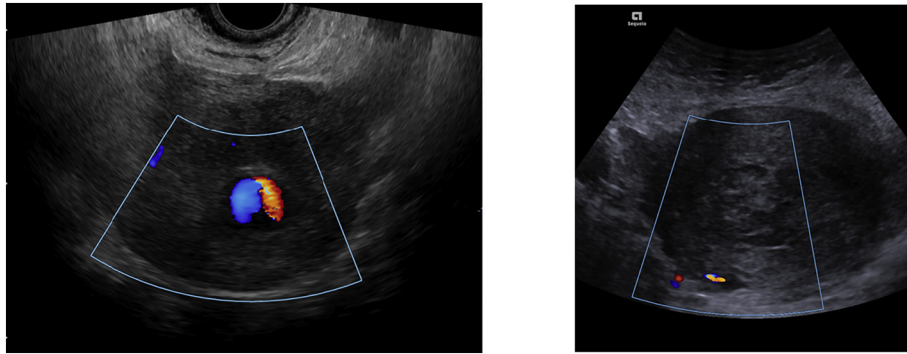


Fig. 1. A. Pelvic U/S doppler prior to thrombin injection showing the “to and fro” or “yin yang” sign, with bidirectional flow due to the swirling of blood within the pseudoaneurysm. B. Pseudoaneurysm on U/S doppler after thrombin injection with diminished activity.

2. Case Presentation

A 21-year-old woman presented with heavy vaginal bleeding on post-operative day 10 after a primary low transverse cesarean section. She reported passing palm-sized “blood clots” and endorsed mild pelvic pain. Her cesarean section was uncomplicated with an estimated blood loss of 900 mL. Her immediate postpartum course was complicated by symptomatic anemia believed to be from intraoperative blood loss (postoperative Hgb 6.6 g/dL) for which she received four units of packed red blood cells. She stabilized following transfusion and was discharged home on post-operative day 3 with a Hgb 7.8 g/dL.

At the time of re-admission, the patient's hemoglobin was 9.7 g/dL and she denied dizziness, palpitations, weakness or syncope. Her vital signs were unremarkable except for a mild tachycardia; her incision was well healed and her fundus was contracted and appropriately sized for a postpartum uterus.

To better image the uterus, both transabdominal and transvaginal ultrasound scans were performed. These showed a 2 cm anechoic structure in the upper fundal region of the endometrium with internal vascular flow consistent with a UAP. This diagnosis was substantiated by CT imaging of the abdomen and pelvis, with contrast (Figs. 2 and 3).

The patient strongly desired to maintain her future fertility and she was hesitant to undergo angiography with UAE. Interdisciplinary consultation between obstetrics/gynecology and radiology was convened to discuss interventions in a clinically stable patient with a known UAP. On hospital day 2 she had a 300 mL vaginal bleed, but remained hemodynamically stable. The decision was made to attempt a

transabdominal ultrasound-guided injection of thrombin into the pseudoaneurysm as a minimally invasive measure to control hemorrhage. If unsuccessful, a UAE would be performed. Thrombin injection resulted in immediate clotting of the pseudoaneurysm, as confirmed by doppler ultrasound imaging (Fig. 1). However, approximately 15 min after the procedure, the patient experienced profuse vaginal bleeding and became hemodynamically unstable, passing an estimated



Fig. 2. CT sagittal view.



Fig. 3. CT axial view.

1000 mL of blood. She received methergine 0.2 mg IM, tranexamic acid 1 g IV, 1 L of normal saline IV and two units of packed red blood cells.

Although the pseudoaneurysm on ultrasound appeared thrombosed at the time of vaginal hemorrhage (after the thrombin injection), due to the patient's instability a bilateral UAE was performed. The left uterine artery was noted to be vasoconstricted, likely from her hypovolemia, and therefore embolization was achieved using Embosphere particle beads. The right uterine artery was embolized with Gelfoam. Successful embolization was confirmed by angiography, which demonstrated complete peripheral embolization with minimal preserved slow flow to the main trunks of the right and left uterine arteries. The patient remained hemodynamically stable with minimal vaginal bleeding throughout the rest of her hospital stay and was discharged on post-operative day 2.

3. Discussion

UAPs typically develop as a result of iatrogenic surgical trauma after a miscarriage, pregnancy or cesarean section [5]. When UAPs rupture, the blood extravasates through the uterine tissue until reaching the uterine cavity, causing heavy vaginal bleeding, which may be associated with abdominal pain.

Although our patient was symptomatic, she was hemodynamically stable at the time of presentation. The clinical challenge of whether or not to intervene in a clinically stable patient who desires future fertility must be balanced against the known high morbidity and mortality associated with untreated UAP. An observational analysis of 24 studies shows a pregnancy rate of 40.5% after UAE with a pooled pregnancy loss rate of 33.5%, mainly due to first-trimester miscarriages [6]. Although reassuring, there is also data demonstrating a decrease in ovarian reserve following UAE. Loss of ovarian function has been attributed to the reflux of injected particles from the uterine artery to ovarian vessels via collateral circulation [7]. The clinical consensus based upon limited published evidence supports management with UAE in order to avoid surgery.

The initial choice of direct thrombin injection was based upon the belief that it posed the least threat to the patient's future fertility since thrombin is associated with a lower risk to native vessels, is well tolerated and has been highly successful when treating pseudoaneurysms in various locations throughout the body [8]. While this decision prioritized the patient's wish to avoid angiography and UAE, it potentially posed serious complications from continued hemorrhage if unsuccessful. Furthermore, the initial imaging identified a region of hypervascularity within the uterine fundus which also may have been consistent with an AVM. Therefore, whether an AVM or UAP, the artery could have ruptured from the back-pressure after attempting coagulation of the pseudoaneurysm. It is unlikely that the uterine artery was punctured during the thrombin injection as it was performed under direct ultrasound guidance and the injection was noted to be far away from the feeding vessels.

Various UAE methods were considered in the attempt to avoid surgery, including the use of Gelfoam, Embosphere particle beads and coils. Gelfoam is a temporary form of embolization that dissolves within three months and might better promote fertility, as it is less likely to reflux and disperse distally compared with particles [9]. The left uterine artery lumen in this case was smaller and more tortuous than the right side, likely secondary to her hypovolemia and the uterotronics administered. Consequently, the microcatheter could not be advanced deep enough into the left uterine artery to administer Gelfoam. Therefore, Embosphere was chosen since the microspheres travel more distal in the vessel and could be applied with more precision to effect hemostasis. As the right uterine artery lumen was larger, Gelfoam was used on that side and successfully placed.

4. Conclusion

This case report demonstrates the diagnostic and clinical challenges posed by UAP and offers nonsurgical management alternatives utilizing the skills of interventional radiologists working in close collaboration with obstetricians and gynecologists. Management was based on shared decision making with patient, who hoped to preserve her future fertility. Contingency plans that anticipate potential complications that occur as a result of the failure of direct thrombin injection by placing cannulas for UAE, judicious use of uterotronics and having blood available in the room should further reduce morbidity. UAPs are rare and unpredictable. Therefore, they should be treated in a timely manner to minimize morbidity and mortality from potential catastrophic vaginal hemorrhage.

Contributors

All authors made a substantive contribution to the material submitted for publication.

Conflict of Interest

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

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