CASE REPORT

Acquired uterine arteriovenous malformation after cesarean section

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Article Info

ABSTRACT

Objective: To demonstrate that embolization is a viable and well-established treatment for acquired arteriovenous malformations (AVMs), offering a safe, effective, and less invasive option for patients seeking to preserve fertility.

Case Report: A 20-year-old female presented with recurrent massive vaginal bleeding. Her medical history included a previous cesarean section complicated by a wound infection that necessitated resuturing. Initial diagnostic evaluation with transvaginal color Doppler ultrasound revealed hypervascularity in the uterus surrounding the surgical scar, raising suspicion for a uterine AVM. This diagnosis was subsequently confirmed through angiography. Given the patient's desire to maintain fertility, uterine artery embolization (UAE) was chosen as the treatment modality. The patient underwent multiple embolization sessions, during which embolic agents were administered to occlude the abnormal arteriovenous connections. The procedures were well-tolerated, and post-procedural monitoring indicated a significant reduction in uterine blood flow and resolution of hypervascularity. Follow-up assessments showed complete resolution of symptoms and no further episodes of bleeding. Importantly, the patient's reproductive potential was preserved, and she reported a return to normal menstrual cycles.

Conclusion: Acquired uterine arteriovenous malformation (AVM) is an uncommon but serious complication that can arise following cesarean section and should be considered in cases of persistent postpartum bleeding. This case highlights the efficacy of uterine artery embolization as a treatment for AVMs, offering a minimally invasive alternative to hysterectomy that effectively relieves symptoms while preserving fertility. Early recognition and timely intervention with embolization techniques can significantly improve patient outcomes in similar clinical condition.


Highlights:

1. Acquired uterine arteriovenous malformation (AVM) is an uncommon sequela of cesarean section, warranting consideration in instances of persistent uterine bleeding in the puerperium.
2. Embolization represents a viable and well-established treatment modality for AVM, providing a safe and efficacious intervention that serves as an alternative, less invasive modality for patients desiring fertility preservation.
INTRODUCTION

Uterine arteriovenous malformation (AVM) is a vascular abnormality characterized by a direct connection between the arterial and venous system within the uterus without any capillary network contribution. Arteriovenous malformation (AVM) is an exceptionally uncommon condition with a reported incidence of approximately 150 cases. Despite its rarity, this disease poses a significant risk of life-threatening complications. This rare condition can be a congenital or acquired lesion.

Congenital uterine AVM is believed to arise from a failure of differentiation during fetal angiogenesis. The embryological arrest or failure in the primitive capillary plexus differentiation results in anomalous capillary speciation and abnormal communication between arteries and veins. Congenital AVM exhibit a propensity for the presence of numerous feeding arteries, which contribute to their extensive vascularization, as well as a considerable number of large draining veins. Consequently, these AVM commonly demonstrate an enlargement beyond the boundaries of the uterus and an invasion into the adjacent pelvic region.

The acquired AVM is often associated with previous uterine traumatic procedures such as caesarean section, curettage or pelvic surgery. Acquired AVM can present a wide variety of symptoms. Its incidence and prevalence are difficult to determine because it is often misdiagnosed with retained products of conception and placenta accreta. However, it can cause massive vaginal bleeding, which is potentially life-threatening.

Imaging modalities including ultrasonography, computed tomography (CT), magnetic resonance imaging (MRI), and computerized tomography coronary angiogram (CTA) play a pivotal role in the diagnosis, treatment, and follow-up of uterine arteriovenous malformations (AVMs). Ultrasonography is capable of detecting the presence of anechoic or hypoechoic tubular or sponge-like areas within the normal myometrium and endometrium. However, it is important to note that other conditions may exhibit similar imaging characteristics, such as retained products of conception, gestational trophoblastic disease, or hydrosalpinx. The utilization of color doppler ultrasonography is crucial in obtaining more precise and accurate information in the evaluation of uterine AVMs.

Historically, the management of symptomatic acquired uterine arteriovenous malformations (AVMs) involved the use of hysterectomy. However, advancements in endovascular techniques have offered an alternative and minimally invasive treatment approach for patients who wish to maintain their fertility. In this case report, we present a detailed account of a 20-year-old female patient who experienced recurrent severe bleeding after undergoing a caesarean section. This challenging condition was successfully addressed through a series of uterine artery embolization procedures, leading to favorable outcomes. This report showed that embolization represents a viable and well-established treatment modality for acquired AVM, providing a safe and efficacious intervention, less invasive modality for patients desiring fertility preservation.

CASE REPORT

A 20-year-old Asian woman, multigravida, presented to the emergency room with massive vaginal bleeding. She had a history of caesarean section surgery in the previous four months during her last pregnancy. The patient was also diagnosed with wound infection a month after her surgery and underwent resutting. No history of hypertension and diabetes. On initial assessment, the patient presented shock symptoms (BP: 91/58 mmHg, HR 118 bpm; RR 26) with moderate pallor. Bleeding was present through the cervix based on speculum examination.

Color doppler ultrasonography showed dominance of pale shades during both systole and diastole represented low-impedance, high-velocity flow within the lesion and a colored mosaic pattern representing turbulent flow was noted. Spectral analysis of the vessels within the lesion confirmed high-velocity flow during both systole and diastole, and a low resistance index. The spectral waveform trace also showed spectral broadening consistent with turbulence and the spectral envelope was irregular. These findings indicated arteriovenous shunts and marked turbulence within the arteriovenous malformation. Spectral analysis of the venous flow revealed high flow velocities and systolic velocity peaks similar to an arterial pattern. The uterine artery velocity waveforms were characterized by high flow velocity and a low resistance index hypervascularity in the uterus around the surgical lesion with RI 0.32 and PSV 138 cm/s suggesting a uterine arterio-venous malformation (Figure 1). The hematological test result was a decline in hemoglobin (8.4 d/dL), and red blood cell (3.04 x 10⁹/mm³) level.
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Figure 1. Color Doppler ultrasonography showed a hypervascularity in the uterus around the surgical lesion, suggesting a uterine arterio-venous malformation.

Figure 2. The result of CT angiography confirmed communication with anomalous vessels refer to left uterine arterio-venous malformation (arrow).

The patient's condition was stable after being treated with a blood transfusion. She was moved to the ward for further investigation. The next day, she underwent computed tomography angiography (CTA), which revealed that in the arterial phase, the AVM nidus in the uterus appeared with irregular and inhomogeneous edges. CTA showed hypervascularity of the uterine artery, a branch of the left internal iliac artery, confirming a left uterine arterio-venous malformation (Figure 2).

The patient was scheduled for left uterine artery embolization. Before the embolization, the patient's vital signs deteriorated (BP: 90/60 mmHg, HR 108 bpm; RR 24) with a declining hemoglobin level (4.5 g/dL). The patient was transferred to the ICU for stabilization and subsequently sent to the Cath lab for emergency embolization. The patient's condition improved afterwards. Subsequently, the patient was referred to the interventional radiology department to undergo a computed tomography angiography (CTA) and uterine artery embolization. The procedure involved accessing the femoral artery and performing super-selective microcatheterization of the uterine arteries. During this process, an anomalous vascular pattern was identified in the left uterine artery, characterized by distal angiodysplasia and arteriovenous fistula.
The uterine artery embolization (UAE) procedure was conducted under moderate sedation, utilizing the left transfemoral approach. Initially, a direct approach was employed to gain access to the left common and internal iliac artery using a 5 French (F) Roberts Uterine Catheter (Cook Medical LLC, 750 Daniels Way, P.O. Box 489, Bloomington, IN 47402-0489 USA). Under the guidance of fluoroscopy, selective embolization was performed into the distal left uterine artery (Figure 3(A)). Non-spherical polyvinyl alcohol 700 microns (PVA) particles were initially chosen, considering the apparently low fistula debit. Subsequent angiographic assessments confirmed satisfactory occlusion of the fistula flow following PVA embolization. Post-embolization angiography demonstrated successful occlusion of the aberrant vessels without any observed vascular complications (Figure 3(B)).

Forty days after the embolization procedure, the patient underwent a follow-up transvaginal ultrasound, which revealed no abnormal findings. Two months later, pelvis magnetic resonance imaging (MRI) was performed, revealing no detectable abnormalities. The patient approved all of the work to be published that includes the personal information and all relevant family members have been informed and have consented to the publication of this information.

**DISCUSSION**

Around 1 to 2% of cases are attributed to secondary postpartum hemorrhage, which is characterized by causes such as uterine subinvolution, retained products of conception, endometritis, and retained placenta. Less commonly encountered etiologies of secondary postpartum hemorrhage encompass cervical cancer, submucous fibroids, placental adherence, cesarean scar dehiscence, uterine pseudoaneurysm, and uterine rupture. Uterine vascular abnormalities, such as arteriovenous malformations (AVMs), and congenital coagulopathies, are implicated in the occurrence of postnatal hemorrhage. Uterine arteriovenous malformation (UAVM), an infrequent and potentially life-threatening condition, serves as a significant cause of excessive bleeding following childbirth. UAVM is characterized by an anomalous connection between the uterine arteries and the venous system. Reported cases of UAVM are limited to fewer than 200 instances. This condition can be classified as either congenital or acquired, with the acquired form often associated with prior cesarean section, curettage, or pelvic surgery. In this particular case, the patient's medical history revealed a previous cesarean section performed four months ago, which suggests that the acquired form of uterine arteriovenous malformation (AVM) may be the underlying condition. It is important to note that congenital uterine AVMs and acquired uterine AVMs exhibit distinct pathophysiological processes. Congenital AVMs are believed to arise from the arrest or failure of embryological differentiation of the primitive capillary plexus, resulting in abnormalities in capillary formation and aberrant arteriovenous communication. This leads to the presence of numerous vascularized arteries and enlarged drainage veins in congenital AVMs, often causing them to extend beyond the confines of the uterus and invade the surrounding pelvic structures.

Acquired uterine arteriovenous malformations (AVMs) manifest as a result of previous uterine interventions, including pelvic surgery, therapeutic abortion, curettage procedures, or cesarean sections. These interventions contribute to the occurrence of abnormal communi-
cations between arteries and veins, specifically within the venous sinuses of scar tissue where necrotic villi are associated. The healing process following these interventions provides an opportunity for the development of aberrant vascular connections, often characterized by the fusion of arterial and venous vessels. Additionally, acquired AVMs frequently exhibit multiple vascular connections and demonstrate the capacity to invade adjacent anatomical structures.12-13

Uterine arteriovenous malformation (AVM) presents with a wide array of clinical manifestations. Vaginal bleeding is the primary and most prevalent symptom, spanning from intermittent spotting to profuse hemorrhage. Other reported presenting symptoms include uterine abnormal bleeding and postcoital bleeding. These symptoms can lead to the development of severe anemia. Acquired uterine AVMs commonly exhibit vaginal bleeding in women of childbearing age, especially those who are in the postpartum period or have a history of previous uterine interventions. Accurate diagnosis plays a crucial role in initiating appropriate management strategies. Imaging modalities are indispensable for early detection and effective management, as the diagnosis of uterine AVM cannot be solely established based on clinical evaluation.14-15

Ultrasound imaging is a valuable tool for detecting the presence of multiple anechoic or hypoechoic tubular areas, commonly referred to as "sponges," within the myometrium of the normal endometrium. These tubular structures can be visualized and characterized through their distinct ultrasound appearance. However, it is crucial to consider that other pathological conditions may also manifest similar sonographic features. These conditions include retained products of conception, hemangioma, gestational trophoblastic disease, or hydrosalpinx. Therefore, a comprehensive diagnostic approach, including clinical correlation and potentially additional imaging modalities or histopathological evaluation, is necessary to differentiate between these entities and arrive at an accurate diagnosis.16

Color Doppler ultrasonography is an essential modality for obtaining more precise and accurate information in medical imaging. In the context of evaluating uterine pathology, such as uterine arteriovenous malformations (AVM), color Doppler ultrasound provides valuable insights. In a normal myometrial signal, color Doppler parameters exhibit specific ranges. These include a peak systolic velocity (PSV) ranging from 9-44 cm/s and a resistive index (RI) ranging from 0.6-0.8. These values serve as reference points for assessing normal vascular flow in the myometrium. However, in the presence of a uterine AVM, color Doppler ultrasound reveals distinct and characteristic features. These include intense vascularization and multidirectional flow pattern. The visualization of juxtaposed red and blue areas indicates the presence of multiple tortuous blood vessels with varying orientations. This phenomenon is a hallmark of uterine AVMs. With color Doppler ultrasound, specific hemodynamic parameters can be assessed to differentiate uterine AVMs from other conditions. Uterine AVMs typically exhibit high velocity flow (mean PSV: 136 cm/s), low resistance flow (mean RI: 0.3), low pulsatility of the arterial waveform, and high velocity and pulsatile venous waveforms. These findings are indicative of abnormal vascular flow pattern associated with uterine AVMs. It is important to note that distinguishing between venous and arterial waveforms can be challenging in the context of uterine AVMs. Additionally, it is not uncommon to observe pulsatile flow in pelvic veins distal to AVMs, deviating from the expected monophasic flow pattern. Initial investigation is often performed by US Color Doppler, which suggests hypervascularity, and this test should include flow velocity measurement.17-18

CT angiography (CTA) plays a pivotal role in the comprehensive management of uterine arteriovenous malformations (AVMs), encompassing diagnosis, treatment, and follow-up. It is widely regarded as the "gold standard" for diagnosing AVMs due to its high accuracy and detailed imaging capabilities. CTA allows for the identification and characterization of crucial anatomical and hemodynamic features associated with uterine AVMs. These include bilateral hypertrophy, visualization of feeding uterine arteries, identification of tortuous hypertrophic arterial masses along with large accessory vessels, and early visualization of drainage into hypertrophic veins. These findings provide valuable information for accurate diagnosis and subsequent treatment planning. Beyond its diagnostic capabilities, CTA offers several notable advantages. One such advantage is its rapid acquisition time, allowing for efficient and time-sensitive evaluation of uterine AVMs. Additionally, CTA is widely available, enabling its utilization in various clinical settings, thus facilitating prompt diagnosis and timely intervention.19-20 The patient underwent a US color Doppler to investigate the bleeding etiology. The evaluation documented a hypervascularity structure above the previous uterus caesarean section scar with an apparent flow reversal suggesting turbulent high-velocity flow, which indicates a suspicion of uterine AVM. Therefore, the patient was scheduled for CT angiography for further investigation. Ultimately, the uterine AVM diagnosis was made by the CTA findings.21

A study by Timmerman et al. revealed that ultrasound examination had a low positive predictive value. From 30 cases declared AVM based on color Doppler examination, only three were declared AVM based on
gold standard examination. These results were different from El's findings. Gawad et al., where USG and CT-Angiography had a sensitivity of 100%. However, this study only used angiography as the gold standard for diagnosing AVM. However, both examinations have their respective roles and functions in diagnosing or determining therapy in AVM case.

The hemodynamic stability of the patients, as well as their desire to preserve fertility, determine therapeutic options for uterine AVM. Resuscitation, emphasizing achieving hemostasis and maintaining tissue perfusion, is performed in post-partum hemorrhage.

Hysterecomy and embolization are the remaining primary treatment options in UAVM. The consideration can be made from the patient's condition and the reproduction expectancy. Hysterecomy represents the definitive therapeutic approach for uterine arteriovenous malformations (AVMs). However, its application is primarily reserved for resource-limited environments or instances where uterine embolization is contra-indicated; it remains a viable alternative for individuals who have discontinued their fertility aspirations or in cases where embolization proves ineffective in resolving bleeding. Preservation of reproductive function is typically prioritized, especially in younger patients. Importantly, it should be noted that hemodynamic instability does not represent a contraindication for this procedure. The patient's condition deteriorated, and underwent an emergency embolization. The embolization was performed using N-butyl cyanoacrylate (NBCA) as an embolant agent. Embolization using N-butyl cyanoacrylate (NBCA) material can be considered in patients with active bleeding, unstable hemodynamics, and failed embolization with a gelatin sponge. This material works by polymerizing when in contact with blood and can embolize even in cases of coagulopathy. Trans-arterial embolization is the first-line endovascular therapy used to treat AVMs, especially when multiple arteriovenous shunts exist. If necessary, trans-arterial embolization can also be performed repeatedly. Even though the patient's condition improved after the first embolization, the follow-up US color doppler documented residual UAVM. Hence, the patient required another embolization management. After another embolization series, complete embolization was achieved, and the patient did not experience vaginal bleeding afterwards. Some cases that can be treated with embolization are bleeding due to uterine atony, birth canal lacerations, placental abnormalities, and AVM. This method is fast, can be performed repeatedly and does not require general anesthesia. Apart from that, this method can maintain the function and anatomy of the uterus.

The rebleeding rate in bleeding patients treated with arterial embolization is 5.2-13.5%. The main cause of rebleeding after embolization is recanalization of the embolized artery followed by collateral formation, and it is necessary to look for spontaneous arterial Anastomoses such as from the ovarian artery, rotundum artery, middle rectal artery or inferior mesenteric artery. Re-embolization is an effective and safe procedure to do.

In the past, this case could only be managed using arterial ligation or hysterectomy. Since its introduction in 1982, treatment using the arterial embolization method has become more common. This method is less invasive compared to ligation and hysterectomy methods. Apart from that, this method can also have advantages, especially for the patient's fertility function. Several other advantages of this method are shorter hospital stays, CTA-guided examination so that embolization can occur in the right artery, and minimal post-operative injuries.

There is limitation to this study. It is a weakness that we only had one case which was fully documented, while other cases of acquired AVM were not fully documented and no surgical intervention had been performed. Our proficiency lies in the thorough documentation and evaluation of the AVM case, encompassing a comprehensive analysis from the pre-embolization phase to the post-embolization phase.

CONCLUSION

Uterine arteriovenous malformation (AVM) is an abnormality of the vasculature characterized by a direct connection between the arterial and venous systems within the uterus, devoid of any capillary network involvement. Uterine AVMs are a rare etiology of profuse and potentially catastrophic vaginal hemorrhage. While hysterectomy represents the definitive therapeutic approach, its implementation is typically avoided in younger patients who still desire fertility. Uterine artery embolization stands as the prevailing treatment modality for symptomatic uterine AVMs. However, obstetricians should be prepared to intervene or repeat the procedure if embolization fails to achieve the desired resolution of bleeding.

DISCLOSURES

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Conflicts of interest

The authors declare no conflicts of interest regarding the publication of this paper.

Patient consent for publication

The patient approved all of the work to be published that includes the personal information and all relevant family members have been informed and have consented to the publication of this information.

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Author contribution

All authors contributed to all processes in this research, including preparation, data gathering and analysis, drafting and approval for publication of this manuscript.

REFERENCES