

Surgical Therapy for Uterine Arteriovenous Malformation

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ABSTRACT Arteriovenous malformations (AVMs) entail direct connections between arteries and veins. Uterine AVMs, in particular, are rare gynecologic vascular pathologies that can manifest either asymptotically or with life-threatening bleeding. They may arise congenitally or, more commonly, post-myometrial trauma. While Doppler ultrasonography serves as the primary diagnostic tool today, computed tomography, magnetic resonance imaging, or angiography may also be employed. Treatment selection should be tailored to each patient, considering symptoms and fertility expectations. For patients desiring fertility, monitoring, medical therapy, or angiographic interventions are favored, whereas for those without fertility expectations, advanced age, and symptomatic disease, surgery emerges as the definitive treatment. In our case, a 45-year-old woman with a history of one previous cesarean section presented with prolonged and uncomfortable vaginal bleeding and no fertility expectations. She underwent a total laparoscopic hysterectomy and bilateral salpingo-oophorectomy, oophorectomy was performed according to the patient's request.

Keywords: Arteriovenous malformations; ultrasonography, doppler; uterine hemorrhage; uterine artery embolization

Uterine arteriovenous malformations (AVM) are rare gynecologic diagnoses characterized by direct arteriovenous shunts within the myometrium, which can occur congenitally or acquired. They were first described in the 1920s.¹ In addition to, the recent increase in cesarean delivery rate and uterine interventions such as myomectomy and endometrial curettage, the development of Doppler ultrasonography (USG) technology, the frequency of diagnosis has increased.^{2,3}

Patients may exhibit a range of clinical symptoms depending on the size and location of the shunt, with vaginal bleeding being the most common complaint. Asymptomatic cases may be incidentally discovered during transvaginal USG examinations, where heightened hypoechoic vascular shadows in the uterus and increased vascular flow velocity (>20 cm/s) on Doppler examination

serve as diagnostic markers. Additionally, the severity of shunt flow can lead to the development of heart failure.⁴

While angiography was previously the gold standard diagnostic method, Doppler USG has now come to the forefront due to its ease of use, high sensitivity, and cost-effectiveness. Computed tomography (CT) and magnetic resonance imaging (MRI) can also be used for diagnosis.^{4,5}

In determining the appropriate treatment approach, it is crucial to conduct individualized evaluations for each patient. For young, asymptomatic women seeking fertility preservation, a watchful waiting strategy may be suitable. However, in symptomatic cases, uterine artery embolization (UAE) stands out as the most common treatment option, boasting a clinical success rate of 93%. Nonetheless, accessibility to UAE may be limited due to cost im-

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plications and the need for a multidisciplinary approach.⁶

In instances where UAE is not feasible, various medical treatments such as progesterone, methylergonovine, and methotrexate have demonstrated success rates of 88%.⁷ Moreover, successful outcomes have been documented with operative hysteroscopy in specific case series in the literature.⁸

For symptomatic patients who do not desire fertility preservation or are of advanced age, hysterectomy may be considered as a definitive treatment method. However, it's essential that hysterectomy surgeries be performed by surgeons experienced in surgical hemostasis techniques, such as uterine compression sutures, iliac artery ligation, or hypogastric artery ligation, due to the heightened risk of bleeding associated with these procedures.

CASE REPORT

Forty five years old, known comorbidity Arrhythmia, G1P1, former cesarean section (C/S), presented with vaginal bleeding. On admission, general condition was good and vitals were stable. On examination, dark vaginal bleeding was present, no tenderness was observed on vaginal palpation, and the uterus was voluminous on bimanual examination. Transvaginal USG showed hypoechoic large vascular areas near the fundus, colour Doppler USG showed increased blood flow compatible with diffuse AVM, and Doppler flow peak systolic velocity was 46 cm/s. Pelvic MRI showed lesions consistent with uterine volume and dilated vascular structures in the parenchyma. Dilatation was observed in bilateral pelvic veins and may support the uterine AVM reported in the preliminary diagnosis (Figure 1). Laboratory tests revealed negative tumour markers, negative beta human chorionic gonadotropin, haemoglobin: 13.2 mg/dL, haematocrit: 41%. Hysterectomy and bilateral salpingo-oophorectomy were planned after the evaluation with the patient who had no fertility desire.

The pathological examination revealed AVM in the myometrium (Figure 2).

The patient has given informed consent for this case report.

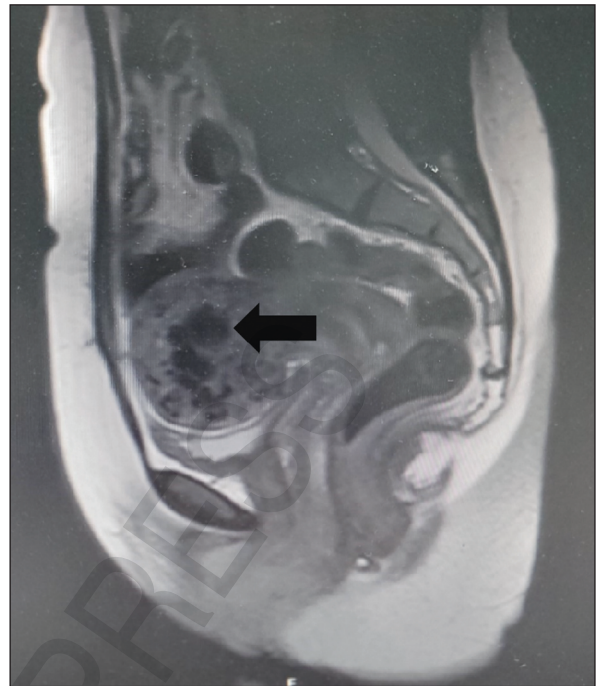


FIGURE 1: Sagittal plane of the pelvic magnetic resonance image showed the uterus is voluminous, and lesions consistent with dilated vascular structures are observed within the parenchyma.

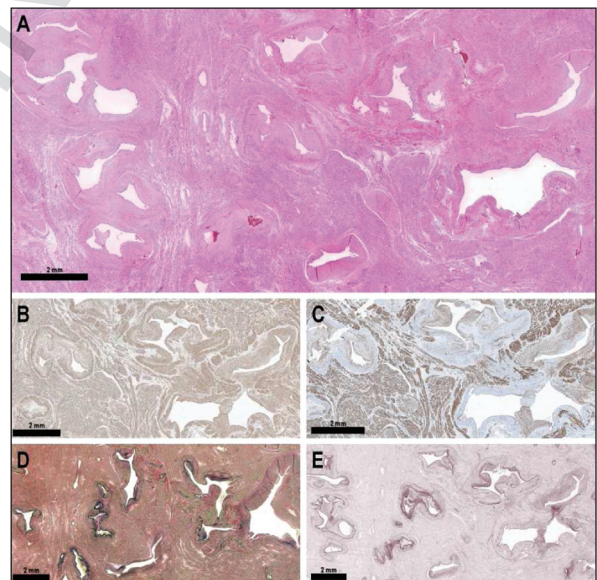


FIGURE 2: Some of the thick-walled vasculat structures exhibit irregularities in the vessel walls and loss of elastic layers (A-Hematoxylin and eosin staining, B-Smooth muscle actin, C-Desmin, D- Elastic van gieson, E- Orcein).

DISCUSSION

Acquired uterine AVMs often stem from various procedures, notably curettage, C/S, myomectomy, and interventions for endometrial and gestational neoplasms.

sia. The escalating rate of cesarean deliveries, coupled with an increase in placental invasion anomalies leading to greater myometrial trauma, has notably contributed to the rising prevalence of these malformations. Postpartum and post-abortion evaluations reveal a frequency of 0.63% for uterine vascular malformations, with uterine AVMs occurring at a frequency of 0.1%.^{2-4,9}

Cesarean scar pregnancies represent a predominant cause linked to uterine AVMs, necessitating a high rate of surgical intervention. Methotrexate emerges as a valuable treatment option due to its efficacy in managing both cesarean scar pregnancies and uterine AVMs.⁴

In cases of vaginal bleeding with a history of myometrial injury, caution is warranted. If diagnostic endometrial curettage is contemplated, it is imperative to assess uterine vascular structures using Doppler USG. A reevaluation of the necessity for curettage should be conducted, considering the potential risk of precipitating severe bleeding.⁴

Recent literature indicates a shift away from utilizing angiography for diagnostic purposes in cases where therapeutic intervention is not planned. Instead, emphasis is placed on non-invasive imaging modalities such as transvaginal USG and color Doppler USG. Hypoechoic vascular echogenicity areas detected on transvaginal USG, along with increased vascular flow velocity (e.g., ≥ 20 cm/s) on color Doppler USG, demonstrate high sensitivity in diagnosing AVMs.⁴

Furthermore, studies have shown that patients with uterine AVMs experiencing spontaneous resolution typically exhibit an average flow velocity of 58.5 cm/s, whereas those requiring treatment manifest an average flow velocity of 85.2 cm/s.⁴

Additionally, CT and MRI have emerged as valuable diagnostic tools. CT angiography offers high sensitivity for detecting uterine AVMs, while MRI is particularly useful for evaluating other potential pathologies within the myometrium.⁵

In the selection of treatment, decisions should be made based not only on the severity of abnormal uterine bleeding but also on the patient's age and desire for fertility. Observation may be preferred in asym-

ptomatic patients with stable complete blood counts and Doppler flow velocity < 40 cm/s.⁴ In symptomatic patients desiring fertility, angiographic UAE emerges as the most extensively researched treatment method, with a success rate of 88-93%.^{4,6} The experience of the radiology team is important in this treatment. Operative hysteroscopy has also reported successful results reaching 100%, but studies are limited.⁸ In medical treatment, successful results have been reported with synthetic Progesterone derivatives such as norethisterone, methylegonovine, mifepristone, methotrexate, and danazol, with a success rate averaging 88%.^{4,7} For patients without a desire of fertility or advanced age, and excessive bleeding, hysterectomy is the definitive treatment method. Since there is a possibility of developing severe hemorrhages, surgery should be performed by surgical teams experienced in advanced hemostasis techniques such as uterine compression sutures, iliac artery ligation, or hypogastric artery ligation. In our case, surgical treatment was decided due to increased vaginal bleeding and no desire for fertility, and during the surgery; prior to hysterectomy, 5 mm clamps were placed on both uterine arteries to control bleeding, and the operation was successfully completed. If hysterectomy is to be performed laparoscopically, the use of a uterine manipulator and placing clamps on the uterine arteries before surgery may be beneficial in terms of preventing the massive bleeding.²

In light of the available data, there is no clear treatment algorithm. Still, the treatment method should be tailored to individualized evaluation for each patient, and especially surgical treatment should be performed in healthcare facilities where there are experienced teams under adequate conditions. Although rare, uterine AVMs, which can lead to serious problems such as massive bleeding, should be prevented by avoiding unnecessary uterine interventions.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: Cihan Mutu, Günay Gahramanlı, Gürdeniz Serin, Ali Akdemir; **Design:** Cihan Mutu, Günay Gahramanlı, Gürdeniz

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