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INSTRUMENTATION AND TECHNIQUES

Uterine Arteriovenous Vascular Malformation Masked by Partial Molar Pregnancy: Diagnostic Challenge and Subsequent Embolic Treatment

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About the Author

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Introduction

Uterine arteriovenous vascular malformations (UAVM) are uncommon and usually present in women of reproductive age. When these occur in a pregnant patient or in the immediate postpartum period, clinical presentation may overlap with several other entities such as retained products of conception (RPOC), postpartum endometritis, as well as gestational trophoblastic disease (GTD) [1]. We present a case of a UAVM that presented with life-threatening vaginal bleeding several months after dilatation and curettage (D&C) of a molar pregnancy. The UAVM was diagnosed with ultrasound (US) and magnetic resonance imaging (MRI), and subsequently treated with transarterial embolization (TAE).

Case

Case reports are exempt from institutional review board approval at our institution. A 37-year-old G2P0020 female was referred with complaints of heavy vaginal bleeding for 1 week. She reported passing "pancake sized clots" and stated that she was changing her pad every 20 min. She also reported vaginal cramping and abdominal pain.

The patient's first pregnancy resulted in a spontaneous abortion 2 years ago. Four months prior to presentation, she was diagnosed with a partial molar pregnancy based on US findings (Fig. 1) and a beta-hCG level of 262,000 IU/L.

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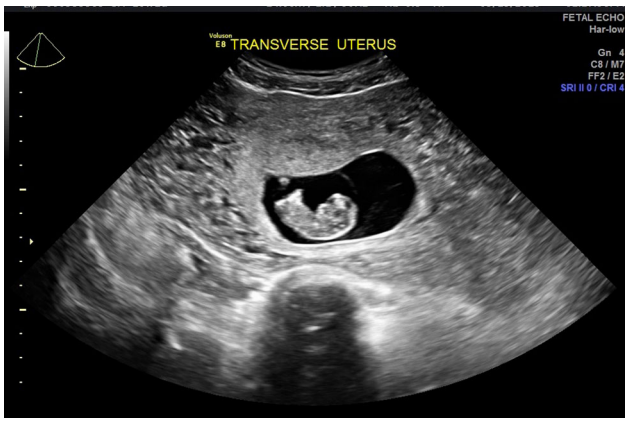

Fig. 1 Transvaginal US image of the uterus shows a gestational sac and an intrauterine pregnancy. There are also several cystic spaces in the uterus concerning for a molar pregnancy

She underwent a D&C for treatment. Three months following her discharge from the hospital, she presented with 3 days of vaginal bleeding. During this hospital stay, the bleeding stopped, and she was managed expectantly and discharged home. At this time, no imaging was done. One month later, she presented to our institution with heavy vaginal bleeding, as described above.

On physical exam at the time of admission, she appeared pale and diaphoretic. Her abdomen was soft and there was a palpable mass 5 cm inferior to the umbilicus. Her uterus was 12 cm and anteverted. There was a moderate amount of dark red blood in the vaginal vault. Her beta-hCG had trended down from the time of her partial molar pregnancy to 2 IU/L. Her hemoglobin was 6.7 g/dL and she was transfused with two units of packed red blood cells. An US of the pelvis showed a fluid-filled endometrial canal and several cystic structures within the uterus (Fig. 2a). This finding was similar to her US 2 months prior (Fig. 1). However, on color Doppler interrogation, there was color flow within these anechoic cystic spaces (Fig. 2b). Spectral analysis revealed a rapid arterial flow of up to 80 cm/s with a low resistance index (RI) of 0.23 (Fig. 2c). Given these findings, a vascular malformation was suspected. MRI of the pelvis demonstrated an enlarged uterus with several flow voids within the fundus and body of the uterus, consistent with a high-flow arteriovenous malformation (AVM) (Fig. 3). The patient was managed conservatively

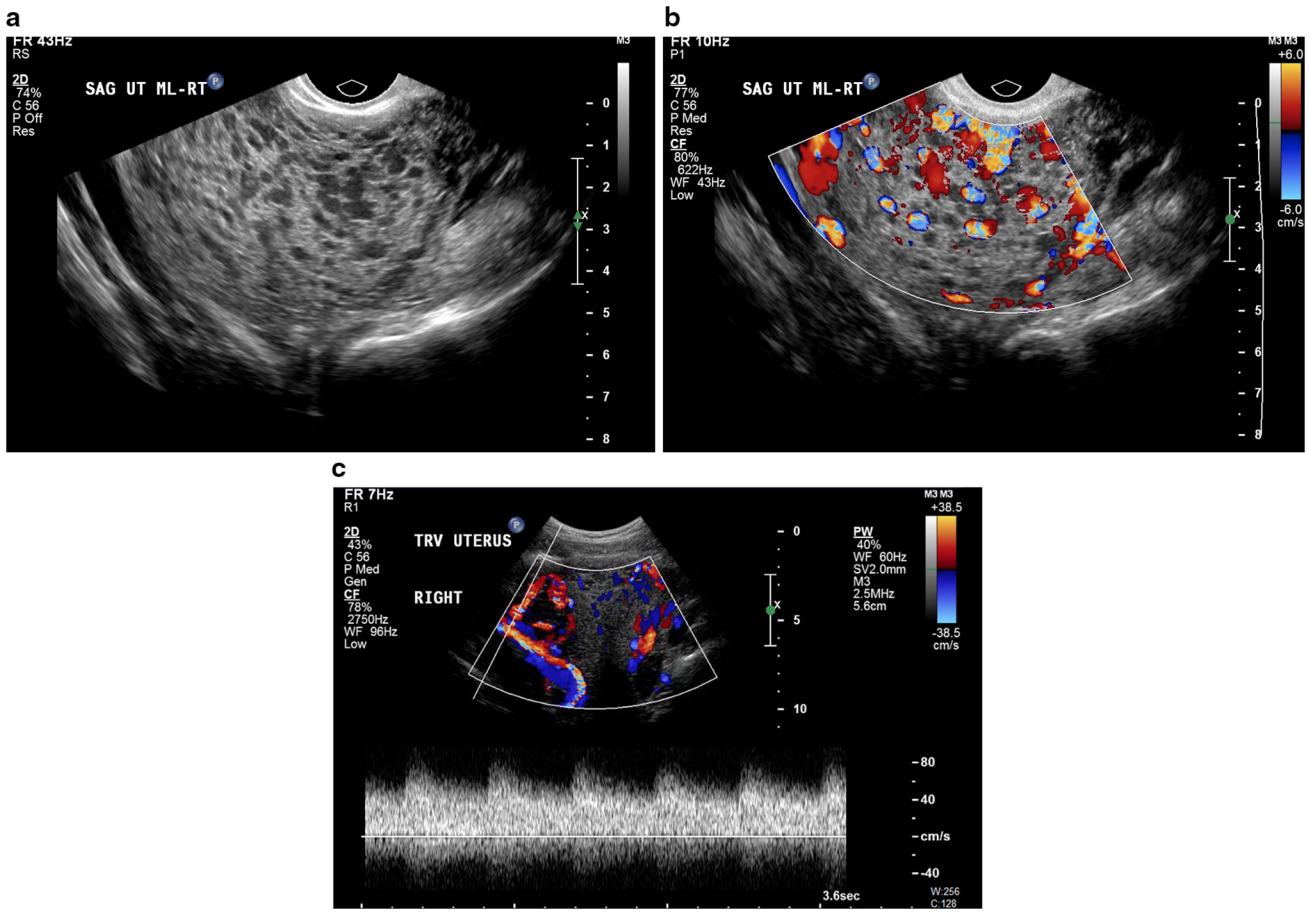

Fig. 2 **a** Transvaginal US image of the uterus showing several cystic structures within the uterus. **b** Color Doppler US image of the uterus showing flow within the cystic spaces. **c** Color Doppler US image of the uterus showing peak systolic velocity of 80 cm/s

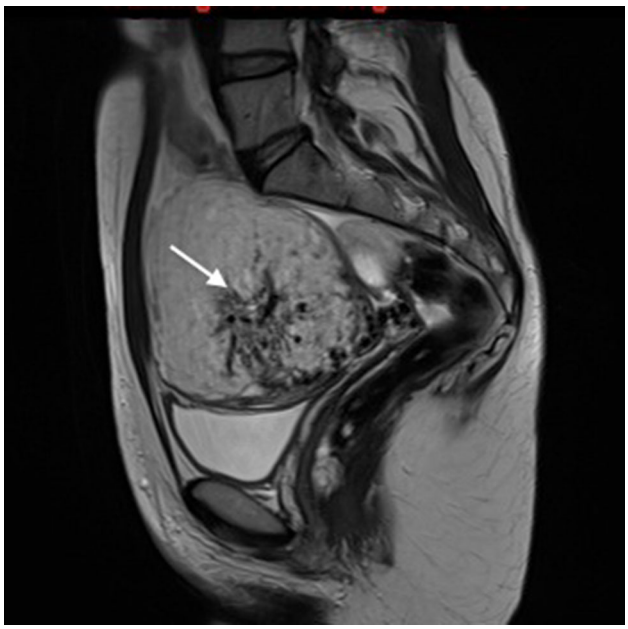

Fig. 3 Sagittal T2 weighted MR image of the pelvis shows numerous flow voids within the uterine body (*arrow*), consistent with a vascular malformation

with vaginal packing for 2 days, however, her bleeding persisted and she was referred for angiography and possible embolization.

A pelvic angiogram demonstrated a large AVM in the uterus supplied by both uterine arteries and multiple smaller feeding vessels with its nidus supplied mainly by the right uterine artery (Fig. 4a). Early venous filling of the right internal iliac vein was also seen (Fig. 4b). A 5F SOS Omni Selective Catheter (Angiodynamics, Latham, NY) was then used to select the right uterine artery. A 3F microcatheter (Renegade Hi-Flo, Boston Scientific, Natick, MA) was then placed distally into the right uterine artery and into the nidus of the AVM. The vessel was then embolized with a 1:1 mixture of N-butyl cyanoacrylate (N-BCA, TRUFILL®, Miami Lakes, FL) and Ethiodol (Guerbet LLC, Bloomington, IN). 1.4 cc of embolic was used. Post-embolization angiogram demonstrated decreased flow to the AVM (Fig. 4c). Subsequently, the left uterine artery was selected using the 5F SOS catheter (Angiodynamics, Latham, NY) and the 3F microcatheter (Renegade Hi-Flo, Boston Scientific, Natick, MA). However, only a limited supply to the AVM from the left uterine artery was seen (Fig. 4d). This artery was embolized with 500–700 μm embospheres (Merit Medical, South Jordan, UT). One vial of the embolic mixed in 10 cc of Isovue 300 was used. Post-embolization aortogram demonstrated exclusion of flow to the AVM (Fig. 4e). The patient had no further episodes of vaginal bleeding post-procedure and did not require any additional transfusions. She was discharged on post-procedure day #3. She was seen in the gynecology clinic 3 months post-procedure with resumption of her normal menstrual cycles.

Discussion

Uterine arteriovenous vascular malformations (UAVMs) are uncommon gynecologic vascular lesions that often present as unexplained, intermittent, vaginal bleeding of sudden onset [2–5] occurring with or without pelvic pain or dyspareunia [4, 6]. The bleeding may be profuse, long lasting, and can lead to life-threatening hemorrhage. The resulting hemodynamic instability makes the diagnosis of UAVM an important consideration in any patient presenting with vaginal bleeding as prompt diagnosis and therapy are essential for favorable outcomes. These may occur as congenital lesions that grow with puberty or pregnancy, or they may develop as acquired lesions.

The development of acquired UAVMs often follows pregnancy or uterine trauma, such as D&C, pelvic surgery, endometrial and cervical carcinoma in situ, maternal diethylstilbestrol exposure, infection, RPOC, GTD, or choriocarcinoma [3, 7–10]. Typically, acquired AVMs consist of multiple small arteriovenous fistulas, while congenital AVMs have an intervening nidus with multiple feeding arteries and draining veins [2, 3]. In our case, imaging findings were consistent with a congenital AVM, and we hypothesize that the growth and subsequent bleeding of the AVM was a result of the hormonal variations in her molar pregnancy [11].

The true incidence of UAVM is unknown, although rough estimates have been reported with 3.4–4.5 % of women with abnormal premenopausal bleeding being diagnosed with a UAVM [12, 13]. While a UAVM can present at any age, they typically affect women of childbearing age and rarely occur in nulligravid women [7, 11].

The diagnosis of UAVM is challenging given the rarity of the condition and the nonspecific US findings of heterogeneous, anechoic spaces within the myometrium. Included in the differential diagnosis are RPOC, GTD [3, 5, 8, 14], hemangioma, varicosities, uterine malignancies [3, 5], subinvolution [5, 14], fluid-filled bowel loops, mulit-locular ovarian cysts, and hydrosalpinx [3]. The use of color Doppler with spectral analysis can characterize the gray-scale findings further [8]. With UAVMs, continuous high flow throughout systole and diastole with systolic flows four to six times that of normal are demonstrated. Irregular spectral broadening of the waveform reflects increased turbulence. RI measurements are low, and mixing of arterial and venous waveforms is considered diagnostic. Waveforms of pelvic veins distal to the UAVM demonstrate pulsatile flow in contrast to normal monophasic flow [3]. High-flow arterial velocity with peak velocity values

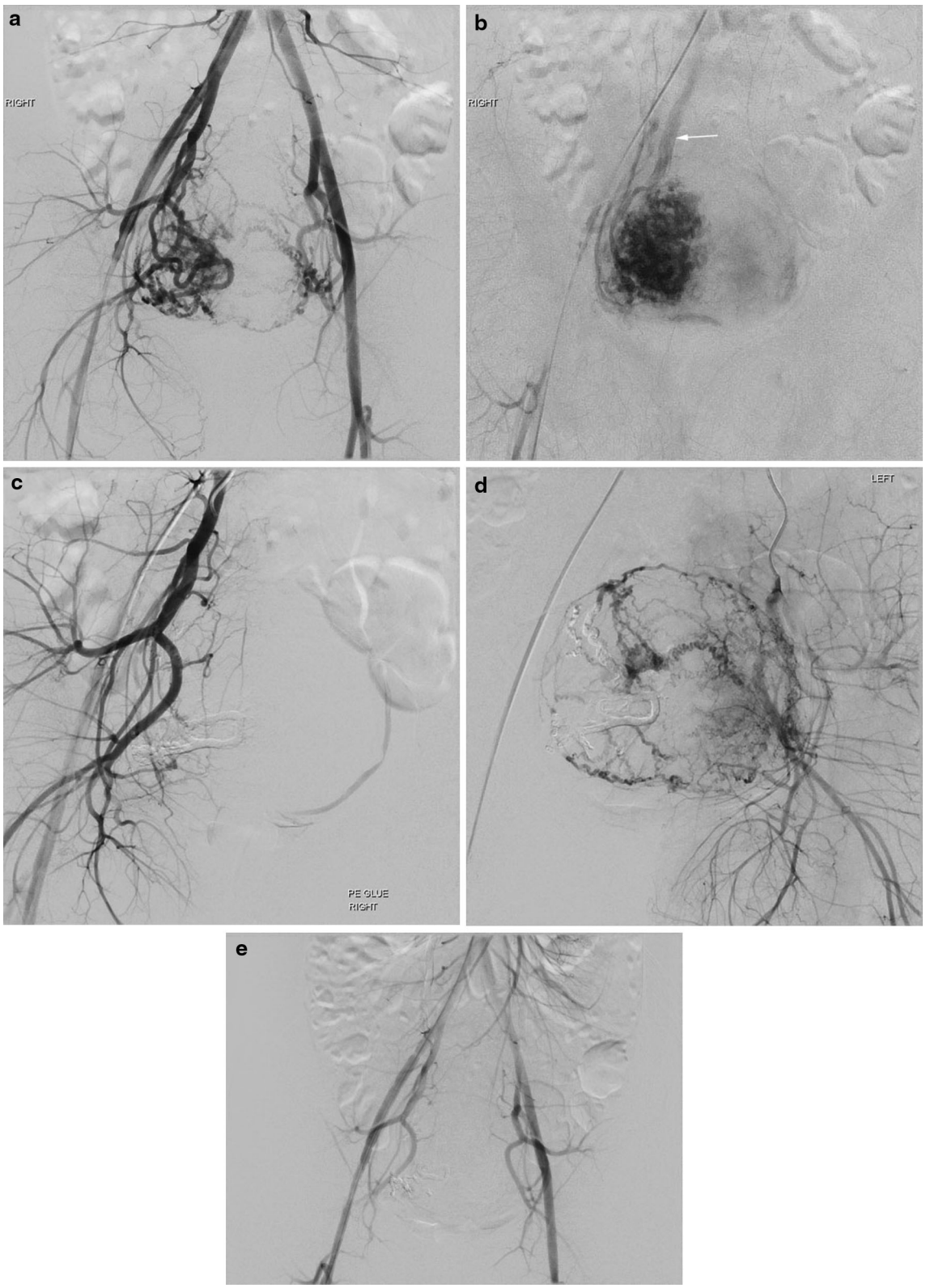

Fig. 4 Right uterine arteriogram demonstrates **a** a large arteriovenous malformation supplied by the right uterine artery and multiple smaller feeding vessels, **b** early venous filling of the right internal iliac vein (*arrow*), and **c** exclusion of flow to the vascular malformation. **d** Left uterine arteriogram demonstrates several feeding vessels to the vascular malformation. **e** Post-embolization aortogram demonstrates exclusion of flow to the vascular malformation

ranging from 40 to 96 cm/s and low RI values ranging from 0.25 to 0.55 are characteristic of UAVMs [7, 8]. If values within these ranges are found, it may give a more precise diagnosis of a UAVM. Like UAVMs, RPOC may also give a hypervascular appearance with turbulent flow on color Doppler imaging. However, beta-hCG levels may be used to distinguish pregnancy-related conditions (GTD, RPOC, and abnormal placentation) from the diagnosis of a UAVM [8].

Due to the similarity in appearance of molar pregnancies and UAVMs on US and color Doppler, it may be worthwhile to perform color Doppler with spectral analysis in these patients followed by angiography for a definitive diagnosis, particularly if they have a past history of a spontaneous abortion or other uterine trauma [7, 8]. In this particular case, we suspect that the molar pregnancy was masking the patient's congenital UAVM, the growth of which was stimulated by molar-related hormonal fluctuations. In our case, the patient did not have color Doppler imaging at the time of her molar pregnancy or when she initially presented with vaginal bleeding. As a result, it is possible that the UAVM could have been diagnosed earlier if these imaging studies were performed. If a UAVM was in fact present, then earlier management options, such as an angiography with TAE could have been utilized. However, as angiography is an invasive procedure, it can be reserved for those with low beta-hCG levels and color Doppler and spectral analysis findings which correspond to a UAVM in patients with persistent and severe vaginal bleeding. In our case, the patient reported persistent vaginal bleeding despite a downward trending beta-hCG. This presentation coupled with her later US and color Doppler findings led us to investigate further with an MRI. Typical MR findings are a bulky uterus with absence of a defined mass and the presence of serpiginous and dilated vessels within myometrium or parametrium [1]. Given her history, the patient likely had growth of a congenital UAVM secondary to hormonal changes induced by her molar pregnancy.

Transarterial embolization (TAE) can be used to confirm the diagnosis angiographically and used to treat the UAVM. Embolization is the preferred treatment for UAVMs as it is effective, can be done without general anesthesia, and can maintain fertility [6, 7, 15]. Hysterectomy, once the treatment of choice, and the most definitive treatment, is now reserved for post-menopausal women or those who need emergent therapy for life-threatening hemorrhage [6] when TAE is unavailable [10], or when patient choice dictates it [9]. In summary, UAVM is a rare vascular lesion that can lead to life-threatening vaginal bleeding. A high index of suspicion for UAVM should be maintained in a patient presenting with unexplained profuse vaginal bleeding with a history of prior pregnancy or uterine trauma. US with color Doppler and spectral analysis, as well as MRI, can aid in the prompt diagnosis of this condition. Angiography, the gold standard for diagnosis, can bridge the gap between diagnosis and definitive treatment. The success of TAE in treating this lesion and in maintaining fertility has led to its ascendance as the treatment modality of choice.

Compliance with Ethical Requirements The Institutional Review Board does not mandate approval for case reports such as this, thus this report meets the Ethical standards as stated in the 1964 Declaration of Helsinki and its subsequent amendments. While a specific consent from the patient for this report was not obtained, our procedure consents obtained prior to each procedure state that we may use images such as the ones here for teaching and illustrative purposes. All patient identifiers in the report and images have been removed.

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