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Arteriovenous malformation of the uterus associated with secondary postpartum hemorrhage

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KEYWORDS: arteriovenous malformation; color Doppler ultrasonography; arterial embolization

ABSTRACT

We present the case of a young woman with persistent secondary postpartum hemorrhage. Transvaginal imaging demonstrated an irregular pulsatile lesion in the anterior myometrium. Color Doppler analysis revealed the presence of abnormal vessels consistent with an arteriovenous malformation. Typically this vascular abnormality had a turbulent pattern of arterial and venous flow with high peak velocities and low resistance. The patient was treated with selective arterial embolization leading to a full recovery. Copyright © 2003 ISUOG. Published by John Wiley & Sons, Ltd.

INTRODUCTION

Arteriovenous malformations of the uterus are rare lesions although it is likely that hitherto they have been underreported. Dubreuil and Loubat[1] reported the first case in 1926 and there are around 30 more cases reported in the literature. In most cases the lesions are acquired and have been attributed to trophoblastic disease, previous pelvic surgery or curettage, and cervical or endometrial malignancy[2,3]. Some cases are thought to be congenital, and in this situation the lesions may be multiple involving other organs of the body.

The classical presentation of uterine arteriovenous malformations is often one of severe uterine bleeding with no obvious cause. Although a definitive diagnosis is usually made by pelvic angiography, transvaginal scanning with color Doppler provides a valuable, non-invasive method of diagnosis[4,5].

This report describes the association of an arteriovenous malformation with secondary postpartum hemorrhage in a young woman 1 month following Cesarean section.

CASE REPORT

This 36-year-old woman achieved a twin pregnancy following her first cycle of *in vitro* fertilization for male factor infertility. Her antenatal course was complicated by one hospital admission for hyperemesis gravidarum at 10 weeks' gestation. Then at 23 weeks' gestation a routine ultrasound scan detected a shortened cervix with obvious funnelling. For this reason an emergency cervical cerclage was performed without complication. The pregnancy continued until 31 weeks' gestation when the patient presented in preterm labor. In view of the patient's history and at the parents' request an emergency Cesarean section was performed. Both female fetuses were delivered in good condition and were transferred to the neonatal unit and were discharged approximately 4 weeks later.

The patient herself experienced a moderate intraoperative blood loss but despite her postoperative hemoglobin falling to 7.7 g/dL she declined transfusion. She was discharged on the sixth postoperative day.

The patient was readmitted 19 days postoperatively with a sudden and profuse vaginal bleed, which settled spontaneously. At this point her hemoglobin was reported as 8.5 g/dL. Based on clinical findings a presumptive diagnosis of endometritis with possible retained products of conception was made. Treatment in the form of intravenous antibiotics was instituted (the patient again declined a blood transfusion). A pelvic ultrasound scan revealed the presence of a small area of probable retained products of conception. An evacuation of the uterus was advised but as the bleeding had appeared to have settled the patient declined and continued conservative treatment and was discharged. She was readmitted 24 h later with a further profuse bleed. The hemoglobin was reported at 7.2 g/dL and the patient agreed to both transfusion and evacuation of the uterus. The evacuation procedure was performed with gentle digital exploration only and a small amount of organized blood clot was removed. There were

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no specific products of conception removed and sharp curettage was not performed. Three days later whilst visiting her babies in the neonatal unit she experienced another large vaginal bleed, became hypotensive and required a further blood transfusion. A transvaginal scan performed demonstrated a large cystic area in the anterior myometrium, which with the addition of color Doppler showed typical features of an arteriovenous malformation. Figure 1 shows the cystic spaces as seen on gray scale imaging. Figure 2 shows the color Doppler image demonstrating the arteriovenous malformation. Pulsed Doppler analysis of the blood flow pattern revealed typical features as seen with arteriovenous fistulae formation. The waveform showed a broad spectral envelope with continuous high-velocity flow throughout systole and diastole (Figure 3). The peak systolic velocity (PSV) was elevated (PSV = 97 cm/s) and the pulsatility index (PI) low (PI = 0.39), illustrating a typical high flow, low resistance, blood flow pattern (resistance index (RI) = 0.32). The patient underwent a pelvic angiogram with a view to selective embolization of the arteriovenous malformation (Figure 4). It was discovered that the malformation was fed predominantly by the right uterine artery, which was selectively embolized with a combination of Gelfoam® (Upjohn, Kalamazoo, MI, •USA) and Embosphere Microspheres® (BioSphere Medical, Inc., Rockland, MA, •USA). There appeared to be no contribution from the left uterine artery therefore this was not embolized.

The patient experienced some mild lower abdominal discomfort and a low-grade fever for 2 days after the procedure but made a full and uneventful recovery. A repeat transvaginal scan performed 1 week later demonstrated a significant change in appearance of the malformation on gray scale imaging (Figure 5) and a markedly reduced blood flow pattern with color Doppler (Figure 6).

DISCUSSION

Arteriovenous malformations arise by definition from an abnormal communication between an artery and a vein. Histological examination of these malformations usually reveals a localized proliferation of both arterial and venous vessels with interconnecting fistulae. Intertwining these muscular vessels there are many thin-walled capillary-type vessels. It has also been recognized that the various proportions of different vessel types may vary[6,7]. This fact most likely explains some of the variations seen in Doppler waveform profiles from the various reported cases of uterine arteriovenous malformations.

Although these vascular anomalies have been reported in both adolescence and following the menopause, they tend to occur predominantly in women of reproductive age and very rarely in women who have not been pregnant. In fact pregnancy appears to have an important role in the pathogenesis of uterine arteriovenous malformations[8]. It is postulated that these malformations may arise when venous sinuses become incorporated in scars within the myometrium after necrosis of the chorionic villi. This is also perhaps why there is an association with trophoblastic disease, in particular following treatment with chemotherapeutic agents.

From a clinical perspective these vascular anomalies most commonly present with abnormal uterine bleeding. Rarely the patient may present with a pulsatile mass in the pelvis. The bleeding may be severe and result in

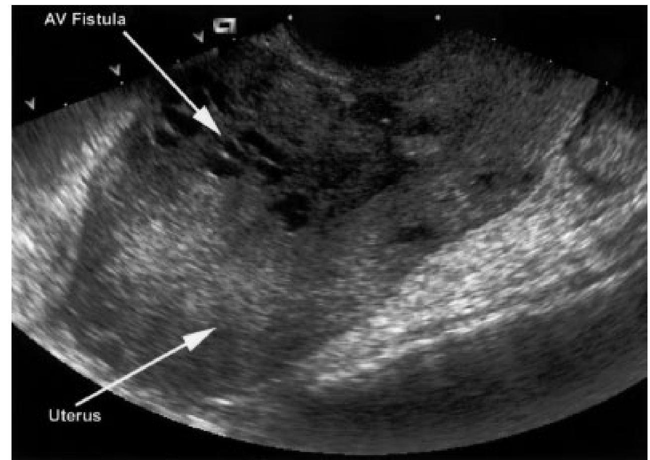

Figure 1 Gray scale image of the arteriovenous malformation with prominent cystic spaces within the myometrium.

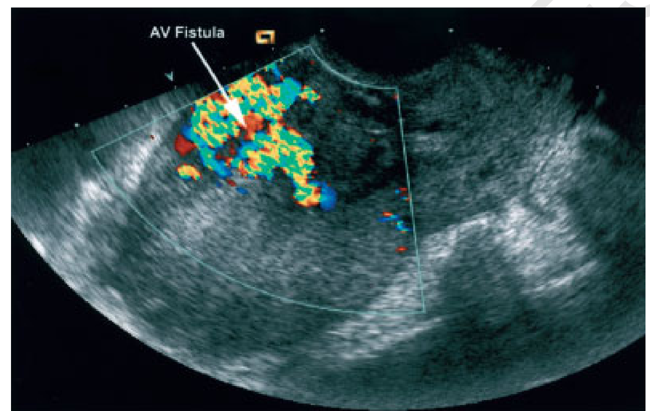

Figure 2 Color Doppler image of the arteriovenous malformation.

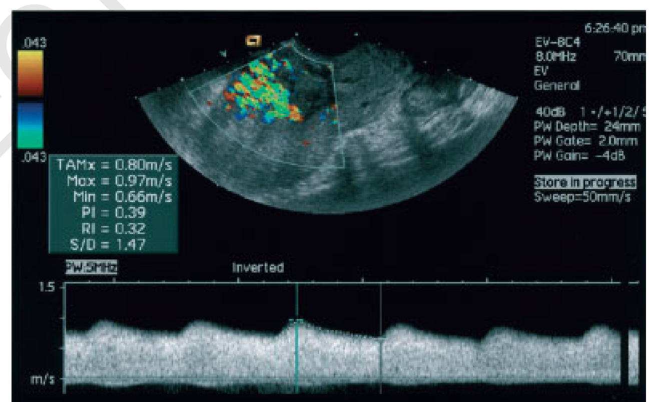

Figure 3 Pulsed Doppler image showing the broad spectral waveform with continuous flow throughout systole and diastole.

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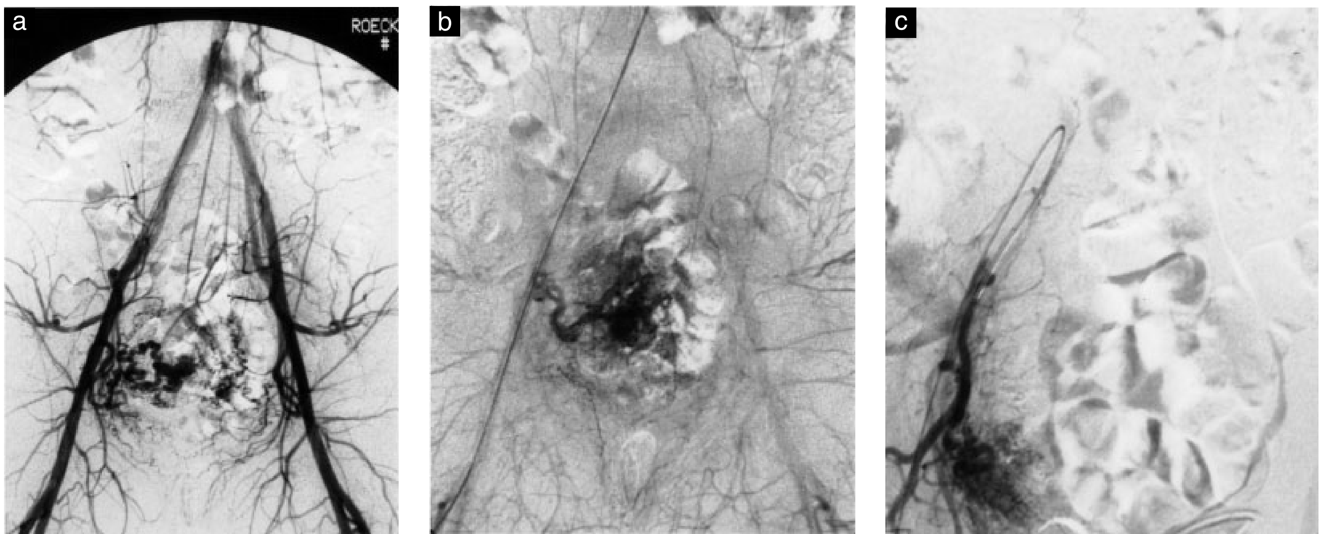

Figure 4 Arteriograms pre- and post-embolization. (a) Early phase arteriogram showing filling of the uterine artery and the arteriovenous malformation. (b) Later phase of the same injection showing early filling of a prominent draining vein from the uterus. The tip of the catheter in (a) and (b) is in the tight internal iliac artery. (c) Post-embolization there is absence of filling of the occluded uterine artery.

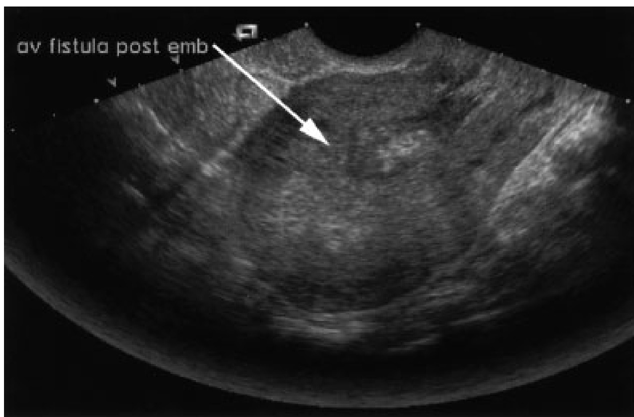

Figure 5 Gray scale image of the arteriovenous malformation following selective embolization.

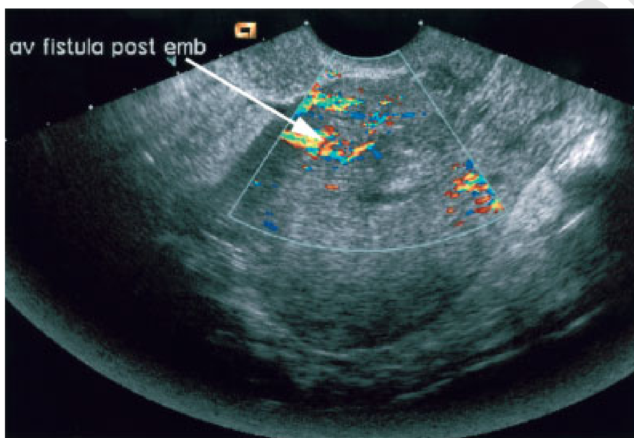

Figure 6 Color Doppler image following embolization showing the markedly reduced blood flow.

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significant anemia or even shock. Torrential bleeding may follow uterine curettage as there is often only a thin layer of endometrium overlying the malformation, which may be disrupted. Treatment has often been in the form of hysterectomy.

Diagnosis of uterine arteriovenous malformations has traditionally proved difficult. Standard diagnostic procedures such as pelvic examination, hysteroscopy and gray scale ultrasonography have not proved to be adequately robust. The current gold standard method of diagnosis is pelvic angiography. This invasive procedure allows confirmation of the diagnosis but also helps to identify the leading 'feeder' vessels where embolization may be indicated as a conservative treatment option[9,10].

Gray scale ultrasonography alone may play a role in diagnosis but the features of arteriovenous malformations may be similar to other pelvic structures or pathologies[11]. The addition of color Doppler improves the diagnostic ability of ultrasonography[12]. A localized area of increased vascularity within the myometrium itself typifies these lesions. Pulsed Doppler evaluation of the identified area will normally reveal a low-resistance blood flow with high peak velocities and evidence of turbulence. The waveform is usually broad with an irregular spectral envelope indicating a turbulent flow resulting from the many direct arteriovenous connections that are present. There is usually continuous high blood flow throughout both the systolic and diastolic components of the cardiac cycle. Analysis of the waveform will show a typically high PSV with low values for both the RI and PI.

Therefore the prime advantage of Doppler ultrasound is that it provides a non-invasive method of diagnosis of uterine arteriovenous malformations. Angiography can be reserved as a therapeutic option where selective embolization of the vessels feeding the malformation can be performed. The patient can then be asked to give consent for embolization prior to angiography, thereby avoiding the need for a second therapeutic procedure. Additionally, localization of the lesion by

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prior Doppler ultrasound may aid the embolization procedure.

In our patient, the diagnosis of arteriovenous malformation by color Doppler determined the correct approach to management, which was uterine artery embolization. Without a scan and color Doppler assessment it is probable that curettage would have been performed on the basis of the likely presence of retained products of conception with the risk of massive uterine hemorrhage. It is recommended that a transvaginal scan and color Doppler assessment should be performed on any woman with moderate to severe secondary postpartum hemorrhage to exclude this rare but dangerous abnormality.

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