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ORIGINAL ARTICLE

Uterine Arteriovenous Malformation: Case Series and Literature Review

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About the Author

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Abstract

Background Uterine AV malformation is a rare cause of torrential post-abortal hemorrhage, which can present with varying grades of severity. Diagnosis requires a high degree of suspicion and is done with ultrasound and Doppler.

Case Series In our institution, during the period 2008–2013, five cases of symptomatic uterine AVMs have been reported. All of them were in the reproductive age group (22–36 years), presenting with a history of miscarriage or termination of pregnancy for which curettage was done. The presentation was with recurrent bouts of torrential bleeding, some triggered by second curettage, and not controllable with regular measures. Diagnosis was by ultrasound-gray scale, color Doppler, and spectral Doppler. The time interval between the onset of symptoms and the primary curettage was 8–89 days; four patients underwent selective arterial embolization, and one patient opted for hysterectomy. On follow-up, all the four patients are presently free of symptoms; two of them conceived within 2 years of the procedure and carried the pregnancy to term—one resulting in a live-birth and the other intrauterine death.

Conclusion Uterine AV malformation should be thought of as a differential diagnosis in all cases presenting with bleeding after miscarriage or curettage, since diagnosis is simple and treatment by selective arterial embolization saves morbidity of surgery and anesthesia, and more importantly reduces hospital stay and the absence from work.

Keywords Arteriovenous malformation · Uterine artery embolization · Vascular malformations · Gelfoam

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Introduction

Uterine AV malformation is a rare and under diagnosed cause of hemorrhage after diagnostic or therapeutic curettage. It is a potentially life-threatening condition, and diagnosis requires a high degree of suspicion on the part of the caregiver—and prompt diagnosis could save many a uterus and thus preserve fertility, due to the availability of conservative alternatives to hysterectomy.

Objective

To study the clinical features, presentation, diagnostic, and treatment options in a series of five cases of symptomatic uterine AV malformations, who presented to the general Gynaecology OP of our hospital during the period 2008–2013.

Methods

A retrospective analysis of case records during 2008–2013 revealed five cases of symptomatic uterine AV malformation. All of them presented with recurrent bouts of torrential bleeding following curettage. Some had been treated with second curettage by the first attending doctor that only aggravated the hemorrhage. Complete hemogram and serum beta HCG were done. All were anemic at presentation, requiring blood transfusion. Ultrasound and Doppler were used for diagnosis, which was confirmed by angiography prior to uterine artery embolization (four cases), and one patient opted for hysterectomy.

Case Series

Case 1

A 32-year-old, P1L1A1, was admitted to the hospital on July 17, 2008 following profuse bleeding pv for 1 week/changing 6–7 pads per day. Her last pregnancy was a missed abortion 2 weeks earlier for which she had undergone curettage with sterilization. Her hemoglobin was 6.6 g/dl, and ultrasound showed irregular hypo-echoic area of size 1.8 × 1.0 cm in the posterior myometrium which had continuity with the uterine arteries on both sides. Despite counseling for embolization, she opted for hysterectomy, and TAH with RSO was done.

Case 2

A 36-year-old, P1L1A1, with previous LSCS, was admitted with bleeding pv × 10 days on December 10, 2008. She had an induced abortion in September and after having a normal cycle in October, went to the local doctor for increased bleeding in November—curettage was done on December 2 following which she had increased bleeding. Hemoglobin on admission was 8.4 g/dl; ultrasound showed AV malformation in the upper anterior part of the uterus. Per-operative angiogram showed AVM with feeders from both uterine arteries and right ovarian artery (Fig. 1). The embolization of the uterine and ovarian arteries were carried out through selective retrograde cannulation from both femoral arteries. The procedure was completed after the final angiogram showed complete occlusion (Fig. 2). She conceived spontaneously in February 2009, and was admitted on November 28, 2009 at term with IUD and delivered vaginally a 2.92-kg baby.

Case 3

A 22-year-old, P1L1A1, was admitted on August 13, 2010 with H/O profuse bleeding pv after curettage one-and-a-half month earlier. She had been admitted to a local hospital with similar complaints, where her Hb was 6.9 g/dl and she was transfused with 2 units of blood, and then referred to us due to failed medical management. USG showed echo-poor lesion of size 3.7 × 2.8 × 3.3 cm in the left side of fundus with florid vascularity (Fig. 3). She was transfused two more units of blood, and embolization of left uterine artery was done on the same day.

Case 4

A 29-year-old lady with H/O 3, previous abortions, the latest one on November 7, 2010 was admitted in our hospital on December 29, 2010 with profuse bleeding pv for the last 20 days. Hb on admission was 9.7 g/dl, and USG showed AVM on the right side (Figs. 4, 5). Per-op angiogram confirmed the findings (Fig. 6), and embolization of right uterine artery was done. She had a normal vaginal delivery at term in 2012.

Case 5

A 34-year-old, P3L3A3, with previous history of one LSCS and three terminations was admitted on September 11, 2013 with profuse bleeding and last curettage in August. USG showed serpiginous tubular anechoic spaces within the myometrium largest 2.2 × 1.8 cm with high-velocity low-resistance flow (Fig. 7). Hb on admission was 4.6 g/dl, she was transfused 2 units of PRBC, and taken up for treatment the next day. Per-op angiogram showed a large AVM with feeders from both uterine arteries and anterior division of left internal iliac artery. Left uterine and anterior division of internal iliac artery were embolized with 0.35 × 5 × 3 mm coil, and right uterine artery embolized with gel foam.

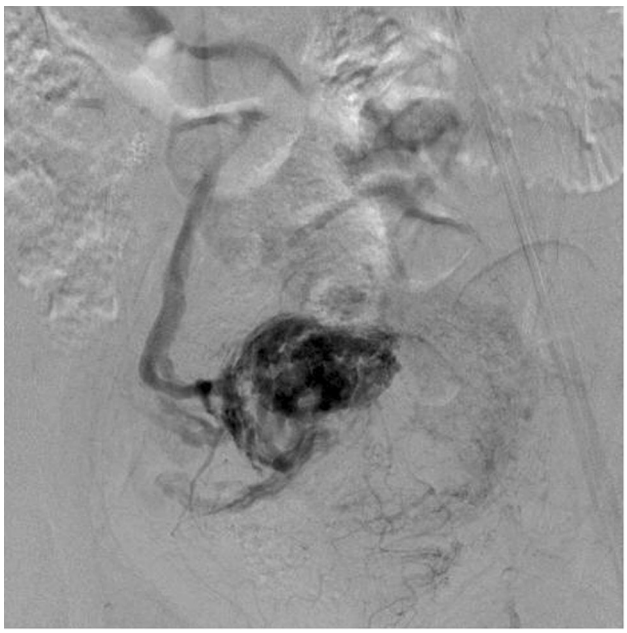

Fig. 1 Angiographic picture of Case 2 showing AV malformation

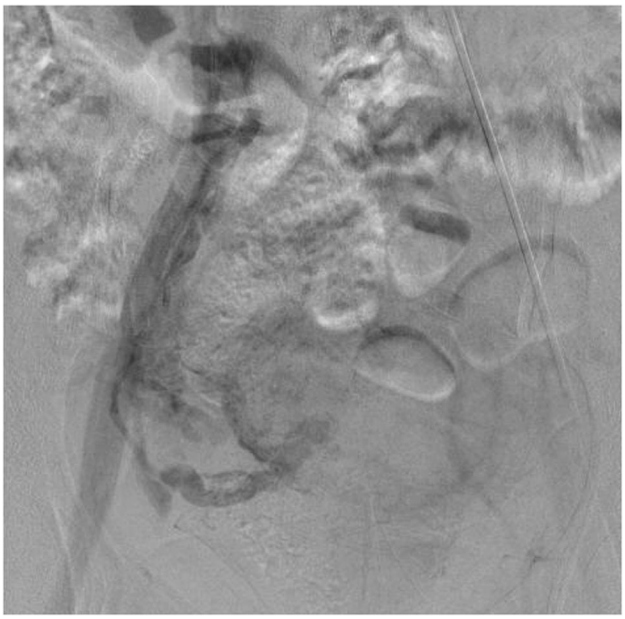

Fig. 2 Angiographic picture of Case 2 showing cessation of blood flow following embolization

Results

All patients were offered the option of selective arterial embolization. Our first patient refused embolization and chose to have hysterectomy instead. The other four patients were taken up for embolization after obtaining informed consent and addressing fertility issues. The procedures were

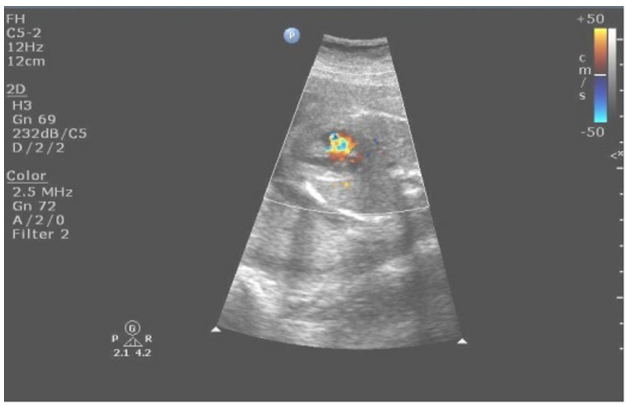

Fig. 3 USG picture of Case 3 showing AV malformation close to the fundus

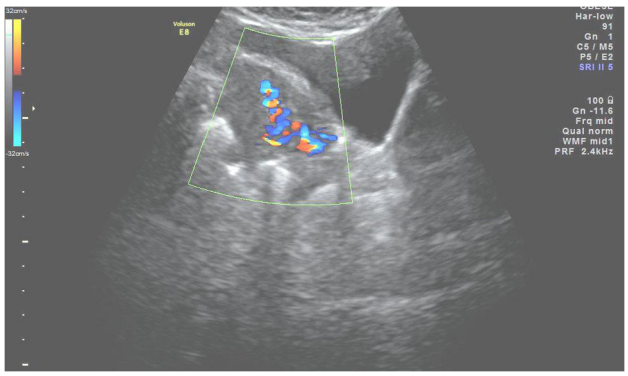

Fig. 4 USG picture of Case 4 showing the serpiginous vessels on the left side

performed by two cardiologists trained in interventional Cardiology. Under local anesthesia and access through femoral artery, the target vessel was embolized with gel foam, and cessation of flow was confirmed. The patients showed improvement and were discharged in 2 days.

Discussion

Uterine arteriovenous malformations may be congenital or acquired; acquired or traumatic AVMs represent multiple small AV fistulae in the myometrium and may have unilateral/bilateral uterine artery feeders [1]. AVM is a rare entity, may be due to under reporting. O'Brien et al. put the incidence at 4.5 % [1]. Typically, the patient presents with torrential vaginal bleeding, following a D&C, therapeutic abortion, or uterine surgery. All the patients in our series had a prior curettage for termination/incomplete miscarriage. Diagnosis is by clinical suspicion. Diagnostic modalities include ultrasound (with Doppler [2], spectral Doppler, and 3D power doppler [3, 4]).

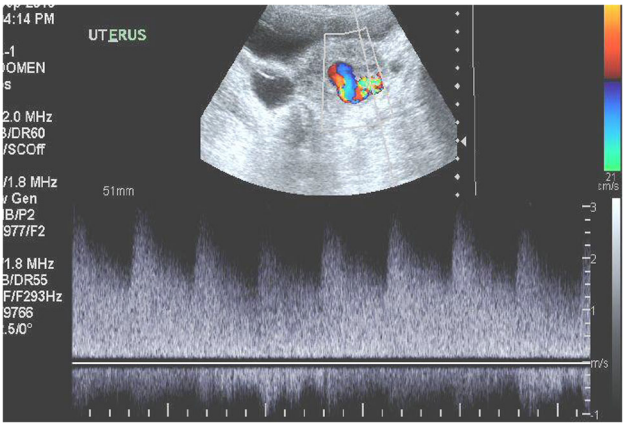

Fig. 5 USG picture of Case 4 showing Doppler flow with low-resistance high-velocity flow

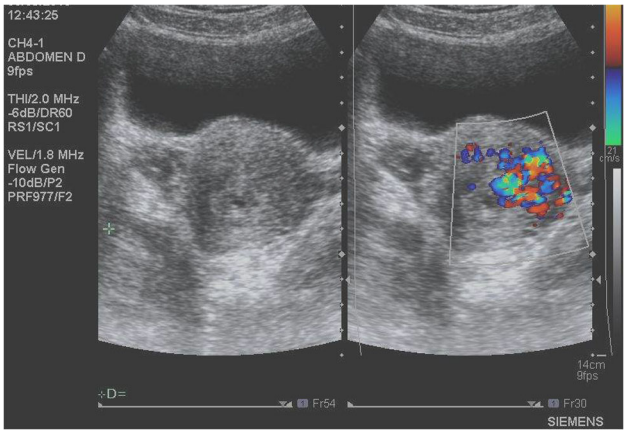

Fig. 7 USG picture of Case 5 showing AV malformation with feeder vessels

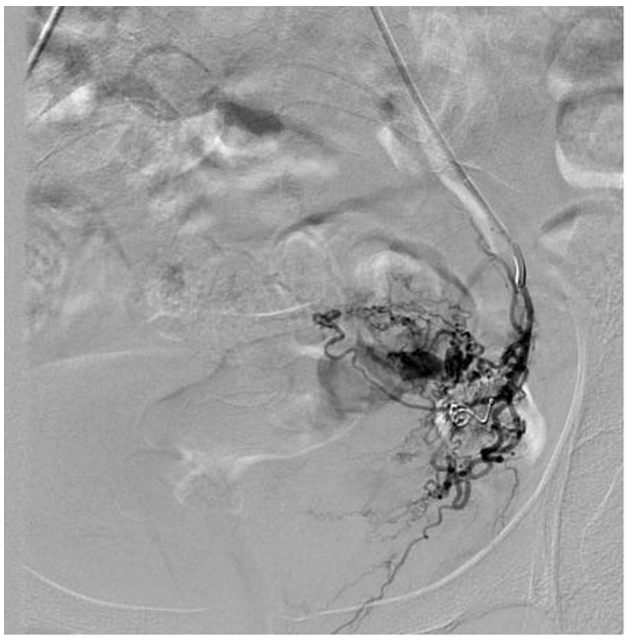

Fig. 6 Angiographic image of Case 4 showing large AV malformation in the right uterine artery

Gray scale imaging may reveal subtle myometrial heterogeneities or anechoic spaces [1]; Color Doppler shows a tangle of vessels with high velocity flow; spectral Doppler shows high-velocity, low-resistance flow with RI (resistance index) values ranging from 0.25 to 0.55 and peak systolic velocity (PSV) values in the range of 40–100 cm/s [4]. The diagnosis in all our cases was made by TAS/TVS with Doppler.

Uterine AVM may be symptomatic or asymptomatic—regression of asymptomatic AVMs have been reported [5, 6]. Peitsidis et al. [5] report spontaneous resolution in 6 % of patients; Sellers et al. [7] report conservative management of uterine AVM. Timmermann et al. [2] state that conservative management is effective in two-thirds of patients with a sonographically diagnosed AVM, particularly when PSV <40 cm/s; in cases where PSV >80 cm/s and early venous filling was demonstrated on angiography, definitive treatment was necessary. 14/21 cases reported by O'Brien et al. were symptomatic requiring embolization. Symptomatic AVMs require intervention in the form of artery embolization [8, 9], uterine artery ligation, or hysterectomy. All the five cases in our series were symptomatic. Four were treated with selective arterial embolization and one by hysterectomy. In their systematic review of 85 case reports involving 100 patients, Peitsidis et al. [5] reported that uterine artery embolization (UAE) was the commonest treatment option (59 %), followed by TAH (29 %). Common agents used for embolization include gel foam [1, 8] PVA, or glue (*N*-butyl cyanoacrylate) [9]. Ghai et al. [9] report a technical success rate of 100 %, and clinical success rate of 93 % in their series.

The embolization procedures in our hospital were performed by two cardiologists trained in interventional cardiology. Through femoral artery access, pre-procedure angiography was done for confirming the presence of AVM. Gel foam was the agent used in all the four cases. One case required an additional coil placement due to the complex nature of AVM. Cessation of flow was confirmed at the end of the procedure.

Complications which are often reported include pelvic pain, local hematoma, and rarely, skin sloughing (in the case of internal iliac artery embolization or the use of glue). Pregnancies have been reported following embolization [5, 8–12], proving that an adequate collateral supply can develop to support a full-term pregnancy. Peitsidis et al. report a 27 % pregnancy rate following bilateral UAE.

Two out of four patients in our series conceived in a year following embolization. Pregnancy proceeded to full

term in both cases—one resulting in a live birth and the other intrauterine death.

Conclusion

Uterine AV malformation should be thought of as a differential diagnosis in all cases presenting with bleeding after miscarriage or curettage, since diagnosis is simple; treatment by selective arterial embolization reduces morbidity of surgery and anesthesia, and is cost effective; reduces hospital stay and the absence from work.

Acknowledgments The authors acknowledge the services of all the staff who helped us in the management of these cases.

Compliance with ethical requirements and Conflict of interest All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008(5). Informed consent was obtained from all patients for the surgery. An ethical clearance has also been taken from the institutional ethical committee. The authors declare that they have no conflict of interest.

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