Selective Arterial Embolisation of Uterine Arteriovenous Malformation in an Infertile Patient, subsequent Term Pregnancy and Postpartum Recurrence

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ABSTRACT

Uterine arteriovenous malformation (AVM) is a rare entity. This report describes selective catheterisation and embolisation of a uterine AVM in an infertile woman, subsequent spontaneous pregnancy and postpartum recurrence of the AVM treated with a second embolisation procedure. A 22-year woman presented with menometrorrhagia and failure to conceive. Pelvic magnetic resonance imaging showed a large uterine AVM. Selective catheterisation and embolisation of the AVM was performed. The patient conceived spontaneously two months later and delivered vaginally. A second embolisation was performed due to recurrence of uterine AVM at six months postpartum.

This is the first case reporting postpartum recurrence of a pre-conceptionally treated uterine AVM. Selective catheterisation and embolisation is a minimally-invasive, fertility-preserving procedure that successfully treats uterine AVM and should be the treatment of choice when the patient desires pregnancy. Successful pregnancy and vaginal delivery following embolisation is possible; however, an increased awareness of postpartum AVM recurrence is required.

Key Words: *Fertility, Pregnancy, Therapeutic embolisation, Uterus, Arteriovenous malformation.*

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INTRODUCTION

Uterine arteriovenous malformations (AVMs) are abnormal communications of arteries and veins by-passing capillaries. Although the true incidence of uterine AVM is unknown, it is a very rare condition, with less than 100 cases reported in the literature.[1] One prospective study from Japan investigated the incidence of uterine AVM in women who had an abortion or delivery and reported this to be approximately 0.10%.[2]

Doppler ultrasound examination is the initial step for uterine AVM diagnosis. Subsequent magnetic resonance imaging (MRI) allows for accurate evaluation of the AVM.[1] Since most women with uterine AVM are of reproductive age, the desire to maintain fertility influences the choice of available treatment options. Conservative management with embolisation treats uterine AVM and preserves the uterus. However, reports of successful pregnancy following this technique are scarce.[3]

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This case report describes selective arterial catheterisation and embolisation of a uterine AVM in a subfertile woman with subsequent spontaneous pregnancy leading to a live birth. Six months postpartum, there was a recurrence of the AVM which was successfully treated with a second embolisation procedure.

CASE REPORT

A 22-year woman, gravida 1, para 0 with a previous molar pregnancy, presented with heavy menometrorrhagia of eight months duration and failure to conceive. She had undergone uterine curettage for suspected mole one year prior to presentation and histopathological examination confirmed a diagnosis of complete hydatidiform mole (CHM). She had a history of referral to multiple clinics for vaginal bleeding and had been treated with estrogen and progesterone regimens. However, the bleeding was unresponsive to hormone therapy.

Color Doppler ultrasound examination showed a large, uterine AVM on the left side. Pelvic MRI revealed multiple, vascular-like, serpiginous structures within the myometrium (Figure 1 a-c). As the patient desired future fertility, selective embolisation was planned. Patient's written informed consent for the procedure was obtained. Angiography revealed a 5×3 cm AVM involving the left uterine artery (Figure 2a). Selective catheterisation of the left uterine artery was performed and the AVM was successfully occluded using 900-1200 µm and 700-900 µm acrylamido polyvinyl alcohol microspheres (a-PVAM) (BeadBlock; Biocom-

patibles, Farnham, UK). Control angiography demonstrated complete occlusion of the uterine AVM with no residual flow through the arteriovenous shunt (Figure 2b). Her postoperative recovery was uneventful and she was discharged home on post-operative day 2. The patient's menstrual cycles subsequently regularised and this was accompanied by decreased menstrual blood loss.

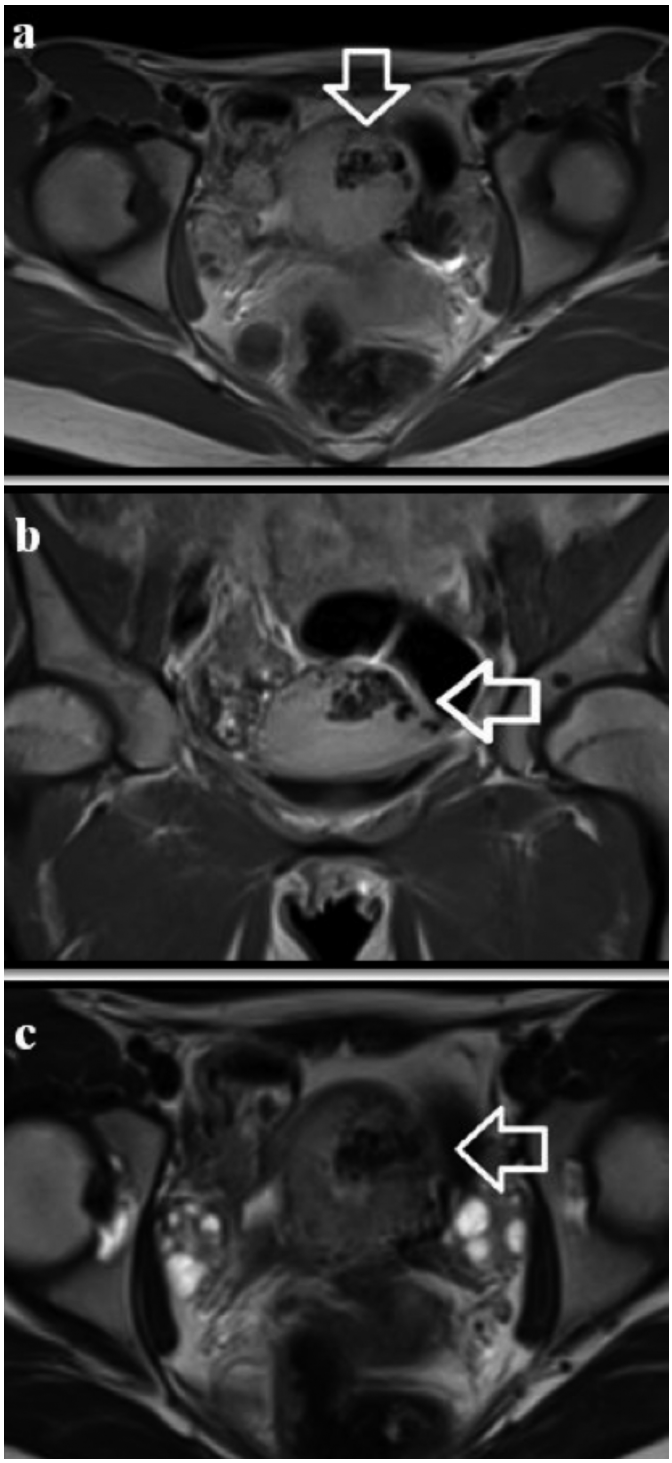


Figure 1: (a,b,c) Pelvic MRI- (a) axial, (b) coronal, (c) post-contrast T1-weighted sequences, T2-weighted TSE sequences. The images show multiple vascular-like serpiginious structures characterised by flow void in the lateral wall of the fundus (arrow).

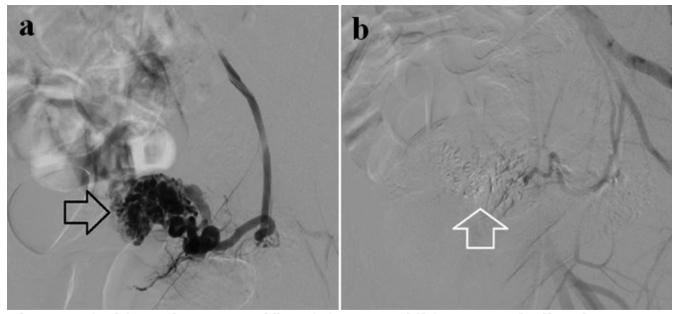


Figure 2: (a,b) Angiograms of first (a) pre- and (b) post-embolisation procedure. The pre-embolisation arteriogram showed a tangle of dilated vascular structures (black arrow), and post-embolisation images showed occluded vascular malformation and embolising agent (white arrow).

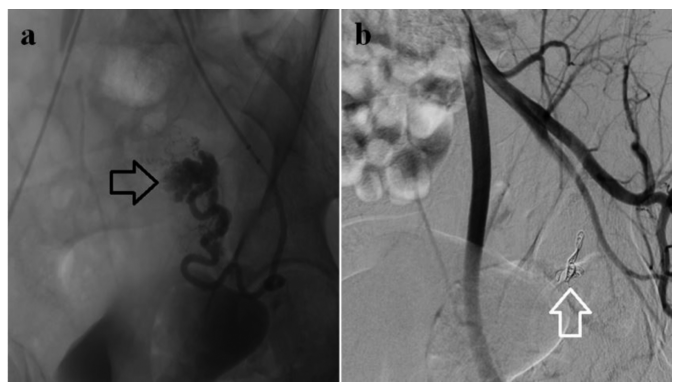


Figure 3: (a,b) Angiograms of the second (a) pre- and (b) post-embolisation procedure. The pre-embolisation arteriogram showed a tangle of vascular structures (black arrow), and post-embolisation images showed occluded left uterine iliac artery and vascular malformation with an embolisation coil (white arrow). Resolution of AVM is evident.

The patient conceived spontaneously two months after the procedure. She had an uncomplicated pregnancy and labour began at 40 weeks. A healthy newborn, weighing 2,750 g with a 9-10-10 Apgar score, was delivered vaginally. Both the volume of postpartum bleeding and the postpartum recovery were normal. Six months after the birth, the patient presented to our emergency department with profuse vaginal bleeding and symptomatic anemia. Her hemoglobin concentration was 8.1 g/dL and serum beta-human chorionic gonadotropin (β-hCG) was negative. Color Doppler ultrasound revealed a 43×28 mm myometrial lesion in left lateral wall of the uterus with hypoechoic areas demonstrating high velocity blood flow which indicated recurrence of the AVM. She received two units of red blood cells preoperatively. A second angiography, showing a tangle of dilated vessels, confirmed the presence of a recurrent AVM. Selective catheterisation of the left uterine artery was performed and AVM was occluded with 900-1200 µm a-PVAM followed by placement of a Target 360 Ultra 4 mm ×10 cm coil (Stryker, Fremont, CA., USA). Post-embolisation angiography demonstrated complete embolisation of the AVM (Figure 3a and 3b). Her postoperative hemoglobin concentration was 10.2 g/dL. She had an uneventful post-operative course and reported regular periods with light menstrual bleeding at six month follow-up.

DISCUSSION

Acquired AVM formation has been associated with uterine surgery and gestational trophoblastic disease.[1,4] In this case,

the patient experienced profuse vaginal bleeding after undergoing uterine evacuation for CHM, suggesting that the molar pregnancy and/or uterine curettage may have led directly to the acquired AVM.

Failure to conceive was the chief complaint together with vaginal bleeding in this case. In addition, the patient conceived spontaneously two months after embolisation. There are a few other reports of pregnancy shortly after embolisation of a uterine AVM.[3] Thus, it is plausible that the presence of a uterine AVM has a negative effect on fertility, although robust evidence for this causality is scarce.

There have only been a few previous reports of pregnancy following uterine AVM embolisation in the literature.[3] Placental abnormalities and postpartum hemorrhage have been reported.[5,6] Poor vascularisation of the treated area affecting placentation has been implicated as a potential cause.[5] However, our case had an uneventful pregnancy course and an uncomplicated vaginal delivery. There are few other case reports describing successful pregnancy outcome.[7] In the current case report, selective embolisation of the AVM and preservation of other arteries may have contributed to improve obstetric outcome.[3]

Recurrence of the AVM occurred at six months postpartum in our case. High recurrence rates up to 16.7% have been reported in the literature.[8] However, this is the first case reporting postpartum recurrence of a pre-conceptionally treated uterine AVM. The pathogenesis of uterine AVMs has been associated with pregnancy and accompanying hormonal changes.[6] Thus postpartum recurrence may be expected.

Clinicans should suspect a uterine AVM, especially in patients with unexplained persistent vaginal bleeding. Besides life-threatening bleeding, uterine AVMs can also affect fertility. Selective catheterisation and embolisation is a minimally invasive, conservative alternative to hysterectomy and successfully treats uterine AVM. Successful pregnancy and vaginal delivery following embolisation is possible. However, clinicians should be aware of the recurrence of the AVM postpartum.

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PATIENT'S CONSENT:
Written patient consent was obtained for publication of the case report.

CONFLICT OF INTEREST:
The authors declared no conflict of interest.

AUTHORS' CONTRIBUTION:
EC: Conception and design.
OC: Data collection, manuscript writing.
OSYC: Manuscript writing, submission, proofreading.
IC: Patient recruitment, data collection.

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