



Case Report

Fertility outcomes following pelvic embolization in women with acquired uterine arteriovenous malformation

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ABSTRACT

Objective: Acquired uterine arteriovenous malformation (UAVM) is a rare, life-threatening disease. Angiography with uterine arterial embolization (UAE) is the diagnostic tool and a choice of fertility-sparing treatment. Here, we present a series of five successful pregnancies after embolization of UAVM. **Case reports:** Three reproductive aged women were treated for UAVM, resulting in five successful pregnancies. Their past history suggested that three cases had had previous uterine procedures, including second trimester abortion and elective dilatation and curettage. Intermittent heavy vaginal bleeding was the primary symptom of UAVM. One patient with anemia had two ineffective embolizations and achieved a singleton pregnancy after the third embolization. However, intrauterine fetal demise with severe fetal growth retardation was noted on the 28th gestation week. The other two women had temporary ovulation disorder after UAE. After Clomiphene Citrate (CC) treatment, successful pregnancies were achieved and carried to term uneventfully. **Conclusion:** UAE is an acceptable method for preserving fertility and treatment in women with symptomatic UAVMs.

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Introduction

Uterine arteriovenous malformation (UAVM) is a rare condition that affects mostly 20–40 year-old women with life-threatening uterine bleeding [1]. Acquired UAVM may result from uterine trauma, previous abortion, or neoplastic disease [2]. Congenital forms are extremely rare. The main symptoms are mild intermittent vaginal bleeding to profuse menstrual flow. In 30% of cases, the bleeding is so heavy that blood transfusions are needed [3]. Recurrent pregnancy loss (RPL) has been reported in some UAVM cases [1,4]. Small cystic lesions detected in the myometrium or below the endometrium via gray scale trans-vaginal ultrasound (TVUS) would be a sign arousing suspicion. Abnormal hyper-vascular area with blood vessels in the myometrium via color Doppler ultrasound is the characteristic finding of arteriovenous malformation [3,5]. A low-impedance and high-flow feature

presented in ultrasound is also an important clue [3]. According to Timmerman et al. in 2006, the ultrasonographic features may aid in determining the management of UAVMs for which inappropriate uterine curettage could result in catastrophic bleeding [1,6].

Pelvic angiography is the gold standard diagnostic tool. The positive findings are two typical traits: engorgement of uterine vessels and rapid venous return. Previous case reports classified them into two categories: true AVMs and non-AVMs [6]. Non-AVMs lack typical angiographic traits and result from the sub-involution of the endometrial bed, and their incidence is much higher than true AVMs (83%: 17%; n = 15:3) [6]. Uterine arterial embolization (UAE) could be an effective and safe therapy for UAVM, although 17% of patients needed repeated embolization [1,7,8]. However, the preservation of fertility after embolization remains controversial [9–11]. In 2013, the American College of Obstetrics and Gynecology (ACOG) declared that UAE in treating uterine myoma or post-partum hemorrhage may affect further fertility [12]. Other case reports for true UAVMs after UAE are scattered and seldom mention their fertility outcomes. In a systemic review in 2011, 17 of 59 (29%) patients had been reported to have subsequent pregnancies after UAE for UAVM [1]. Therefore, it is important to share the experience

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of management of those cases that, in their reproductive age, attempt pregnancy. Here, we report our case series of women with UAVM after UAE and discuss fertility outcomes.

UAE methods

This case series was conducted retrospectively. All cases were enrolled based on angiographic diagnosis with the typical angiographic features: rapid venous filling of engorged uterine vessels.

All patients presented with heavy or intermittent vaginal bleeding between 2008 and 2016. In our series, UAVM was initially diagnosed with trans-vaginal Doppler ultrasound and confirmed by pelvic angiography. UAE was performed in a dedicated angiography suite by qualified interventional radiologists under sedation and local anesthetic. In all cases, one or the other side of the common femoral artery was punctured according to the disease location. Under aseptic conditions, a 6 Fr. guiding catheter was introduced in one side of the internal iliac artery. Two microcatheters were placed in the branches of the bilateral internal iliac vessels, respectively, and were advanced to as near the vessel structure as possible. Once a stable position of the catheter was set up, embolizations were performed with glue polymerization (N-Butyl Cyanoacrylate/NBCA).

All patients after UAE were followed until cessation of vaginal bleeding and their deliveries. Data were collected through electronic or paper chart records with informed consent from all patients.

Case reports

Case 1

A 22-year-old female, gravida 3, abortion 3, had received second trimester termination at 15 weeks of gestation due to preterm premature rupture of the membrane (PPROM) at a local clinic. Before that episode, she had undergone two elective abortions at 8

weeks' gestation. Menorrhagia occurred 2 months after the last abortion. TVUS revealed a 2.6 cm heterogeneous uterine lesion at the anterior fundus with strong turbulent flow. The serum beta-hCG level showed below 5 mIU/ml. With the suspicion of uterine AVM, selective pelvic angiography was performed to confirm the diagnosis. Then embolization was performed by supra-selection of bilateral uterine arteries with glue (25% n-Butyl Cyanoacrylate/NBCA). Three months after the procedure, TVUS revealed complete resolution of the AVM. However, anovulation bleeding was noticed for 3 months. Clomiphene Citrate 100 mg per day from day 3 to day 7 was given for 5 cycles. Serial monitoring of the follicle size and subcutaneous injection of human chorionic gonadotropin (Ovidrel 6500IU) was performed. Eleven months after UAE and 5 cycles of CC, she achieved pregnancy and had an uneventful term vaginal delivery. Three years later, she had another pregnancy without medical assistance. However, the second birth was accompanied by precipitate labor, post-partum hemorrhage and a small gestational age (Tables 1 and 2).

Case 2

A 38-year-old female had had three elective abortions over the previous 10 years. She had suffered from hypo-menorrhea after the most recent abortion. In recent months, she had experienced severe abdominal pain accompanied by heavy vaginal bleeding after strenuous activity. According to her statement, the condition became worse gradually. TVUS revealed a 3.6 cm heterogeneous cystic myometrial mass with strong turbulent flow at the posterior fundus. Waveform study also showed a decreased resistant index (RI). A posterior UAVM was diagnosed by angiography and selective UAE was performed through supra-selection of both uterine arteries. Post-embolization suprapubic pain was controlled by morphine. Complete stasis of AVM was noted by TVUS one month after embolization. However, dysfunctional uterine bleeding (DUB) was noted for 2 months after UAE. She attended our hospital for a failed trial of pregnancy

Table 1
Clinical characteristics of three women with true UAVM, treatment and outcome.

Case	Abortions/uterine surgery history	Age (year-old)	Initial symptoms	UAE	Fertility outcome
1.	SA1AA2 (SA at 15 weeks of pregnancy)	22	Menorrhagia 2 months after abortion, 2.6 cm AVM at anterior fundus, nidus at midline.	Supraselection of bilateral uterine arteries with NBCA 25%, complete resolution.	Pregnancy achieved by clomiphene citrate (CC), 11 months after embolization. The 2nd pregnancy achieved without medical assistance.
2.	AA3	38	Severe abdominal pain and menorrhagia, 3.6 cm AVM at posterior fundus, nidus at midline.	Supraselection of bilateral uterine arteries with NBCA 30%, complete resolution, post-TAE ischemic pain.	Pregnancy achieved 2 years after embolization. The 2nd pregnancy achieved without medical assistance. All were term with vaginal delivery.
3.	AA1	34	Massive vaginal bleeding attacked 3 years after D&C, 3.8 cm AVM at posterior fundus, nidus deviation to right side.	Bilateral internal iliac arteries with NBCA and lipidol, TAE completed after 3 trials	Pregnancy achieved 1 year after last embolization. A 28 weeks of pregnancy complicated with IUGF, IUGR and pre-eclampsia. (Birth weight 600 g).

AA (artificial abortion), DUB (dysfunctional uterine bleeding), SA (spontaneous abortion), UAE (uterine arterial embolization).

Table 2
Pregnancy outcome after embolization of uterine arteriovenous malformation in the study.

Case	Maternal Age at pregnancy (years)	Gestational Age (weeks)	Gender	Weight	Apgar score	Complications
1.	24	39	F	3020 g	9' → 9'	N/A
	25	39	F	2370 g	9' → 9'	SGA, Post-partum hemorrhage
2.	40	37	F	2892 g	9' → 9'	N/A
	42	37	M	2850 g	9' → 9'	N/A
3.	36	28	F	600 g	0' → 0'	IUGR and IUGF

SGA (small for gestational age).

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