CASE REPORT

Successful Pregnancy and Live Birth after Uterine Artery Embolization for Uterine AV Malformation—A Rare Entity: A Case Report

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Received on: 30 January 2022; *Accepted on*: 18 April 2023; *Published on*: 16 September 2023

Abstract

Objective: This paper reports on a rare case of pregnancy after uterine artery embolization (UAE) for uterine arteriovenous (AV) malformation (AVM) following septic abortion. Traditionally, Pelvic AVMs were surgically treated with hysterectomy, abolishing the opportunity for future pregnancies endovascular treatments like UAE are the only fertility-preserving procedure for such life-threatening conditions in young women. There are many concerns about fertility issues after UAE. Embolization after symptomatic AVM not only resolves the symptoms but also gives the possibility to carry a pregnancy. Persistent amenorrhea and reduced ovarian reserve can occur due to this technique. However, the pathogenesis of such adverse effects that occur after the UAE is still not well explored.

Case description: A 31-year-old young woman was admitted to our department after being unwell and with heavy bleeding following an elective termination of pregnancy by suction and evacuation. Transvaginal ultrasound and Doppler studies detected uterine AVM at the posterior wall and fundus.

Conclusion: The peculiarity of this case was that spontaneous pregnancy occurred within a few years of follow-up after UAE and she gave birth to a live healthy baby at term with normal vaginal delivery with no complications.

Keywords: Angiography, Case report, Fertility, Uterine artery embolization, Uterine arteriovenous malformation.

Journal of South Asian Federation of Obstetrics and Gynaecology (2023): 10.5005/jp-journals-10006-2239

Introduction

Uterine arteriovenous malformations (AVMs) are rare, but they are potentially life-threatening conditions.[1] They are abnormally connected vessels, often arteries and veins interconnected by fistulae. Acquired AVM occurs most commonly following dilatation and curettage (D&C), cesarean section, myomectomy, and spontaneous abortions. Rarely, AVM can also arise from trophoblastic disease, malignancy, and infection of the uterine cavity. Acquired uterine AVMs (UAVMs) were further classified into high-flow or true UAVMs and low-flow or non-UAVMs based on the angiographic diagnosis.[1] Although there are diverse symptoms for this infrequent condition, heavy menstrual bleeding is the most frequent presenting affliction. Of these, 30% of women require massive blood transfusions.[2] Transvaginal ultrasonography with color and spectral Doppler is the preferred primary investigation in patients with suspected UAVM. Magnetic resonance imaging (MRI) is also used to assess the extent of AVM, arterial feeders, and their relationship with the myometrium. However, digital subtraction angiography is the gold standard technique to confirm the diagnosis. Although hysterectomy was considered the treatment of choice earlier, uterine artery embolization (UAE) is considered the only minimally invasive and conservative approach in young women wanting to preserve fertility. We report this case of true acquired UAVM following septic abortion in a young woman, managed conservatively with UAE with a successful spontaneous conception during the follow-up. The purpose of presenting this case is not only to highlight the clinical features of this rare condition but also to underline the safety and benefits of UAE in young women wanting to preserve fertility.

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How to cite this article: Dhandapani KS, Selvaraj P. Successful Pregnancy and Live Birth after Uterine Artery Embolization for Uterine AV Malformation—A Rare Entity: A Case Report. J South Asian Feder Obst Gynae 2023;15(4):465–467.

Source of support: Nil

Conflict of interest: None

Patient consent statement: The author(s) have obtained written informed consent from the patient for publication of the case report details and related images.

Case Description

A 31-year-old P1L1A1 woman presented with complaints of heavy bleeding with high-grade fever with chills and rigors and several episodes of diarrhea and vomiting since 2 days after an elective termination of pregnancy for 8 weeks intrauterine gestation by suction and evacuation 2 days back. On admission, she was unwell, pale, and dehydrated with hypotension and impending circulatory failure. Per abdominal examination revealed vague tenderness in the right iliac region. Bimanual examination revealed that the uterus was anteverted and slightly enlarged with forniceal tenderness. Blood investigations, such as full blood count, blood grouping, Rhesus typing

and crossmatching, and kidney and liver function tests with serum electrolytes were done, and blood clotting and bleeding time were estimated. High vaginal and cervical swabs were collected.

The woman was transferred to high-dependency unit and resuscitation was done. Higher antibiotics, intravenous fluids, and antifibrinolytics were started immediately along with two units of packed red cell transfusion. The provisional diagnosis was established as septic abortion. The patient's condition improved over the next 24 hours. Beta HCG showed a decreasing trend. Transvaginal ultrasound revealed the uterus size of 9.14 × 5.11 × 4.12 cm with posterior wall multiple serpiginous cystic spaces present from the fundus extending to the cervix (Fig. 1). Bilateral ovaries were cystic but normal size. Color Doppler showed a tangle of vessels with multidirectional high-velocity flow and color mosaic pattern suggestive of UAVM (Fig. 2). Patient was counseled about the condition, disease progression, and conservative management options. Transcatheter embolization of the arteriovenous fistula and UAVM were bilaterally done through right femoral artery access. A 5-French Roberts uterine artery catheter was used for injecting Histoacryl (Braun) mixed with Lipiodol at a concentration of 50% to the left and 25% to the right. This procedure resulted in complete devascularization of the plexus. The final angiogram post-procedure shows *total occlusion of the lesion with no residual flow.* Repeated ultrasound with Doppler studies revealed the uterus size of 7.7 × 3.85 × 3.1 cm with a heterogeneous texture of myometrium with a few cystic spaces. The patient was kept on a 3-month follow-up. She had complaints of irregular and scanty menses post-procedure for 1 year. Anemia was corrected with parenteral iron therapy. After 2 years, she had regular ovulatory cycles. Four years after the procedure, the patient wished to complete her family. Folliculometry with timed sexual intercourse proved fruitful after 4 months. She delivered a full-term healthy female baby vaginally of 3500 grams birth weight. A blood loss of 350 mL was observed. Placenta showed no histopathological alterations. Her immediate and late postpartum period was uneventful.

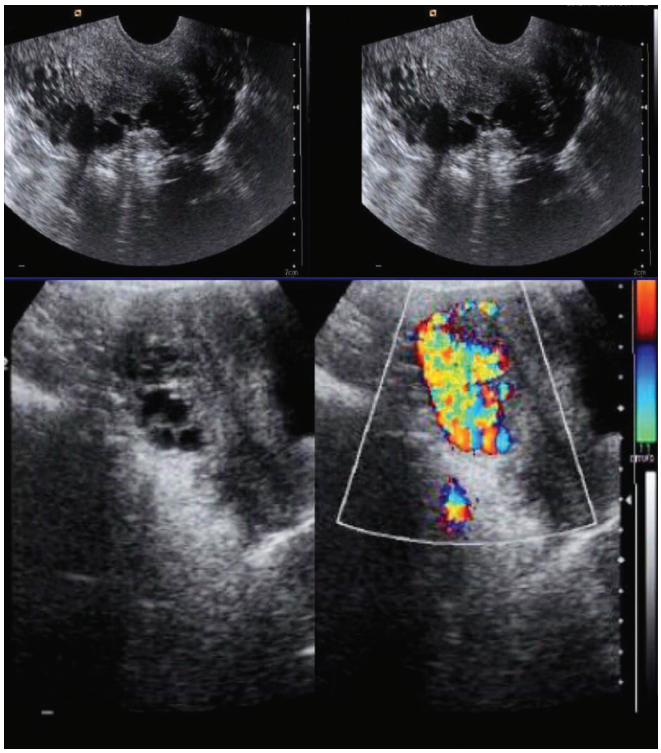

Fig. 1: Figures on the top show transvaginal ultrasound, which reveal heterogeneous myometrium and anechoic cystic spaces in myometrium. The figures below show color Doppler imaging (transabdominal, a tangle of vessels with the multidirectional high-velocity flow and a color mosaic pattern

Discussion

The most common cause of abnormal uterine bleeding in women of reproductive age-group is mostly pregnancy related. Although UAVM is rare, it must be considered in patients with untoward bleeding after abortion after ruling out retained products and gestational trophoblastic disease. The course of this rare condition and management was less explored until the early 1980s. The UAE for UAVM was first described by Forssmann et al. back in 1982.[2] Burbank et al. proposed that the stasis of blood flow caused by UAE induces clot formation in the uterus.[3] Fibrinolysis occurs in the normal myometrial vasculature followed by reperfusion. However, UAVMs, which are neovascular masses, lack proper

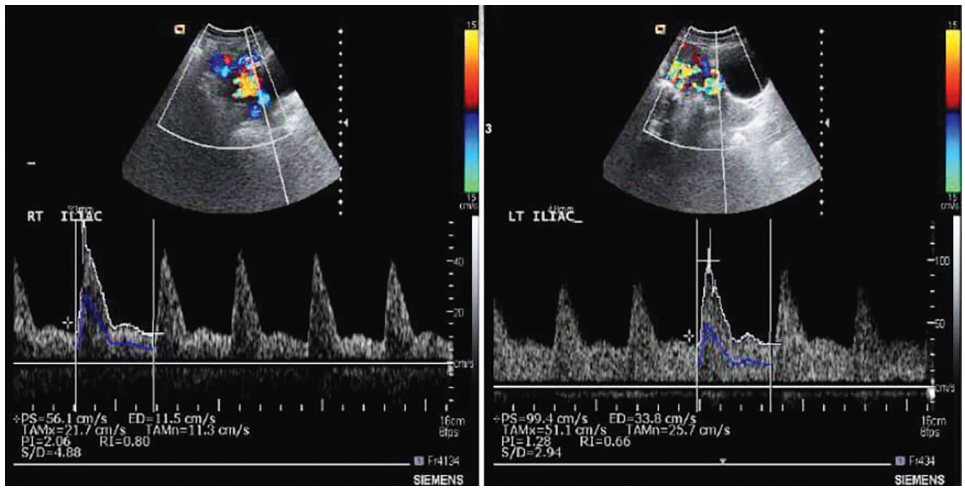

Fig. 2: Color Doppler studies show peak systolic value (PS) in right and left iliac arteries. PS more than 40 suggestive of true AVM. In our case, the PS of the right iliac artery is 56.1 cm/second and the left iliac artery is 99.4 cm/second

fibrinolytic pathways leading to ongoing clots, stasis, and eventual regression.[3] Acquired or traumatic AVM represents the multiple small arteriovenous fistulae between intramural arteries and myometrial venous plexus, that is, a single artery joining a single vein. Acquired AVM in our case is likely to result from an aberration at the molecular level, which is due to trauma from the curettage resulting in improper regression of the placental bed, thus causing the venous sinuses to be incorporated in the myometrium.[3] This activates the myometrial vascular endothelial growth factor, pro-inflammatory, and angiogenesis factors thereby creating a local immune response.

In our case, UAE was the default treatment and she desired fertility, as there were no other standard conservative medical treatment protocols for UAVM. Many studies claim that UAE is safe and effective for women desiring fertility but certain case series describes placental abnormality, such as placenta previa, morbidly adherent placenta, and abruption placenta.[2] But no such complications were present in our case. The fertility concern after UAE for UAVM is a less explored topic in modern gynecology and no robust evidence comparing the fertility, pregnancy, and perinatal issues is present. However, selective embolization avoids ovarian vascularity preserving fertility. The most recent guidance from the American College of Obstetricians and Gynecologists (2004) recognizes the UAE as safe and effective.[4]

Local complications are rare. Spasms of the uterine artery during the procedure can be avoided by meticulous techniques and by using microcatheters. Persistent spasms can result in incomplete embolization. Post-embolization syndrome is an early complication. It includes pain, nausea, fever, malaise, and raised white cell count. Usually, it is self-limiting and managed with analgesic and anti-inflammatory medication. Other early complications are deep venous thrombosis and urinary tract infections are very rare. Late complications (beyond 30 days) include vaginal discharge, endometritis, and amenorrhea. The incidence of amenorrhea is age-related being much less common (<1%) in women under 40 years of age. However, treatable abnormal uterine bleeding was noticed in our case in the first 2 years, later on, followed by regular ovulatory cycles. This makes the UAE the most preferable treatment for women seeking fertility. Repeated UAE for AVM for residual lesions and its impact on fertility has not been fully understood.

Conclusion

Uterine ateriovenous malformations are rare but are dangerous clinical conditions with complex investigations and management options. It must be kept in mind that for patient with heavy bleeding following an abortion, the possibility of AVM must be considered as a differential diagnosis. Due to its proven efficiency in treating UAVM, the technique of UAE must be the first option for women seeking fertility in modern gynecology.

References

1. Ho CY, Seow KM, Huang LW, et al. Fertility outcomes following pelvic embolization in women with acquired uterine arteriovenous malformation. Taiwan J Obstet Gynecol 56(6) 831–835. DOI: 10.1016/j.tjog.2017.10.023.
2. Vladimir J, Miroslav M, Magdalena M, et al. Term pregnancy after unilateral uterine artery embolization for aggressive uterine arteriovenous malformation. 2020;8(2):AJBSR.MS.ID.001255. DOI: 10.34297/AJBSR.2020.08.001255.
3. Vilos AG, Vilos GA, de Vrijer B, et al. Fertility and pregnancy outcomes following uterine artery embolization (UAE) for uterine arteriovenous malformation (AVM). Gynecol Surg2010;7(4):401–405. DOI: 10.1007/s10397-009-0503-5.
4. Committee on Gynecologic Practice, American College of Obstetricians and Gynecologists. ACOG Committee opinion: Uterine artery embolization. Obstet Gynecol 2004;103(2):403–404. DOI: 10.1097/00006250-200402000-00044.