

Case report of recurrent uterine arteriovenous malformation managed conservatively

Hoo P.S., Norhaslinda A.R., Shah Reza J.N.

ABSTRACT

Introduction: Uterine arteriovenous malformation (AVM) is one of the rare conditions in gynecology. Conservative management such as uterine artery embolization can be tried to preserve the uterus. We have successfully treated a patient with recurrent uterine arteriovenous malformation with three times of uterine artery embolization. **Case Report:** A 35-year-old female presented with heavy vaginal bleeding. Uterine arteriovenous malformation was suspected by Doppler ultrasonography and it was confirmed with angiography. The first uterine artery embolization procedure was performed using polyvinyl alcohol but it recurred after one year and another recurrence after three years. Finally she was successfully treated with third uterine artery embolization. Otherwise, patient remained asymptomatic and keen for pregnancy. **Conclusion:** Uterine Artery embolization is a safe and effective treatment for recurrent uterine arteriovenous malformation.

Keywords: Angiography, Color Doppler ultrasound, Post-partum hemorrhage, Uterine arteriovenous malformation, Uterine artery embolization

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INTRODUCTION

Uterine arteriovenous malformation (AVM) is one of the rare conditions in gynecology, with fewer than 100 cases reported in literature [1]. It can be life-threatening especially when patient present with profuse genital tract bleeding. Color Doppler ultrasound (US) is commonly used as a non-invasive test for provisional diagnosis and confirmation can be done with diagnostic angiography. Uterine artery embolization can be used as conservative treatment to avoid hysterectomy.

We report a case of recurrent uterine arteriovenous malformation (AVM) which successfully treated with multiple times of embolization procedure.

CASE REPORT

A 35-year-old female presented to us with secondary post-partum hemorrhage after 12 days delivery of

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macerated stillbirth (MSB) at term via induction of labor. The delivery was uneventful and she was discharged two days later. She has previous history of cesarean section four years ago due to placenta previa, which complicated with massive post-partum hemorrhage, required blood transfusions.

Upon examination, she was hemodynamically stable and afebrile. However, she looked pale with a hemoglobin level of 6.7 g/dL. Uterus was palpable at 20 week size with normal cervix and parous os. She was transfused with 4 pints packed cells. Transabdominal scan showed the endometrial thickness of 11.2 cm, irregular margin and mixed echogenicity. The provisional diagnosis was endometritis and broad spectrum antibiotics were commenced but the symptoms did not improve. Repeat ultrasound with Doppler showed solitary intrauterine mass 15.7 mm x 33.4 mm with areas of high vascularity within myometrium, differential diagnosis of retained product of conception, placenta accreta or placenta site trophoblastic tumor (PSTT) were made but serial serum beta human chorionic gonadotropin (hCG) level showed decreasing in trend from 125.7–20.1 mIU/mL.

Surgical evacuation of uterine cavity removed blood clots only with no product of conception. MRI scan of pelvis showed a bulky uterus, widened with heterogeneous, highly vascular lesion with extend into the myometrium with clear demarcation with surrounding area. Features are suggestive of hypervascular lesion of the anterior uterus. Impression of uterine arteriovenous malformation or PSTT was made. She was referred to intervention radiologist for pelvic angiogram and embolization of the lesion for the intractable genital tract bleeding.

The uterine artery angiogram revealed arteriovenous malformation in both hemipelvis. The first embolization procedure with polyvinyl alcohol (PVA) 355–500 microns was performed to block the bilateral uterine arteries. The genital tract bleeding was reduced remarkably.

One year later, this patient presented to us with new episode of profuse genital tract bleeding, pelvic MRI scan was done to confirm the AVM recurrence, and it did show evidence of residual AVM. Angiography demonstrated new fine feeders from the right uterine artery, which was successfully embolized using polyvinyl alcohol.

This lady remained asymptomatic until three years later with latest pelvic ultrasound showed bilateral abnormal and prominent vessel on right and left myometrium with prominent draining vein more on the right side (Figure 1). She was explained that her uterine AVM was likely acquired from her previous cesarean or a regrowth of the AVMs as result of persistence of unknown inherent factors. They may regress spontaneously or rupture, which may cause genital tract bleeding and complicating future pregnancy. Treatment options were discussed and she chose to undergo third uterine artery embolization. The uterine artery angiogram confirmed the presence of an arteriovenous malformation in the right lateral wall region, originating from right uterine

artery (Figure 2A), which was successfully treated by embolization (Figure 2B). The patient was discharged well after the successful procedure. She is still under our gynecological outpatient follow-up.

DISCUSSION

Arteriovenous malformations can occur in any organ in the body, including the pelvic vasculature and rarely in the uterus. Fewer than 100 cases of uterine AVM reported in literature [1]. The first case of AVM was reported in 1926. Uterine AVMs result in sudden and massive vaginal bleeding that may be life-threatening. It can occur during postpartum period, post abortion or menstrual abnormality especially heavy menstrual bleeding.

Uterine AVMs can be divided into congenital or acquired.

Congenital AVMs have multiple vascular connections, may manifest at other sites, and involve surrounding structures. Congenital uterine AVMs arise from an abnormality in the embryologic development of primitive vascular structures, resulting in multiple abnormal communications between arteries and veins. Whereas

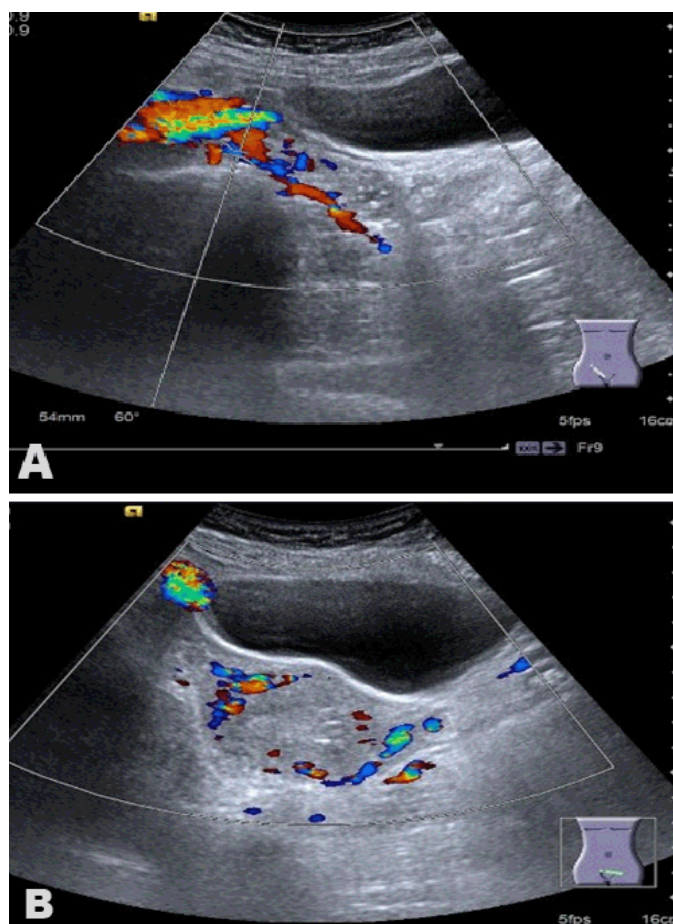


Figure 1: (A) Bilateral abnormal and prominent vessel on right and left myometrium, and (B) Prominent draining vein more on the right side.

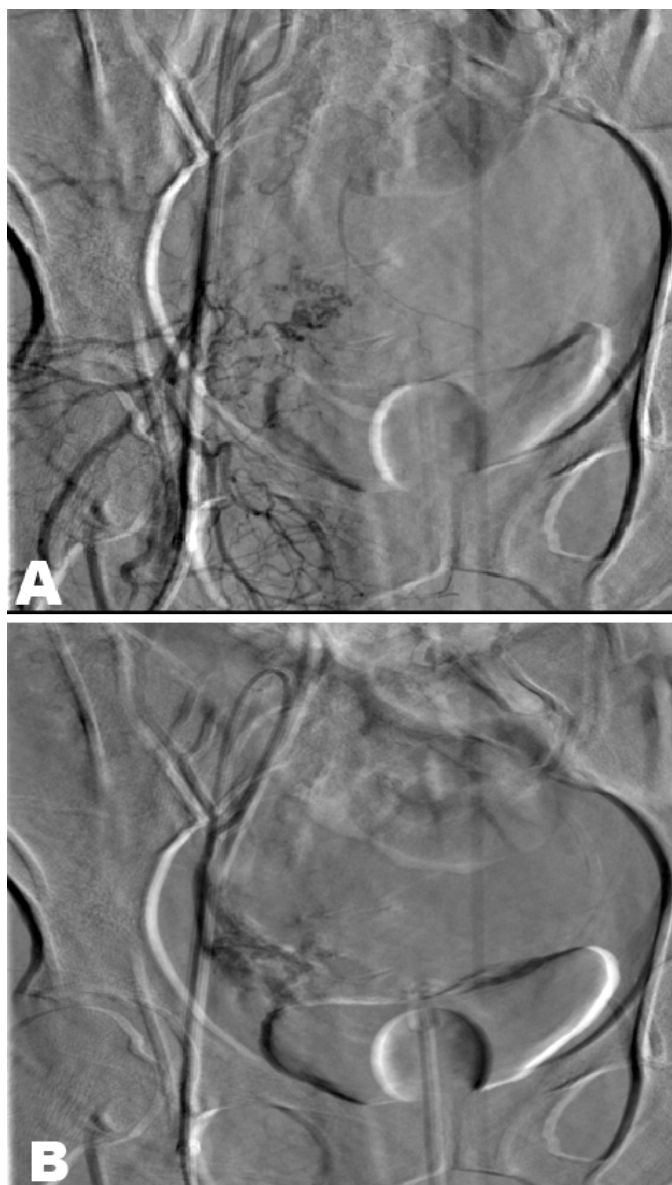


Figure 2: (A) Presence of an arteriovenous malformation in the right lateral wall region, originating from right uterine artery and (B) Post-embolization showed complete embolization of the AVM.

acquired AVMs usually have a single connection between an artery and a vein [2]. Acquired uterine AVMs are usually traumatic, resulting from uterine curettage, cesarean section, direct uterine trauma and less commonly from gestational trophoblastic disease, endometrial carcinoma and cervical carcinoma. Small arteriovenous fistulas between intramural arterial branches and the myometrial venous plexus are the one responsible to form the AVM and appear as a vascular tangle. In cases of trauma cause of AVM, healing process and pregnancy will aggravate in forming abnormal communication between arteries and veins. AVMs are hormone-sensitive. This makes the congenital AVM appears during puberty or after a pregnancy [3].

In this case, it is possible that this is an acquired form of uterine AVM due to previous cesarean section.

Historically, the diagnosis was made after laparotomy or histopathologically after hysterectomy. Subsequently, angiography became the gold standard tool for diagnosis. Currently, transvaginal color Doppler ultrasound is the most commonly used tool for diagnosis, and angiography is reserved for patients submitted to surgical treatment or therapeutic embolization [4]. As in this case, AVM was diagnosed with color Doppler ultrasound and angiography.

Gray scale ultrasound is the first step investigation, able to detect the presence of multiple tubular or “spongy” anechoic or hypoechoic areas within the myometrium of a normal endometrium [5–7]. Differential diagnosis with similar sonographic findings includes gestational trophoblastic disease, retained conception products, hemangioma, multilocular ovarian cysts, hydrosalpinx or abnormal placentation.

Doppler ultrasound able to demonstrate arteriovenous shunt with low-resistance and high-velocity flow. Spectral analysis may predict the degree of the vascular lesion arterializations and aid in the definition of the treatment [8]. Although Doppler ultrasound able to show the presence of AVM, but the ability to assess the lesion extent in the pelvis may be limited.

Magnetic resonance imaging scan is an advanced, modern non-invasive tool to assess the disease extent and aid to confirm the diagnosis [7]. The characteristic features of diagnosis include a bulky uterus with a focal mass, disruption of the junctional zones, multiple serpiginous flow-related signal voids within the lesion, and prominent parametrial vessels [6, 7]. Pelvis MRI scan of this patient showed similar findings as given in literatures.

Digital subtraction angiography (DSA) is an invasive investigation tool to diagnose AVM. It remains the gold standard in diagnosing AVM. Characteristic findings of diagnosis include bilateral hypertrophy of uterine arteries that feed a tortuous, hypertrophic arterial mass with large accessory feeding vessels, and early drainage into enlarged hypertrophic veins [4]. Due to its invasive nature, DSA is usually reserved when a patient requires surgical intervention or embolization [6].

The management of uterine AVM depends on multiple factors such as hemodynamic stability, severity of bleeding, patient’s age, her desire to preserve fertility, availability of treatment options and trained man-power. Acute management includes initial stabilization, uterine tamponade with Foley’s catheter or rolled gauze packing to stop the massive bleeding and medical therapies like estrogens, progestins, methylergonovine, danazol, and 15-methyl-prostaglandin F₂alpha [2, 9–11].

Expectant management may have a role in stable patient. Timmerman et al. [12] showed that of 265 patients with abnormal premenopausal bleeding, nine had uterine AVMs diagnosed on ultrasonography. Of these, six had spontaneous resolution, two patients with hydatidiform mole needed chemotherapy and the AVMs resolved after chemotherapy, and only one required embolization. If the

bleeding is not severe, long-term medical management may be used and includes estrogens and progestins, methylergonovine, danazol, 15-methyl-prostaglandin F₂alpha, oral contraceptives, and intramuscular followed by oral methylergonovine maleate [2, 10, 11].

Traditionally, hysterectomy was performed for symptomatic AVMs, especially if fertility was not a concern. Since uterine AVM is commonly diagnosed in women of childbearing age, angiographic embolization has made hysterectomy no longer necessary. However, hysterectomy remains the treatment of choice in post-menopausal patients or as an emergency treatment in life-threatening situations [13].

Uterine artery embolization has changed the modern management of uterine AVMs. Rosch et al. reported first use of arterial embolization in 1972 for acute gastrointestinal bleeding [14]. Later on, it was further used in trauma-related pelvic bleeding. Uterine artery embolization has been commonly used in the emergency setting as well as less urgent circumstances, since the first description of a successful embolization treatment for uterine AVM in 1986. Many types of embolic materials have been used, for example, polyvinyl alcohol, histoacryl (glue), stainless steel coils, detachable balloons, haemostatic gelatin, etc. [6].

From literatures, sixteen cases were reported after 10 years of using this procedure. Eleven were managed by a single procedure, two of them required repeat embolization, and three eventually required hysterectomy [2]. Yang et al. [15] reported the long-term success rate of embolization for uterine AVMs as 79% (11 of 14 patients) and Kwon et al. [16] reported as 90% (22 of 24 patients). The overall efficacy was reported as 93% by Ghai et al. [17] in a retrospective review of 15 patients over 10 years (1992 to 2002). A total of 25 embolization procedures were performed in these 15 patients, six required repeat embolization (one patient underwent embolization six times and was pregnant when this series was reported, and five underwent embolization twice for recurrence of bleeding). Only one patient underwent a hysterectomy [18–21].

Transcatheter arterial embolization has many advantages especially the outstanding success rates, low complication rates, avoidance of surgical risks, and preservation of fertility [22]. Some authors reported cases of post-embolization pregnancy. The common side effects of the procedure, such as low-grade temperature, pain, infection, or symptoms, have been documented and can be tolerable. However, pelvic pain is the main side effect, some patients requiring strong analgesia such as opiate and non-steroidal analgesia. Disadvantages of this procedure especially the insufficient embolization which need a repeat procedure and this can be costly and time consuming. Lim et al. reported that one of their patients experienced self-resolving buttock and lower-limb claudication, could be result of extensive and multiple embolization in the pelvic vessels [23]. So far, our patient has no these post embolization side effects.

From the literatures, the use of liquid embolization materials or very small particles are more commonly causing lower limb neurological defect [24]. Some authors did reported case series which causing serious complications, such as perineal skin sloughing, uterovaginal and recto-vesico-vaginal fistulae, and bladder necrosis. They found that most of the cases involving embolization of the internal iliac arteries with cyanoacrylate [24].

Experienced interventional radiology experts may avoid the rare complications of the transcatheter arterial embolization. There are multiple factors contributing the failure of embolization, which can be the type of embolic material used, expertise of the intervention radiologist, or a regrowth of the AVMs the result of persistence of unknown inherent factors. More case series or bigger sample size study are needed to identify the factors causing the failure. Our patient did have multiple recurrence due to unknown reasons, possibly similar to the above literatures.

Nowadays, more young women or those who desire future fertility will prefer angiographic uterine artery embolization for uterine AVMs especially with the benefit of non- interfering their menstrual cycle or pregnancy. Ghosh had reported seven cases of UAE for AVMs, resulting six successful procedures, two of them experienced recurrent bleeding and had successful re-embolization, and three of them became pregnant. However, two of them chose to continue their pregnancies and both had normal deliveries [10]. O'Brien et al. [8] showed that normal menstrual cycle returned within two months in all eleven women of UAE, and surprisingly five of them became pregnant.

There are some reported uncommon surgical managements such as the coagulation of AVM under hysteroscopic guidance, surgical removal of AVM, laparoscopic bipolar coagulation of uterine vessels, and ligation of the uterine artery [2, 20, 21]. Modern approach of AVMs treatment indicates that hysterectomy is only for women who do not need fertility preservation, poor resource area, or failed embolization therapy.

Long-term follow-up after an apparently successful embolization may reveal more failures as we know that the AVMs might regrow. Successful results have been reported after a shorter period of follow-up (six weeks), long-term follow-up is needed will reveal the true success rate of these procedures .

This case report highlights the use of US and DSA for diagnosing uterine AVM in a patient of childbearing age who is initially presented with secondary post-partum hemorrhage. The woman remains no symptoms, however, during follow-up suggestive of residual AVM. As she desires further pregnancy, subsequent uterine artery embolizations were done without complications. It also highlights that uterine artery embolization of uterine AVMs is a safe and effective treatment as alternative to surgical therapy.

CONCLUSION

Minimally invasive management is an option in recurrent uterine arteriovenous malformation.

Author Contributions

Hoo P.S. – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Norhaslinda A.R. – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published.

Shah Reza J.N. – Substantial contributions to conception and design, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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