Letter to the Editor

# Congenital uterine arteriovenous malformation treated by hysterectomy: a description of two cases

**Beichen Zhang[1,2]^, Xianqing Wu[1,2], Tianyu Zhu[1,2]^, Xiaoshan Chai[1,2]^**

[1]Department of Obstetrics and Gynecology, The Second Xiangya Hospital of Central South University, Changsha, China; [2]Clinical Research Center for Gynecological Disease in Hunan Province, Changsha, China

*Correspondence to:* Xiaoshan Chai, MD. Department of Obstetrics and Gynecology, The Second Xiangya Hospital of Central South University, 139 Renmin Middle Road, Furong District, Changsha 410011, China; Clinical Research Center for Gynecological Disease in Hunan Province, Changsha, China. Email: chaixiaoshan@csu.edu.cn.

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## Introduction

Uterine arteriovenous malformation (UAVM) is a disease caused by an abnormal connection between uterine arteries and veins, which can lead to life-threatening vaginal bleeding (1). UAVM can be divided into congenital and acquired types. We report the case of a 71-year-old postmenopausal woman who suddenly experienced severe vaginal bleeding, with no history of surgery or intrauterine procedures. The diagnosis of UAVM was confirmed through three-dimensional (3D) computed tomography angiography (CTA). As the patient had been postmenopausal for over 20 years and did not require fertility preservation, she underwent a safe and successful hysterectomy. Another patient, a 34-year-old woman, also presented with severe vaginal bleeding. Prior to her visit, she had experienced two episodes of heavy vaginal bleeding leading to moderate hypochromic anemia and had undergone two uterine arterial embolization (UAE) treatments. Angiography confirmed her condition as congenital UAVM. Considering her age, the patient initially chose arterial embolization therapy, but after the embolization, she again experienced severe vaginal bleeding and ultimately opted for a hysterectomy.

## Case presentation

### *Case 1*

A 71-year-old female, gravida 4, para 4, was admitted to the Second Xiangya Hospital of Central South University with a 5-day history of vaginal bleeding and lower abdominal pain. The patient had experienced intermittent vaginal bleeding without any obvious trigger 5 days earlier. The bleeding varied in quantity, sometimes being heavier than her normal menstrual periods, and included large blood clots. She also reported tolerable, position-independent, dull lower abdominal pain. Before menopause 20 years ago, she had completed four normal deliveries, with no history of miscarriage or other intrauterine procedures. Her menstrual flow had been heavy in the past. She denied any other medical history. Upon admission, her vital signs were stable, with an appearance of anemia. Bimanual examination revealed an enlarged uterus, approximately the size of a 2-month pregnancy. Laboratory tests showed hemoglobin at 77 g/L; liver and kidney functions, coagulation profile, and erythrocyte sedimentation rate were essentially normal. Pelvic ultrasound revealed an enlarged uterus with multiple tubular echoic areas in the myometrium and

^ ORCID: Beichen Zhang, 0009-0004-4849-0421; Tianyu Zhu, 0009-0001-4538-8315; Xiaoshan Chai, 0000-0003-3264-0726.

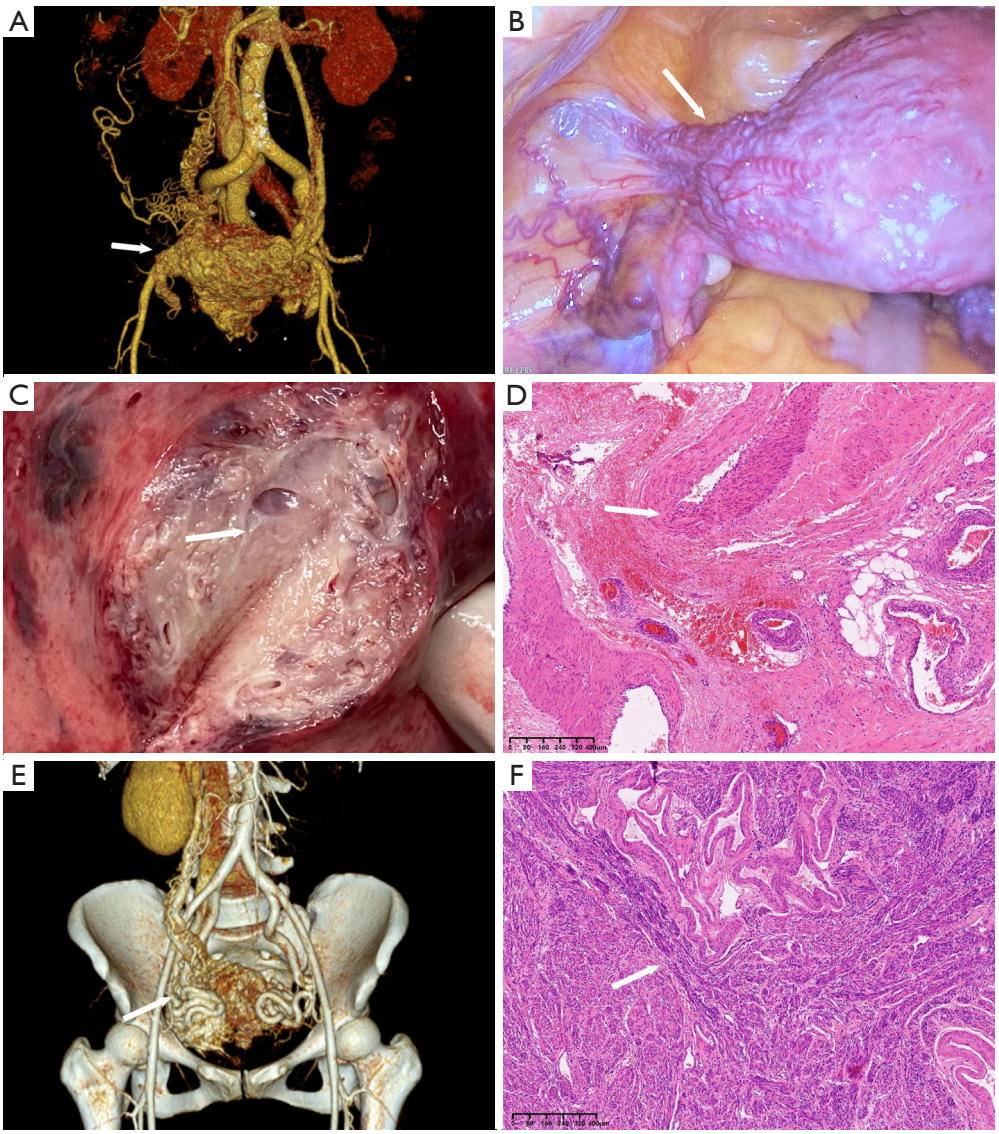


**Figure 1** Combined overview of imaging, surgical, and pathological findings for Case 1 (A-D) and Case 2 (E,F). (A) Contrast-enhanced CT scan: multiple serpiginous and dilated branches of the internal iliac artery are distributed in the uterus and parametrium (arrow). The right ovarian vein is tortuous and dilated. (B) Intraoperative findings: numerous, dilated vessels around the uterine corpus (arrow), cervix, external iliac, and ovarian arteries. (C) Sectional view: multiple venous sinus in the myometrium (arrow). (D) Pathological section by HE staining (×200): multiple vessels and sinuses in the myometrium and parametrium with thicker endothelium (arrow). (E) CTA showed twisted vascular masses near the uterine fundus (arrow), mainly from uterine arteries, with dilated right ovarian veins refluxing into the inferior vena cava. (F) Pathological features by HE staining (×200). Arteriovenous dilation and tortuosity in the uterine myometrium, with some vessels showing mucoid degeneration (arrow). CT, computed tomography; HE, hematoxylin and eosin; CTA, computed tomography angiography.

adnexal region. Magnetic resonance imaging (MRI) showed numerous abnormal vessels in the uterus and adnexal areas and abnormal signals in the myometrium. CTA revealed multiple tortuous and dilated branches of the internal iliac artery surrounding the uterine body, with dilated draining veins entering the internal iliac vein (*Figure 1A*). After confirming the diagnosis of UAVM, we consulted with interventional radiology staff to assess the feasibility of UAE. Given the extensive range of the UAVM, the difficulty and high recurrence risk of embolization, and the fact that

the patient had been postmenopausal for over 20 years, hysterectomy was deemed a better option. Ultimately, with the patient's consent and after thorough preoperative preparation, we performed a hysterectomy. Intraoperatively, the uterus was found to be anteriorly positioned, and about the size of a 2-month pregnancy. Tortuous and dilated vessels were present on both sides of the infundibulopelvic ligaments, parauterine areas, and lateral walls of the uterus (*Figure 1B*). On incising the uterus, the myometrium showed numerous venous sinuses (*Figure 1C*). The postoperative pathological examination revealed numerous abnormal, thick-walled vessels within the uterine smooth muscle, without endothelial cell proliferation (*Figure 1D*). Based on a comprehensive analysis of the imaging findings, intraoperative observations, and pathological results, it was concluded that the patient's condition aligned with the defining features of a congenital arteriovenous malformation. The patient was discharged uneventfully without any vaginal bleeding after 6 months of follow-up.

### *Case 2*

A 34-year-old woman, gravida 4, para 2, with a history of two cesarean sections and two induced abortions, presented to the outpatient clinic with vaginal bleeding. During the examination, the patient's vital signs were stable. The patient exhibited a pale complexion and signs indicative of anemia. Bimanual examination further revealed an enlarged uterus. Laboratory tests indicated a hemoglobin level of 87 g/L; liver and kidney functions, electrolytes, lipids, and coagulation profile were found to be within normal ranges. The patient's β-human chorionic gonadotropin (β-HCG) level was less than 3.1 μg/L, ruling out pregnancy-related bleeding. Further pelvic CTA revealed multiple tortuous vascular masses at the uterine fundus, predominantly from the uterine arteries, with tortuous and dilated right ovarian veins refluxing to the inferior vena cava, confirming the diagnosis of UAVM (*Figure 1E*). Before this visit, she had experienced two episodes of heavy vaginal bleeding causing moderate hypochromic anemia and had undergone two arterial embolization treatments. The most recent embolization was four months prior to this visit, with severe vaginal bleeding reoccurring one day ago. Her menstrual cycle was regular. She had a history of left nephrectomy (specific reason unknown). Considering her young age and desire to preserve her uterus, arterial embolization treatment was recommended. She underwent UAE at another hospital, but experienced severe vaginal bleeding again after two months and underwent a hysterectomy. During surgery, the pelvic peritoneum was shown to be covered with tortuous vessels; the uterus was enlarged, about the size of a 50-day pregnancy, with numerous tortuous and engorged veins on the uterine surface and around the uterus. The histopathological examination showed arteriovenous dilation and tortuosity in the uterine myometrium, with some vessels showing mucoid degeneration (*Figure 1F*). The patient recovered well after surgery, and was discharged smoothly. Six months of postoperative follow-up recorded no vaginal bleeding.

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was provided by the two patients for publication of this article and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

## Discussion

UAVMs are rare and usually diagnosed in patients with menorrhagia or abnormal uterine bleeding, hence the true incidence is unknown (2). Their characteristic feature is the abnormal growth and connection of arteries and veins without a capillary bed, leading to high and low flow areas that are fragile and prone to bleeding. UAVM is a potentially fatal condition that lacks specific clinical symptoms, with the most common symptom being vaginal bleeding. Patients with UAVM may also experience menorrhagia or intermenstrual bleeding (3).

UAVMs can be classified into congenital and acquired types. Congenital UAVMs are embryological developmental defects of the primitive capillary plexus, leading to multiple arteriovenous connections, which are extremely rare (4). Acquired UAVMs are more common, primarily resulting from gynecological surgeries and other traumatic factors, such as curettage, direct trauma, and so on. Normal vaginal delivery may also trigger the formation of UAVMs. Retained products of conception, gestational trophoblastic disease, cancer, infections, and exposure to diethylstilbestrol are also associated with acquired UAVMs. The cause is the disruption and defective healing of the uterine vascular system (5).

There is a fundamental difference between congenital and acquired arteriovenous malformations, and correctly differentiating between them is crucial for choosing

 

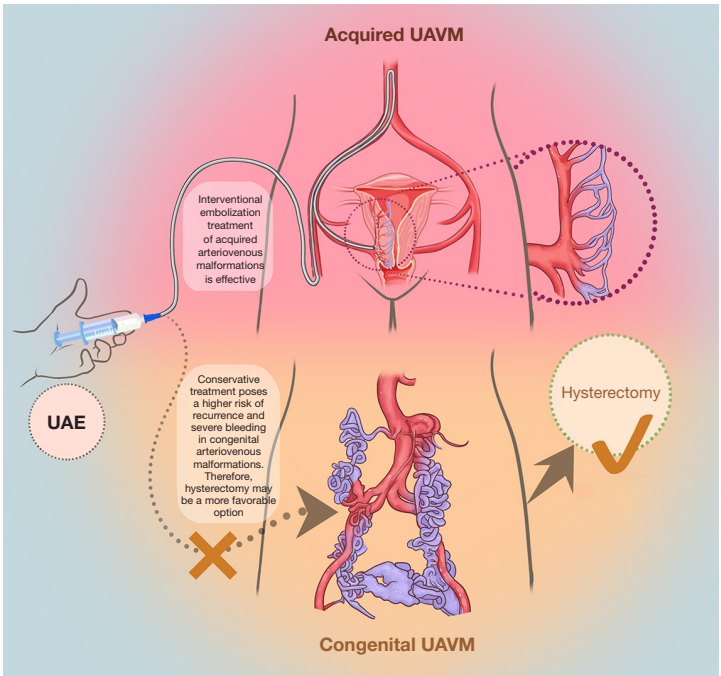


**Figure 2** The differences between congenital and acquired UAVM and the selection of different treatment modalities. UAVM, uterine arteriovenous malformation; UAE, uterine artery embolization.

treatment strategies (*Figure 2*). There are no clear standards for this differentiation in previous literature, and many case reports have not clarified whether they are congenital. Some cases are inappropriately determined based solely on the history of gynecological surgery or trauma, because these factors not only cause acquired UAVMs, but also may promote the original congenital UAVMs. Radiographically, congenital UAVMs typically present with multiple feeding arteries, a central lesion, and multiple draining veins, which are often enlarged above the uterus and invade the surrounding pelvis (6,7). Their extent is far greater than that of the acquired type, often involving the iliac artery, with a spiraled, basket-like pattern interwoven with the uterus. Historically, congenital UAVMs often accompany vascular malformations in other parts of the body, but sometimes are limited to the uterus, complicating their differentiation from acquired lesions (6). Pathologically, congenital UAVMs usually do not have endothelial cell proliferation or endothelial cellular stroma (8). In contrast, acquired UAVMs are usually caused by reactive angiogenesis, characterized by direct fistulas between intramural arterial branches and uterine venous plexuses (9). Histologically, congenital arteriovenous malformations often appear as sprawling, with multiple dilated vessels in a spiral pattern, whereas acquired arteriovenous malformations may present a cavernous sinusoid type, including a single arterial vessel supplying many small connecting fistulas (10). They can also present different clinical symptoms; congenital UAVMs often lead to severe vaginal bleeding that is refractory to conventional treatments (11).

UAVM is a rare cause of abnormal bleeding. There are only a few reports in postmenopausal women, and reports of congenital UAVM in postmenopausal women are even rarer. The patient in Case 1, who had been postmenopausal for many years with no history of abnormal vaginal bleeding, experienced sudden massive vaginal bleeding without any obvious trigger, which might be associated with concomitant infection. The patient in Case 2, a woman of childbearing age, presented with recurrent massive abnormal vaginal bleeding. She underwent three failed UAE treatments and subsequently chose hysterectomy due to reoccurring massive vaginal bleeding. Her imaging suggested a wide range of vascular malformations, and she had never experienced vaginal bleeding before pregnancy, possibly because her UAVM was not superficial enough for the vessels to rupture and bleed. Agarwal *et al.* have reported similar cases (1).

UAVM may be a possible cause of postpartum hemorrhage

(PPH). Among the congenital UAVM cases we summarized, two cases had no history of uterine-related surgeries and experienced postpartum bleeding (12,13); one case (12) occurred as secondary PPH; the other (13) occurred after the expulsion of placenta following an inevitable miscarriage. In both cases, a single UAE did not achieve satisfactory results, requiring repeated embolization. Salmeri *et al.* (14) summarized 11 cases of UAVM-related PPH, among whom 81.8% underwent bilateral UAE. In one out of two cases treated by unilateral UAE, emergency total hysterectomy was performed for a sudden hemodynamic instability. However, due to lack of follow-up, it is unknown whether patients who underwent embolization experienced recurrence of UAVM-related bleeding or other symptoms. Both cases we reported had a history of childbirth but did not experience PPH, possibly because the vascular malformations at that time were not large or superficial enough. As the lesion gradually enlarges, patients may experience recurrent vaginal bleeding symptoms.

The diagnosis of UAVM has significant clinical implications. The most commonly used imaging method is ultrasound, particularly transvaginal color Doppler ultrasound. This non-invasive and cost-effective method can be widely applied, and it avoids the potential dangers of radiation and contrast agents to the patient. However, ultrasound has limitations in detecting smaller or deeper lesions. Another imaging technique, MRI, provides the most detailed anatomical information and soft tissue characteristics. Multiplanar imaging and contrast enhancement further improve diagnostic accuracy. However, MRI is limited by its high cost and extended scanning time, restricting its application. Computed tomography (CT) scans are useful in determining the extent of involvement, excluding external uterine effects, differentiating between congenital and acquired UAVMs, and identifying the supplying vessels or uterine arteries (15). CTA allows rapid 3D image reconstruction, providing high-resolution vascular anatomy and insights into anomalies. Therefore, it is the preferred choice for patients with critical bleeding. Digital subtraction angiography (DSA) is the gold standard for confirming UAVM. It accurately assesses vascular anatomy and flow dynamics, thus identifying the primary vessels to target during embolization treatment.

The treatment of UAVM is influenced by several factors, including the severity of the disease, the patient's age, comorbidities, and fertility goals (12). UAE is suitable for women with frequent or severe bleeding, as well as those who are hemodynamically unstable but wish to preserve fertility. Hysterectomy remains a common choice for postmenopausal women, those not considering fertility preservation, in emergency situations, or when embolization treatment is ineffective. During the operation, attention should be paid to the anatomical structure, and excessive exposure of vascular stumps avoided. Additionally, conservative measures and medical interventions, including observation and hormone therapy, are effective for stable patients and those with occasional or minor vaginal bleeding, helping to alleviate symptoms and reduce the size of the lesion. Finally, other surgical methods such as UAVM coagulation, laparoscopic uterine vessel bipolar coagulation, and UAVM excision can also be considered.

UAVM can result in compromised fertility. In our literature review, many patients were reported to have experienced miscarriages (1,11,13,16,17); unfortunately, these studies did not provide a summary of fertility outcomes post-treatment. Delplanque *et al.* (18) examined the fertility outcomes of 22 patients with UAVM following treatment. Their findings revealed that UAE did not significantly affect the subsequent fertility of patients, and the prognosis for the fetus was relatively positive. For UAVM patients planning pregnancy within their reproductive years, UAE for fertility preservation stands as the preferred treatment option. However, it is crucial to note that trauma, hormonal effects, or incomplete treatment of residual UAVM lesions may all contribute to accelerated UAVM growth (19). Therefore, patients desiring fertility preservation should be thoroughly informed about the potential, even life-threatening, risks of sudden bleeding. In cases of recurrent bleeding after UAE or for patients with no fertility needs, hysterectomy should be recommended.

Based on the two cases presented in this article, we believe that the type of UAVM also influences the choice of treatment method. As congenital UAVMs are very rare, there is currently a lack of literature summarizing the effects of various treatment methods for congenital UAVMs. We reviewed literature published in the last 15 years and found only 11 cases with clear pathological and angiographic diagnosis of congenital arteriovenous malformation, summarized in *Table 1*. Of these congenital UAVM patients, two underwent radical hysterectomy, and one, due to the wide extent of the vascular malformation and the difficulty of the surgery, underwent uterine artery ligation. The long-term outcomes of the remaining patients, who were treated with uterine-preserving methods such as vascular embolization, are unclear due to short follow-up periods. As acquired UAVMs are usually caused by trauma and typically

 

**Table 1** Literature review

| Number | Authors | Post date | Age (years old) | Gravidity/ parity | Surgical history | CP | Pretreatment | Treatment | Outcome | FUT |
|---|---|---|---|---|---|---|---|---|---|---|
| 1 (12) | Afaf Alsharif | 2023 | 23 | G1P1 | No | AUB | UAE | Repeated UAE | Relieved | 2 years |
| 2 (13) | Bin Lv | 2022 | 29 | G1P0 | No | Postpartum hemorrhage | Oxytocin and ergonovine | UAE | Relieved | – |
| 3 (11) | Tertu Nakashololo | 2021 | 22 | G1P0 | D & C | AUB | Oral contraceptive | UAE | Relieved | 1 month |
| 4 (20) | Shui-Qing Liu | 2019 | 47 | – | Cervical polypectomy | AUB | Curettage | UAE | Relieved | 3 months |
| 5 (16) | Xiaoqing Zhu | 2019 | 49 | G5P1 | – | AUB | UAE | Hysterectomy | Relieved | – |
| 6 (1) | Neha Agarwal | 2017 | 25 | G4P2 | No | Postcoital bleed | No | UAE | Relieved | 12 weeks |
| 7 (17) | Emi Sato | 2015 | 51 | G3P2 | Cesarean | AUB | UAE twice | Hysterectomy | Relieved | 1 year |
| 8 (3) | Hiroyuki Yazawa | 2013 | 38 | G2P0 | Myomectomy, D & C | AUB | Medical treatment | UAE | – | – |
| 9 (9) | Tae-Hee Kim | 2010 | 25 | G0P0 | No | AUB | No | UAE | Relieved | 6 months |
| 10 (21) | Daisaku Yokomine | 2009 | 54 | – | Cesarean | AUB | – | Arterial ligations | Relieved | 1 year |
| 11 (22) | Kaei Nasu | 2008 | 30 | G1P1 | No | AUB | – | UAE | Tubal pregnancy | 3 years |

CP, clinical presentation; FUT, follow-up time; AUB, abnormal uterine bleeding; UAE, uterine artery embolization.

present as a single arteriovenous fistula, whereas congenital UAVMs consist of multiple vessels, the prognosis of medical treatment and UAE is poorer for congenital than it is for acquired forms. Both cases of congenital UAVM reported in this article ultimately underwent hysterectomy. Especially in Case 2, where the patient had an extensive lesion, three embolization attempts failed, necessitating a hysterectomy. Congenital UAVMs, with their wide and diffuse arteriovenous communications, have a higher risk of recurrence and severe bleeding, and conservative treatment often fails. Even if the UAE of an UAVM is initially very successful, long-term follow-up often shows recurrence due to the presence of collateral circulation. Hysterectomy may be the most effective treatment method for congenital UAVMs.

## Conclusions

The treatment decision for UAVMs should be personalized based on the individual patient's clinical status, hemodynamic stability, and fertility preferences. Additionally, it is crucial to distinguish between congenital and acquired UAVMs, as they exhibit significant differences in anatomical structure, clinical presentation, and treatment outcomes. Congenital UAVMs continue to grow due to high blood flow between arteries and veins (19). In cases of congenital UAVMs, conservative treatment poses a higher risk of recurrence and severe bleeding. Therefore, hysterectomy may be a more favorable option. Women in their childbearing years who do not respond well to embolization treatment should be vigilant about the potential presence of congenital UAVMs. Conservative approaches may be less effective in such cases, emphasizing the importance of thorough assessment before determining the appropriate treatment.

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## Footnote

*Conflicts of Interest:* All authors have completed the ICMJE uniform disclosure form (available at https://qims.

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*Ethical Statement:* The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was provided by the two patients for publication of this article and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.



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