Successful treatment of uterine arteriovenous malformation by conservative management: Case report

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ABSTRACT

Uterine arteriovenous malformation (UAVM) is a potentially fatal disorder caused by abnormal and nonfunctional connections between veins and arteries in the myometrium. It’s consistently related to uterine trauma or prior pregnancy. A 32-year-old patient presented with severe vaginal bleeding as well as a 30-day history of abnormal vaginal bleeding. She was diagnosed with UAVM by transabdominal ultrasound color Doppler. MRI confirmed the diagnosis of UAVM. Therefore, she was given oral progesterone 5 mg three times a day for 21 days as a conservative treatment. She was followed for a year, remained asymptomatic and conceived spontaneously, and had a low-risk pregnancy, with good maternal and fetal outcomes. Since the treatment, the patient has not suffered any serious uterine hemorrhage.

Keywords: Uterine arteriovenous malformation; conservative management; transabdominal ultrasound; Mutilation; Magnetic resonance imaging

1. INTRODUCTION

Uterine arteriovenous malformation (UAVM), previously, was known by other names, including arteriovenous aneurysm, cirsoid aneurysm, and arteriovenous fistula. It’s described as nonfunctional and anomalous connections between the veins and arteries of the uterus without the presence of a capillary network in between. It’s a rare but potentially fatal condition, with only approximately 100 cases published in the scientific literature (Hickey and Fraser, 2000). There are two types of UAVM: congenital and acquired. Congenital presentation is rare and arises from a defect in the production of primitive vascular systems during embryonic development, resulting in the creation of abnormal vascular connections between veins and arteries that are likely to expand beyond the uterus’s boundaries into the pelvis (Halperin et al., 2007). Acquired UAVM predominantly occur mostly after uterine surgical procedures such as therapeutic abortion, dilation and curettage (D/C), cesarean section, trophoblastic diseases, endometrial or cervix cancer, and direct uterine trauma. Acquired UAVM is on the rise, and it affects women of reproductive age more commonly than those after menopause. Clinically, it has varied presentation, from irregular to profuse
and sometimes life-threatening vaginal bleeding in young women (Cura et al., 2009).

The diagnosis of this rare condition is easily made by transabdominal ultrasound with spectral and color Doppler, which is the preferred noninvasive diagnostic imaging modality for the initial diagnosis of UAVM. In many cases of UAVM is confirmed diagnosis by further imaging techniques such as angiography, Computed Tomography (CT), and Magnetic resonance imaging (MRI) (Calzolari et al., 2017). Treatment for UAVM is determined by the patient’s age, desire to have children in the future, and hemodynamic status, amount of hemorrhage, size, and localization of the lesion. Conservative management or embolization is a better option than a hysterectomy, particularly in women of fertile age (Hashim and Nawawi, 2013).

The aim of highlighting this case report is to emphasize the evolution of knowledge and the contribution of diagnostic imaging in making the diagnosis of this rare gynecological condition which was effectively managed with conservative management.

2. CASE PRESENTATION

32-year-old patient, para3, all of the babies were born healthy and of typical weight (3.1–3.5 kg) via vaginal delivery at full term, with one miscarriage in the first trimester treated with dilatation and evacuation three months ago. She presented to the emergency department with severe vaginal bleeding and a 30-day history of abnormal vaginal bleeding. There was no previous history of gynecologic cancer or bleeding disorders in the family. Upon admission, on physical examination, she seemed frail and pallid, but afebrile and had a normal heart rate and blood pressure. Her hemoglobin concentration (HGB) was 11.6 g/dL, and the level of the hormone chorionic gonadotropin (UCG) was less than 2mIU/mL besides the histology report revealed no signs of trophoblastic disease. The pelvic examination revealed that the uterus was normal in size and shape with a small amount of blood at the uterus’s external orifice, but there was no active bleed.

Following American Institute of Ultrasound in Medicine (AIUM) guidelines, a gynecological transabdominal pelvic ultrasound with a full bladder showed a globular-shaped bulky uterus with an anterior myometrial echo pattern which was heterogeneous with numerous an-echoic cystic & tubular areas inside the uterus and normal endometrial thickness (Figure 1). Doppler color flow reported increased vascularity with a mosaic pattern and aliasing of blood flow inside these an-echoic cystic areas. At the feeding arteries, spectral Doppler ultrasonography reveals high velocity and moderately high resistive arterial flow. Both uterine arteries flowed in a low-resistance pattern, contrary to the nongravid uterus, which had a high resistive flow pattern. Venous flow is seen by spectral tracing in the draining veins (Figure 2). A diagnosis of UAVM was suspected and an MRI study was required for further confirmation. Axial T2 & coronal T1-weighted fat-suppressed imaging of the pelvis revealed multiple convoluted vascular flow voids in the myometrium and abutting the endometrium, which corresponds with the high-velocity vascular channels. Sagittal & coronal fat-saturated gadolinium-enhanced T1-weighted MR images of the pelvic revealed lobulated tortuous strongly enhancing lesion within the myometrium & parametrium (Figure 3).

Figure 1 Transabdominal ultrasonography of the pelvis shows a bulky uterus with an anterior myometrial echo pattern which was heterogeneous with numerous an-echoic cystic & tubular areas inside the uterus.
**Figure 2** Spectral Doppler ultrasonography reveals uterine arteries flowed in a low-resistance pattern and venous flow is seen by spectral tracing in the draining veins.

**Figure 3** (A) Axial T2 & (B) coronal T1-weighted fat-suppressed imaging of the pelvis shows numerous tortuous vascular flow voids in the myometrium. (C) Coronal & (D) sagittal fat-saturated gadolinium-enhanced T1-weighted MR images of the pelvic shows lobulated tortuous lesion within the myometrium & parametrium that is strongly enhancing.
Uterine arteriovenous malformation was proposed as a diagnosis based on these entire results. The patient was admitted to the hospital and scheduled for uterine artery embolization (UAE), after a consultation with an interventional radiologist. Although this procedure is safe, the patient declined this management because she has a desire for future pregnancy and she is afraid of the risk of UAE after pregnancy such as malpresentation, postpartum hemorrhage, and cesarean delivery (Goldberg et al., 2002). Therefore, she was given oral progesterone 5 mg three times a day for 21 days as a conservative treatment. She was followed for a year, remained asymptomatic and conceived spontaneously, and had a low-risk pregnancy with satisfactory maternal and fetal results.

3. DISCUSSION

Despite its rarity, UAVM has a substantial impact on gynecology, because of the potential for severe bleeding and hemodynamic instability, which can be fatal in some patients. In 1926, Dubreuil and Loubat published the first UAVM case. There have only been a few 100 cases described in the literature thus far. UAVM is a congenital or acquired condition, with acquired being the more frequent. Congenital UAVM occurs when the embryological differentiation of primitive blood vessel structures fails, leading to the creation of anomalous vascular connections that expand outside the uterus’ borders and into the pelvis, while the cause of acquired UAVM is unclear (Aiyappan, 2014). Nevertheless, it most commonly occurs as a complication of uterine trauma (uterine curettage, uterine operations), uterine infections, uterine malignancy, myomas, and gestational trophoblastic disease, which can result in the creation of anomalous vascular connections between veins and arteries throughout the healing process.

In the current case, it is likely that an acquired form of UAVM was present in the patient given the previous history of uterine curettage three months ago. Radiographic imaging is necessary for prompt detection and management of UAVM. The optimal first diagnostic imaging modality for diagnosis is transabdominal ultrasonography of the pelvis with color Doppler and spectral analysis. MR angiography is a powerful diagnostic tool for determining the relationship between UAVMs and nearby organs, as well as distinguishing these lesions from gestational trophoblastic disorders. In this case, classical pelvic ultrasound with color Doppler and spectral analysis results allow us to consider the probability of UAVM as our preliminary diagnosis. MRI was done to confirm the diagnosis and also allow for assessment of the extent of the UAVM and surrounding pelvic organs.

The treatment of UAVM depends on patient age, desire to have children in the future, and hemodynamic status, amount of hemorrhage, size, and localization of the lesion. Hysterectomy or embolization of the uterine arteries has been the standard treatment for UAVM. According to several reports, in asymptomatic patients, conservative management is effective (Van Den Bosch et al., 2002; Van Schoubroeck et al., 2004). Patients with mild bleeding have been treated with methylergonovine maleate, danazol, and gonadotropin-releasing hormone analogues, according to certain reports (Takeuchi et al., 2003; Nonaka et al., 2011).

In the present case, the patient is hemodynamically stable and has a desire for future pregnancy besides she is afraid of the risk of UAE after pregnancy such as malpresentation, postpartum hemorrhage and cesarean delivery. Therefore, long-term medical management was the treatment of choice.

4. CONCLUSION

UAVM is a rare and potentially dangerous vascular disease. Bleeding from the uterus is one of the most prevalent symptoms among women, and it can be life-threatening in some cases. The greatest diagnostic tools that provide the most precise information are pelvic ultrasound with color Doppler and spectral analysis. Conservative management is a relatively safe and effective treatment option for hemodynamically stable patients and in childbearing age. The patient must be closely monitored for the UAVM’s reactivation and the reappearance of profuse uterine bleeding. If it shows up again, prompt and suitable treatment will be required as the patient desires for future pregnancy.

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Author’s contribution
Samih Kajoak prepared, revised the manuscript, and agreed with the final version.

Informed Consent
Written & oral informed consent was obtained from the patient in this case report. Additional informed consent was obtained from all individuals from whom identifying information is included in this case report.
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Conflicts of interest
The authors declare that there are no conflicts of interests.

Data and materials availability
All data associated with this study are present in the paper.

REFERENCES AND NOTES


