Arteriovenous Malformation in a Bicornuate Uterus Associated with Dilatation and Curettage: Case Report

Dilatasyon Küretaj ile İlişkili Bikornuat Uterusta Saptanan Arteriovenöz Malformasyon

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ABSTRACT Uterine arteriovenous malformation (AVM) is a life-threatening condition due to vaginal bleeding. It may be congenital or acquired. It may be associated with uterine traumas like dilatation and curettage (D&C), uterine surgery and therapeutic abortion. In the present case report, a 24-year-old G3 P1 A1 woman presented to our gynecology clinic with complaints of heavy vaginal bleeding and passage of blood clots. On history, she had two similar episodes of heavy vaginal bleeding in one week after she underwent a D&C procedure for a 12-week missed abortus three months ago. In our clinic, the patient was diagnosed with AVM located in one of the cavities of the bicornuate uterus and associated with D&C, and she was treated with intrauterine balloon replacement.

Key Words: Arteriovenous malformations; uterine balloon tamponade; dilatation and curettage

ÖZET Uterin arteriyovenöz malformasyonlar (AVM) özellikle ciddi vajinal kanamalara sebep olması nedeniyle hayatı tehdit edebilir. AVM konjenital veya edinsel olabilir. Dilatasyon küretaj (D&C), uterin cerrahi ve terapötik küretaj gibi uterin travmaya neden olan durumlarda gözlenebilir. Yirmidört yaşında G3 P1 A1 hasta, yoğun vajinal kanama ve pıhtı düşürme şikayetiyle, jinekoloji kliniğine başvurdu. Hasta öncesinde bir hafta içerisinde bu şekilde iki kez yoğun vajinal kanama şikayeti bildirmekte idi. Ayrıca hastaya üç ay önce, 12 haftalık intrauterin ex fetus nedeniyle D&C işlemi uygulandığı öğrenildi. Bikornuat uterusu mevcut olan vakada, uterin kavitelerin birinde AVM saptanmış ve intrauterin balon yerleştirilmesi yöntemiyle tedavi edilmiştir.

Anahtar Kelimeler: Arteriyovenöz malformasyonlar; uterin balon tamponadi; dilatasyon ve küretaj

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Uterine arteriovenous malformation (AVM) is a potentially life-threatening condition as patients may present with severe vaginal bleeding. In the literature, fewer than 100 cases have been reported.[1] Most of the cases were women aged between 20 and 40 years.[2]

Uterine AVM may be congenital or acquired.[3] The congenital form is very rare. It is the result of a defect in embryonic vascular differentiation or a premature arrest in the development of the capillary plexus leading to multiple abnormal connections between arteries and veins.[4] Recently, there has been a rise in the reported number of acquired cases of AVM appearing after pregnancy, abortion, and curettage. Besides, in most of these

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cases, spontaneous resolution of vascular lesions during follow-up has been reported.[5] Here, a case of AVM located in one of the cavities of a bicornuate uterus, associated with D&C and treated with intrauterine balloon replacement is presented.

CASE REPORT

A 24-year-old G2 P1 A1 woman presented to our gynecology clinic with complaints of heavy vaginal bleeding and passage of blood clots. On history, she had two similar episodes of heavy vaginal bleeding in one week after she had a D&C procedure for a 12-week missed abortus at an another hospital three months ago. When she had one more episode of bleeding in the preceding 2 months, D&C was repeated at the same hospital.

On examination, she was afebrile and hemodynamically stable with a hemoglobin (Hb) level of 11.8 g/dL. Vaginal examination showed a large amount of blood at the external os of the uterine cervix and active bleeding. Besides, bleeding intermittently increased. The human chorionic gonadotropin (hCG) level was less than 1.2 mIU/mL. Transabdominal ultrasonography of the pelvis showed a bulky uterus measuring 76x44x58 mm with an endometrial thickness of 15 mm. There was increased vascularity of the uterus with a prominent vessel seen on the posterior wall of the uterus. The sagittal endovaginal image of the uterus showed intracavitary lesions in the posterior myometrium (Figure 1a). Color and spectral Doppler image showed multiple tortuous vessels with a multidirectional high-velocity, low-resistance flow producing a color mosaic pattern (Figure 1b).

She was hospitalized and immediate hemodynamic resuscitation was performed, with intravenous fluids supplement and blood component therapy. Pelvic angiography was planned, but the clinician who was experienced in angiography and embolization was off duty. The hemorrhage was not responsive to medical therapy such as transamines and uterotonics (methylergonovine, oxytocin). Subsequently, uterine tamponade with a 16 French Foley balloon, under ultrasonography guidance, was performed. About 40cc saline was infused into balloon for effective cavity tamponate. Intermittent heavy bleeding dramatically stopped. In the clinic, the patient was followed for two days with her intracavitary balloon catheter. She was uneventfully discharged. In the follow up visit two weeks after discharge, she reported to have a regular menstrual cycle and remained asymptomatic. The patient also indicated her desire to conceive again, but she was recommended contraception for minimum six months in the future. Written informed consent was obtained from the patient for present case presentation.

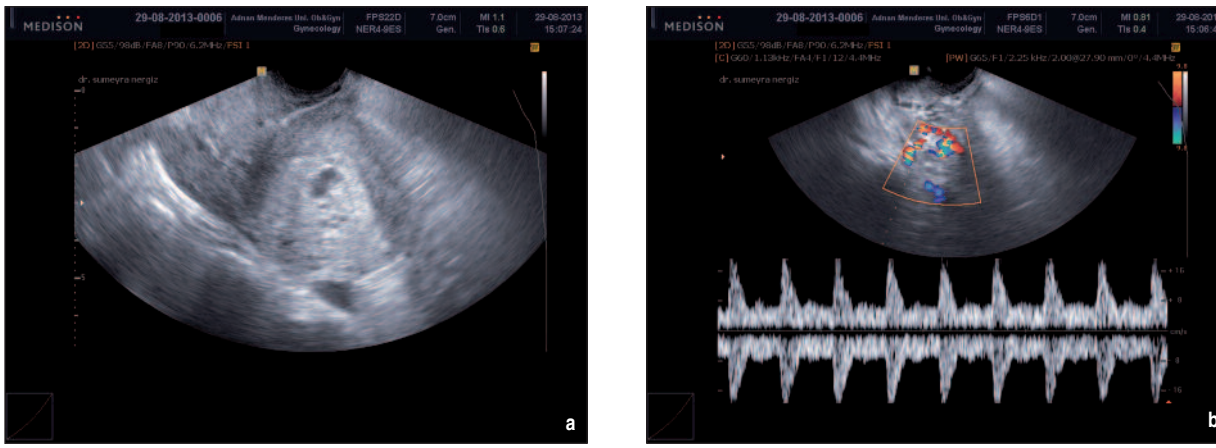

FİGURE 1: (a) The sagittal endovaginal image of the uterus shows intracavitary lesions in the posterior myometrium. **(b)** Color and spectral Doppler image shows multiple tortuous vessels with a multidirectional high-velocity, low-resistance flow producing a color mosaic pattern.

DISCUSSION

Uterine AVM had an incidence of 4.5% in 464 pelvic sonographic examinations performed for pelvic bleeding.[6] Prior D&C, therapeutic abortion, uterine surgery, or direct uterine traumas, less commonly, diethylstilbestrol exposure, endometrial carcinoma, cervical carcinoma, and gestational trophoblastic disease have been incriminated for the causes of acquired uterine AVM.[7] In the present case, the patient had prior D&C three months ago on history.

Patients affected by uterine AVM mostly present with menorrhagia or menometrorrhagia. Symptoms may occur very slowly or suddenly.[3] The pattern of bleeding is intermittent and torrential, suggestive of arterial hemorrhage. Uterine bleeding is thought to occur when superficial vessels of the AVM are exposed from sloughing of the endometrium iatrogenically during D&C or menses.[8] Lower abdominal pain, dyspareunia and anemia secondary to blood loss are the other symptoms. In very severe AVMs, shunting may cause cardiovascular symptoms such as dyspnea, fatigue, and even heart decompensation.[3] In the present case, cardiac decompensation was not observed, but the patient presented with intermittently increasing vaginal bleeding, abdominal pain, and fatigue, which is consistent with the literature.

In the diagnostic evaluation of AVM, although angiography remains the gold standard method, ultrasound is the most common form of initial and available investigation.[9] Color and duplex Doppler ultrasound is a valuable, noninvasive tool for diagnosis of uterine AVM.[10] Color Doppler ultrasonography shows multiple tortuous vessels with a multidirectional flow and apparent flow reversals of juxtaposed reds and blues with different flow velocities giving a mosaic pattern similar to the present case. Duplex Doppler ultrasonography shows the classic features of arteriovenous shunting including a high velocity arterial flow with low resistance, high peak systolic velocity (PSV), an arterial spectral waveform with a high diastolic component, and a pulsatile high velocity venous waveform with little variation in systolic-diastolic velocities. Vascular malformations with a PSV value of ≥0.83 m/s are potentially dangerous and PSV values <0.39 m/s are considered as safe.[11] In the present case, the PSV value was 0.76 m/s, which was moderately safe.

Necrotic placental tissue due to retained products of conception may also be associated with low-resistance, high velocity pulsating flow in the endometrial cavity or the myometrium, similar to an AVM on spectral Doppler analysis. Besides, gestational trophoblastic disease must be considered in the differential diagnosis in cases of AVM. Therefore, accurate initial differential diagnosis is critical to prevent D&C as a treatment for abnormal uterine bleeding, which may cause severe bleeding and is contraindicated in uterine AVM.[12] However, in the present case, D&C was repeated in another hospital due to bleeding two months after the first D&C session. There have been no report about pathological examination.

Management of uterine AVM depends on the hemodynamic status, degree of bleeding, patient age, and desire for future fertility. Acute treatment involves stabilizing the patient's hemodynamic status, and stopping blood loss. Traditionally, hysterectomy is the treatment of choice. However, in stable patients who are appropriate for close follow-up, long-term medical management may be preferred. Moreover, oral contraception as well as intramuscular and subsequent oral methylergonovine maleate has been shown to be associated with regression of lesions based on US.[8] Besides, Nonaka et al. reported that the uterine AVM lesion completely disappeared after 6 months of treatment with a GnRH agonist and that the patient subsequently had a successful pregnancy and delivery.[13] Parenteral estrogen, progesterone, PGF2 alpha and danazol were included in medical management by Patel et al.[12] The case presented here was recommended methlylergonovine maleate, tranexamic acid and oral contraceptives by different clinicians and she used them approximately for one month, but vaginal bleeding gradually increased.

In addition to medical management, conservative treatment includes selective uterine arterial embolization (UAE) and uterine vascular ligation and uterine AVM lesion resection through bipolar coagulation under laparoscopy.[14] Failed embolization, high recurrence rates up to 16.7%, repetitive embolization rates up to 21.4%, postembolization syndrome, decreased rates of successful pregnancy, increased pregnancy complications such as spontaneous abortions, intrauterine growth restriction, postpartum uterine atony and postpartum hemorrhage due to poor vascularization and improper placental implantation because of uterine contractions may be observed in patients treated with UAE.[5,12,13] In the present case, uterine tamponade with a 16 French Foley balloon, under ultrasonography guidance, was performed. Intermittent heavy bleeding dramatically stopped.

In conclusion, clinicians should have a high level of suspicion about the presence of uterine AVM if a woman has recurrent, unexplained, massive genital bleeding after D&C despite medical treatment. It should be kept in mind that Doppler ultrasonography helps to differentiate uterine AVM from rest placenta or trophoblastic diseases, which prevents unnecessary, hazardous recurrent D&C in uterine AVM. Besides, clinicians should decide carefully UAE in management of uterine AVM as it is not free of complications. Intrauterine Foley catheter application is a good alternative method in management in that it is inexpensive, readily available and easily inserted into the uterine cavity without any special techniques. The procedure is valuable and effective especially when there is not an opportunity to perform a definitive treatment such as embolization or hysterectomy or when patients desire future fertility.

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