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Case Report

Uterine arteriovenous malformation: a rare cause of abnormal uterine bleeding

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ABSTRACT

Uterine arteriovenous malformation (AVM) is a rare gynecological entity that usually presents with vaginal bleeding of variable spectrum. High level of suspicion aided by color Doppler ultrasound is needed to confirm the diagnosis. This case report describes a 52-year-old woman G7P6A1 who presented with irregular pervaginal bleeding for 5 years. Her symptom was recurrent, on and off and refractory to hormone therapy. She was diagnosed with uterine AVM on pelvic color Doppler ultrasound that revealed a dilated and hypervascular cystic mass of 6.2×4.1 cm located at right uterine wall where blood flow was bidirectional. As a definitive treatment, open abdominal hysterectomy was performed successfully. This report reminds gynecologists to consider uterine AVM as a rare differential diagnosis of abnormal uterine bleeding (AUB).

Keywords: Abnormal uterine bleeding, Color Doppler ultrasound, Hysterectomy, Uterine arteriovenous malformation

INTRODUCTION

Uterine arteriovenous malformations (AVMs) are rare and abnormal direct connections between the uterine arteries and veins.[1] These can be either congenital or acquired.[2] Congenital AVMs are extremely rare and represent defective vascular development during embryogenesis, whereas acquired AVMs may follow dilatation and curettage (D&C), gestational trophoblastic diseases (GTDs), direct uterine trauma, normal pregnancy or rarely after hysterectomy.[3,4] Uterine AVMs are usually found in reproductive age women; rare in premenopausal group and very rare after menopause.[1] Most of patients typically presents with abnormal vaginal bleeding of variable spectrum that may sometimes end up with potentially life-threatening brisk hemorrhage.[5] To diagnose uterine AVM, color flow Doppler ultrasound serves as a reliable and non-invasive imaging tool especially in a low-resource set-up. Digital subtraction angiography (DSA), computed tomography (CT) angiography, MRI or magnetic resonance (MR) angiography can be used further for better delineation of uterine AVMs.[4] Though uterine artery embolization (UAE) is a preferable conservative method of treatment in patients of child-bearing age; hysterectomy remains definitive surgical approach to treat uterine AVMs and that can be performed either as first-line therapy or after failure of embolotherapy.[6,7]

We encountered a case of uterine AVM as a rare cause of abnormal uterine bleeding (AUB), which was successfully treated with open abdominal hysterectomy.

CASE REPORT

A 52-year-old rural housewife, gravida 7, para 6, abortion 1 presented at a peripheral private clinic with history of irregular pervaginal bleeding for last 5 years. All of her viable births were spontaneous home delivery at full term with average birth weights. Her last issue was intrauterine death (IUD) at 38 weeks gestation that was delivered vaginally at home. She also had a history of missed abortion at 16 weeks prior to her last pregnancy 18 years back, for which D&C was performed in a non-recognized centre.

With initial history of irregular pervaginal bleeding for 6 months, the patient seeked gynecological consultation in a rural clinic and treated with oral cyclical norethisterone (5 mg twice daily) for 3 months. As her symptoms persisted after 3 months, she again visited to gynecologist and was prescribed oral norethisterone (5 mg thrice daily) for another 3 months and advised for pelvic ultrasonography but refused. Due to recurring symptoms after 6 months of hormonal therapy, she did a pelvic ultrasonography as self-referred and a complex mixed echogenic mass of 3.9×3.5 cm size was found in right uterine wall with grossly heterogenous nature of rest uterine myometrium. Unfortunately, thereafter the patient did not seek any further consultation; so was not possible to elucidate the specific pathology at that time. But the patient continued previously prescribed oral hormone preparations irregularly on and on by her own for next 1.5 years and however; bleeding was controlled at once. On the day of her current presentation, she again complains of irregular pervaginal bleeding for last 2 years despite taking hormones and consulted to me in a peripheral private clinic. She was found mildly anemic, normotensive, non-diabetic and euthyroid. She had no significant past medical or surgical illness.

On bimanual examination, uterus was found mild bulky, soft in consistency, non-tender, freely mobile and vaginal fornices were free. Transvaginal ultrasonography (TVS) was advised and revealed a 6.2×4.1 cm cystic mass located at right uterine wall with heterogenous myometrial echotexture. Color flow Doppler study showed dilated vascular channels with increased vascularity in right half of uterus and blood flow was bidirectional. Endo-myometrial junction was well-defined with no intracavitary collection and both adnexa found apparently normal. Thus, based on these sonographic findings, arteriovenous malformation of uterus was diagnosed. Other investigation findings were hemoglobin level 10.3 g/dl, haematocrit 36.80%, serum thyroid stimulating hormone (TSH) 0.99 iIU/ml, FT_4 1.14 ng/dl, serum beta hCG <1.20 mIU/ml and VIA negative.

Owing to patient's advanced age, completed family and being symptomatic, she was advised for total abdominal hysterectomy as a definitive treatment. Upon her informed written consent, elective surgery was scheduled under subarachnoid block (SAB) with arrangements of 2 units of whole blood. Intraoperatively, numerous dilated, tortuous, prominent pulsatile vessels were found along the right half of uterus, consistent with the sonographic findings. During clamping, dilated vessels were ligated and cut cautiously along with meticulous care. Special attention was given to avoid any untoward vascular or ureteral injury. Thus surprisingly, an uncomplicated total abdominal hysterectomy was performed with minimal bleeding in the operative field; although significant intraoperative blood loss was anticipated. Patient's immediate postoperative period was unremarkable and was discharged home at 6th POD with healthy abdominal wound. During the follow-up 6 weeks later, the patient had no further episodes of pervaginal bleeding.

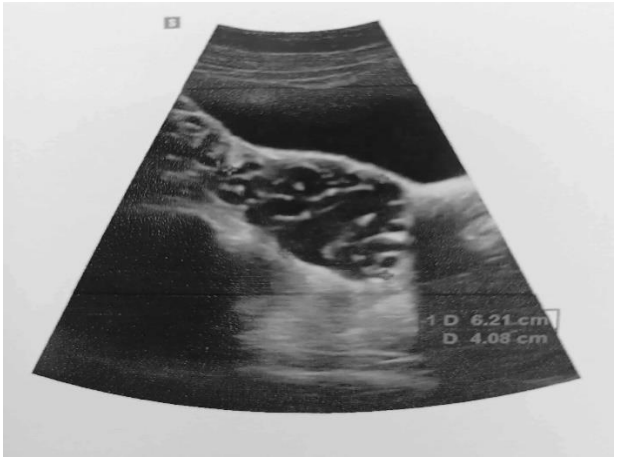

Figure 1: Gray scale image of uterus shows 6.2×4.1 cm cystic mass located at right uterine wall.

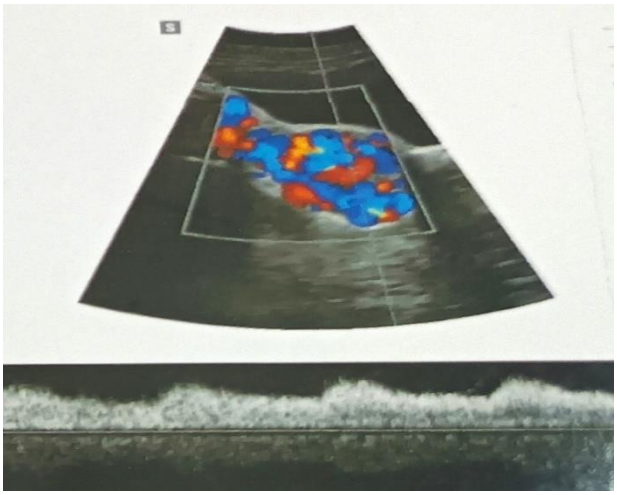

Figure 2: Color Doppler image shows dilated vascular channels with increased vascularity in right half of uterus and blood flow was bidirectional.

DISCUSSION

Uterine AVMs are extremely rare entities in gynecology defined as abnormal connections between uterine arteries and veins. These can be either congenital or acquired

(traumatic) lesions. Congenital AVMs are suspected to arise from anomalous embryologic differentiation of primitive vascular structures resulting in abnormal communications between arteries and veins. Alongside, congenital lesions may extend beyond the uterus to invade surrounding structures and more commonly have multiple vascular connections.[8] In contrast, acquired uterine AVMs are usually iatrogenic, more common than congenital AVMs and may form after D&C, GTDs, direct uterine trauma, uterine surgery, uterine malignancy, normal pregnancy, pathologic pregnancy-related conditions or rarely after hysterectomy.[9] The present case possibly was an acquired form of uterine AVM given the confined nature of lesion not extending beyond the uterus (sonographically evident), did not present earlier in life and the presence of prior contributing risk factors; previous one D&C and repeated childbirths. Moreover, this case was not considered unique as prolonged interval intervenes from her past D&C or last confinement to onset of symptoms. Some authors reported symptoms of AVMs presenting from 3 months to 35 years after hysterectomy.[10,11]

Uterine AVM is a rare cause of AUB that encompasses a variable spectrum of clinical presentation ranging from vaginal spotting to life-threatening profuse bleeding in some patients. A high level of suspicion is required to document the existence of uterine AVM in a patient with persistent, recurrent and unexplained vaginal bleeding.[9] Thus, uterine AVM was considered to be a differential in this premenopausal woman who presented with recurrent irregular pervaginal bleeding that persisted despite hormone therapy and had a suspicious complex mixed echogenic mass in right uterine wall identified on ordinary pelvic ultrasound.

In current practice, uterine AVM can easily and reliably be diagnosed using color Doppler ultrasonography. Gray scale ultrasound findings include a nonhomogenous mass with multiple tubular or cystic structures within the myometrium. In addition, color Doppler study demonstrates multidirectional, hypervascular appearance with turbulent flow.[4] As, retained products of conception (RPOC) and GTDs may also give similar Doppler findings, keeping remote possibility in mind, estimation of serum beta hCG level was considered in this patient to rule out these conditions and the level was found normal (<1.20 mIU/ml).

Although, digital subtraction angiography (DSA) remains the gold standard for the diagnosis of uterine AVMs, it is rarely performed for diagnosis alone because of its invasive nature. DSA findings include bilateral hypertrophy of uterine arteries feeding a tortuous, hypertrophic arterial mass with large accessory feeding vessels, and early drainage into enlarged hypertrophic veins.[4] Several other diagnostic imaging tools including CT angiography, MRI or MR angiography may be helpful to better delineate the anatomy and relation of uterine AVMs to the neighboring organs.[2] None the less, to obtain definitive diagnosis of uterine AVM in our case, we had to rely on color Doppler sonography in a low-resource set-up like us; because of its easy availability, noninvasiveness and considering patient's economic constraints.

Management of uterine AVM depends on degree of bleeding, patient's hemodynamic status, age, desire for future fertility and localization and size of the lesion. A diverse range of therapeutic modalities exist. For treatment of asymptomatic uterine AVMs, although, there is currently no clear agreement, conservative approach has been suggested in some studies.[12,13] Some documented modalities for expectant or medical management of patients with mild haemorrhage include parenteral estrogen and progestin, methylergonovine maleate, gonadotropin releasing hormone (GnRH) analogues and danazol.[4] Uterine artery embolization (UAE) or hysterectomy is the mainstay for management of uterine AVM. UAE remains the well-recognized initial treatment option for women at reproductive age desiring future fertility.[14] However, not only technical and clinical efficacy but also difficulties and failures of UAE have been shown in several literatures. A systemic review of 40 studies that included 54 patients with acquired uterine AVMs who underwent transcatheter embolization found a primary and secondary success rate of 61% and 91% respectively.[15] In contrast, selective arterial embolization may also have a higher failure rate due to incomplete obliteration and recruitment of collateral vessels leading to recurrence.[16]

Hysterectomy is the definitive surgical therapy for uterine AVMs and may be performed either first-line or after failure of embolotherapy. However, it may not always be desirable as a first-line treatment due to its invasiveness and incapability to retain future fertility. Regardless of all possible complications of traditional hysterectomy, Smith reported that complete surgical resection of AVM appears to be permanently successful, though risk of large volume blood loss at the time of surgery.[17] Given patient's advanced age, being symptomatic, refractory to hormone therapy, localized lesion to uterus and completed family, hysterectomy was considered as a definitive surgical treatment in this instance.

CONCLUSION

This case report highlights the use of color Doppler ultrasound to diagnose uterine AVM in a premenopausal woman presenting with irregular pervaginal bleeding and has history of repeated childbirths and D&C in remote past. Hysterectomy can be considered as a comparatively safe and effective therapeutic option to manage this age group of women. Our case serves as a reminder to consider uterine AVM in differentials as a rare cause of AUB.

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