CASE REPORT

Uterine arterio-venous malformation, an uncommon life-threatening condition: a case report

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Abstract

Uterine Arteriovenous Malformation is a rare gynaecological disorder which commonly presents with profuse vaginal bleeding. This case report presents a patient referred to the Military Hospital, Rawalpindi from Pakistan Aeronautical Complex Hospital Kamra, a peripheral secondary care hospital. Patient was diagnosed as a case of Uterine Arteriovenous Malformation at the Military Hospital and was successfully treated with uterine artery embolization.

Keywords: Angiography, Arteriovenous Malformations, Uterine Artery Embolization, Uterine Haemorrhage
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Introduction

Uterine Arteriovenous Malformation (UAVM) is an abnormal, direct communication between arteries and the veins of the uterus without intervening capillaries.[1] These are heterogeneous sized high flow vessels.[2] This vascular malformation is a rare finding and a potentially life-threatening condition. One hundred cases have been reported in the literature.[3]

Patients with UAVM presents with variable symptoms. The most common being irregular, intermittent uterine bleeding which is usually profuse with no obvious cause, it is diagnosed when uterine bleeding remains uncontrolled despite medical measures.[4] Timely diagnosis of this condition is crucial as instrumentation required to find out other causes of the uterine bleeding can cause massive haemorrhage.[5] In this case report, we present our experience with a patient having a similar condition.

Case Report

A 32-year-old female, Para1+2 presented in the Obstetrics & Gynaecology (OBGYN) outpatient department (OPD) of the Pakistan aeronautical complex (PAC) hospital, Kamra in March, 2018. She was then referred to the Military Hospital (MH), Rawalpindi (Rwp). Patient had one male child, four years of age, delivered by an uneventful caesarean section (CS). Her second pregnancy ended in the first trimester as a missed miscarriage, treated by evacuation and curettage. Her third pregnancy was diagnosed as gestational trophoblastic disease (GTD) and was terminated by suction and curettage at 12 weeks. She remained asymptomatic after the procedure. Her serum β Human Chorionic Gonadotrophin (β-HCG) dropped to a non-pregnant range (<5mIU/L) in four weeks. Histopathology report revealed a complete mole.

She reported again at the PAC Hospital, six weeks later with profuse vaginal bleeding for the past two days. Her haemoglobin (Hb) was 4.8g/dl (Normal Range 12.0 - 15.5 g/dl) with a normal coagulation profile and β HCG noted to be in a non-pregnant range. She was managed with transfusion of red cell concentrate (RCC), injectable tranexamic acid and mefenamic acid. The gynaecologist at the PAC hospital performed a transvaginal ultrasound (TVU) but no abnormality was detected. Patient had similar episodes of heavy bleeding and required repeated admissions. She had multiple transfusions of RCC and fresh frozen plasma (FFP).

After about five months, her endometrial biopsy was taken which revealed a non-specific endometritis. Patient was given broad spectrum antibiotics followed by Norethisterone 15mg/day for 21 days. Despite treatment, her symptoms did not settle down. TVU repeated by the radiologist, showed hypo-echoic areas of about 2.00-2.50cm in the posterior myometrium reaching the endometrium. These areas showed high vascularity on Doppler ultrasound. These findings were interpreted by a radiologist as invasive mole but βHCG was not in accordance with it. The diagnosis remained inconclusive and the patient was referred to MH.

On admission at MH, she had no active vaginal bleeding, with haemoglobin of 6.0 g/dl and normal β-HCG. TVU revealed hypo-echoic, tubular spaces in posterior myometrium. Colour Doppler showed these spaces to be highly vascular with raised diastolic flow in multiple

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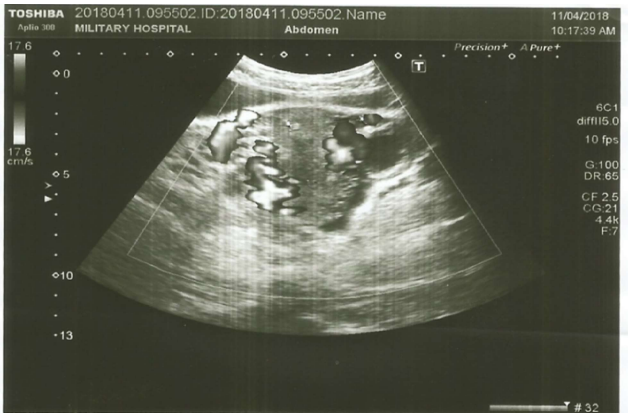

Figure-1: Uterine artery Doppler showing increased vascularity and multidirectional flow in posterior myometrium.

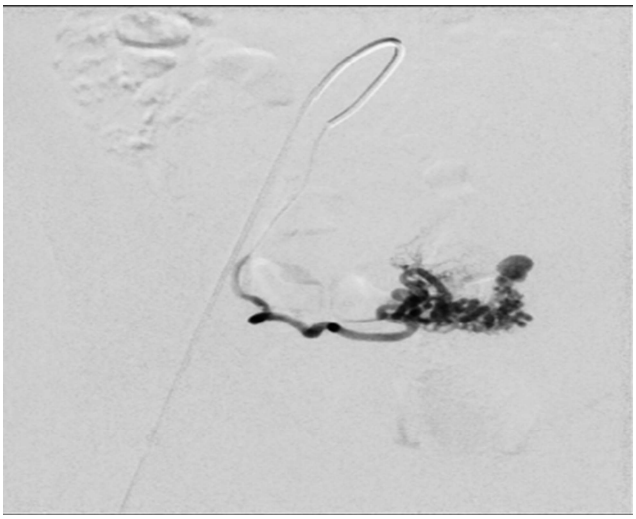

Figure-2: Uterine artery angiography confirming the diagnosis of UAVM.

directions and low resistance, concluding it to be a sub-endometrial arterio-venous malformation. Diagnosis was confirmed by a uterine artery angiography. Colour Doppler ultrasonogram is shown in Figure-1 and uterine artery angiography is shown in Figure-2.

To preserve the fertility of the patient, uterine artery embolization was planned instead of hysterectomy. Bilateral Uterine artery embolization was performed by an Interventional Radiologist through the femoral arteries, on 18th of April 2018. Polyvinyl alcohol (PVA) particles were used for embolization. During her admission, she was transfused with 8 RCC and 6 FFP. Patient experienced mild lower abdominal pain for a few days. She was discharged after five days. Consent was taken from the couple to publish their case report. TVU was done two weeks after the embolization. A hypo-echoic lesion of 1.00 x 1.50 cm was found at the site of UAVM. She resumed her normal menstrual cycles after two months. Patient is now asymptomatic and is on regular follow up every 3 months, in a gynaecology OPD of the PAC hospital.

Discussion

UAVM is a rare condition. Literature review shows its incidence to be around 0.10%.[1] It is an important differential diagnosis in patients of reproductive age group with unexplained vaginal bleeding.[6]

First case of UAVM was reported in 1926 by Dubreil and Loubat.[7] It is classified into two types, congenital and acquired. Congenital UAVM is caused by an abnormal differentiation of the primitive vascular structure in which capillary bed is not formed leading to a direct communication between the arteries and the veins. Congenital UAVM can be extensive and may involve pelvic walls. Acquired UAVM is much more common. It occurs due to disruption and abnormal healing of the vasculature. Patients usually have a history of uterine damage such as a CS, endometrial curettage, endometritis, endometriosis or gestational trophoblastic disease. Acquired UAVM usually has single or bilateral supply exclusively from the uterine arteries.[8]

UAVM may be symptomatic or asymptomatic. Its most common symptom is periodic, profuse vaginal bleeding indicating it to be arterial in origin. It has been reported in the postmenopausal women as well.[9]

The reported case is of an acquired UAVM as indicated by the patient's history of multiple events causing uterine damage such as CS, suction with evacuation and a repeated endometrial curettage. The probable cause of this malformation was the first endometrial curettage done after her missed miscarriage followed by the second curettage, damaging the vessels and endometrium covering them, eventually leading to a heavy vaginal bleeding.

Different imaging modalities are used to diagnose UAVM. Angiography is the standard investigation for an AVM. It shows complex tangle of the vessels being supplied by large arteries, the feeding arteries. There is an early venous drainage along with stasis of the contrast medium in the abnormal vasculature.[10] Angiography being an invasive procedure cannot be used as a method of choice for investigation purposes. Currently, ultrasound pelvis, especially TVU is a useful, non-invasive investigation which

shows nonspecific findings like an echoic area, tubular spaces or masses in the myometrium with a normal endometrium. Colour and spectral Doppler provide more accurate information of the intense vasculature with multidirectional flow and low resistance with Resistive index and Pulsatility index of 0.25-0.55 and 0.3-0.6, respectively.

Although the diagnosis of UAVM seems simple but it is quite often missed and confused with GTD or retained products of conception due to similar findings on the ultrasound. Its diagnosis requires a high degree of suspicion. In this case, the initial TAU was done at a peripheral hospital. Due to a limited experience and rarity of the condition, the radiologist could not detect the malformation and misdiagnosed it as an invasive mole.

There are many factors that determine the management of UAVM, the most important being the haemodynamic stability, the desire for future fertility and the availability of expertise. Treatment modalities include expectant, medical and surgical options; however, expectant and medical treatment is uncommon. Agents used for medical management are Progestin, parenteral Estrogen, Danazol and Methylergonovine. Angiographic embolization of uterine arteries has now replaced hysterectomy which was previously considered the treatment of choice for a bleeding UAVM. Several successful pregnancies have been reported after uterine artery embolization.[11] It can be used in emergency setting as well as an elective procedure. Different materials are available for embolization including Polyvinyl alcohol, detachable balloons, histoacryl glue, coils and haemostatic gelatin.

Conclusion

This case highlights the need to consider UAVM as an important differential diagnosis among the patients of reproductive age group presenting with irregular heavy vaginal bleeding, particularly if the β-HCG level is within the normal limits. It also shows the importance of experience at interpretation of pelvic ultrasound before attempting any instrumentation in such patients. This becomes more important if patient has a history of procedure that may have caused uterine/endometrial damage. During the management of this patient, the procedure of uterine artery embolization was observed and learnt.

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