

A Rare Case of Association of Uterine Arteriovenous Malformation with Positive Serum Beta HCG.

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Abstract:

Background: Uterine arteriovenous malformations are more frequent than diagnosed. AVM are seen in majority after curettage post abortion. Rarely an association between AVM and medical abortion is seen.

Case: A, P2L2A3, patient presented following medical abortion, with continuous bleeding per vaginum for 23 days. TVS doppler revealed ill-defined irregular heterogenous anechoic channels showing vascularity in posterolateral aspect of lower uterine segment of size 3.5x3.0x 2.4 cm, with vascular channels showing arterial and venous components. Findings were suggestive of arteriovenous malformation with a differential diagnosis gestational trophoblastic disease. Her serum beta hCG was 9500 IU/L. MRI pelvis confirmed AVM and so, selective uterine artery embolization was done. Patient responded well and was followed up with serum beta hCG, which reached non-pregnant values in 4 months.

Conclusion: Uterine AVM should be a differential diagnosis in all cases presenting with abnormal bleeding after abortion even in the absence of instrumentation and uterine artery embolization is an effective means of combating any complications.

Keywords: uterine arteriovenous malformation, spontaneous abortion, beta hCG, embolization

Introduction:

Uterine Arteriovenous malformation (AVM) is defined as multiple small fistulas in the myometrium and can have either unilateral or bilateral uterine artery supplies or even extra uterine supplies. [1] These lesions can remain asymptomatic or can present as torrential haemorrhage requiring emergency treatment. Less than 100 cases have been reported by 2005, but many due to their varied nature may remain undiagnosed [2]. The incidence of uterine AVM is around 4.5%.¹ Uterine AVM can be congenital or acquired [3]. Usually, any kind of surgery of the uterus like caesarean section or myomectomy or instrumentation in cases of abortions can present as AVM [4, 5, 6]. Very rarely, symptomatic AVM is detected following spontaneous abortion. The diagnosis of AVM in majority of cases, thus necessitate high degree of suspicion. Our case has number of peculiarities; rare diagnosis of AVM, AVM after spontaneous abortion with no history of instrumentation in recent past, high levels of beta hCG on presentation and its slow decline to non-pregnant values even after embolization.

Case:

A 29-year-old para 2 live 2 abortion 3 female presented to the outpatient department of obstetrics and gynaecology of Sir Ganga Ram Hospital, New Delhi with chief complaints of bleeding per vaginum for 23 days, soaking 3-4 pads per day associated with passage of clots. On enquiring, patient gave history of pregnancy of 8 weeks. She had an ultrasound done, which was suggestive of a missed abortion of 7 weeks 1 day with a subchorionic hematoma of size 35x 18mm in superior and posterior aspect of the sac. She was prescribed medical abortion in view of the

above-mentioned findings. But patient continued to bleed thereafter. Ultrasound was done again in view of continued bleeding and was suggestive of retained product of conception of size 20x16x16mm (2.8ml) and was again given vaginal misoprostol. Patient continued to bleed and presented to me. She had regular menses prior to this conception with normal flow since menarche. Her obstetric history was two missed abortion at 2 and a half months amenorrhea 7 years back and other at 3 and a half month amenorrhea 5 years back. Both these cases were followed by dilation and curettage, not associated with any excessive bleeding. Last abortion was as mentioned above. She had two live births of 6 years and 4 years, both born by lower segment caesarean sections. Both these cases were also not associated with postpartum haemorrhage or need for blood transfusion.

On examination, her general condition was fair, her vitals were pulse rate 92 beats per minute, blood pressure 120.70 mmHg, respiratory rate 14 per minute and temperature normal. Pallor was present. There was no icterus, cyanosis, pedal edema or lymphadenopathy. Cardiovascular and respiratory system examination was within normal limits. On per abdominal examination, a transverse scar was seen around 2 cm above pubic symphysis, there was no pelvic mass or hepato-splenomegaly. There was no guarding, tenderness or rigidity. On per speculum examination, cervix was obscured due to bleeding, external cervical os closed and vagina healthy looking. On per vaginum examination, cervix upwards, uterus retroverted, bulky, firm with restricted mobility with bilateral fornices free and non-tender.

I decided to get a transvaginal ultrasound (TVS) with colour doppler along with all baseline investigations and serum beta hCG. TVS doppler revealed ill-defined irregular heterogenous anechoic channels showing

vascularity in posterolateral aspect of lower uterine segment of size 3.5x3.0x 2.4 cm, bulging into the lower endometrial cavity with vascular channels showing arterial and venous components, as seen in figure 1.

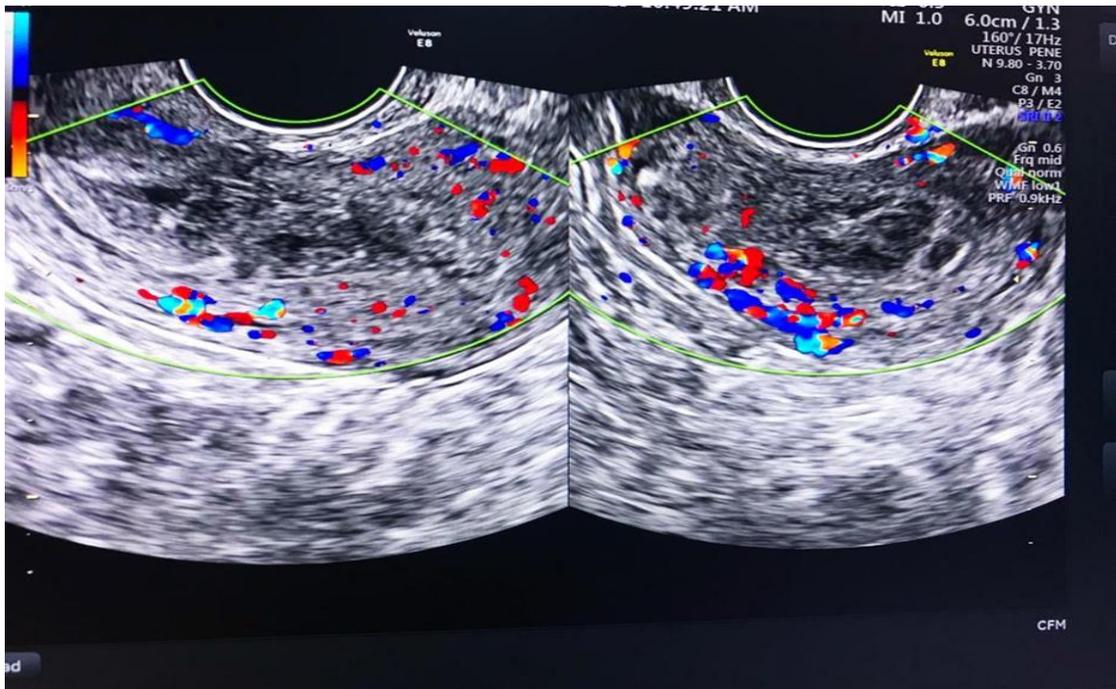


Figure 1: TVS showing ill-defined mass with enhanced myometrial vascularity

Findings were suggestive of arteriovenous malformation with a differential diagnosis gestational trophoblastic disease. Her baseline investigations were, hemoglobin 9.5gm%, total leucocyte counts 10,500/ml, platelet counts 1.54 lakhs, serum beta hCG 9500 IU/L, INR 1.0, liver and kidney function tests were within normal limits. MRI with contrast was planned to reach a definitive diagnosis. It showed lobulated bunch of vascular flow voids with surrounding T1 isointense and T2 STIR bright signals size of 3.9x3.4x2.4cm involving postero-lateral aspect of lower uterine body bulging into the lower endometrial lumen with adequate curvilinear enhancement on dynamic post-contrast study. Angio-venography showed lobulated bunch of tortuous enhancing vessels involving posterolateral aspects of lower body of the uterus fed by left uterine artery and drained by left vein. After consultation with intervention radiologist, patient was explained about the need to undergo selective uterine artery embolization and follow up thereafter. Fluoroscopic guided selective uterine artery embolization was done using polyvinyl alcohol particles. Both right and left uterine arteries (figure 2, 3, 4) were seen supplying the mass and thus were embolised.

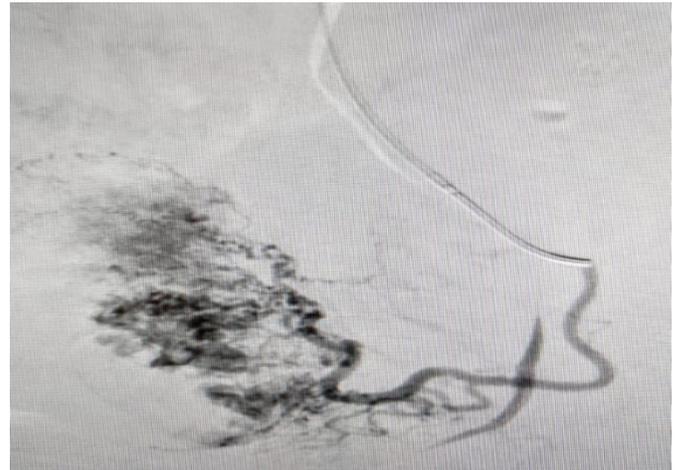


Figure 2: Branch of left uterine artery supplying the mass.

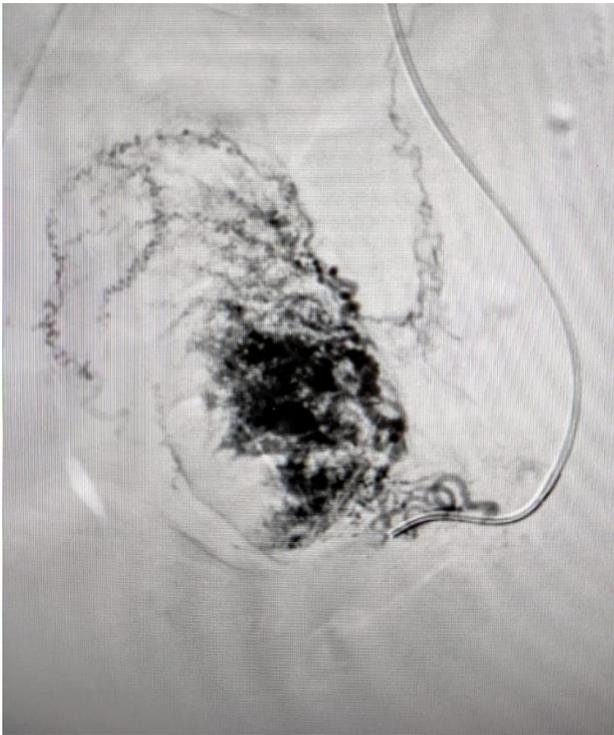


Figure 3: Selective cannulation and embolization of left uterine artery.

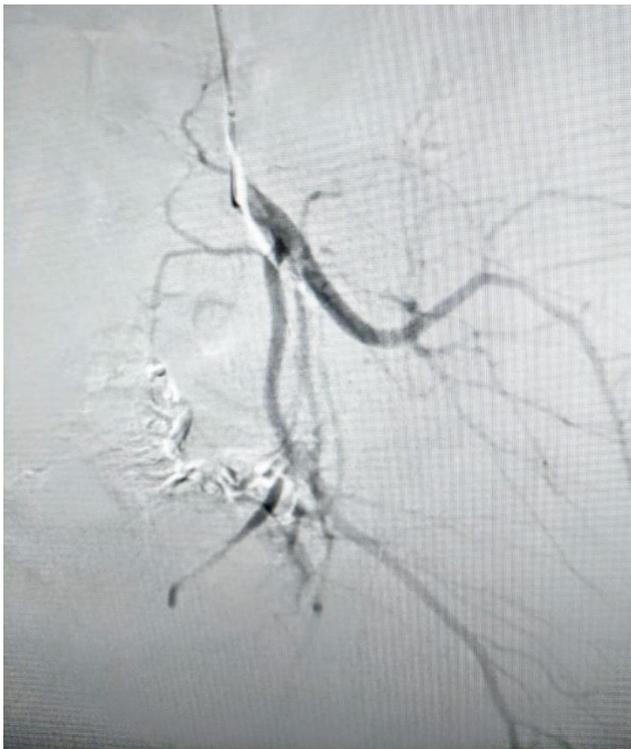


Figure 4: Post-embolization- No flow observed in the lesion

Patient stood the procedure well and her bleeding per vaginum resolved. She was followed up weekly with beta hCG till negative and transvaginal ultrasound monthly. After embolization, serum beta hCG reduced to 2006 IU/L, thus ruling out the possibility of invasive mole. Her beta hCG became negative in four months. In the fifth and sixth month of follow

up, patient had irregular cycles with normal flow and so an ultrasound was repeated was done 6 months after embolization, which revealed normal size uterus with endometrial thickness of 7.6mm and normal bilateral adnexa. She was prescribed cyclic progesterone therapy for 3 months and now patient is doing well.

Discussion:

AVM are thin walled vessels which can get disrupted either naturally or following instrumentation. In majority, uterine AVM presents after dilatation and curettage following abortion. But in few cases, it is seen even after spontaneous abortion [7] AVM can also be seen in association with gestational trophoblastic disease, gynaecological malignancy, uterine infections and rarely maternal exposure to diethylstilbestrol [8]. AVM may have varied presentations from undiagnosed to asymptomatic and even massive haemorrhage in nearly 12 % cases [9].

AVM can itself lead to miscarriages as can be suspected in our case. Hypoxic environment is necessary for the production of vascular endothelial growth factors, which in turn starts the process of angiogenesis and vasculogenesis. This helps in placental formation and thus growth of fetus. In cases of uterine AVM, this process is missing. There is excessive vascularization which produces an increased rate of oxygen supply in the fetus. This can be deleterious to the embryo and thus leading to pregnancy loss [10].

The diagnosis of AVM requires a high degree of suspicion. In the past, the diagnosis was an intraoperative diagnosis during laparotomy and later angiography came. Nowadays, transvaginal ultrasonography and color doppler has emerged as a simple and accessible diagnostic modality. The findings seen commonly are multidirectional flow that produces color mosaic pattern in the uterine myometrium [1]. Spectral analysis done shows high velocity arterial flow within the lesions with a low resistive index [11]. Peak systolic velocity (PSV) is calculated and can help in deciding the mode of treatment. In AVM with PSV less than 40 cm/sec, conservative management can be opted and in cases with PSV more than 80 cm/sec, definitive treatment is done. Magnetic resonance imaging or CT scan can add to the information on lesion size, location, vascularity and involvement of adjacent organs if any. In spite of all the diagnostic modalities available, the diagnosis of AVM can be challenging.

The type of treatment depends on the patient's clinical status, amount of vaginal bleeding and her desire for future pregnancy. Proper informed consent is a must. Asymptomatic cases can be managed expectantly with regular follow ups. Selective uterine artery embolization is an effective modality in symptomatic cases, which is a fertility sparing procedure as well [1]. It is done using gel foam, polyvinyl alcohol particles or glue. The success rate of arterial embolization is around 90% and have low complication rates, helps in avoidance of surgery or need of blood transfusion thus preserving fertility as well as reducing morbidity [12]. The last resort is hysterectomy in cases with significant haemorrhage causing hemodynamic instability, unavailability of facility of embolization, where conservative management has failed or patient's desire.

There are cases reported of uterine AVM with positive beta hCG. In a case reported by Su Mi Kim et al, patient presented with a suspicious hyper vascular uterine mass of around 8cm with beta hCG of 496 m IU/ml. Angiography was done and confirmed AVM. Bilateral uterine arteries and extra feeder vessels from internal iliac artery and inferior epigastric artery were embolized. Patient was followed up with repeated TVS and serum beta hCG. uterine mass disappeared in 3 months and beta hCG became negative in one month [13]. This is in contrary to our case, where the size of the mass was much smaller and serum beta hCG was high. There was possibility of gestational trophoblastic disease (GTD) owing to high levels of beta hCG, but angiography done prior to embolization confirmed AVM. Beta hCG done after 1 and 2 weeks were 2006 IU/L and 998 IU/L respectively, ruling out the possibility of GTD. It took 6 months and 4 months respectively to revert to normal TVS findings with

disappearance of uterine mass and negative non-pregnant beta hCG values. The persistence of beta hCG can be attributed to abnormal retention of trophoblasts with vascular invasion. In another case reported by Francisco Seller, patient presented with spontaneous miscarriage followed by intermittent spotting. TVS doppler revealed a vascular mass and serum beta hCG was 125 IU/ml. This patient was kept on conservative management, as bleeding stopped and beta hCG declined to non-pregnant values in 7 weeks post abortion. The TVS revealed normal findings with disappearance of mass in 8 months of follow up [14]. Our case was unique in more than one way, AVM after spontaneous abortion, being symptomatic and thus requiring embolization, high levels of serum beta hCG on presentation making the diagnosis difficult and delayed regression to normal findings.

Conclusion:

Uterine AVM should be a differential diagnosis in all cases presenting with abnormal bleeding after abortion even in the absence of instrumentation. Early identification and timely management of such patients help in combating serious complications. Uterine artery embolization is an effective means to reduce morbidity of the patient.

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