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A successful natural conception and childbirth after embolization of uterine arteriovenous malformation – A case report

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CASE STUDY

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ABSTRACT

Uterine AV malformation is dreadful condition due to vaginal bleeding. In past the only option was hysterectomy which leads to cessation of reproductive function. Here we are presenting a case where we preserved the reproductive function through Transarterial Embolization (TAE) technique. Post TAE patient resumed average flow regular menses, conceived naturally and delivered by elective caesarean section.

Key Words
Uterine AV malformation, embolization, natural conception

Implications for Practice:

1. What is known about this subject?
During treatment of uterine AV malformation in pregnancy are usually reproductive capacity is compromised.

2. What new information is offered in this case study?
We used a relatively novel method for treatment of uterine AV malformation and helped to preserve the reproductive ability of patient.

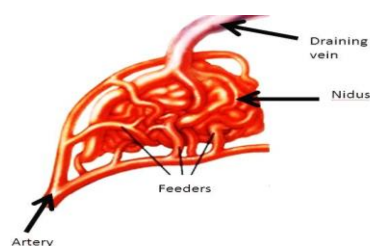
3. What are the implications for research, policy, or practice?
If this technique is used the many cases of iatrogenic sterility can be prevented by preserving reproductive ability of patients.

Background

Arterio-venous Malformation (AVM) is the communication between artery supplying and vein draining the organ. Due to high velocity of blood flow vascular fistula forms. In gynecological cases, there may be possibility of life threatening hemorrhage.[1,2] This may either be congenital or iatrogenic.[3,4]

In congenital, anomalous differentiation in primitive capillary plexus is usually multiple in numbers. Acquired AVMs are usually iatrogenic.[5-8] The true incidence rate remains unknown. As per literature this is a rare entity with different prevalence of 0.10 per cent, 4.5 per cent & 1–2 per cent.[9-11]

Figure 1: AV Malformation



AVMs as shown in Figure 1, are silent due to their slow growth before becoming symptomatic. Clinically symptoms are gradual or sudden ranging from abdominal pain, urinary symptoms, heavy uterine bleeding to congestive cardiac failure. Pelvic examination reveals poorly defined pulsatile adnexal mass or a soft and enlarged uterus transmitting pulsations. It is an important differential diagnosis in women of reproductive age with unexplained vaginal bleeding and in post-menopausal women for anechoic structures on ultrasonography.[3]

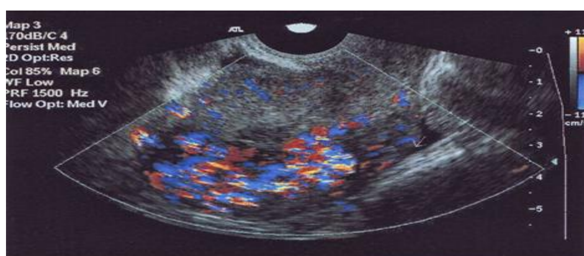
Since last few years trans-arterial embolization has become the treatment of choice. It is recommended as a technique prior to surgery for intractable haemorrhage due to its efficacy and safety.[12] TAE is quick procedure, even under local anaesthesia, can control obstetrical haemorrhages and preserve future fertility[13,14] Angiography is the gold standard for diagnosis and may serve therapeutically.[15] Here we describe a case of uterine AVM treated with embolization with the intent of pregnancy followed by natural conception and childbirth.

Case details

A 31-year-old female gravida 2, para 1, with 1 abortion 2 months back came to us with complain of bleeding per vagina. This had started after dilatation & curettage done after incomplete abortion 2 months ago. It did not respond to hormonal therapy. Patient was treated with ethamsylate and plasma expanders. Pelvic ultrasonography revealed multiple varying size hypoechoic lesions on posterior uterine wall. She had normal, regular periods previously. Family history was negative for any bleeding disorders. Her previous delivery was 2 years ago by emergency caesarean section.

At examination patient appeared severely anaemic with pulse rate of 100 per minute and blood pressure of 100/60 mm Hg. Per abdomen examination reveals a pfannenstiel scar, softness on palpation and no organomegaly. On speculum examination minimal bleeding was observed. Per vaginal examination revealed minimal active bleeding with closed os, normal sized uterus and clear fornices. At the time of admission her blood parameters were as follows Hb=4.8gm/dl, PCV=15 per cent, WBC=5500/Cmm, Platelets= 1,80,000/Cmm, PTT=50/30, Beta HCG=28.1mIU/L. An urgent transvaginal scan which is shown in Figure 2, was carried out to rule out ectopic but it showed tubular hypoechoic foci within the posterior myometrium with high vascularity measuring approximately 4 X 3 cm. This confirmed the diagnosis of uterine arteriovenous malformation.

Figure 2: AV malformation on ultrasound examination

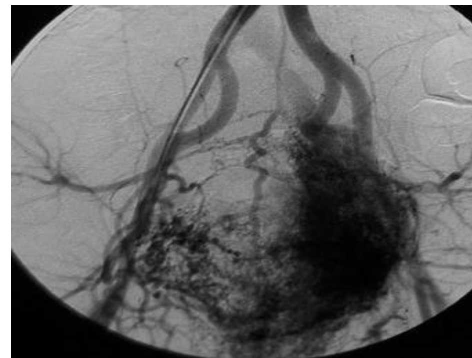


Doppler ultrasonography shows hypervascular lesion with multidirectional turbulent flow in the endometrium and myometrium of the posterior uterine wall

She was treated with 4 units of packed cells, 4 units of fresh frozen plasma (FFP), 1 unit of cryoprecipitate over 2 days. This was to correct her volume loss as well as to prevent disseminated intravascular coagulation which was suggested by abnormal PTT. Post transfusion her parameters were restored to Hb=9.5gm/dl, WBC=10,000/Cmm, Platelet=1,63,000/Cmm, PT=18, PTT=36 INR=1. She was taken up for superselective trans-arterial embolization (TAE) of uterine artery. The procedure is performed by interventional radiologist. This procedure was done under up-to-date C-arm fluoroscopic unit. It is equipped with road mapping and dose reduction mode. The necessary consent for the procedure was obtained from patient after explaining the pros and cons of the procedure in the language best understood by patient.

Venous access was established. Monitors for blood pressure, oxygen saturation, pulse, sedation score and respiratory rate were set. Resuscitation facilities were available along with competent support staff (Figure 3).

Figure 3: TAE for AV Malformation



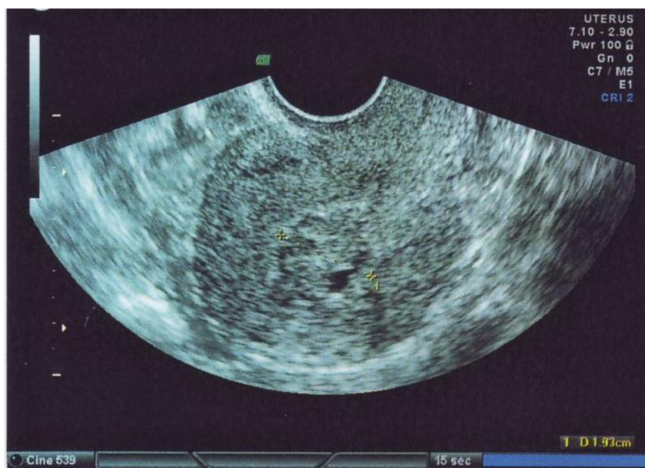
We used right sided per cutaneous femoral arterial access. The 5 French catheter manipulated under fluoroscopic guidance via anterior division of internal iliac artery. The polyvinyl particles are then injected once the main feeder is identified. The opposite uterine is reached and injected in the same manner.

Patient withstood procedure well. Post procedure her blood parameters continued to be within normal range. Patient experienced moderate pelvic pain for which oral nonsteroidal anti-inflammatory (NSAIDs) drugs were prescribed.

On day two of procedure repeat ultrasound examination Figure 4 showed a well-defined nonvascular lesion reduced to 1.9×1.1 cm with otherwise normally perfused posterior uterine wall.

There was no more vaginal bleeding during hospitalization. Hence AVM was successfully treated with superselective endovascular intervention. Thirty five days after procedure menstrual bleeding has started. Follow-up examination with ultrasonography shows no abnormal blood flow. Patient had natural conception two years later and delivered by full term elective caesarean section in view of previous caesarean section.

Figure 4: AV malformation after TAE



Doppler ultrasonography shows a well-defined nonvascular lesion on day 2 of the TAE procedure

Discussion

Uterine AVM is rare but the life threatening event due to massive blood loss that can go unrecognized. Medical literature also has very few reports of uterine AVMs. There is history of miscarriage, surgical abortion or D&C. Thus some way or the other most of them are related to pregnancy. Some are also reported to be related to infection, trophoblastic diseases and malignancy. [16, 17] Hormonal changes are also thought to be one of the causes of uterine AVMs as they can cause bleeding episodes especially in the age group 20-40.[18]

Clinically these AVMs presented in the childbearing age with mean age of presentation 30 years.[19] Nulliparous women without any history of uterine trauma due to surgical/gynecological procedures are rarely affected by uterine AVMs.[20] Most common presenting symptoms are per vaginal bleeding either intermittent or torrential and signs are menorrhagia and metrorrhagia. The cause of bleeding may be the different in pressure gradient across the arterial and venous system.[16] Due to acute and severe blood loss these patients may present with hypotension and/or anaemia. Some patients present with urinary symptoms like polyuria, incontinence or even dyspareunia. Congestive cardiac failure is also reported in some cases may be due to shunting the blood to venous system.[21]

In past diagnosis of AVMs was made pathologically after hysterectomy.[22] Recently various techniques are used including some minimally invasive ones. The easily available first line cost effective imaging modality is ultrasound; it shows a hypoechoic region and mild bulging of uterine wall and with colour Doppler it shows abnormal vascularity with high flow rates and low impedance flow on spectral Doppler. Identification of uterine high velocity blood flow with low impedance by Doppler ultrasound is highly suggestive for a uterine AVM[23] Subinvolution of the placental bed and adenomyosis in the setting of menorrhagia can mimic AVMs leading to overdiagnosis of Uterine AVMs. This can be differentiated by MRI scan. Better tissue contrast and delineation of surrounding pelvic organ involvement.[24] Computed tomography scans can be done but due to use of ionizing contrasts one should be cautious about its use.[25] A single direct fistulous communication to the venous structures may be identified. This finding is more common in acquired AVM than in congenital AVM, where contrast filling of a vascular plexus or nidus is more commonly seen.[22] Acquired AVM can be differentiated from retained products of conception as AVM primarily involves myometrium while the other one involves endometrium. Diagnosis of imaging studies is confirmed by maternal serum hCG which has slow decline in levels in retained products of conception than acquired AVM.[24]

The gold standard investigation is angiography that shows arterialized venous pouch of uterine parenchyma. Other one is hysteroscopy which provides examination under direct vision that shows bluish purple coloured tangled pulsating mass of irregular vessels. Recently, trans-vaginal ultrasound scanning (TVUS) has emerged as an efficient, simple and accessible diagnostic modality to detect and follow the vascular pattern of the AVM with the use of blood velocity and blood flow indices.[22,26]

Conclusion

Traditionally the treatment was hysterectomy for symptomatic patients. Recently medical as well as surgical treatments are available. The modality is chosen depending upon clinical status of the patient, age of the patient, site and size of the lesion, and future pregnancy aspirations. Conservative management is advised for patients with isolated episodes who are hemodynamically stable.[27]

For medical management estrogen, methylergonovine, danazol or prostaglandins have been tried.[13] Rational

behind this is to cover the bleeding vessels with proliferative endometrium. This is thought to be facilitated by estrogen therapy and methylergonovine reduce the blood flow to AVM and helps to collapse it. Conception and childbirth has been reported in the literature.[27,28]

Commonly used surgical techniques are ligation of feeding vessels, resection of the uterine lesion, and oversewing of the lesion. Sometimes coagulation of the AVMs is also done. Hysterectomy was done in past but at present it is reserved for those who do not wish or need to prevent their fertility.[29] TAE is the relatively recent modality of treatment. This is an interventional method preferred over others as it prevents the normal menstrual pattern and fertility as well.[30] In present case as patient was young and concerned of future fertility TAE was preferred. TAE is advantageous over others in that it helps in retaining fertility, can be done when patient is unfit for major exploration. Also dysfunctional uterine bleeding, intramural fibroids and adenomyosis are considered as indications for TAE.

Polyvinyl alcohol, cyanoacrylate, micro-fibrillar collagen & micro-coils are used as permanent particles while gelatin sponge is used as temporary. This decreases p/v bleeding in 80 per cent cases & uterine size in 40–70 per cent cases. Successful pregnancies are also reported but the risk is high in cases of previous uterine surgery due to chances of scar thinning & scar rupture especially in 3rd trimester. Success rate of TAE ranges from 79 per cent–90 per cent.[30] TAE is usually safe though some minor complications arise like hematoma, urinary tract infection, retention of urine, and vessel or nerve injury at the vascular puncture site are common and require only mild supportive care or careful observation.[31,32]

Overall adverse effects are extreme pain due to ischemic necrosis and vascular injury, post procedural infection, allergic reaction to dye, sciatica, acute arterial thrombosis of lower limb, coagulopathy and spasm of uterine artery can occur.

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CONFLICTS OF INTEREST

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PATIENT CONSENT

The authors, *Adhe A, Laliwala D*, declare that:

1. They have obtained written, informed consent for the publication of the details relating to the patient(s) in this report.
2. All possible steps have been taken to safeguard the identity of the patient(s).
3. This submission is compliant with the requirements of local research ethics committees.