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Case Report

Uterine arteriovenous malformation-beyond surgery: a case series

Uma A. Pankar*, Indusekhar S., Pannag Desai, Vidya Bhargavi

Department of Interventional Radiology, Healthcare Global Enterprises Ltd. Bangalore, Karnataka, India

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***Correspondence:**
Dr. Uma A. Pankar,
E-mail: pnk.uma86@gmail.com

ABSTRACT

Uterine arteriovenous malformations (AVM) are a rare entity, presenting in women of reproductive age. The presentation may vary with bleeding being the chief presenting symptom. Though traditionally hysterectomy has been a definitive form of treatment, trans-catheter embolization is being considered as a good treatment option. We present three cases with uterine AVM who presented with heavy bleeding per vaginam. Three patients of reproductive age group (29-33 years) presented with heavy bleeding per vaginam from January 2017- May 2017. Two had previous miscarriages for which curettage was done and one had undergone a Caesarean section previously. Diagnosis of uterine AVM was made by ultrasound-grey scale, colour Doppler, followed by MRI. All patients underwent trans-catheter angiogram followed by selective embolization of the AVM with n-BCA (n- Butyl Cyanoacrylate) and polyvinyl alcohol (PVA) particles. Successful embolization was done in all of our patients with technical success rate of 100%. On follow-up, all patients are currently asymptomatic. Trans-catheter arterial embolization of uterine AVM is a simple and effective treatment alternative with less morbidity associated with anaesthesia and surgery in turn reducing hospital stay. It has to be considered in patients who need to preserve their fertility.

Keywords: Embolization, Trans-catheter angiogram, Uterine AVM

INTRODUCTION

Uterine arteriovenous malformations are abnormal communications between uterine arterial branches and myometrial venous plexus. Enhanced myometrial vascularity is the recent terminology being used for Uterine AVMs.[1] It is a rare entity, presenting in women of reproductive age and may be congenital or acquired.[2,3] Acquired uterine AVMs can develop following unsuccessful pregnancies, surgery or instrumentation viz dilatation and curettage, termination of pregnancy, caesarean section, endometrial carcinoma and gestational trophoblastic disease.[4]

The congenital and acquired AVMs may be indistinguishable radiologically and therefore obtaining good patient history is of utmost importance to distinguish the two.

The symptoms may vary with bleeding being the chief presenting symptom in majority of the cases.[5] Life threatening haemorrhage can result from trauma, surgical intervention or pre-existing pathological uterine process. Clinical presentation of Uterine AVMs in a pregnant patient or postpartum period may overlap with retained products of conception, postpartum endometritis and gestational trophoblastic disease.[6]

Grey scale USG, Colour and spectral Doppler and contrast enhanced MRI are used for diagnosis. Though traditionally hysterectomy has been a definitive form of treatment, trans-catheter embolization is being considered

as a good treatment option.[7] We present three cases of uterine arterio- venous malformation that presented with heavy bleeding per vaginam and underwent trans arterial embolization for the same.

CASE REPORT

We present three patients of reproductive age group with uterine AVM. The diagnosis was made by ultrasound-grey scale, colour Doppler, followed by MRI.

Following pre procedural workup (coagulation profile and serum creatinine) and informed consent all the patients underwent angiogram followed by selective embolization of the AVM with N-butyl cyanoacrylate (nBCA) and polyvinyl alcohol (PVA) particles after thorough discussion of each case with the Obstetricians. All the procedures were done under local anesthesia. The left common femoral artery was accessed using 5 F sheath (Radifocus, Terumo Corporation, Tokyo Japan). A 5 F Uterine Artery catheter (Merit medical systems Inc., Utah, USA) and 0.032" glide wire (Radifocus Terumo Corporation, Tokyo Japan) combination was used to perform internal iliac artery angiogram on either side which demonstrated hypertrophied uterine artery branches and early filling of draining veins (dominant supply from the right). Right uterine artery was accessed using the same catheter and arterial feeders were embolised using n-BCA (N-butyl cyanoacrylate, Histoacryl; B. Braun Surgical, Rubi, Spain). The n-BCA was mixed with lipiodol (Geurbet, USA) in a ratio of 1:2-1:3 and was delivered in the nidus of the AVM. The glue was injected in small aliquots in combination with 25% dextrose using a sandwich technique with the help of 3 way stopcock attached to the catheter hub. (Dextrose-glue-dextrose). A total of 2-3 ml of the mixture was injected, which formed casts within the network arteries. This was followed by embolization using 500-710 um PVA (Merit medical systems Inc., Utah, USA) particles until persistent capillary filling stasis or maximum reduction of flow was noted in the AVM. Similarly, the left uterine artery was accessed and capillary network of the AVM was embolised using 500-710 um PVA particles till stasis was achieved. Catheter and vascular sheath were removed and hemostasis was achieved at the puncture site.

The patients were discharged 24-48 hours after the procedure with antibiotics, anti-inflammatory and analgesic drugs.

Case 1

29 years old, P2L2 woman presented with urinary incontinence and menorrhagia for a duration of 3 months. She had undergone a caesarean section followed by tubectomy two months prior to onset of symptoms. No excessive intrapartum haemorrhage was encountered in either of her prior pregnancies.

Trans abdominal USG demonstrated bulky uterus with increased vascularity and high flow vessels on colour Doppler. Further evaluation with contrast enhanced MRI showed focal heterogeneous signal intensity myometrial mass in the antero-inferior portion of the uterus with multiple serpiginous flow voids (Figure 1 a, b, c). A diagnosis of uterine AVM was made and embolization of the AVM was planned.

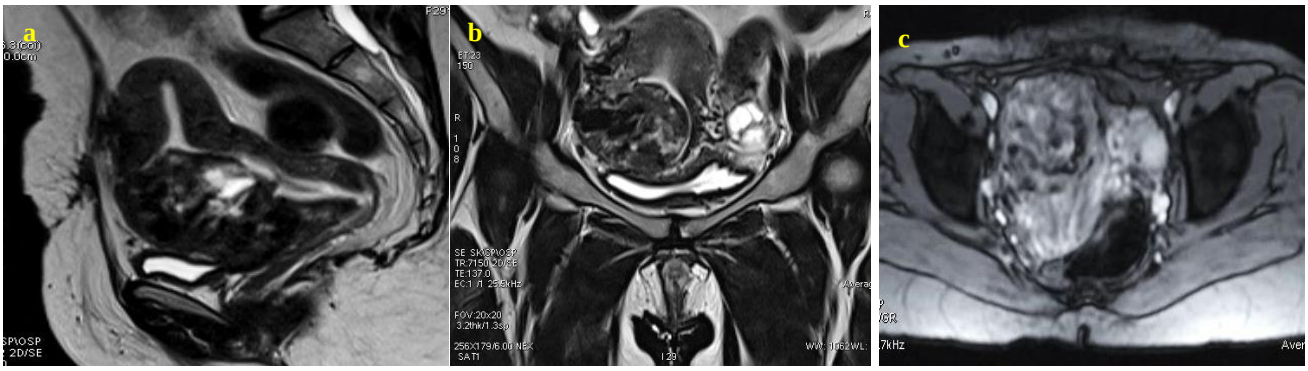

Figure 1: T2 weighted sagittal (a) and coronal (b) images demonstrating heterogeneous signal intensity mass with flow voids in the antero-inferior wall of uterus causing distortion of endometrial cavity with multiple foci of blooming on axial T2* images (c).

Pelvic angiogram demonstrated hypertrophied bilateral uterine arteries supplying a large AVM in the uterus and multiple smaller feeding vessels in the nidus. Early venous drainage was noted into the internal iliac veins. (Figure 2 a, b) Indentation on the right lateral wall of the urinary bladder was noted by the mass of entangled vessels. The patient underwent embolization with glue and nBCA particles for the same (Figure 2 c, d ,e, f).

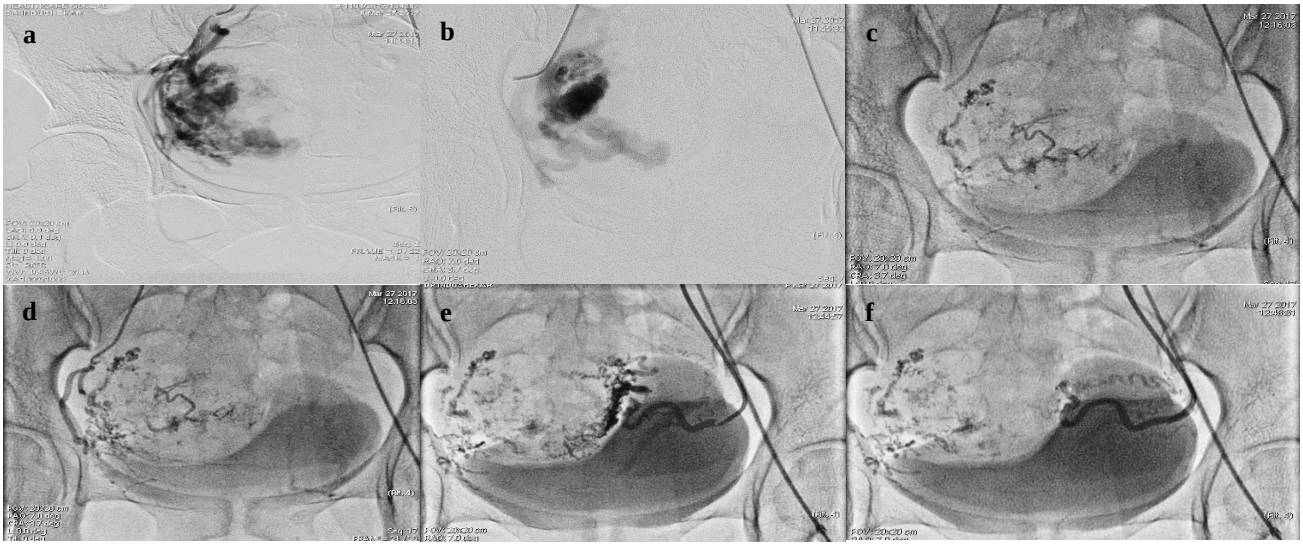

Figure 2: (a) Selective right internal artery angiogram demonstrates large uterine AVM with multiple small feeding arteries with early filling into draining veins (b), (c) DSA image post nBCA injection demonstrating glue cast within the vessel bed (d) Post embolization image showing reduction of flow in the AVM. Note the indentation on urinary bladder by the AVM, (e) Selective angiogram through left uterine artery demonstrating feeder arterial branches to the AVM (f) Post embolisation angiogram showing reduction of flow in the AVM with glue cast within the vessels.

The patient presented with recurrence of symptoms one month post procedure and underwent repeat embolization. No significant flow was noted in the AVM through right uterine branches. (Figure 3 a) Left uterine artery angiogram using 5 F Robertson Uterine Curve Catheter demonstrated tortuous vessels supplying the AVM arising from left uterine artery, draining into a large venous channel. Embolization was done with cyanoacrylate-glue which formed casts within the network of arteries arising from left uterine artery. Significant reduction of flow in the AVM was noted post embolization (Figure 3b, Figure 4).

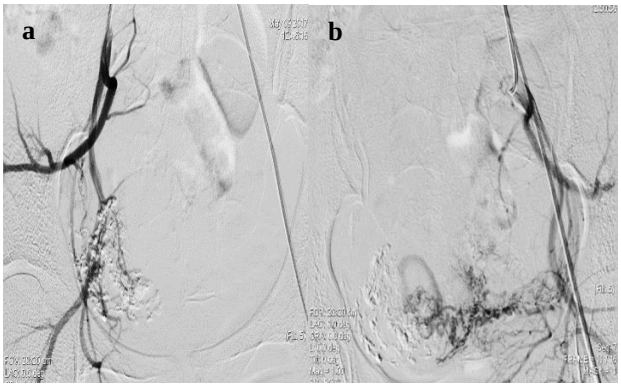

Figure 3: (a) Repeat angiogram six weeks later through right internal iliac artery demonstrating no flow in the AVM. (b) Selective angiogram through left internal iliac artery demonstrating tortuous vessel arising from left uterine artery branches supplying the AVM with early filling of the draining vein.

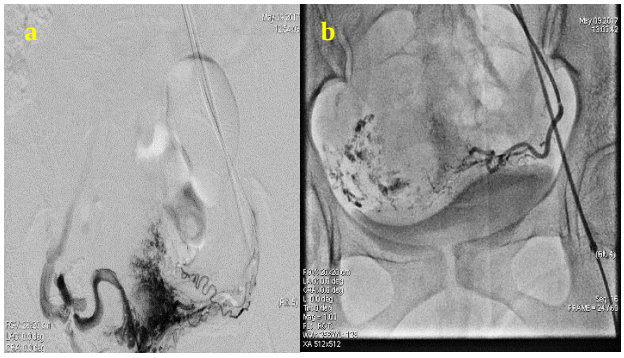

Figure 4: (a) Selective angiogram through left internal iliac artery demonstrating tortuous vessel arising from left uterine artery branches, tangle of capillary network and draining veins. (b) Post embolization angiogram demonstrating marked reduction of flow in AVM with glue cast within the vessels.

Case 2

33 year old patient, P1L1A1, underwent Caesarean section for her first pregnancy. Dilatation and curettage was done for the second pregnancy resulting in heavy bleeding which was managed medically. Medical termination of pregnancy was advised in December 2016 for the third pregnancy as the foetus was in the lower uterine segment. Dilatation and curettage was done which resulted in heavy bleeding. The patient was investigated further with Transvaginal USG and colour Doppler. Bulky uterus with increased vascularity predominantly in the posterolateral wall was noted. MRI demonstrated 6.2x5.2x5 cm lesion with prominent flow voids in right

posterolateral wall of uterine body- indenting and displacing endometrium.

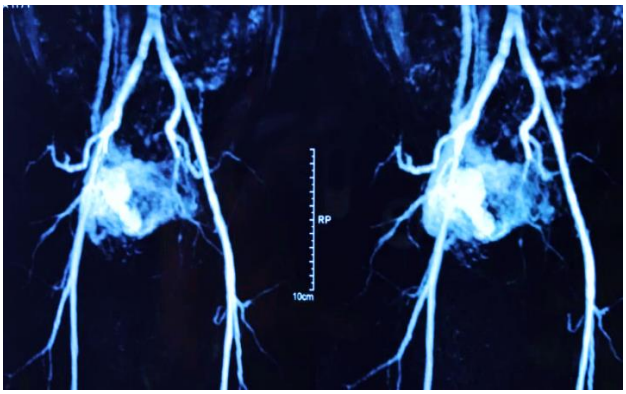

Figure 5: MR angiogram - tangle of complex serpentine abnormal vessels with supply from uterine artery branches from both sides and early venous return representing the AVM.

Rapid arterial phase contrast uptake from right internal iliac artery branches and early venous drainage in right internal iliac vein was also noted (Figure 5). Diagnosis of high flow arteriovenous malformation was done.

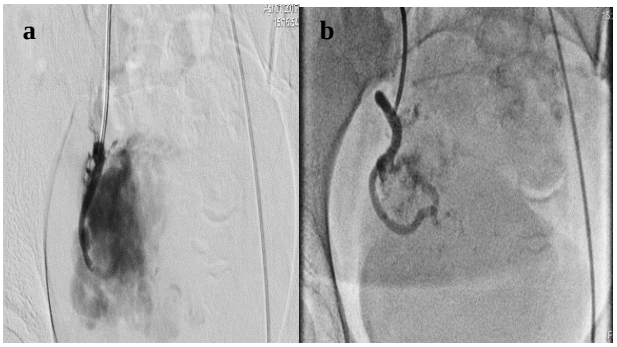

Figure 6: Selective angiogram through right uterine artery (a) demonstrates a large AVM with a nidus. (b) Post embolisation angiogram showing significant reduction of flow in the AVM.

Pelvic angiogram showed hypertrophied bilateral uterine arteries supplying a large AVM with cluster of entangled feeding vessels demonstrating early venous drainage into the internal iliac veins. The patient underwent embolization with glue and PVA particles for the same (Figure 6,7).

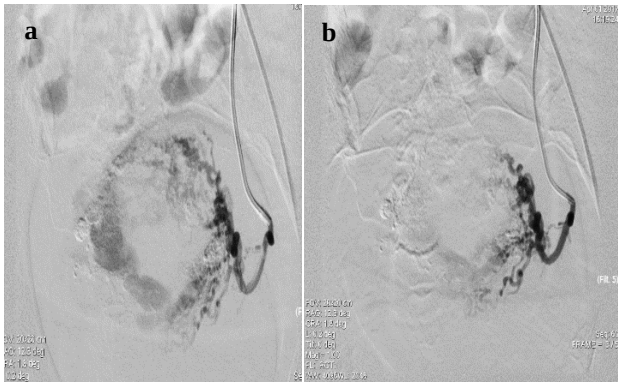

Figure 7: (a) Selective angiogram through left uterine artery demonstrating feeder arteries to the AVM from left uterine artery. (b) Post embolization angiogram shows reduction of flow in AVM with glue cast within the vessels.

Case 3

24 year old G4A2E1 had spontaneous abortion in first pregnancy which was managed medically.

The patient had ruptured ectopic pregnancy the second time and underwent laparotomy for the same.

She had excessive bleeding during first trimester of third pregnancy. Missed abortion was diagnosed and patient underwent suction evacuation for the same. A month later she came with fourth pregnancy. Beta HCG was high (1527 IU/ml) and she was evaluated further with imaging.

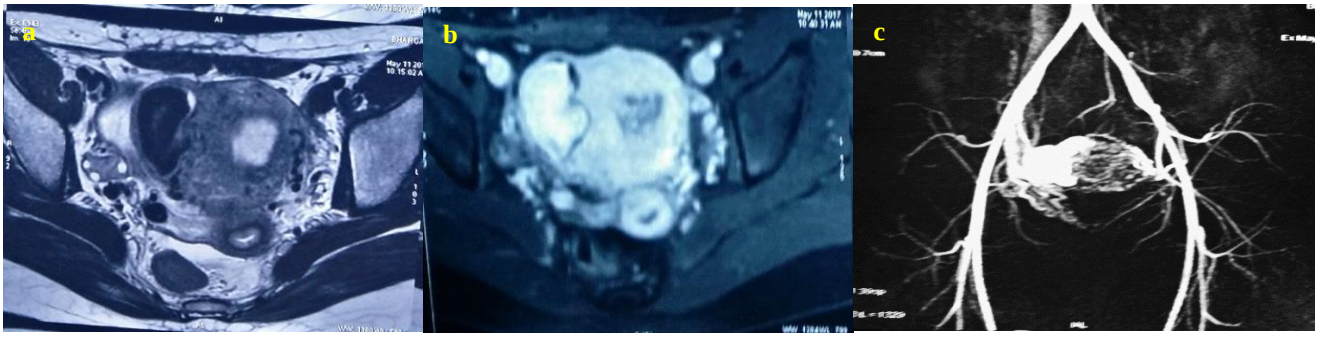

Figure 8: (a) T2 weighted axial MRI demonstarting a large signal void on the right lateral wall of uterus (b) showing intense enhancement with contrast on axial T1W fatstat contrast enhanced image (c) MR angiogram demonstrating hypertrophied uterine artery branches communicating with large venous lake.

USG demonstrated mildly bulky uterus thick echogenic endometrium. Hypoechoic lesion/area with multiple vascular channels with cystic spaces showing internal vascularity in the right lateral wall of uterus suggestive of AVM was noted.

MRI showed multiple tortuous flow voids predominantly on the right lateral wall of the uterus with intense contrast enhancement of the flow voids on post contrast images (Figure 8).

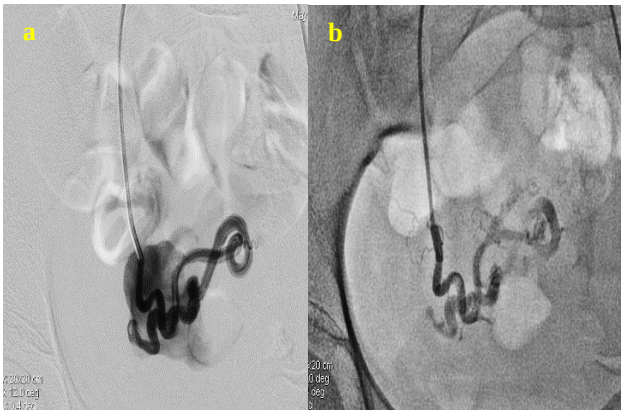

Figure 9: (a) Right uterine artery angiogram shows hypertrophied, tortuous right uterine artery branch draining into a large vascular lake. (b) Post embolisation angiogram shows significant reduction of flow in AVM with absent filling of the vascular lake.

Pelvic angiogram demonstrated a large AVM in the uterus supplied by both uterine arteries. A large vascular lake was noted which was supplied by multiple smaller feeding vessels from hypertrophied uterine arterial branches. Early venous drainage was noted into the internal iliac veins. The patient underwent embolization with glue and PVA particles for the same (Figure 9, 10).

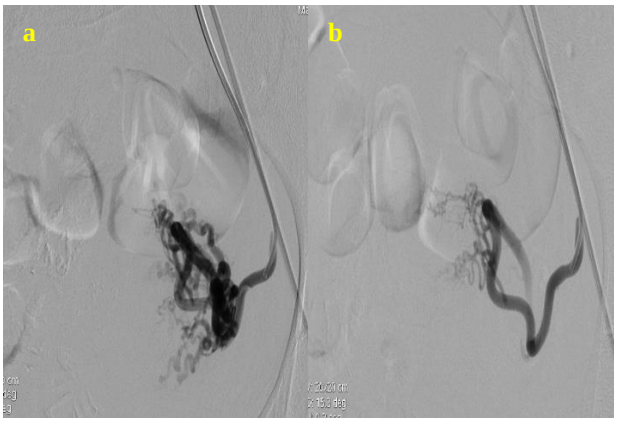

Figure 10: (a) Left uterine artery angiogram multiple tortuous feeder arteries from uterine artery (b) Post embolisation angiogram shows significant reduction of flow through feeder arteries.

DISCUSSION

Uterine vascular malformations are rare gynaecological entities representing around 2 % of all causes of genital and intraperitoneal haemorrhages.[5]

However, in view of chances of profuse and potentially life threatening haemorrhage; it cannot be ignored and needs to be listed as an important differential diagnosis in women of reproductive age with unexplained vaginal bleeding and menorrhagia.

Though the exact incidence is not known, more than 100 cases of uterine AVM have been reported in literature after the first case being reported by Dubreuil and Loubat in 1926.[2,8]

Uterine AVMs can develop congenitally or may be acquired.[2,3] They are basically a proliferation of vascular channels of varying sizes with fistula formation along with tangle of capillary like vessels.

Congenital UAVMs arise because of hampered vascular embryological development. A central nidus with multiple feeding arteries and draining veins is characteristically noted.[9] Congenital AVMs are more likely to extend beyond the confines of the uterus with supply from extra uterine pelvic arterial branches.[8]

Acquired AVMs are more common of the two. They are abnormal communications between intramural arterial branches and myometrial venous plexus and thus essentially represent multiple tiny arteriovenous fistulas. [9] The acquired AVMs can develop following surgery or instrumentation viz dilatation and curettage, termination of pregnancy and caesarean section. As the healing progresses post trauma; there is likelihood of developing abnormal communication between the arteries and veins.[4]

Gestational trophoblastic disease (GTD), retained products of conception, post-partum endometritis and several gynaecological malignancies have clinical and imaging features which may overlap with acquired arteriovenous malformations.

In our series of three cases, though presence of congenital form of AVM cannot be completely ruled out, it seems less likely as the patients did not present at earlier stage of life. Also, the patients having undergone a caesarean section or uterine instrumentation previously without any complications of bleeding nearly rules out the possibility of pre-existence of congenital AVM. Absence of associated pelvic AVM and pelvic (extra uterine) arterial supply is yet another feature pointing towards the acquired nature of the AVM.

Acquired Uterine AVMs generally present in the women of reproductive age group as seen in all three of our patients with most common presenting complaint being menorrhagia or metrorrhagia. Pressure symptoms

resulting in urinary incontinence was also noted in one of our patient. A large uterine AVM can result in vascular steal ultimately leading to congestive heart failure.[10]

Clinical Examination is usually unremarkable. Very rarely a palpable mass may be noted at vaginal examination.[11]

Diagnosis is usually made with USG with colour Doppler followed by contrast enhanced MRI with angiography. USG appearance of uterine AVM is non- specific ranging from uterine, endometrial or cervical mass, myometrial heterogeneity or tubular serpiginous spaces within the myometrium. Colour Doppler will demonstrate aliasing due to high velocity multidirectional flow with high peak systolic velocities and low resistive indices pointing towards arteriovenous shunting.[12]

On MRI it appears as heterogeneous signal intensity mass lesion seen intramuraly, in the parametrium and within endometrial cavity with multiple dilated tortuous flow voids. Post contrast there is intense enhancement of the serpentine vessels with early venous return.[13]

Gestational trophoblastic disease can co-exist and/or present with similar clinical and imaging appearance, which necessitates serial monitoring of Beta HCG levels as was noted in one of our patients.

However, gold standard for diagnosis of uterine AVM is a Digital Subtraction Angiogram. Hypertrophied uterine arteries with a large mass of arterial accessory feeding vessels and early draining hypertrophied veins are characteristic of AVM.[4] A supply from extra uterine arterial branches may also be encountered.[8]

The various catheters which can be used for the purpose of embolization of the AVM are uterine artery catheter, RUC catheter, cobra catheter, renal curve catheter and co-axial microcathters. The embolization materials used are polyvinyl alcohol particles, nBCA and coils. The basis of selection of embolic material depends on the anatomic appearance of the vessels with rate of blood flow and expertise of the operator.[8]

Major complications of the procedure include non-target embolization resulting in end organ damage, (viz pulmonary embolism), uterine necrosis, abscess formation and ovarian failure.

The ovarian failure may be attributed to the inadvertent flow of embolic material into the ovarian artery especially in cases where small sized PVA particles are used (< 500 microns)

Minor complications like persistent pain, puncture site hematoma and puncture site AV fistulas may be seen.[8,14]

The severity of hemorrhage in patients with uterine AVM determines the choice of management. The hemodynamic status of the patient at time of presentation essentially governs the timing of the procedure (emergency or elective). The other factors which should be taken into consideration are patient's age and the need to preserve future fertility. [8]

In present case series successful embolization was done on elective basis in all patients with technical success rate of 100%. One patient required repeat embolization one and half month later in view of recurrence of symptoms. All patients are currently asymptomatic on follow-up of four to six months.

CONCLUSION

Uterine AVMS can be often missed as the presentation may overlap with GTD, postpartum haemorrhage or retained products of conception. It should be considered as an important differential diagnosis in any patient of reproductive age group presenting with excessive bleeding post abortion, caesarean section and uterine instrumentation. Trans-catheter arterial embolization of uterine AVM is a simple and effective treatment alternative with less morbidity associated with anaesthesia and surgery in turn reducing hospital stay. It has to be considered in patients who need to preserve their fertility.

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