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Original research article

Uterine arteriovenous malformations: A rare cause of abnormal uterine bleeding-case series

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Abstract

Introduction: Uterine arteriovenous malformations is a rare but possible cause of abnormal uterine bleeding, which commonly presents with vaginal bleeding that can be profuse enough to cause hemodynamic instability. It is considered as one differential in women of reproductive age presenting with unexplained vaginal bleeding, when the ultrasound reveals anechoic structures with low resistance blood flow.

Case series: In our institution, during the study period, five cases of symptomatic uterine arteriovenous malformations have been reported. All were in the reproductive age group ranging from 24-29 years, four cases presented with a history of miscarriage for which curettage was done and one presented following LSCS. They presented with recurrent bouts of torrential bleeding, not controllable by regular measures. The time interval between the onset of symptoms and the primary event. D&E or LSCS ranged from one to four months. Diagnosis was done by pelvic ultrasound along with color Doppler. Gestational trophoblastic disease and retained products of conception were ruled out. All of them underwent selective uterine arterial embolization by interventional radiologist and got relieved of their abnormal uterine bleeding

Conclusion: Uterine AV malformation should be considered as a differential diagnosis in all cases presenting with bleeding after miscarriage or curettage, since diagnosis is simple and treatment by selective arterial embolization saves morbidity of surgery and anesthesia and also preserves the fertility.

Keywords: Uterine arteriovenous malformations, uterine artery embolization, angiography, hemorrhage

Introduction

Uterine vascular abnormality is a rare but potentially life-threatening condition, which commonly presents with vaginal bleeding that can be profuse and may lead to hemodynamic instability. It is considered as one differential in women of reproductive age presenting with unexplained vaginal bleeding, when the ultrasound reveals anechoic structures with low resistance blood flow [1].

The first case was reported in 1926, by Dubreil and Loubart as a cricoid aneurism of the uterus, since then, it had been referred as an arteriovenous aneurysm or arteriovenous malformations [2]. The true incidence is unknown, but with the increasing use of ultrasound to evaluate abnormal vaginal bleeding, Obrien *et al*. proposed a rough, predicted incidence of 4.5% of all genital and intraperitoneal hemorrhages [3].

AVM can be congenital or acquired [4]. Acquired uterine arteriovenous malformations occur mostly after damage to uterine tissue and are associated with conditions, such as pregnancy, uterine surgical procedure (caesarean section, curettage) [5, 6] and less commonly, cervical or endometrial carcinoma, infection, gestational trophoblastic disease and exposure to diethylstilbestrol [7]. Arteriovenous malformations consist of abnormal growths and connections between arteries and veins without a capillary bed, which are fragile and prone to bleed [8]. Congenital arteriovenous malformations are rare and occur due to an abnormality in the embryological development of primitive vascular structures, resulting in multiple abnormal communications between arteries and veins that invade the surrounding structures while the acquired results from trauma thus remain confined within the myometrium or endometrium, showing direct communication between the intramural branches of the uterine arteries and myometrial veins [9]. This is the basic difference between congenital and acquired forms.

The common presentation of arteriovenous malformation is usually in women of reproductive age, with a previous history of uterine surgery or procedures and vaginal bleeding, which may be intermittent or abundant, leading to hemodynamic instability. Sometimes, pelvic pain is associated with vaginal bleeding [8]. Clinical examination can be unremarkable or with heavy vaginal bleeding.

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Pelvic ultrasonography may reveal subtle myometrial heterogeneities or a mass with multiple hypo/anechoic tubular structures of multiple sizes [3]; Focal endometrial and myometrial thickenings can also be diagnostic. Color Doppler shows a tangle of vessels with high velocity flow; spectral Doppler shows high-velocity, low-resistance flow with RI (resistance index) values ranging from 0.25 to 0.55 and peak systolic velocity (PSV) values in the range of 40-100 cm/s [10]. Timmermann *et al*. [11] stated that conservative management is effective patients with a Sonographic diagnosed AVM, particularly when PSV is < 40 cm/s; in cases where PSV is >80 cm/s and early venous filling was demonstrated on angiography, definitive treatment was necessary.

The typical MRI finding of AVMs are the presence of serpiginous and dilated vessels within the myometrium or parametrium [8]. Angiography, being the gold standard technique for diagnosing AVM, shows enlarged, dilated irregular vascular spaces supplied by enlarged uterine arteries with high-flow dynamics [9]. Hysteroscopy imaging of uterine AVM has been described as normal appearing endometrium [12], pulsatile vessels bulging of the endometrial cavity [13, 14] and more frequently as highly vascular masses indistinguishable from retained products of conception [15, 16, 17].

Two conditions, i.e., retained products of conception and gestational trophoblastic disease mimic AVMs clinically; both should be ruled out before making the final diagnosis. Serum beta-HCG is recommended to exclude a gestational trophoblastic disease.

The treatment of uterine arteriovenous malformation mainly depends on the patient's age, symptoms, desire for fertility, and site of uterine arteriovenous malformation lesion.

The definitive treatment of arteriovenous malformation consists of hysterectomy whereas conservative treatment is selective uterine arterial embolization [17, 18, 19]. Uterine artery embolization is preferred treatment modality in women of reproductive age who want to conserve future fertility [17]. Common agents used for embolization include gel foam, [3, 18] PVA, and glue (N butyl cyanoacrylate) [22]. Ghai *et al*. report a technical success rate of 100%, and clinical success rate of 93% in their series [21]. Complications which are reported include pelvic pain, local hematoma, and rarely, skin sloughing. Pregnancies have been reported following embolization [18, 22], proving that an adequate collateral supply can develop to support a full-term pregnancy. Peitsidis *et al*. report a 27% pregnancy rate following bilateral UAE [22].

Another recently reported approach is the hysteroscopic resection of the uterine lesion by Stefano *et al*., from Italy [23], whereby 11 cases of arteriovenous malformations were managed successfully by operative hysteroscopy and four of them achieved a pregnancy that carried to term. This novel approach can be a breakthrough in the management of uterine arteriovenous malformations, as the author reported a 100% success rate with no surgical complications, high fertility outcomes, and short hospital stay. Further studies are required to declare it as the standard treatment.

Other conservative options i.e., medical treatment with combined oral contraceptive pills and progestogens is reported in some case reports for asymptomatic patients or with mild hemorrhage [24].

Aims and Objective

To study the clinical presentation, diagnostic, and treatment options in a case series of five patients with symptomatic uterine AV malformations, who presented to the Gynaecology OPD of our hospital from January 2021 to November 2022.

Material and Method

This is a retrospective study of cases of uterine AVM managed at our institution Mahatma Gandhi University of Medical Sciences and Technology from January 2021 to November 2022. Patients were referred to our institute for evaluation of menorrhagia or metrorrhagia not responding to D&E and medical management.

After history taking and general physical and local examination transabdominal and pelvic ultrasonography was performed looking for the of size of the uterus, the appearance of the myometrium, endometrium and the uterine cavity. AVM is characterized ultrasonographic as a mass of multiple cystic or tubular hypoechoic areas in the myometrial or in the endometrium or at the endo-myometrial junctions. Addition of color Doppler demonstrates a tangle of vessels showing a characteristic mosaic pattern and vessels with high-flow velocities and low resistance index. Resistance index values in case of AVMs range from 0.25 to 0.55 and peak systolic velocity (PSV) values lie in the range of 40-100 cm/s [10].

Patients age, parity, presenting symptoms and its severity, preceding event, elapsed time to the occurrence of the obstetric event, ultrasonographic and doppler findings were recorded. Gestational trophoblastic disease and retained products of conception were ruled out by doing serum beta human chorionic gonadotropin level < 5 mIU/mL and absence of retained products of conception confirmed by ultrasonography.

After above workup and confirming the diagnosis of uterine arteriovenous malformation, all five patients were taken for uterine artery embolization after taking consent.

The embolization procedures in our hospital were performed by interventional radiologist. Through femoral artery access, pre-procedure angiography was done for confirming the presence of AVM. Gel foam was the agent used in all the five cases. Cessation of flow was confirmed at the end of the procedure.

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All 5 patients were taken up for embolization after obtaining informed consent. All the patients had a follow-up sonogram at 1 month after the UAE, to evaluate the healing process and the resolution of the uterine lesion and its vascularity, any extra treatment needed after UAE for recurrence of symptoms was also noted.

Case 1: 28-year-old female P2L2A1, both FTND, presented to gynae OPD with complaints of bleeding per vagina off and on for 3 months, bleeding was associated with passage of clots. She had undergone D & E three months back for an incomplete abortion. Ultrasonography showed 30×25 mm hetero echoic and hyper vascular area in myometrium, with RI 0.30 and PSV 80 cm/second. She was referred to higher center in view of hypervascularity of products of conception. Meanwhile she had 3-4 episodes of profuse bleeding which were self-limiting.
On examination-pallor was present, PR-92/min, BP-112/70 mmHg. Abdomen was soft. On per speculum examination cervix and vagina were normal, no local lesion seen, on per-vaginal examination-uterus was normal in size, retroverted and retroflexed, soft mobile and non-tender. Beta HCG <2 MIU/ml. Transvaginal sonography with color Doppler showed a large vascular malformation (Fig. 1). Angiogram showed multiple feeder vessels from uterine artery supplying the AVM (Fig. 2). She was taken for embolization after informed consent. The procedure was uneventful. After embolization the patient got symptomatic relief (Fig. 3).

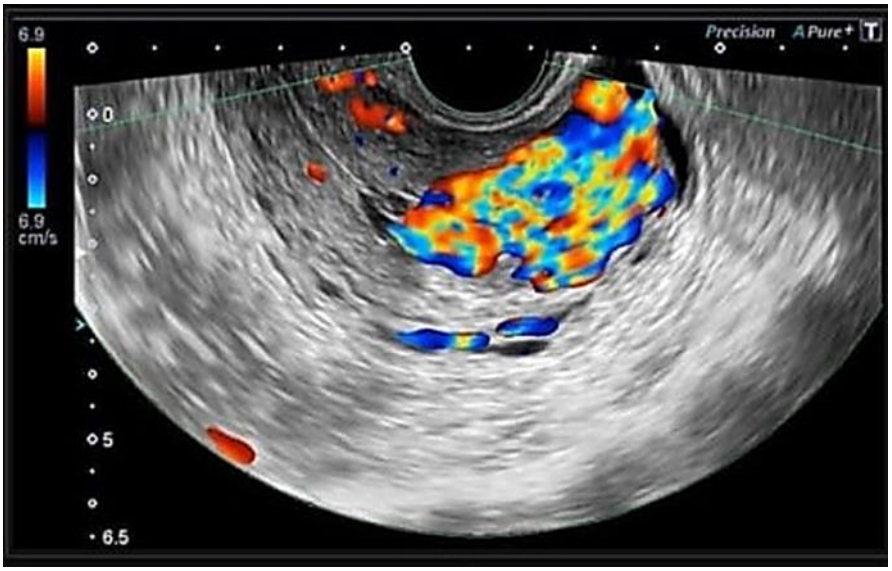

Fig 1: Pelvic ultrasound showing large vascular malformation

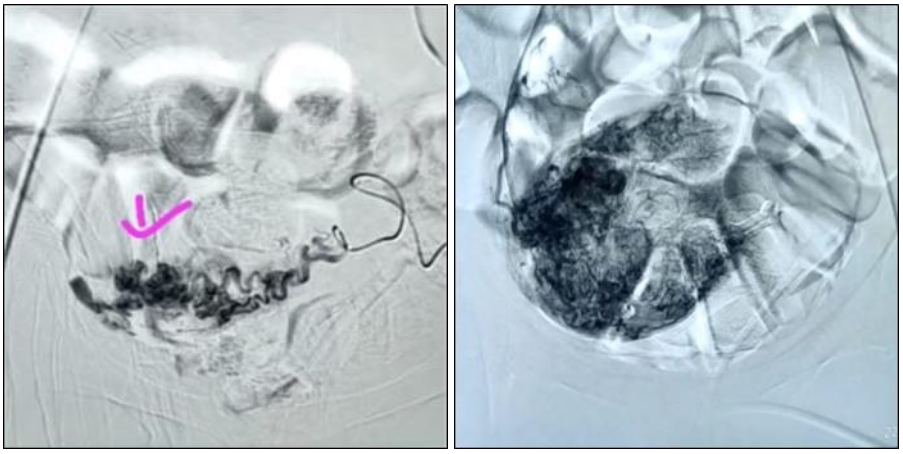

Fig 2: Multiple feeders supplying to AVM/AVF from both uterine arteries

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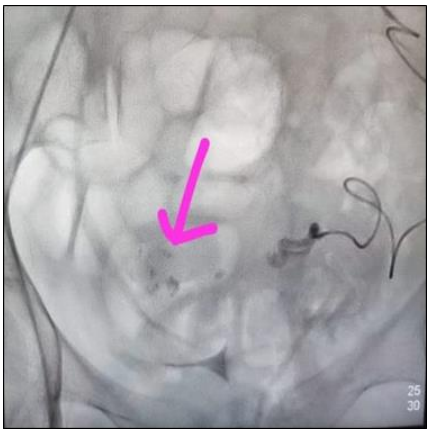

Fig 3: Post embolization angiogram showing no flow

Case 2: -24 year old P2L2 previous LSCS one month back, presented with 2 episodes of heavy bleeding per vaginum 3 week apart. First episode was on seventh post op day for which she has undergone D & E where no RPOCS were found and only clots were removed from uterine cavity she also received 6 units of PRBCS, 2 weeks after this episode on post op 25 she again had second episode of heavy bleeding and presented to our hospital.
On examination patient was vitally stable, afebrile to touch, PR 107/min, BP 128/88 mm Hg, RR 18/min. on P/A-abdomen was soft. On Per speculum examination-approximately 50 cc of clots were present in vagina but no active bleeding. On per vaginal examination Os was patulous, uterus was approximately 10 weeks.

On investigation: Her Haemoglobin was 9 gram/dl. Her beta Hcg was-<5 m IU/ml, on transvaginal sonography-uterus was 59×45×40 mm, ET -3.7 mm. It also showed a heterogeneous and highly vascular area measuring 9×9 mms in the left lateral wall of lower uterine segment, involving endometrium and adjacent myometrium, with low RI value being 0.27 and PSV being 78 cm/second suggestive of AVM. On the basis of these findings and clinical presentation of the patient, she was posted for uterine artery embolization and stood the procedure well.

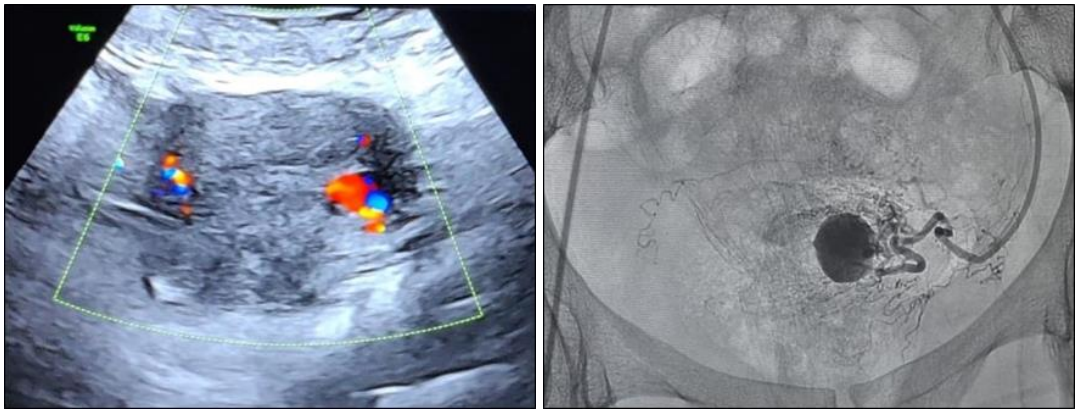

Fig 4: Pre embolization ultrasonography and angiogram showing AVM

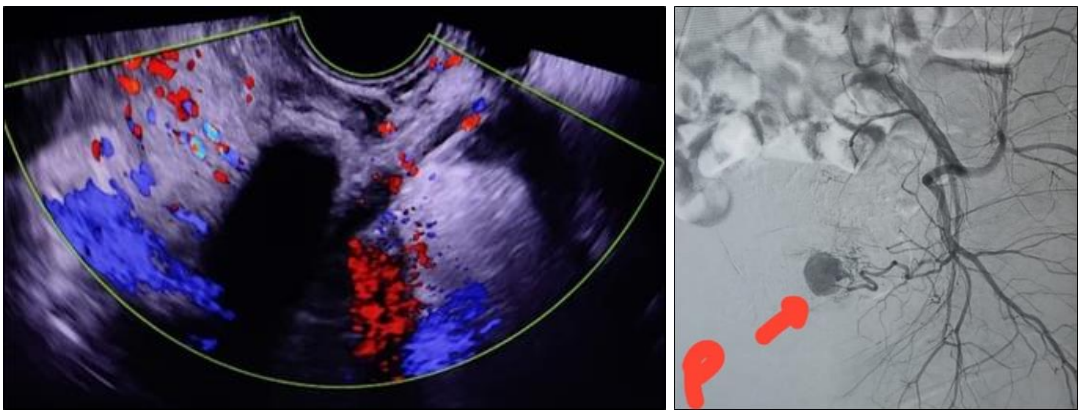

Fig 5: Post procedure angiogram and pelvic ultrasound showing absent flow in the AVM

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Case 3: -29-year-old P2L2A1, previous 2 full term vaginal deliveries, 7.5 years and 2.5 years back respectively. She has undergone second trimester abortion at 4 months of pregnancy in view of down syndrome in the fete 2 months back. She started having complaints of bleeding PV seven days after D & E that continued for 2 months with 2 episodes of heavy bleeding in between. She has undergone repeat D & E for the same 15 days back but not relieved. There is no history of any bleeding disorders. No family history of Gynaecological malignancy in family on examination patient was hemodynamically stable, PR-82/min, BP-138/70 mm hg, RR-20/min. On per abdominal examination-abdomen was soft. On per speculum examination cervix and vagina was healthy, mild blood mixed mucoid discharge was present. On PVl examination Os was patulous, uterus was bulky, anteverted, anteflexed, mobile and non-tender, bilateral adnexa were clear.

On investigation her Hb was 9.7 mg/dl, platelet count and coagulation profile were normal. Her serum beta HCG was less than 5 MIU/ml. Pelvic ultrasonography showed multiple vascular channels present within the endometrium which were communicating with posterior myometrial vessels. On color Doppler the peak systolic velocity was high (PSV-128cm/sec) and RI was 0.40. Angiographic image showed feeder vessels coming from both uterine arteries. She underwent uterine artery embolization under local anesthesia, the procedure was uneventful.

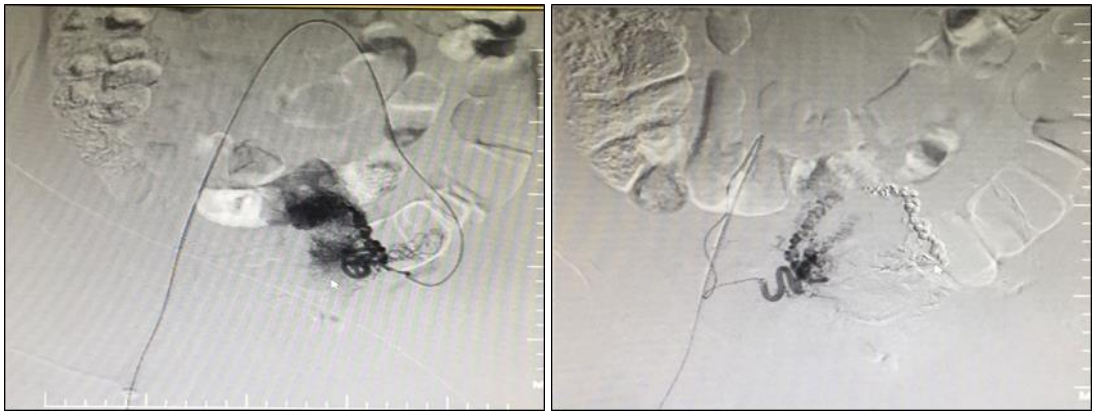

Fig 6: Angiogram showing large AVM/AVF supplied by feeders from left and right uterine artery

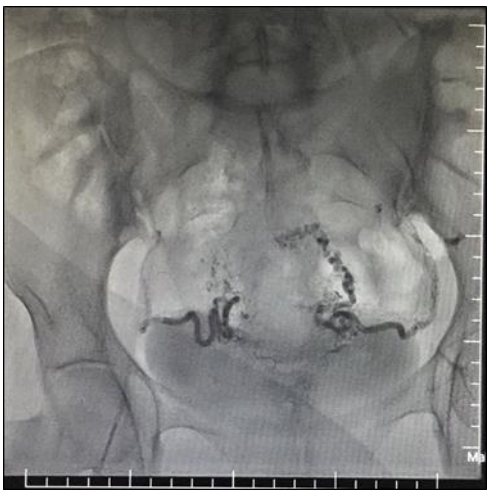

Fig 7: Angiographic image showing bilateral glue cast (post embolization)

Case 4: -29 yr. old female, P2L2A2 with previous 2 normal vaginal deliveries followed by one spontaneous abortion at 2month amenorrhea for which she has undergone D & E 3 months back. She complained of continuous bleeding per vaginum since the D & E not responding to conservative management.

On examination she was vitally stable, pallor was present. On per abdominal examination abdomen was soft, on per speculum examination dark altered blood coloured bleeding was present. Cervix and vagina were healthy. On per vaginal examination uterus was normal in size anteverted and bilateral fornices were free.

On investigation her haemoglobin was 8.9 mg/dl, platelet count and coagulation profile were normal. Pelvic ultrasonography showed 28×28×29 mm hypoechoic area in endometrial cavity in fundal region with tubular spaces within myometrium and prominent parametrial vessels. On colour Doppler these channels showed low resistance and high flow pattern, RI being 0.40 and PSV being 90 cm/second, suggestive of arteriovenous malformation. Uterine artery embolization was performed under local anaesthesia. Embolization was done with injection lipiodol mixed with N-Butyl Cyanoacrylate. Post embolization no

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flow was seen. Vascular sheath was removed and the procedure was uneventful.

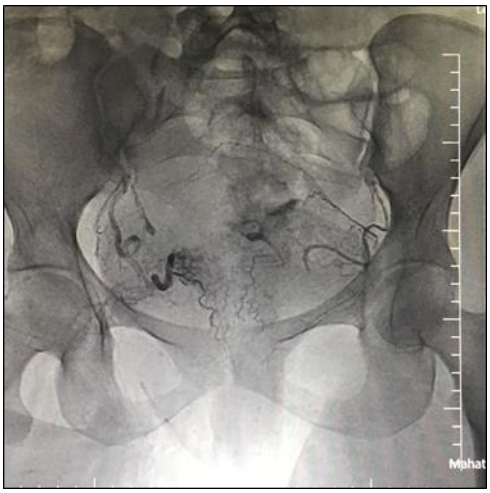

Fig 9: AVM/AVF arising from uterine artery

Case 5: -22-year-old woman P0L0A3 presented with heavy bleeding PV for 4 months. She had 3 abortions each approximately at 2.5 to 3 months of amenorrhea and undergone D & E in each abortion. Her last D & E was 4 months back. Her ultrasound report showed multiple anechoic spaces in myometrium. Doppler study demonstrated markedly increased vascularity in myometrium forming a mosaic pattern, flow parameters suggestive of uterine arterio-venous malformation RI being 0.45 and PSV 89 cm/second (fig 10).

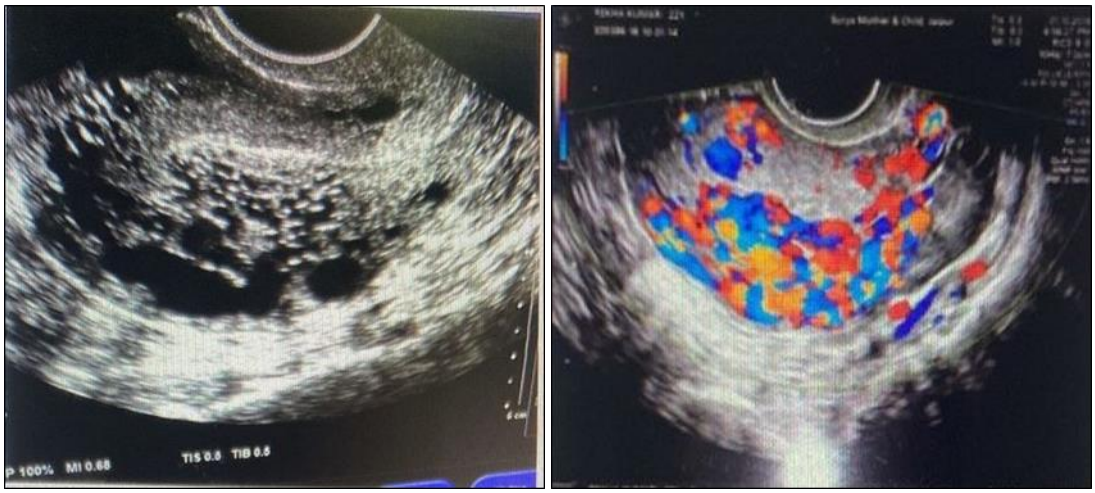

Fig 10: Pelvic ultrasound showing multiple anechoic spaces in the myometrium with markedly increased blood flow forming a mosaic pattern on colour Doppler

Results

S. No.	Age	Parity [P]	Live issue[L]	Abortion [A]	Antecedent event	Time since presentation	Size of lesion (mm)	RI	PSV cm/second
1.	28	P2	L2	A1	D&E	3 month	30X25	0.30	80
2.	24	P2	L2	0	LSCS	1 months	9X9	0.27	78
3.	29	P2	L2	A1	D & E	2 months	9X10	0.40	128
4.	29	P2	L2	A2	D& E	3 months	28X28X29	0.40	90
5.	22	P0	L0	A3	D& E	4 months	10X15	0.45	89

We found 5 cases of uterine AVM during the study period. All of them were in the reproductive age group ranging from 24-29 years. 4 cases out of 5 bleeding occurred following D & E done for abortion; one patient had history of LSCS. All patients presented with in 2 to 3 months of the antecedent event. RI values in all 5 AVMs ranged from 0.27 to 0.45 and PSV ranged from 78 to 128 cm/second. All patients responded well to uterine artery embolization.

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Discussion

Our reported cases presented with the classical pictures of AVMs, i.e., women of reproductive age with history of procedures like dilatation and evacuation presenting with profuse vaginal bleeding all the five cases in our series were symptomatic, presented with moderate to heavy bleeding per vaginal requiring blood transfusion. This case series highlights the importance of considering an arteriovenous malformation in a patient of childbearing age with heavy vaginal bleeding after a uterine procedure. Four out of five patients in our series had history of prior D & E and one patient had history of LSCS. The gestational trophoblastic disease was ruled out in our five cases by serum beta-HCG. Ultrasonography along with Doppler confirmed the diagnosis of arteriovenous malformation and we could take a quick decision to perform selective uterine artery embolization. All 5 were treated with selective uterine arterial embolization, bilateral in few cases. The procedure went uneventful in all and they improved symptomatically. The prognosis after UAE was excellent, and future fertility was conserved. Ghai *et al*. [20] report a technical success rate of 100%, and clinical success rate of 93% in their series. Complications which are often reported include pelvic pain, local hematoma, and rarely, skin sloughing (in the case of internal iliac artery embolization or the use of glue). None of our patient suffered any complication.

In their systematic review of 85 case reports involving 100 patients, Peitsidis *et al*. reported that uterine artery embolization (UAE) was the commonest treatment option (59%), followed by TAH (29%) [21]. We report only five cases. More cases are needed to see the diversity of the clinical presentation of the disease.

Conclusion

Uterine arteriovenous malformations are rare and life-threatening clinical entities in patients presenting with AUB, along with a history of prior uterine instrumentation. Uterine bleeding, being the most common presenting symptom. Doppler ultrasonography provides the most accurate information for confirming the diagnosis. Due to the availability of uterine artery embolization, hysterectomy can be avoided. Based on the available literature, UAE leads to normal periods and without much increase in pregnancy complications.

References

1. Polat P, Suma S, Kantarcy M, Alper F, Levent A. Colour Doppler ultrasound in the evaluation of uterine vascular abnormalities. Radiographics. 2002l22:47-53.
2. Fleming H, Ostor A, Pickel H, Fortune D. Arteriovenous malformations of the uterus. Obstet Gynaecol. 1989;73:209-213.
3. O'Brien P, Neyatatani A, Buckley AR, Chang SD, Legiehn GM. Uterine arteriovenous malformations from diagnosis to treatment. J Ultrasound Med. 2006;25:1387-1392.
4. Grivell RM, Reid KM, Mellor A. Uterine arteriovenous malformation: a review of the current literature. Obstet Gynecol Surv. 2005;60:761-767.
5. Diwan RV, Brennan JN, Selim MA, McGrew TL, Rashad FA, Rustia MU, *et al*. Sonographic diagnosis of arteriovenous malformation of the uterus and pelvis. J Clin. Ultrasound. 1983;11:295-298.
6. Huang MW, Muradali D, Thurston WA, Burns PN, Wilson SR. Uterine arteriovenous malformations: gray scale and Doppler with ultrasound features MR imaging correlation. Radiology. 1998;206:115-123.
7. Ghi T, Giunchi S, Rossi C, Pilu G, Savelli L, Mollo F, *et al*. Three-dimensional power Doppler sonography in the diagnosis of arteriovenous malformation of the uterus. J Ultrasound Med. 2005;24:727-731.
8. Cura M, Martinez N, Cura A, Dalsaso TJ, Elmerhi F. Arteriovenous malformations of the uterus. Acta Radiol. 2009;50:823-829.
9. Nicholson AA, Turnbull LW, Coady AM, Guthrie K: Diagnosis and management of uterine arterio-venous malformations. Clin. Radiol. 1999;54:265-269.
10. Tullio G, Susanna G, Cristina R, *et al*. Three-dimensional Power Doppler sonography in the diagnosis of arteriovenous malformation of the uterus. J Ultrasound Med. 2005;24:727-31.
11. Timmerman D, Wauters J, Van Calenbergh S, *et al*. Colour Doppler imaging is a valuable tool for the diagnosis and management of uterine vascular malformations. Ultrasound Obstet Gynecol. 2003;21:570-577.
12. Corticelli A, Podestà M, Pedretti L, Papadia A, Francescangeli E, Valenzano M. Uterine arteriovenous malformation: a case report diagnosed by sonohysterography. Clin. Exp. Obstet. Gynecol. 2005;32(2):132-4.
13. Rosa E Silva JC, De Aguiar FM, De Sá Rosa E Silva AC, Candido Dos Reis FJ, Poli Neto OB, Nogueira AA. Conservative management of large uterine arteriovenous malformation: case report. Fertil. Steril. 2008 Dec;90(6):2406-7.
14. Scioscia M, Zantedeschi B, Trivella G, Fratelli N, Cosma S, Minelli L. A suggestive diagnosis of uterine arteriovenous fistula based on ultrasonography and hysteroscopy. Eur. J Obstet Gynecol Reprod Biol. 2012 Jan;160(1):116-7.

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15. Taylor E, Hitkari J. Hysteroscopic identification of a uterine arteriovenous malformation. J Obstet Gynaecol. Can. 2009 Dec;31(12):1117-8.
16. Chittawar PB, Patel K, Agrawal P, Bhandari S. Hysteroscopic diagnosis and successful management of an acquired uterine arteriovenous malformation by percutaneous embolotherapy. J Midlife Health. 2013 Jan;4(1):57-9.
17. Goyal S, Goyal A, Mahajan S, Sharma S, Dev G. Acquired uterine arteriovenous malformation developing in retained products of conception: a diagnostic dilemma. J Obstet Gynaecol. Res. 2014 Jan;40(1):271-4.
18. Vogelzang RL, Nemcek AA Jr., Skrtic Z, Gorrell J, Lurain JR. Uterine arteriovenous malformations: primary treatment with therapeutic embolization. J Vasc. Interv. Radiol. 1991;2:517-522.
19. Brown JV 3rd, Asrat T, Epstein HD, Oglevie S, Goldstein BH. Contemporary diagnosis and management of a uterine arteriovenous malformation. Obstet Gynecol. 2008;112:467-470.
20. Nicholson AA, Turnbull LW, Coady AM, Guthrie K. Diagnosis and management of uterine arterio-venous malformations. Clin. Radiol. 1999;54:265-269.
21. Ghai S, Rajan DK, Asch MR, *et al*. Efficacy of embolizations in traumatic uterine vascular malformations. J Vasc. Interv. Radiol. 2003;14:1401-8.
22. Peitsidis P, Manolakos E, Tsekoura V, *et al*. Uterine arteriovenous malformations induced after diagnostic curettage: a systematic review. Arch Gynecol Obstet. 2011;284:1137-51.
23. Calzolari S, Cozzolino M, Castellacci E, Dubini V, Farruggia A, Sisti G. Hysteroscopic management of uterine arteriovenous malformation. JSLS. 2017;21:e2016.
24. Onoyama I, Fukuhara M, Okuma A, Watanabe Y, Nakamura GI. Successful pregnancy after the noninvasive management of uterine arteriovenous malformation. Acta Obstet Gynecol Scand. 2001;80:1148-1149.