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Case Series

Uterine arteriovenous malformations: a life threatening emergency in women

Bullu Priya Oraon*

Department of Obstetrics and Gynaecology, Rajendra Institute of Medical Sciences, Ranchi, Jharkhand, India

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***Correspondence:**
Dr. Bullu Priya Oraon,
E-mail: bullupriya@gmail.com

ABSTRACT

Arteriovenous malformation (AVM) refers to an anomalous, non-functional connection between uterine arteries and veins, potentially posing life-threatening risks due to profuse bleeding. AVM involves the growth of arterial and venous channels, fistula formation, and a combination of capillary-like vessels. This case series sheds light on our institution's experience in diagnosing and managing women with this rare gynecological condition. This retrospective study examined cases of uterine arteriovenous malformation in the Department of Obstetrics and Gynecology at RIMS, Ranchi. All three cases presented with heavy vaginal bleeding. One case had a history of medically induced abortion followed by suction and evacuation. Ultrasound color Doppler confirmed the diagnosis. One patient underwent uterine artery embolization using selective coil and foam agents, while two patients opted for hysterectomy. Two of the cases had a history of previous caesarean sections. Uterine AVM should be considered as a potential differential diagnosis in cases presenting with bleeding after abortion or curettage. The aim of reporting these cases is to highlight the common presentation of this condition and aid in its diagnosis and management.

Keywords: Arteriovenous malformations, Uterine artery embolization, Hysterectomy, Foam

INTRODUCTION

Uterine arteriovenous malformations (AVMs) result from the proliferation of heterogeneous vascular channels with the formation of arteriovenous fistulae.[1] The most common causes of uterine AVM cases are uterine surgery, post-curettage, gestational trophoblastic disease (GTD), or malignancy. The majority of these cases were managed with invasive options including, uterine artery embolization (UAE) (59%), hysterectomy due to uncontrolled bleeding (29%), and major vessel ligation (3%).[2] The incidence of uterine AVM is rare and challenging to quantify, with fewer than 100 cases reported in the literature. Dubreuil and Loubat reported the first case of uterine AVM in 1926.[3] Uterine AVMs may be congenital or acquired. Congenital AVM is believed to arise from arrested vascular embryologic development resulting in anomalous differentiation in the capillaries and abnormal communication, between arteries and veins.[4] Moreover, congenital AVMs can have multiple vascular connections and may invade surrounding structures. They have been found as isolated cases, but have also been reported with AVMs occurring at other sites.[5] Acquired AVM are more common and usually follows a history of previous uterine trauma, such as curettage procedures, caesarean section, or pelvic surgery. Acquired AVMs are more common and typically follow uterine trauma histories, such as curettage procedures, caesarean sections, or pelvic surgeries. The potential to develop abnormal communication between arteries and veins occurs during the healing process, typically when a single artery joins a single vein. Acquired AVM is also associated with infection, retained products of conception, gestational trophoblastic disease, gynaecologic malignancies, and exposure to diethylstilboestrol. Gray scale ultrasound (US) can detect the presence of multiple tubular or “spongy” anechoic or hypoechoic areas within the myometrium of a normal endometrium.[4-6] A colour and spectral Doppler US

is important for obtaining more accurate information. Digital subtraction angiography (DSA) remains the gold standard for the diagnosis of AVM.

Management of uterine AVM depends on the hemodynamic stability, degree of bleeding, patient age, and desire for future fertility. Acute treatment involves stabilizing the patient's hemodynamic status, and stopping blood loss. Earlier, hysterectomy was the treatment of choice. However, the patient's desire for future fertility is an important consideration, as there are now options available to avoid a hysterectomy.

UAE is increasingly becoming one of the preferred treatment modalities in this setting, primarily due to its effectiveness as well as being minimally invasive in nature, with the resultant possibility of preserving uterine function to allow future childbearing [7] and the ready availability of experts to perform these procedures. Various embolic materials have been used, including polyvinyl alcohol, histoacryl (glue), stainless steel coils, detachable balloons, and haemostatic gelatine. Hysterectomy remains the treatment of choice in post-menopausal patients or as an emergency treatment in life-threatening situations.[8] This case series highlights our experience in diagnosis and management in women reported in our institute having this rare gynaecological condition.

CASE SERIES

This was a descriptive retrospective study conducted at the Department of Obstetrics and Gynecology at RIMS, Ranchi, focusing on cases of uterine arteriovenous malformation.

Case 1

A 27-year-old woman, with three previous lower segment cesarean sections (LSCS), was admitted to our hospital on 15 December 2019 with a complaint of heavy vaginal bleeding for one week, occurring on the 20th day of puerperium. On examination, the patient was severely pale, with an Hb% of 6 gm%. Abdominal examination revealed vague fullness 4 fingers above the umbilicus and an ill-defined non-tender lump. A bimanual examination showed a uterus of 14-16 weeks in size. An ultrasound revealed an intrauterine collection of 5×4 cm and a pelvic collection with air foci. Three units of packed cells were transfused, and laparotomy was performed on 21 December 2019. During the surgery, the omentum was adhered to the anterior surface of the uterus, bladder, and anterior abdominal wall up to the fundus of the uterus. Approximately 1 liter of blood-stained purulent foul-smelling discharge was evacuated. Higher antibiotics were administered postoperatively. On the 12th day after laparotomy, the patient experienced another episode of profuse vaginal bleeding. Uterine AVM was suspected, and ultrasound color Doppler confirmed increased vascularity. A cardiology opinion was sought for urgent embolization, and selective coil and foam embolization were performed. The patient also developed a wound gap, which required secondary suturing. She was discharged on 22 January 2022.

Case 2

A 42-year-old woman, with a history of four previous pregnancies and one live birth through LSCS, was admitted with heavy vaginal bleeding for 15 days on 15 June 2022. On admission, she was severely pale, with an Hb% of 4 gm%. She had a history of a spontaneous abortion that was not followed by dilation and evacuation (D&E). Abdominal examination revealed a suprapubic lump of 12-14 weeks in size, firm in consistency, and mobile from side to side but with restricted mobility above and below. Bimanual examination confirmed a lump of 12-14 weeks in size, firm in consistency, with mobility transmitted to the cervix. She received 4 units of packed cells. Ultrasound color Doppler revealed a hypervascular endometrial lesion, most likely an arteriovenous malformation, in the lower uterine segment. As the patient had completed her family and opted for a hysterectomy, she underwent total abdominal hysterectomy and bilateral salpingo-oophorectomy (TAH+BSO) on 30 June 2022. The patient recovered well postoperatively and was discharged on 8 July 2022.

Case 3

A 28-year-old woman, with a history of two previous pregnancies and two live births, was admitted with a complaint of vaginal bleeding for one month on 1 June 2022. She had a history of medically induced abortion followed by D&E in 2019, which was followed by continuous vaginal bleeding. She was diagnosed with uterine AVM at RIMS Ranchi after undergoing a color Doppler ultrasound. Following this, she underwent uterine artery embolization and was discharged symptom-free. In 2021, she spontaneously conceived and delivered her second baby. She returned to LR Emergency RIMS, Ranchi, with heavy vaginal bleeding on 1 June 2022 and was severely anemic, with an Hb% of 4 gm%. Bimanual examination showed an anteverted uterus, 8 weeks in size, mobile, and with free fornices. She received four units of packed cells. A repeat color Doppler ultrasound showed vascularity in the posterior myometrium with both arterial and venous flow, suggestive of a uterine arteriovenous malformation. She underwent TAH on 23 June 2022 and was discharged on 27 June 2022.

Results

Among the cases, two of them were in reproductive age group and one belongs to perimenopausal age group. The presentation was heavy bleeding per vaginum in all the three cases. In one of the case, history of medically induced abortion followed by suction and evacuation was present. Two of the cases had history of previous caesarean section. Diagnosis was done by ultrasound color doppler.

Two patient underwent uterine artery embolization by selective coil and foam agent. Out of these two patients, after UAE, one patient again came symptomatic, and family was also completed, she opted for hysterectomy. So among the cases two patients opted for hysterectomy.

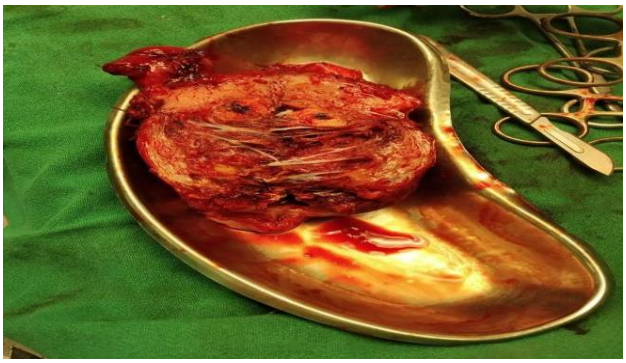

Figure 1: Gross specimen of the uterus with anterior wall cut open showing abnormal multiple arterial and venous channels.

Figure 2: Gross specimen of the uterus with cut section showing arteriovenous malformation in the posterior part of the fundus of the uterus.

DISCUSSION

Uterine arteriovenous malformation results from one or more site of abnormal direct communication between an artery and a vein without an intervening capillary bed.

Typically, the patient presents with heavy vaginal bleeding, following D&E, D&C after therapeutic abortion or uterine surgery. One of patient had H/O prior D&E for incomplete abortion. Diagnosis was done by clinical findings and confirmed by USG color doppler.

The embolization procedure in our hospital were performed by cardiologist trained in interventional cardiology. Gel foam and coil were used in UAE. Two of cases opted for hysterectomy.

In a study conducted by Lalitha et al on topic uterine arteriovenous malformation: case series and literature review at department obstetrics and gyanecology, PSG Institute of Medical Sciences and Research, Coimbatore, India, in 2015 in which all the five cases in case series were symptomatic.[9]

In my study all the cases were symptomatic with chief complain of heavy bleeding per vaginum. Four were treated with selective arterial embolization and one by hysterectomy.[9] Whereas in my study one of the cases underwent uterine artrery embolization and rest two of the cases opted for hysterectomy. All the patients in our series had a prior curettage for termination/incomplete miscarriage.[9] In my study one case had history of incomplete abortion followed by D&E. Rest two cases had history of previous one caesarean section. Diagnosis in all our cases was made by TAS/TVS with Doppler.[9] In my study also diagnostic modality used was ultrasound color Doppler.

In their systematic review of 85 case reports involving 100 patients, Peitsidis et al reported that UAE was the commonest treatment option (59 %), followed by TAH (29 %).[10]

In a study by Nakashololo et al on uterine arteriovenous malformations, clinical and radiological considerations: a report of two cases at Department of Obstetrics and Gynaecology, Faculty of Health Sciences, University of Pretoria, South Africa in 2021,both the cases presented with repeated episodes of profuse vaginal bleeding.[11] In my study all the cases were symptomatic with chief complain of heavy bleeding per vaginum. Ultrasound (US) examination revealed classical signs of uterine arteriovenous malformation (AVM) confirmed on computerized tomography angiography (CTA) and digital subtraction angiography (DSA).[11]

There is a strong association between pregnancy and symptomatic AVM, likely due to these hormonal factors in normal pregnancy, as well as interventions during pregnancy, such as dilation and curettage, prostaglandin E1-induced abortion, and delivery, which have all been listed as primary causes of acquired uterine AVMs.[12,13] Historically, large uterine arteriovenous malformations, first described in the late 1980s, were treated with hysterectomy. Transcatheter embolization has now become the standard of care.[14]

A systematic review analyzing fertility after uterine artery embolization suggested that pregnancy rates were similar to age-adjusted rates in the general population, with similar rates of complications.[15] One of the case in my study underwent UAE, and she conceived spontaneously, delivered vaginally. But she again became symptomatic and her family was also completed so she opted for hysterectomy.

The diagnosis can be challenging in the absence of appropriate imaging studies, as the presenting signs and symptoms are common and non-specific, and vaginal bleeding is common and usually benign for patients presenting to the Emergency department.[16] In the post-

partum period, this can be especially challenging due to many similarities in initial presentation between normal post-partum bleeding and this potentially life-threatening condition. Secondary, or late post-partum hemorrhage is defined as any significant uterine bleeding that occurs between 24 hours and 12 weeks post-partum.[16] While it is usually caused by retained products of conception, subinvolution of the placental bed and infection, less common but potentially dangerous causes include arteriovenous malformation, bleeding diatheses, dehiscence of the cesarean scan and pseudoaneurysms of the uterine, internal pudendal, vaginal, or vulvar labial arteries.[16] Additionally, since vaginal bleeding is normal in the post-partum period, patients may have difficulty differentiating between normal and abnormal bleeding.

CONCLUSION

Uterine AVM is a rare condition which needs to be considered in differential diagnosis of genital tract bleeding. Because proper diagnosis leads to better management. Hysterectomy is the most definitive treatment for such condition in our case series. However, modern management of uterine AVM by uterine artery embolisation is more acceptable these days. Uterine AV malformation should be kept in mind in all the cases presenting with bleeding after abortion or curettage. In our study there is a common presentation of heavy bouts bleeding after D&E and also had history of previous caesarean section. Color doppler USG are the best diagnostic tools that provide the most accurate information. The aim of reporting these cases is to share the common presentation of this condition and in making diagnosis and management of patients.

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