ORIGINAL RESEARCH PAPER

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A RARE CASE OF UTERINE ARTERIO VENOUS MALFORMATION IN A MULTIPARA -A CASE REPORT

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ABSTRACT

Uterine Arterio Venous (AVM) malformations are rare and we came across fewer than hundred cases up till now. Despite it being rare it is a potentially life threatening condition. AVMs are largely acquired lesions. pregnancy plays an important role in their pathogenesis but they may also be congenital. We present a 40 year old female rare case of AVM of the uterus who required radical treatment for excess bleeding PV. AV malformation of the uterus was confirmed on USG Pelvis with Doppler studies. She was initially treated conservatively with recent gadgets like embolization, however failed to respond, had recurrence of symptoms, radical method of hysterectomy was done.

INTRODUCTION

Arterio venous malformation is an abnormal connection between arteries and veins. Uterine arteriovenous malformations (AVMs) are rare. They are usually acquired malformations that follow a pregnancy event and are diagnosed when uterine bleeding remains uncontrolled despite medical measures (3). The bleeding caused by these malformations is episodic and can be torrential, warranting hospital admission and blood transfusions (3). In the past primitive management like hysterectomy was the mainstay of treatment but with advances in technology, conservative management with recent gadgets like embolization is used. This case report highlights our experience with a case having this rare uterine AVM who was treated with hysterectomy, in our medical centre.

CASE REPORT

A 40 years old female patient P2L1D1A1 came with chief complaints of first episode of bleeding per vagina with passage of clots since 10 days with weakness and giddiness. Menstrual history- irregular cycles of 1.5 to 2months with heavy flow with passage of clots. She had one normal delivery 13 years back, one Intra uterine fetal demise and one Abortion. **Per speculum examination:** Vaginal mucosa-healthy, Cervix healthy, Bleeding through the os present. No polyp, erosion, growth or varicosity. **Per vaginal examination: Uterus** anteverted, Uterus bulky, mobile, tenderness +, Fornices free, nontender, Bleeding +. Ultrasonography (USG) was done and it was suggestive of bulky uterus with an Endometrial Thickness (ET) of 10mm and a diagnosis of Dysfunctional Uterine Bleeding was made. The patient was then treated symptomatically with 1-pint PRBC supply,Tranexamic acid and hormonal supplements.

She came back again four months later for the same complaints. **USG was repeated along with Doppler study** which showed anterior myometrium with extensive vascular flow predominantly in fundal region and vascular flow reaching upto endometrium on spectral study where both arterial and venous waveform was demonstrated suggestive of AVM of Uterus. This patient underwent uterine artery embolization in a private hospital as this facility was not available in our institute. However, there were recurrence of symptoms after four months and she was admitted in our hospital again due to failure of embolization. She had no history of infection, inflammation, retained products of conception, gestational trophoblastic disease, gynaecologic malignancies, pelvic trauma, hypertension, diabetes, asthma. **DIAGNOSIS**- P2L1D1A1 with Abnormal Uterine Bleeding (AUB) with AV Malformation of uterus with failure of embolization.

She was treated with further conservative management like methylergonovine maleate, hormonal therapy, PRBC supply, Tranexamic acid.

The said case again failed to respond to the above treatment given. So CECT (contrast enhanced CT scan) Abdomen and Pelvis was done after 3 months. The findings were ...

1. Multiple abnormal serpentine vessels are noted within anterior myometrium reaching into the endometrial cavity.
2. Bilateral parametrium showing intense enhancement in arterial and venous phases suggestive of uterine arterio-venous malformation.
3. No etiology of (e/o) any thrombosis /aneurysm.

She was not affording for a second embolization and not ready for repeated follow ups. There was failure to respond to medical and conservative measures, she was treated with Total abdominal hysterectomy with right sided salpingo-opherectomy with preservation of left ovary.

INTRA OPERATIVE FINDINGS:

Etiology of (E/O) hemoperitoneum ~ 50 ml. No e/o any malformation in pelvic cavity. E/O tortuous vessels feeding the uterine artery coming from internal iliac artery in right parametrium. Left parametrium comparatively less vascular. Left sided ovary was normal. Right sided ovary had a cyst of 1*1 cm which was in bleeding phase, so right sided ovary removed. Difficulty in dissecting urinary bladder on right side due to vascularity as it started bleeding while dissecting.

Blood vessels in right parametrium selectively skeletonized without hurting vessels. Mixter forceps was applied, vessels were cauterized and cut, Free ends tied where ever needed. Bladder found adherent to cervix. Multiple oozers were present. Blood loss was 500ml. Two units of PRBCs given. Rest surgery and post-operative were uneventful.

DISCUSSION

- AV malformations most commonly present to neurosurgeons as the most common organ involved is the brain. However, rarely it may involve the uterus.
- Uterine arteriovenous malformations are rare causes of gynecologic bleeding. The majority of cases are congenital, but at times can occur after obstetric or gynecologic procedures, primarily uterine curettage or rarely hysterectomy. Fewer than 100 cases have been reported in the literature, but with the increasing use of imaging modalities, particularly ultrasound, the incidence is increasing.
- These lesions are diagnosed more commonly in the postpartum period or a few months after a spontaneous miscarriage, surgical evacuation of the uterus for a miscarriage or termination of pregnancy. Acquired AVMs are also associated with infection, inflammation, retained products of conception, gestational trophoblastic disease, gynaecologic malignancies, pelvic trauma and exposure to Diethylstilbestrol. Uterine AVMs may rarely be congenital, when they can be associated with similar AVMs in other organs. Our case appears to have an acquired AVM of Uterus. (3)
- O'Brien et al identified uterine AVMs in 21 women out of 464 pelvic sonographic examinations for uterine bleeding, giving a rough incidence of 4.5%. Conversely, Yazawa et al prospectively followed 959 patients and noted a lower incidence of uterine vascular malformation on ultrasound of 0.6%. (8)
- In the case report in Malaysian Journal Of Medical Sciences by Hilwati Hashim and Ouzeiah Nawawi, a case of uterine artery malformation was treated successfully with bilateral uterine artery embolization with follow up after 1.5 years when the patient did not have recurrences of symptoms. (7) Whereas in our case, she was treated with embolization, however she had recurrence of symptoms after a few months and thus had to be managed surgically.
- In the Case Report by Kyousuke Takeuchi, a case who had acquired AVM after D and C with an extensive lesion and was successfully treated with bilateral uterine artery embolization. (6) In our case, she hadn't undergone D and C and due to failure of response to embolization she had to undergo hysterectomy.
- Another case by Maj Robert M Ore et al described a case of AVM who initially treated with UAE which was then followed by hysterectomy due to intra-abdominal bleeding (2) from the uterine fundus because of a ruptured AVM and thus hysterectomy became necessary because of her clinical presentation. Whereas in our case, there was no rupture of AVM and but she needed hysterectomy.
- One series of 42 patients who underwent Gelfoam embolization for post obstetric uterine AVMs demonstrated successful definitive endovascular treatment in 88% of patients, with the remaining 12% requiring hysterectomy. Another study of 15 patients with uterine AVMs demonstrated an endovascular therapy success rate of 93%, with only one patient requiring hysterectomy (1). Both these facilities are not available in our Institute.

CONCLUSION

Uterine AVMs are a rare cause of uterine haemorrhage. The vast majority resolve spontaneously or with medical treatment. However, a few cases who fail to respond to the recent gadgets like embolization can be successfully treated with old age method of Hysterectomy.

FIGURES

Figure no.1 – CECT showing AVM of the Uterus

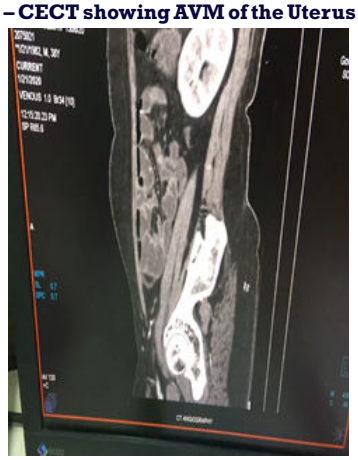


Figure no.2 – Tortuous vessels around the Uterus held by Mixter forceps

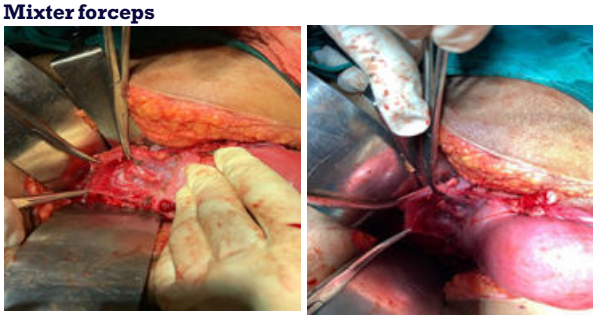
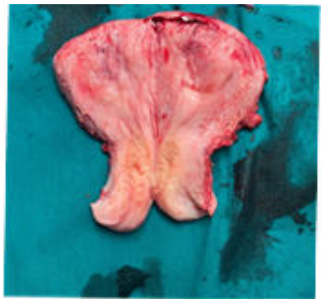


Figure no. 3- Gross specimen of the removed Uterus



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