

## Uterine Arteriovenous Malformation - A Rare Cause Of Uterine Haemorrhage

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Uterine arteriovenous malformations are rare lesions with a considerable risk potential. Clinical presentation varies from no signs over various degree of menorrhagia to massive life threatening vaginal bleeding. We report a case of uterine arteriovenous malformation diagnosed and successfully treated with bilateral internal iliac artery ligation. A married female patient of 25 years old, with two live issues, presented with recurrent excessive menstrual bleeding since her menarche. She had history of spontaneous abortion and D & C. After D & C she was symptom free only for 1 week. She again develop polymenorrhagia.. She was treated with oestrogen progesterone combinations, medroxy progesterone alone for 3 months. Despite she had recurrent uterine bleeding. Her clinical examination was unremarkable except marked pallor. Ultrasonography reveals bulky uterus with dilated, tortuous uterine vessels. Uterine artery colour Doppler revealed bulky uterus with multiple tortuous vascular structures around uterus extending up to both adnexal region. After discussing all possibilities of treatment with patient, bilateral ligation of anterior division of internal iliac artery was done. Her post operative period was uneventful and she was discharged on 7<sup>th</sup> post operative day. She was symptom free in subsequent follow up.

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**Key words:** Arteriovenous fistula, uterus

### Introduction

In women of child bearing age, abnormal bleeding includes any change in menstrual period frequency or duration, or amount of flow, as well as bleeding between cycles.<sup>1, 2, 3</sup> There are seven patterns in the standard classification of abnormal PV bleeding: menorrhagia, hypomenorrhoea, metrorrhagia, polymenorrhoea, menometrorrhagia, oligomenorrhoea and amenorrhoea.<sup>4</sup> The causes of abnormal per-vaginal bleeding (PVB) include a wide range of conditions of the reproductive system. Abnormal PVB is a common problem that affects one in five women during the pre-menopausal years.<sup>2, 4</sup> AVMs can occur in any organ in the body, including the pelvic vasculature and rarely in

the uterus. The first case of AVM was reported in 1926.<sup>5</sup> A uterine arteriovenous malformation (AVM) is a rare cause of uterine bleeding. The lesion has been variably described as cirroid aneurysm, arteriovenous fistula, arteriovenous aneurysm, pulsating angioma, or cavernous angioma. Earlier diagnosis was usually made on angiography,<sup>6</sup> laparotomy, or pathology. However with the advent of newer techniques such as colour doppler sonography, contrast computed tomography (CT) and magnetic resonance imaging (MRI), the detection of this entity has become easier and therefore nowadays even small AVMs can be detected.<sup>7, 8</sup>

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### Case Report

A married women of 25 years old with two live issue and having history of one MR and one abortion (6 months back) presented with excessive menstrual bleeding. Her menarche was at 15th year. Since then she had polymenorrhagia. Past history included spontaneous abortion followed by D & C in November 2009. After D & C she was bleeding free up to one week. Then she developed polymenorrhagia which worsen one month after the curettage. She was treated with oestrogen progesterone combinations, medroxy progesterone alone for 3 months. Despite that she had recurrent uterine bleeding. She had history of three units of blood transfusion as she was presented with life threatening pervaginal bleeding.

On systemic examination there was no abnormality detected. Pallor being predominant during general examination. Per abdominal and per speculum examination was normal. On internal examination uterus was bulky, mobile, non pulsatile, fornices clear.

On investigation hemoglobin was 7.5 gm/dl, coagulation profile was within normal limit. Ultrasonography reveals bulky uterus with dilated, tortuous uterine vessels. Serum  $\beta$ -HCG was  $<1.00$  mIU/ml. Sonography suggested features of uterine arteriovenous malformation. Uterine artery colour Doppler revealed bulky uterus with multiple tortuous vascular structures around uterus extending up to both adnexal region. After discussing all possibilities of treatment with patient bilateral ligation of anterior division of internal iliac artery was done on 2 April 2010. Along with bilateral tubal ligation was done as patient was nulliparous. Her post operative period was uneventful and she was discharged on 7<sup>th</sup> day with hormonal treatment in the form of medroxy progesterone acetate from 15<sup>th</sup> to 25<sup>th</sup> day of menstrual cycle for 3 cycles. Progesterone withdrawal bleeding was not so heavy. She is now on combined oral contraceptive pill & symptom free till June, 2013. She was advised for further follow up.



Figure 1. USG Findings (AV fistula)

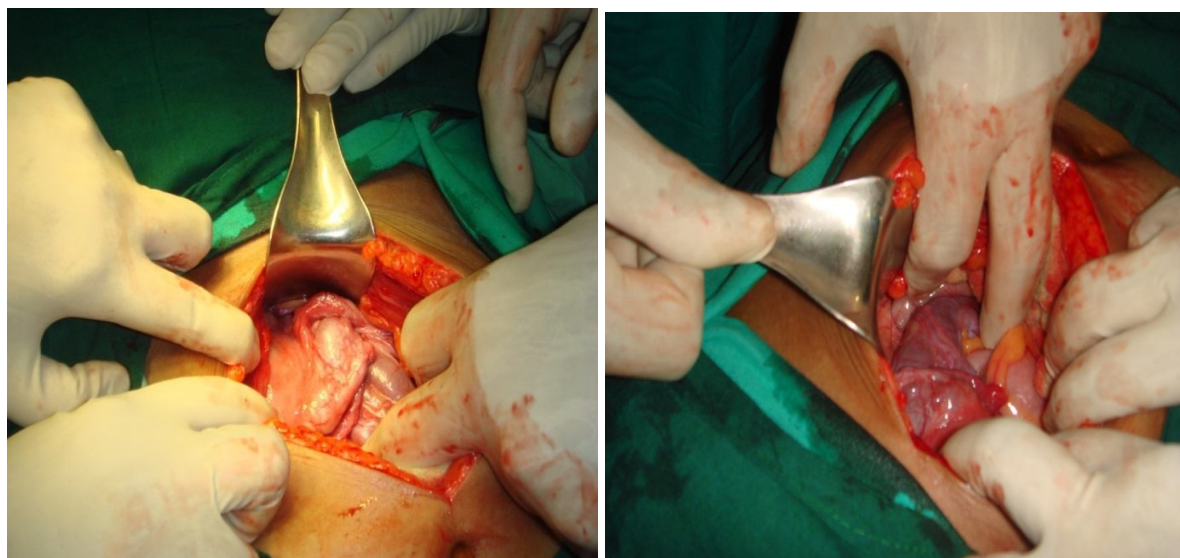


Figure 2. Per-operative findings (AV Mal formation)

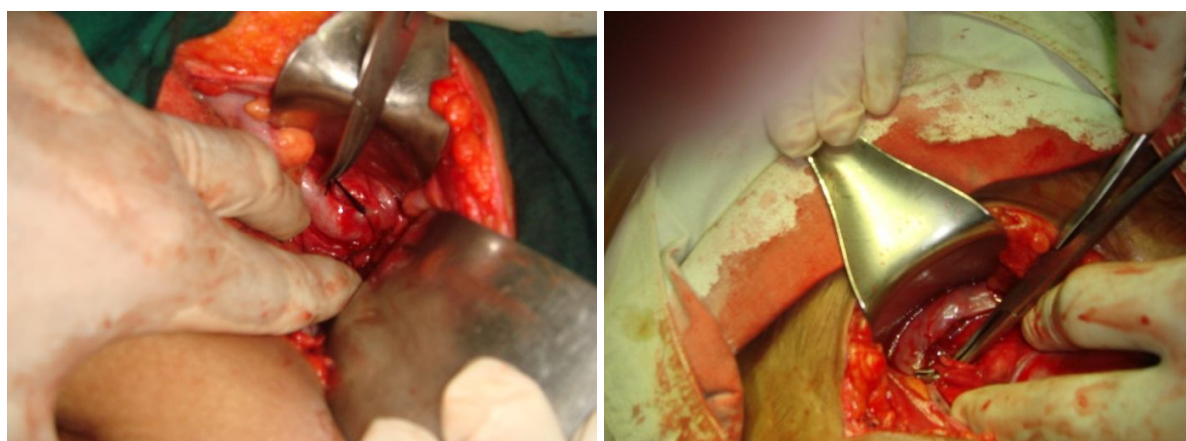


Figure 3. Per-operative findings (Ligation of anterior division of internal illiac artery)

### Discussion

The true prevalence of uterine AVMs is unknown. In a study of 265 patients with abnormal premenopausal bleeding, a diagnosis of uterine vascular malformations was obtained with sonography in 9 patients (3.4%).<sup>9</sup> Uterine arteriovenous malformations (AVMs) are broadly classified as congenital or acquired.<sup>10</sup> Congenital AVMs are rare, whereas acquired or traumatic AVMs are being increasingly diagnosed. Congenital uterine AVMs result from abnormal

embryologic development of primitive vascular structures, which result in multiple abnormal communications between arteries and veins.<sup>11</sup> Congenital AVMs tend to have multiple feeding arteries, a central nidus (a tangle of vessels with histologic characteristics of both arteries and veins), and numerous large draining veins.<sup>12</sup> Conversely, acquired or traumatic uterine AVMs represent multiple small arteriovenous fistulas between intramural arterial branches and the myometrial venous plexus.<sup>13</sup> Acquired AVMs

tend to have single/bilateral uterine artery feeders without an extrauterine arterial supply and do not have a characteristic nidus.<sup>14</sup> Causes of acquired<sup>15</sup> uterine AVM include previous uterine surgery like curettage, caesarean section<sup>16</sup> or hysterectomy,<sup>17</sup> pelvic trauma, previous pregnancy,<sup>18</sup> gestational trophoblastic diseases,<sup>19</sup> exposure to diethyl stilbesterol,<sup>20</sup> endometriosis, fibromyoma, and endometrial or cervical cancers. In this case report patient's having history polymenorrhagia as an adolescent may indicate a congenital etiology. Although a trauma related to post curettage aggravated the condition.

Many methods have been used to diagnose uterine vascular abnormalities. The reference standard for definite diagnosis of AVMs is angiography, with classic features of a complex tangle of vessels supplied by enlarged feeding arteries associated with early venous drainage during the arterial phase and stasis of contrast medium within the abnormal vasculature.<sup>21, 22</sup> Currently, sonography is an important noninvasive modality for the detection of these abnormalities. Sonographic findings in gray scale include nonspecific findings such as subtle myometrial inhomogeneity, tubular spaces within the myometrium, intramural uterine masses, endometrial or cervical masses, and sometimes prominent parametrial vessels. Color Doppler features are more consistent and include intense juxtaposed signals with aliasing. Spectral Doppler sonography reveals high-velocity diastolic components with low-resistance flow (resistive index, 0.25–0.55; pulsatility index, 0.3–0.6) and high PSV greater than 96 cm/s that suggest arteriovenous shunting.<sup>23,24-27</sup> In a study of 30 patients with uterine vascular malformations defined as an abnormal hypervascular area in the myometrium with turbulent flow, PSV values of greater than 83 cm/s were associated with higher probabilities of further treatment such as embolization, and no vascular malformations with PSV values of less than

39 cm/s required embolization.<sup>28</sup> In our case we found that colour doppler features of uterine AVMs are consistent and diagnostic. Management of uterine AVM based on the clinical status of the patient. Patients who are anemic or hemodynamically unstable should be referred for angiography and embolization. Patients with a single episode of bleeding who are hemodynamically stable can be treated conservatively with estrogen, progesterone combination, progesterone, methyl organovine, danazol, 15-methyl prostaglandin F<sub>2</sub> alpha. Many of these patients will remain asymptomatic, suggesting that traumatic AVMs do spontaneously regress. If patients have recurrent bleeding, then embolization is indicated. As shown in this study, the size of an AVM on imaging does not correlate with the need for embolization. This decision is entirely based on the patient's clinical status.<sup>29</sup> Surgical management includes surgical removal of AVM, laparoscopic bipolar coagulation of uterine vessels; coagulation of the AVM may also be done under hysteroscopic guidance, selective uterine artery embolization, bilateral uterine artery ligation and hysterectomy. Our patient presented with profuse bleeding (occasionally) which required blood transfusion. Clinical examination was unremarkable except pallor. Patient was 25 years and not desirous of having child. Moreover, conservative treatment with drugs failed so that decision for ligation of uterine artery was taken. But after laparotomy it was found the extensive AVMs in between uterus bladder & broad ligament and adnexae. Due to the fear of massive injury decision was taken to ligate the anterior division of internal iliac artery which was safely performed. In the last decade, an increasing number of women have been treated conservatively with success and hysterectomy is no longer considered essential.

*Conclusion*

Uterine AVMs are common than previously thought and should be considered as a cause of refractory uterine bleeding. Colour doppler sonography is important for proper assessment of the lesion. Site & size of the lesions probably have a major role in deciding whether the patient needs medical or surgical intervention. So which dealing with a case of refractory type of menorrhagia one should always consider the possibility of uterine AVM.

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