A RARE CASE OF UTERINE VASCULAR MALFORMATION PRESENTING WITH HEAVY MENSTRUAL BLEEDING AND RECURRENT PREGNANCY LOSS

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ABSTRACT: Uterine Arteriovenous Malformation (AVM) is a rare condition, with fewer than 100 cases reported in the literature. It is a potentially life-threatening condition, as patients may present with profuse bleeding. Vascular lesions of the uterus are rare and the vast majority reported in the literature are those of arteriovenous malformations. Uterine AVM can be congenital or acquired. This case reports a woman with Uterine AVM presenting with heavy menstrual bleeding and a history of recurrent pregnancy loss.

KEYWORDS: Uterine arteriovenous malformation, heavy menstrual bleeding, recurrent pregnancy loss.

INTRODUCTION: Uterine Arteriovenous malformation (AVM) is a rare condition, with fewer than 100 cases reported in the literature.¹ Uterine AVM is a rare condition which may be congenital or acquired. Acquired lesions are believed to result from pelvic surgery, trauma, curettage, trophoblastic disease, diethylstilbestrol exposure, neoplasm or infection. Colour Doppler ultrasound (US) provides a non-invasive method for initially diagnosing this rare condition and confirmation can be made using diagnostic angiography. Conservative management or embolisation is a preferable method of treatment in order to avoid a hysterectomy in patients of child-bearing age. This case reports a woman with Uterine AVM presenting with heavy menstrual bleeding and a history of recurrent pregnancy loss and her subsequent management.

CASE REPORT: A 38-year-old woman with history of Recurrent Pregnancy Loss (RPL), presented with numerous episodes of heavy menstrual bleeding (HMB), separated with periods of moderate bleeding. She had seven spontaneous 1st trimester miscarriages out of which four required curettage. Histopathological examination and karyotype were done in the sixth and the seventh time respectively and were found to be within normal limits.

On examination, she was afebrile and hemodynamically stable with a hemoglobin level of 6.5 g/dL. Vaginal examination revealed blood tinge at the external os, but no active bleeding. The human chorionic gonadotropin (hCG) level was < 2 mIU/mL. US (Transabdominal) of the pelvis showed an anteverted bulky uterus having normal shape, outline and echopattern, measuring (9.9×4.5×7.3cm) with an endometrial thickness of 1.5 cm. In Colour Doppler study there was a highly vascular lesion in the posterior uterine wall showing low resistance high velocity flow. The vascular lesion was fed from both uterine arteries. Doppler US showed a Peak Systolic Velocity (PSV) of 51 cm/s and Resistive Index (RI) of 0.66. The vascularity of the endometrium was also increased. Cervix was normal and the internal os was closed. No free fluid was seen in the pouch of douglas. Both adnexa were clear and bilateral ovaries were normal in size, shape, outline and echotexture.
The patient was first stabilized with blood transfusions to increase her hemoglobin status. Laparotomy was planned and despite profuse intraoperative bleeding, bilateral uterine artery ligation along with ligation of bilateral utero-ovarian anastomosis was successful and her postoperative period was uneventful. At follow up she was relieved of HMB. A repeat Colour Doppler US study showed no vascular lesion.

**DISCUSSION:** Uterine AVM results from formation of multiple arteriovenous fistulous communications within the uterus without an intervening capillary network. Distinction between arteries and veins is difficult because of secondary intimal thickening in veins due to increased intraluminal pressure. Though uterine vascular abnormalities are rare, yet they are potentially life-threatening, presenting with vaginal bleeding that may be profuse enough to cause hemodynamic instability. Thus, it is an important differential diagnosis to be considered in women of reproductive age with unexplained vaginal bleeding and in post-menopausal women, when anechoic structures are identified by ultrasound. Dubreuil and Loubat reported the first case of uterine AVM in 1926.

Uterine AVM may be congenital or acquired. Congenital AVM arise from arrested vascular embryologic development, resulting in anomalous differentiation in the capillaries and abnormal communication between arteries and veins. Moreover, congenital AVM can have multiple vascular connections and may invade surrounding structures. They have been found as isolated cases, but have also been reported with AVM occurring at other sites. Acquired AVM are more common and usually follows a history of previous uterine trauma, such as curettage procedures, cesarean section, or pelvic surgery. 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, gynecologic malignancies, and exposure to diethylstilboestrol. In order for the endometrium to bleed, endometrial blood vessels and surface epithelium must both breakdown.

Clinical presentation varies; some may cause life-threatening massive genital bleeding in young women, while these are less common after menopause and post-menopausal bleeding is rarely seen. Congestive heart failure secondary to a vascular steal syndrome may be a less common clinical manifestation with a large uterine AVM. Very rarely they present with recurrent pregnancy loss. In this case, it is possible that an acquired form of uterine AVM was present due to multiple curettages performed and consequent uterine trauma. Also as the patient presented at a later age it points towards an acquired AVM as congenital AVM presents at a younger age. It has been seen that acquired uterine AVM are associated with obstetric disorders and procedures such as multiple pregnancies, spontaneous miscarriages, previous dilation and curettage, therapeutic abortion and cesarean section.

Transcatheter arterial embolization is an excellent treatment option in selected cases but it may not be possible in all centers where radiographic embolization procedures are unavailable. In such cases bilateral uterine artery ligation can serve as an equally effective procedure which is also simple to perform and associated with minimum side effects. The only disadvantage being that a laparotomy being required. Thus, in low resource settings laparotomy and bilateral uterine artery ligation may serve as a lifesaving option in these rare presentations.
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