Successful management of uterine arteriovenous malformation by laparoscopic bilateral uterine artery ligation*

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ABSTRACT

Arteriovenous Malformations are vascular disorders where there is an abnormal communication between an artery and vein. It can occur anywhere in the body not even sparing the uterus. Uterine Arteriovenous Malformations (AV Malformation) is a rare occurrence with less than 100 cases reported in literature. It can cause significant bleeding leading to anemia and even hypovolemic shock. It may be acquired from previous uterine manipulation such as dilatation and curettage and previous uterine surgeries. Diagnosis is made by angiography or doppler ultrasonography. Definitive treatment is hysterectomy however a less invasive, fertility preserving are uterine vascular occlusion techniques, of which the treatment of choice is Uterine Artery Embolization (UAE). We present our experience with 24 year-old G2P2 (1101) with scarred uterus suffering from recurrent profuse vaginal bleeding suspected to have uterine arteriovenous malformation. Laparoscopic bilateral uterine artery ligation, an alternative, more economical, relatively safe and available treatment option was given to the patient.

Keywords: uterine arteriovenous malformation, uterine artery embolization, abnormal uterine bleeding

INTRODUCTION

Arteriovenous malformation is an abnormal communication between an artery and a vein which can occur anywhere in the body. Histologically, there is a localized proliferation of both arterial and venous vessels with interconnecting fistulae. These rare vascular malformations may be congenital or acquired. When they occur in the uterus, which is usually acquired, can cause obstetric or gynaecologic torrential bleeding. Acquired AV Malformations may be postpartum, post-abortion or menstrual. Definitive diagnosis is done by pelvic angiography. This however, is invasive and relatively expensive. Doppler ultrasonography is an alternative method. Uterine AV Malformation may be treated medically or surgically. Uterine artery embolization (UAE) is currently preferred treatment for AV Malformation wherein percutaneous catheterization of the femoral artery is performed.[1] We present a woman experiencing heavy vaginal bleeding from this unusual condition. The patient is young, desirous of pregnancy, financially restricted and is managed in a tertiary government where UAE is not available.

CASE

This 30 year-old G2P2 (1101), underwent Primary LTCS at 30 weeks AOG for cord prolapse 1 year prior to consult at a government hospital, delivered to a live preterm baby boy with an estimated blood loss of 800cc. She tolerated the procedure well but was transfused with 3 units of packed red blood cells (PRBC). No other maternal complications reported.

Patient resumed her regular menstrual cycle 1 month post-partum consuming 2-3 pads per day, moderately soaked lasting for 3 days until after 8 months when the patient experienced prolonged heavy menses consuming 4 pads per day which lasted for 10 days. She sought consult at a government hospital where she was initially managed as a case of abnormal uterine bleeding secondary to coagulopathy. Anemia was corrected.

Patient again had another episode of profuse vaginal bleeding, sought consult at another tertiary government hospital. Transvaginal ultrasound was done revealing a normal sized anteverted uterus with a poorly delineated mass located at the anterior low segment of the myometrial wall measuring 4.3cm x 3.8cm x 3.1cm with marked visuality. AV malformation and gestational trophoblastic neoplasia (GTN) was considered. Beta HCG was normal at 2.75 m/IU. Patient was advised to undergo uterine artery embolization but was referred to other institution due to lack of facilities. She was then discharged and was lost to follow-up until two weeks later, heavy vaginal bleeding recurred. Patient was admitted at another tertiary hospital and was transfused with 2 units PRBC and was managed as a case of abnormal uterine bleeding secondary to AV malformation, anemia secondary. Anemia was corrected however, no surgical intervention was done.

At the Emergency Room, patient came in with 3 days history of heavy vaginal bleeding, tachycardic

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and clinically pale. On internal examination, cervix is closed, uterus was not enlarged with no adnexal mass or tenderness with bleeding per vagina. On work-up, she was anemic with hemoglobin of 68mg/dl. Doppler sonography was done which revealed a normal sized anteverted uterus with a highly vascularised area at the right anterior lower uterine segment measuring 2.99cm x 3.16cm x 2.23cm. Doppler Studies showed very low resistance, RI was 0.24 and PI of 0.31 which was suggestive of AV malformation (Figure 1).

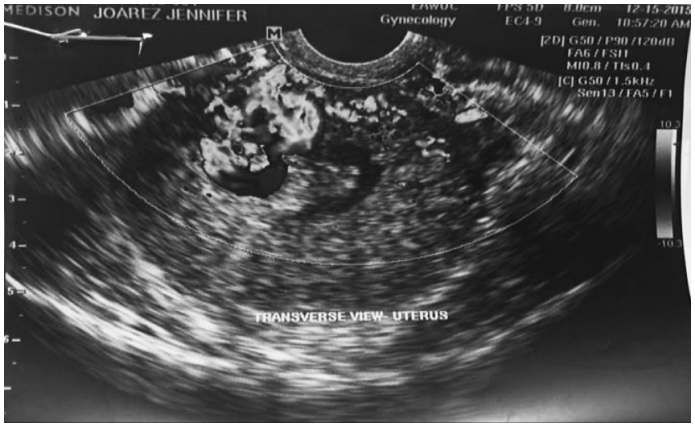

Figure 1. Doppler Ultrasound showing a highly vascularised right anterior lower segment of the uterus

Correction of anemia was done by blood transfusion and patient was started on anti-fibrinolytics and hematinics. Vaginal bleeding subsequently decreased. Patient was transfused with total of 6 units PRBC. Surgical plan for the patient was to do Uterine Artery Embolization however, due to financial constraints and lack of facility for the said procedure in our institution, other uterine vasculature occlusion procedures were discussed with the patient such as uterine artery and hypogastric artery ligation.

The patient underwent laparoscopic bilateral uterine artery ligation at her 13th hospital day. Intraoperatively, the uterus was normal looking, with grossly normal bilateral adnexa. The uterine arteries were explored on the lateral side of the anterior leaf of the broad ligament. (Figure 2) The external iliac, ureters, obliterated hypogastric artery and uterine arteries were identified. The Uterine Arteries were then ligated using Silk 1-0 suture (Figure 3). Patient tolerated the procedure well. Post-operatively, patient had stable vital signs with no episodes of vaginal bleeding. She was then discharged on the second post-op day.

At the patient's 5th post-operative day, transvaginal ultrasound was done wherein the previously noted 2.99cm x 3.16cm x 2.23cm highly vascularised area at the right anterior lower uterine segment has now disappeared with only note of peripheral vascularizations (Figure 4). She

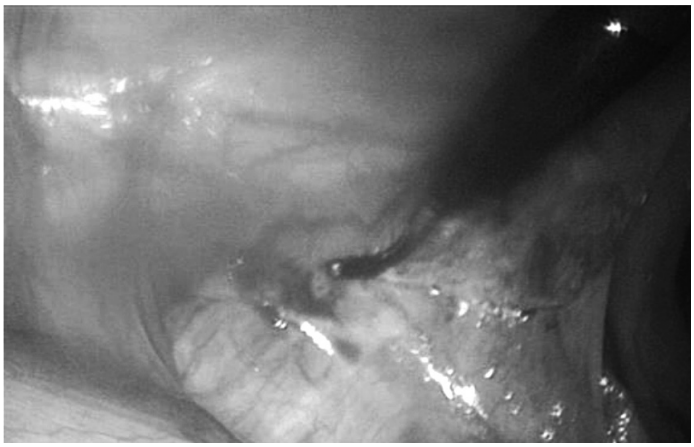

Figure 2. The uterine arteries were explored from the lateral side of the broad ligament

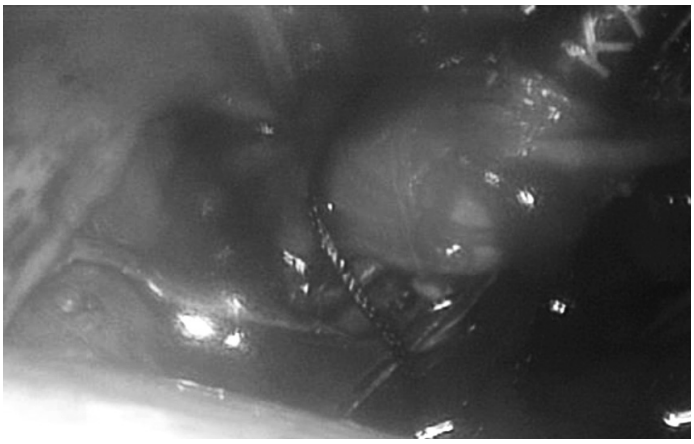

Figure 3. Uterine arteries were dissected and ligated using silk1-0 suture

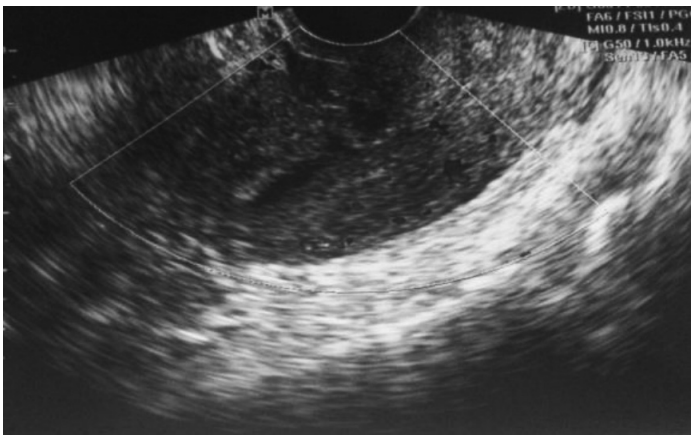

Figure 4. Transvaginal Sonography at 5th post-operative day. The Previously noted AV malformation was not appreciated.

then resumed her menses 1 month after the operation which lasted for 3 days, consumed 2-3 pads per day, moderately soaked. Doppler studies was done on her day 9 of cycle which showed no significant difference from the previous scan. (Figure 5)

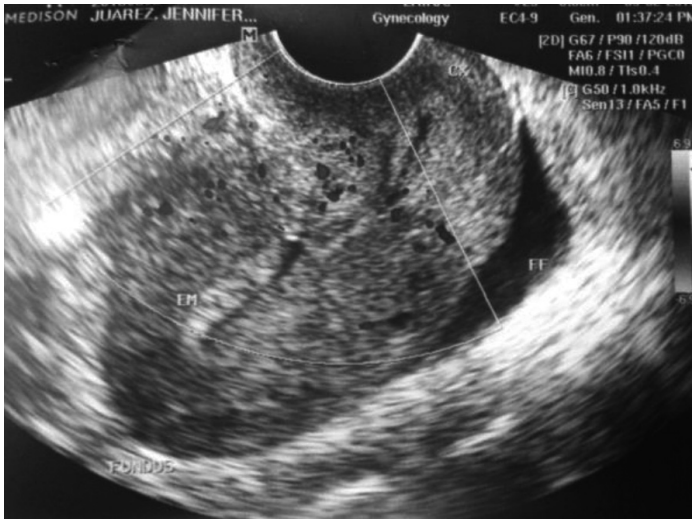

Figure 5. Transvaginal Sonography at 5th post-operative day. The Previously noted AV malformation was not appreciated.

DISCUSSION

Abnormal uterine bleeding (AUB) refers to any considerable changes from the normal menstrual cycle which may be deviation on its regularity, frequency, amount of bleeding. FIGO has devised a classification system, divided to 9 categories arranged according to acronym PALM-COEIN, to help in diagnosis and treatment of AUB. The PALM group are structural entities while the COEIN group are non-structural. Uterine AV malformation belongs to the less common AUB-N category along with other uterine conditions such as chronic endometritis, myometrial hypertrophy.[2]

Uterine Arteriovenous Malformations is a rare condition in gynecology. The first case was reported in 1926 by Dubreuil and Loubat. It can be congenital or acquired. Cases have been reported in our country, however, there are no actual studies regarding its incidence locally.

Clinically, these vascular anomalies most commonly present with abnormal uterine bleeding which can be severe enough to cause anemia and even shock.[3] It is also a rare cause of secondary postpartum hemorrhage occurring in up to 0.5-1.5% of pregnancies. Diagnosis has proved to be difficult however, one should suspect in patients presenting with secondary heavy vaginal bleeding without obvious cause requiring repeated medical attention. Patient has history of previous dilatation and curettage, trauma, uterine surgeries as in the case of this patient, has had caesarean section. Bleeding is usually painless. Uterine bleeding is thought to occur when the vessels of the AVM are exposed from iatrogenic sloughing of the endometrium during D&C or during the menstrual period.

Traditionally, diagnosis of AVM is made after hysterectomy and histopathologic examination. Diagnosis is done by pelvic angiography, the current gold standard. This invasive procedure is effective in identifying the feeders and facilitates Uterine Artery Embolization if a conservative procedure is planned. A good non-invasive method is a Doppler ultrasonography wherein AVMs appear as non-specific heterogenous or anaechoic spaces in the myometrium on gray scale ultrasound. Upon evaluation, the identified area will reveal a low-resistance blood flow with high peak velocities and evidence of turbulence. Its features, however, may be similar to other pelvic structures or pathology as in Gestational Trophoblastic Neoplasia. It may be differentiated from GTD by normal HCG levels as in our patient. In this case, series of Doppler ultrasonography was performed both revealing a highly vascularised area with low resistance. Pelvic angiography was not available in our institution and is expensive thus was not performed. CT Scan and MRI may also be useful in determining the size, extent, vascularity, and involvement of adjacent organs.

Treatment of Uterine AV malformation relies heavily on patient's hemodynamic stability, amount of bleeding, age and desire for fertility. Patient usually presents with torrential bleeding. In the presence of hemodynamic instability, resuscitation with IV fluids and blood products are vital.

In stable patients, expectant management can be considered. Medical management includes estrogens, progestins, methylergonovine, danazol. There has been a reported case of patient with AV malformation who was treated with Intravenous conjugated estrogens and oral Methylergometrine maleate. The bleeding regressed on the fourth day of therapy.[5] Methylergonovine maleate was thus suggested to induce tetanic myometrial contractions and reduce blood flow to the AVM, causing it to collapse; intravenous conjugated estrogens help by covering the hemorrhaging vessels with a proliferative endometrium. Since our patient was hemodynamically unstable and had recurrent episodes of vaginal bleeding, medical management alone was deemed not enough.

Uterine AV malformation has been treated traditionally by uterine artery ligation or hysterectomy. Currently, embolization is the more preferred treatment due its potential to preserve fertility and is minimally invasive. It is especially beneficial in young women because it does not appear to interfere with the menstrual cycle or pregnancy. The first arterial embolization procedure was done in 1972 for acute gastritis and then was subsequently used for trauma-related pelvic bleeding and post-partum hemorrhage. Embolic materials depend on the size and range of AVMs. These may be absorbable gelatine sponge, stainless steel coils, and polyvinyl alcohol. Of the cases reported since its utilization, 69% managed by a single procedure, 12.5% required repeat embolization, and 19%

required hysterectomy. As high as 93% overall efficacy has been reported.[5] Uterine Artery embolization has also been used to manage post-partum hemorrhage. And although this may be considered as the technique of choice by some authors, some practical issues have been considered as the procedure requires a radiologist trained in embolization techniques as well as the appropriate equipments for vascular intervention[6] which may not be readily available in some areas in resource-poor country as in the Philippines. In our patient, she was offered to undergo UAE, however, patient cannot support the procedure financially. The institution is also not equipped for this kind of procedure which is only available in specialized hospitals.

Other surgical managements reported less frequently include the hysteroscopic or laparoscopic guided coagulation of AVM, surgical removal of AVM, laparoscopic bipolar coagulation of uterine vessels, and ligation of the uterine artery.[7]

In 2013, Dr Icasas from Cardinal Santos Medical Center made a case report and literature review on uterine AVM. She reported 2 cases managed at their institution and reviewed 2 cases previously published in local literature. These four cases were managed differently and a flowchart for diagnosis and treatment was created. It was concluded that hysterectomy is still the treatment of choice for symptomatic patients no longer desirous of future pregnancy. Uterine artery embolization is preferred in centers with facilities and training to perform interventional techniques. And in resource-poor countries or when there is limited access to medical facilities, bilateral hypogastric artery ligation followed by excision of mass may be done.[8]

Uterine artery ligation is a relatively safe procedure typically performed in cases of Post-Partum Hemorrhage and also allows future child-bearing. This technique is said to be most successful if hemorrhage is of moderate degree and if originating from the lower segment. It is also useful for lower segment extensions or laceration. No data has been reported comparing the efficacy of uterine artery ligation and Uterine Artery Embolization in treatment of uterine AV malformation. However, reviews done by Sergent et al in 2004 on the role of vascular ligations, peripartum hysterectomy or arterial embolization in intractable post-partum haemorrhages, showed that arterial embolization and uterine artery ligation are equally effective in controlling post-partum hemorrhages with close to 100% success.[9]

Literature reviews showed that in post-partum women, ligation of all visible arterial supply of the uterus did not damage the uterus. A study done in 1964 on three women suffering from menorrhagia underwent ligation of uterine arteries resumed normal menstrual cycles. Question on the effect on fertility arises when Uterine Arterial Occlusion is performed. Successful pregnancies have been documented and prove that pregnancy after ligation of uterine arteries is possible. [10] In our case, uterine vasculature occlusion/ligation was an option considering the age, desire for future fertility, and economic status of the patient.

Diagnostic and Operative Laparoscopy has been a major breakthrough in the field of Gynecology. Throughout the years since it was first described, laparoscopy has evolved and is able to perform different procedures such as adhesiolysis, treatment of endometriosis, tubal sterilization, ovarian cystectomy, oophorectomy, salpingectomy, salpingostomy, and hysterectomy. It is relatively economical and safe.[11]

Laparoscopic uterine artery ligation is utilized as an adjunct procedure for laparoscopic myomectomy to prevent/lessen intraoperative bleeding. It was also used in treatment of symptomatic uterine fibroids and was found to reduce average fibroid volume of 54%. Identification and occlusion of uterine arteries requires expertise in retroperitoneal anatomy and skills. The uterine artery is dissected at the retroperitoneal space above the psoas muscle and lateral to the posterior broad ligament. Some surgeons do transvaginal occlusion of uterine arteries or medially through the posterior leaf of broad ligament close to the place where the uterine artery crosses the ureter. Occlusion can be done by a ligature, vascular clip or electrocautery. No reports on Laparoscopic bilateral uterine artery ligation on AV malformations have been reported however reviews on clinical outcomes when used for uterine leiomyomas improved clinical symptoms in the majority of the patients. [12] This patient underwent ligation of the bilateral uterine arteries laparoscopically, which produced a significant decrease in vascularizations as seen through ultrasound as early as 5th post-operative day.

Laparoscopic Uterine Artery ligation poses as an option in patients with uterine arteriovenous malformation which is relatively safe, more readily available and economical. A return to normal menstrual cycle is possible as evident in the case presented. ■

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