CASE REPORT

UTERINE ARTERIOVENOUS MALFORMATION: A CASE REPORT

Chandrashekar Murthy Y. M¹, Kiran A²

HOW TO CITE THIS ARTICLE:

Chandrashekar Murthy Y. M, Kiran A. "Uterine Arteriovenous Malformation: A Case Report". Journal of Evolution of Medical and Dental Sciences 2014; Vol. 3, Issue 69, December 11; Page: 14877-14882, DOI: 10.14260/jemds/2014/4000

ABSTRACT: Uterine arteriovenous malformation (AVM) is a rare condition, with less than 100 cases reported in the literature. Despite it being rare, it is a potentially life threatening disease. This case report describes 31- year-old women who presented with abnormal uterine bleeding. Trans abdominal sonography, colour and spectral Doppler imaging was performed, diagnosis was confirmed by non- invasive MRI scan. Laparoscopic bilateral uterine artery ligation was done successfully.

KEYWORDS: Arteriovenous malformation, embolisation, abnormal uterine bleeding, ultrasound scan, MRI, uterine artery.

INTRODUCTION: Uterine arteriovenous malformation (AVM) is a rare condition, with fewer than 100 cases reported in the literature.¹ It is a potentially life threatening condition, as patients may present with profuse bleeding. Colour Doppler ultrasound (US) and MRI provides a non-invasive method for diagnosing this rare condition. Conservative management, hysterectomy, uterine artery embolization and uterine artery ligation are the available treatment options. Hysterectomy is performed in woman who has completed family, though uterine artery embolization is a very effective minimally invasive alternative to hysterectomy, this procedure is done in very few hospitals in developing countries like India. Laparoscopic bilateral uterine artery ligation is a minimally invasive, very effective procedure and can be done in all most all hospitals where laparoscopic surgeries are performed.

CASE REPORT: A 31- year –old women presented with intermittent heavy vaginal bleeding since 6 months, she had caesarean delivery 8 years back and it was uneventful and she did not have any miscarriages. Patient denied having any curettage procedure performed.

On examination, she was anaemic with haemoglobin (Hb) level of (8.2 g/dl, vaginal examination showed a small amount of bleeding through external os of the uterus. In addition her serum Beta human chorionic gonadotropin (hCG) level was less than 2mIU/ml.

Transabdominal Ultrasonography of the pelvis showed uterus of 9 cm x 5 cm x 6 cm with endometrial thickness of 0.9 cm. There was increased vascularity at the fundus of the uterus. spectral US showed a peak systolic velocity (PSV) of 51 cm/s and resistance index (RI) of 0.66. uterine arteriovenous malformations (AVMs) was considered and which was diagnosed by MRI angiogram.

CASE REPORT

Fig. 1: MRI angiogram showing uterine arterio-venous malformation

We then proceeded with laparoscopic bilateral uterine artery ligation.

Pneumoperitoneum was created by inserting veres needle, abdomen entered with standard 3 ports technique, bladder dissected and pushed down with monopolar hook, broad ligament anterior leaf was cut and dissected then posterior broad ligament leaf was dissected and main uterine artery ascending branch was ligated with no 1 0 vicryl. The procedure was repeated on the other side.

No complications were encountered. The patients vaginal bleeding decreased post operatively and she was eventually discharged. During the follow up bleeding was completely stopped one week post operatively. The patient also indicated her desire to conceive in future.

DISCUSSION: Uterine vascular abnormalities are rare entities in gynaecology. However, it is a potentially life-threatening disorder in which patients present with vaginal bleeding that may be profuse and 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 transabdominal ultrasonography.²

Uterine arteriovenous malformations (AVMs) are classified as congenital and acquired types.³ Congenital uterine arteriovenous malformations are the results of abnormal embryonic development of primitive vascular structures which results in abnormal communications between arteries and veins,⁴ these are tend to have multiple feeding arteries, a central nidus and large draining veins.⁵

Acquired uterine arterio - venous malformations represents multiple small arterio - venous fistulas between intramural arterial branch and myometrial venous plexus.⁶ Acquired arterio - venous malformations tend to have single or bilateral uterine artery feeder without an extra uterine arterial supply or do not have characteristic nidus.⁷

Acquired uterine arterio-venous malformations are usually traumatic, results of prior dilatation and curratage (D&C), therapeutic abortions, uterine surgeries or direct uterine trauma,^{8,9}

CASE REPORT

&¹⁰. less commonly, endometrial carcinoma, Ca cervix, gestational trophoblastic disease (GTD)^{9,11&12}. Traumatic arteriovenous malformations usually present with intermittent but torrential vaginal bleeding. This pattern of bleeding is suggestive of arterial bleeding. In traumatic arterio venous malformations instrumental D&C may aggravate the condition.

Dubreuil and Loubat reported the first case of uterine arteriovenous malformation (AVM) in 1926.⁹ To date, there are fewer than 100 cases reported in the literature¹. Arteriovenous malformation (AVM) consists of proliferation of arterial and venous channels with fistula formation and a mixture of capillary-like vessels. Distinction between arteries and veins is difficult because secondary intimal thickening occurs in the veins due to increased intraluminal pressure.⁹

In this case, it is possible that a congenital form of uterine arteriovenous malformation (AVM) was present in the patient in the absence of uterine trauma, and presence of small feeding arteries. However, if it was truly congenital rather than acquired, it is unclear why the patient did not present earlier in life. In order for the endometrium to bleed, endometrial blood vessels and surface epithelium must both break down.¹ We can only postulate that the arteriovenous malformation (AVM) was not large or superficial enough for the vessels to be exposed at the endometrium and subsequently bleed.

With the availability of colour Doppler US, a non-invasive method can now be used to detect this rare condition. Nevertheless, digital subtraction angiography remains the gold standard of diagnosis. MRI is another noninvasive effective mode of diagnosis.

Gray scale ultrasound (US) can detect the presence of multiple tubular or “spongy” anechoic or hypoechoic areas within the myometrium of a normal endometrium.^{2,10} However, other conditions may present a similar appearance, such as retained products of conception, hemangioma, gestational trophoblastic disease, multilocular ovarian cysts, or hydrosalpinx. Thus, using colour and spectral Doppler US is important for obtaining more accurate information. A normal myometrial signal will show a PSV of 9–44 cm/s and RI of 0.6–0.8. In addition, uterine arteriovenous malformation (AVM) will exhibit intensely vascular and multidirectional flow (regions of juxtaposed reds and blues caused by multiple tortuous vessels of varying orientations). Spectral Doppler US will show high velocity (mean PSV: 136 cm/s), low resistance (mean RI: 0.3) flow, low pulsatility of the arterial waveform, and pulsatile high-velocity venous waveform.¹⁰

As mentioned above, retained products of conception may also give a hypervascular appearance with turbulent flow. Timmerman et al, reported two cases in which residual placental tissue was removed from patients diagnosed with uterine vascular malformation, thereby complicating the diagnosis further.¹³ Gestational trophoblastic disease (GTD), particularly in patients with associated uterine arteriovenous fistula, may also have similar US findings of increased uterine vascularity with a low RI.¹⁴ This patient did not have elevated hCG levels, which would be expected in GTD. However, a rare form of GTD, called placental site trophoblastic disease (PSTT), does not produce high levels of hCG, and instead produces high levels of human placental lactogen (hPL).¹⁵ However, the hPL levels were not measured in this patient because GTD and PSTT were not considered in the differential diagnosis.

Though Digital subtraction angiography (DSA) remains the gold standard for the diagnosis of AVM, non-invasive colour Doppler and spectral sonography is the effective good imaging modality in patients with abnormal uterine bleeding. MRI scan is also a good noninvasive alternative to DSA.

CASE REPORT

Management of uterine arteriovenous malformation (AVM) depends on the hemodynamic status, degree of bleeding, patient age, and desire for future fertility. Acute treatment involves stabilizing the patient's hemodynamic status, and stopping blood loss. Traditionally, hysterectomy was the treatment of choice. However, the patient's desire for future pregnancy is an important consideration, as there are now options available to avoid a hysterectomy. In stable patients who have the ability for close follow-up, expectant, and long-term medical management may be appropriate. Timmerman et al. presented 10 cases that demonstrated uterine arteriovenous malformation (AVM) features by colour Doppler US; of these, six cases spontaneously resolved.¹³

Because uterine arteriovenous malformation (AVM) is commonly diagnosed in women of childbearing age, angiographic embolisation has made hysterectomy no longer necessary.

However angiographic embolisation option is available in only few centres and only few Radiologists are trained in doing this procedure. Considering these points Laparoscopic uterine artery ligation is another effective and widely accessible alternative to angiographic embolisation.

However, hysterectomy remains the treatment of choice in post-menopausal patients or as an emergency treatment in life-threatening situations.¹⁶

This case report highlights the use of US and MRI for diagnosing uterine arteriovenous malformation (AVM) in a patient of childbearing age who presented with abnormal uterine bleeding. It also highlights our experience in performing laparoscopic bilateral uterine artery ligation in this patient, which provided an alternative option for her to retain her fertility.

Fig. 2: Ultrasonography showing regression of arteriovenous malformation after surgery

REFERENCES:

1. Hickey M, Fraser I. Clinical Implications of Disturbances of Uterine Vascular Morphology and Function. *BaillieresClinObstetGynaecol*.2000; 14(6):937-951.
2. Polat P, Suma S, Kantarcy M, Alper F, Levent A. Colour Doppler Ultrasound in the Evaluation of Uterine Vascular Abnormalities. *Radiographics*. 2002; 22:47-53.

CASE REPORT

3. Volgezang R L, Nemcek JR, Skrtic Z, Gorrel J, Lurain J R. Uterine arteriovenous malformations: primary treatment with therapeutic embolisation. *J Vasc Interv Radiol* 1991; 2: 517- 522.
4. Shiskawa T. Congenital arterio venous malformations involving pelvis and retroperitoneum: a case report. *Angiology* 1979; 30:70-74.
5. Powers J M, Horovpian D S. Central nervous system. In Damajanov I, Linder J (eds). *Anderson's pathology*, 10 th ed. St Louis, MO: Mosby – year book; 1996: 2713-2714.
6. Manolitas T, Hurley V, Gilford E. Uterine AVM: a rare case of uterine hemorrhage. *Aust NZ J Obstet Gynecol* 1994; 34:197-199.
7. Hoffman M K, Meilstrup J W, Shackelford P, Kaminiski PF. Arteriovenous malformations of the uterus: an uncommon cause of vaginal bleeding. *Obstet Gynecol Surv* 1997; 52:736 -740.
8. Diwan R V, Brennan J N, Selim M A et al. Sonographic diagnosis of arterio venous malformations of the uterus and pelvis. *J Clin Ultrasound* 1983; 11:295 -298.
9. Fleming H, Ostor A, Pickel H, Fortune D. Arteriovenous Malformations of the Uterus. *Obstet Gynaecol*. 1989; 73(2):209–213.
10. Huang M, Muradali D, Thurnston W, Burns P, Wilson S. Uterine Arteriovenous Malformations: Gray-Scale and Doppler Ultrasound features with MR Imaging Correlation. *Radiology*. 1998; 206(1):115–123.
11. Cockshot WP, Hendrickse JP. Persistent arteriovenous fistulae following chemotherapy of malignant trophoblastic disease. *Radiology* 1967; 88: 329 -333.
12. Follen MM, Fox H E, Levine RV. Cervical vascular malformations as a cause of antepartum and intrapartum bleeding in three DES exposed pregnancy. *Am J Obstet Gynecol* 1985;153:890-891.
13. Timmerman D, Bosch TVD, Peeraer K. Vascular Malformations in the Uterus: Ultrasonographic Diagnosis and Conservative Management. *Euro J Obstet Gynaecol Reprod Biol*. 2000; 92:171–178.
14. Ichikawa Y, Nakauchi T, Sato T, Oki A, Tsunoda H, Yoshikawa H. Ultrasound diagnosis of uterine arteriovenous fistula associated with placental site trophoblastic tumor. *Ultrasound Obstet Gynecol*. 2003; 21(6):606–608.
15. Gillespie A, Hancock B. Placental Site Trophoblastic Tumour. In: Hancock B, Seckl M, Berkowitz R, Cole L, editors. *Gestational Trophoblastic Disease*. 3rd ed: International Society for the Study of Trophoblastic Diseases; 2009. p. 420–429.
16. Delotte J, Chevallier P, Benoit B, Castillon J, Bongain A. Pregnancy after Embolisation Therapy for Uterine Arteriovenous Malformation. *Fertility and Sterility*. 2006; 85(1):228e1–6.

CASE REPORT

AUTHORS:

1. Chandrashekar Murthy Y. M.
2. Kiran A.

PARTICULARS OF CONTRIBUTORS:

1. Assistant Professor, Department of Obstetrics and Gynaecology, ESIC MC, PGIMSR, Bangalore.
2. Assistant Professor, Department of Obstetrics and Gynaecology, ESIC MC, PGIMSR, Bangalore.

NAME ADDRESS EMAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Chandrashekar Murthy Y. M,
No. 4/173/2, 2nd main,
Bovipalya, Mahalaxmipuram,
Bangalore-560086, Karnataka, India.
Email: drchandruym@gmail.com

Date of Submission: 13/11/2014.
Date of Peer Review: 14/11/2014.
Date of Acceptance: 05/12/2014.
Date of Publishing: 11/12/2014.