

Research Article: Acquired Uterine Arteriovenous Fistula

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Abstract:

Background: Vascular anomalies are a new emerging field involving several medical and surgical specialties. Uterine arteriovenous malformation is a rare condition with fewer than 100 cases reported in the literature. Uterine arteriovenous malformations may cause life-threatening abnormal genital bleeding in women of child bearing age.

Aim: The purpose of this study is to describe the role of high degree of suspicion and sonographic features of uterine arteriovenous malformation and to describe the role and clinical outcome after arterial embolization. Color Doppler ultrasound provides a non-invasive method for diagnosing this rare condition.

Material & Method: A 37 Year female patient presented with severe abdominal pain and episodes of refractory vaginal bleeding in a surgical set-up masquerading as suprapubic pain. A complete work up of the case was done. Trans abdominal ultrasound of the pelvis showed a moderate sized mass of mixed reflectivity in upper posterior myometrium.

Result: The sonographic grey scale findings of uterine Arteriovenous Malformation are nonspecific. The finding was subtle myometrium heterogeneity. Color Doppler sonography showed a tangle of vessels with multidirectional high velocity flow that produced a "Color Mosaic Pattern". Spectral Doppler ultrasonography revealed vascular lesion in the posterior wall of uterus near fundus in case and increased vascularity of the uterus with prominent vessel seen on the left lateral wall of the uterus. Spectral study showed high velocity, low resistance flow with high diastolic component. Peak systolic velocity was approximately 34.5 cm/s while resistive index was 0.39. The UVAM was classified as stage III. Schobinger staging system. She responded to uterine arterial embolization at higher center

Conclusion: Routine use of color and spectral Doppler sonography allows one to confidently make the correct diagnosis. Transcatheter arterial embolization is an excellent treatment option. Endovaginal sonography should be used to monitor post embolization outcomes. Uterine arteriovenous malformations are more common in essential for prompt diagnosis and treatment.

Keywords: Arteriovenous malformations, Doppler spectral sonography, Embolization, Menorrhagia.

I. Introduction

Vascular anomalies are a new emerging field involving several medical surgical specialties. A biological classification of vascular anomalies first proposed in 1982 [1] [2] [3]. Based on clinical finding, natural history, and cellular characteristics, the binary scheme was accepted by the International Society for the Study of Vascular Anomalies (ISSVA) in 1996 [4] [5]. Malformation is divided into slow-flow lesions Capillary malformation (CM), venous malformation (VM), lymphatic malformation. The fast flow arterial malformation is aneurysm, ectasia, stenosis, fistula or Arteriovenous malformation (AVM) [6] [7] [8].

Uterine arteriovenous malformation (UAVM) is a rare vascular condition with less than 100 cases reported in literature [9]. UAVM cause heavy vaginal bleeding, which may be postpartum, post abortion or menstrual and is usually refractory to conventional therapies. Uterine AVM is a dilatation of the intervillous space deep inside the myometrium allowing a direct flow from arterial system towards the venous system [10]. Such condition represents about 1-2% of all genital and intra-peritoneal hemorrhages. UAVM are classified as congenital or acquired. The congenital UAVM tends to have multiple feeding arteries as central nidus, a tangle of vessels with histologic characteristics of both arteries and veins and numerous large draining veins. Acquired UAVM represent multiple small arteriovenous fistulas between intramural arterial branches and the myometrial venous plexus [11] [12] [13]. They typically represent a single artery joining a simple vein.

Acquired UAVMS diseases are associated with conditions:

- ❖ Multiple Pregnancies
- ❖ Miscarriage
- ❖ Previous surgery

- Dilatation and curettage
- Termination of pregnancy
- Caesarean section
- ❖ Gestational trophoblastic neoplasia
- ❖ Endometrial carcinoma

Acquired UAVM is often nomenclature as uterine Arteriovenous Fistula [14] [15] [16] Grey-scale sonographic appearance can be nonspecific and have a range of manifestations including area of subtle myometrium in homogeneity, tubular spaces within the myometrium a intramural uterine, endometrial or cervical mass like region or sometimes as prominent parametrical vessels [17] [18]. The extent of mass effect is however minimal. Color Doppler typically shows serpiginous/tubular structures within the myometrium with a low resistance (RI-0.2-0.5) high velocity flow pattern on color Doppler interrogation.

A diagnosis of an AVM required an angiogram for confirmation. Pelvic MR imaging allows one to confirm the diagnosis of UAVM non-invasively [19]. Multiple serpentine flow related signal voids are typically seen in the uterine wall endometrial cavity and parametrium on T₁ and T₂ weight images. Contrast-enhanced dynamic MR angiography can depict complex serpentine as normal vessels and show early venous return [20].

II. Material And Method

Case Report

A 37 years old women with one live issue presented with bleeding per vagina following MTP done six months back for an approx. three months gestation . Her last child birth was five years ago She was started on hematinics, combined high-dose oral contraceptive pills (ethinyl estradiol 50 mcg plus levonorgestrel 250 mcg). The patient was given once daily from the 5th to 25th day of the cycle and tranexamic acid tablets during menstruation. She continued to have menorrhagia despite this treatment. On examination there was pallor. Per vaginaspeculum she has bleeding from Os and normal size uterus with bilateral fornixes free. A complete work up of the case was done. Her complete blood picture revealed a hemoglobin value of 6.8 gm %. Platelet count was 285,000/mm³ and coagulation profile was normal.

III. Result

Grey Scale Image

B/W USG revealed presence of a moderate sized (Approx. 18.5 x 24.3 mms) mass of mixed reflectivity in upper posterior myometrium .Uterus was normal sized and anteverted with a endometrial thickness(Single layer) of approx. 3.8 mm and small anechoic collection within the endometrium representing hematometra.

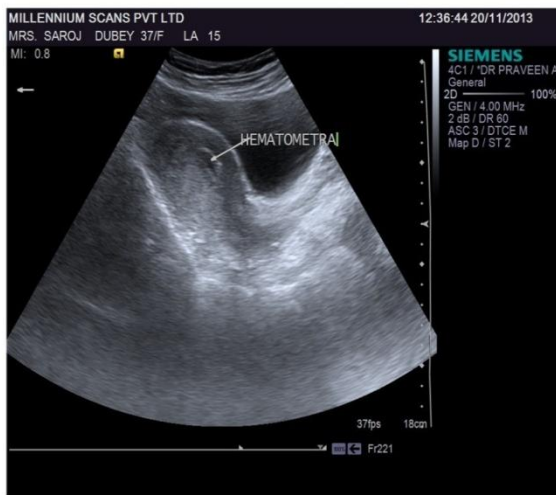


Fig I USG WITH HEMATOMETRA

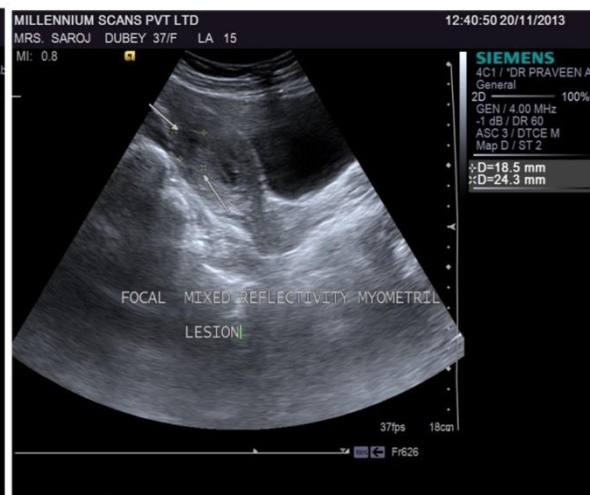


Fig II USG WITH MASS

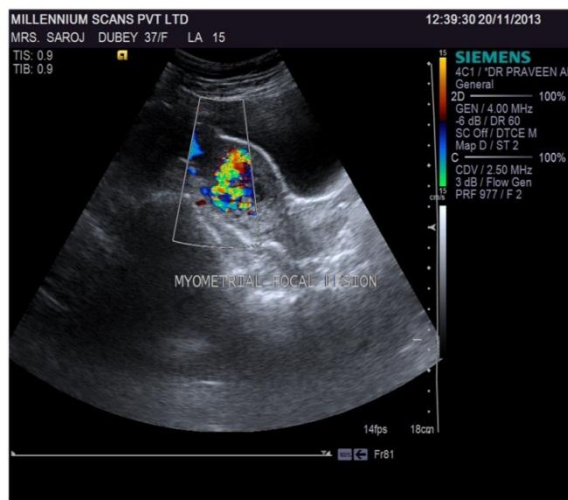
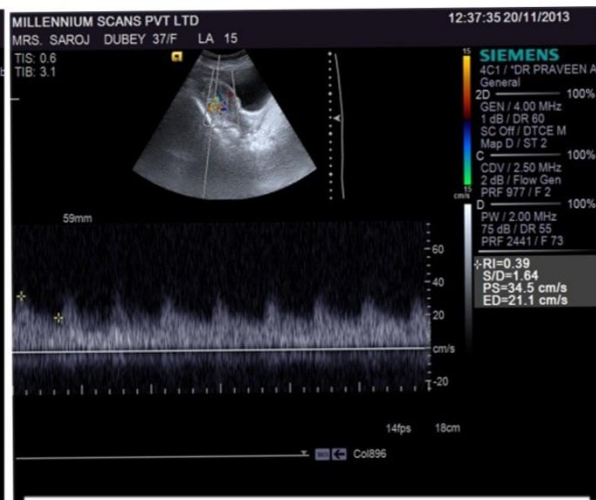


Fig III COLOR FLOW IMAGING



FigIV SPECTRAL STUDY SHOWING “COLOR MOSAIC PATTERN”

Doppler ultrasonography suggested an UAVM. Treatment options were discussed and she chose to undergo embolization at higher center. She was referred to PGIMER, Chandigarh, India and responded to arterial embolization.

IV. Discussion

Uterine vascular abnormalities are rare entities and potentially life-threatening disorders in which patients present with profuse vaginal bleeding and cause hemodynamic instability. Dubreuil and Loubat reported the first case of UVAM in 1926 [21]. UAVM consists of proliferation of arterial and venous channels with fistula formation and a mixture of capillary-like vessels. UAVM may be congenital or acquired. Congenital UAVM is believed to be caused from arrested vascular embryologic development resulting in anomalous differentiation in the capillaries and abnormal communications between arteries and veins. [22] [23] Congenital UAVM can have multiple vascular connections and may invade surrounding structures. Acquired UAVM are more common and usually follows a history of previous uterine trauma, Dilatation&Curettage, Dilation &Evacuation, Caesarean section or Pelvic surgery [24] [25]. The potential to develop abnormal communication between arteries and veins occur during the healing process, typically when a single artery joins a single vein. Acquired UAVM’s are associated with infection, retained POC, gestational trophoblastic disease, gynecological malignancies and exposure to diethylstilboestrol. With the availability of Color Doppler US a noninvasive method, UAVM’s can be detected if high degree of suspicion is present. Grey scale US can detect the presence of multiple tubular of “spongy” anechoic or hypo echoic areas within the myometrium of a normal endometrium [2] [4] [5]. Using Color and spectral Doppler US is important for obtaining more accurate information. A normal myometrium signal will show a PSV of 9-44 cm/s and RI of 0.6-0.8. in additional UAVM will exhibit intensely vascular and multi directional flow. Spectral Doppler US will show high velocity (mean PSV 136 cm/s) low resistance (Mean RI 0.3) flow, low plasticity of arterial waveform and pulsatile high velocity venous waveform [26] [27].

The ArteriovenousMalformation lesion can be classified according to the Schobinger staging system [28]

Table I Schobinger Staging of AVM

STAGE	CLASSIFICATION	CLINICAL FINDING
I	QUIESCENT	WARM, PINK-BLUE, SHUNTING ON DOPPLER
II	EXPANSION	ENLARGEMENT, PULSATION, THRILL, BRUIT, TORTIOUS VEINS
III	DESTRUCTION	DYSTROPHIC SKIN CHANGES, ULCERATION, BLEEDING, PAIN
IV	DECOMPOSITION	CARDIAC FAILURE.

Most AVMS are diagnosed by history and physical examination. If AVM is suspected, the diagnosis should be confirmed by US with color Doppler examination showing fast flow and shunting. MRI is also obtained to (a) Confirm the diagnosis determines the extent of the lesion and (b) Plan treatment. MRI shows dilated feeding arteries and draining veins, enhancement and show void on T-2 weighted imaging. Angiography is indicated if embolization or resection is planned to determine the how dynamics of the lesion [29].

As AVM is often diffuse involving multiple tissue plain and important structures, cure is rare. Goal of treatment is usually to control the malformation Intervention is focused on alleviating symptoms. Management options include remobilization, resection or a combination. Resection offers the best chance for long-term control but the reexpansion rate is high and extirpation may cause worse deformity. Intervention is determined by (a) the size and location of AVM (b) age of the patient and (c) Schobinger stage. Stage III & IV AVMS require intervention to control pain bleeding, ulceration or congestive heart failure [30].

Digital subtraction Angiography remains the gold standard for the diagnosis of AVM.[31] Finding with DSA include bilateral hypertrophy of uterine arteries that feed a tortious hypertrophic arterial mass with large accessory feeding vessels and early drainage into enlarged hypertrophic veins [32]. Management of UAVM depends on the hemodynamic status degree of keeping patient age and desire for future fertility. Acute treatment involves stabilizing the patient's hemodynamic status and stopped blood loss. Traditionally a hysterectomy was the treatment of choice. However the patient's desire for future pregnancy as there is newer option to avoid a hysterectomy [33]. In stable patients who have the ability for close follow-up expectant and long-term medical management may be appropriate more ever oral contraception as well as intramuscular and subsequent oral methyl ergo ovine maleate have been shown to be associated with regression of lesions based on U.S.[34]

Since the first description of a successful embolization treatment for UAVM in 1986, it has been commonly used in the emergency setting as well as less urgent circumstances. Various materials have been used including polyvinyl alcohol, histoacryl (glue), stainless steel coils, detachable balloons and hemostatic gelatin. Some cases may require repeat embolization. However, hysterectomy remains the treatment of choice in post menopause patients or as emergency treatment in life threatening situations [35]. Gonadotropin releasing hormone agonists have been used as a adjunct to embolization and 6 month of therapy reduce the size of UVAM subsequent uterine artery embolization resulted in complete disappearance of the AVM and normal cycles were resumed 3 months later [36]

V. Conclusion

Successful results have been reported after a shorter period of follow-up (6 weeks), but only a long-term follow-up will determine the true success rate of these procedures. The index case is interesting because the woman had menorrhagia and significantly increased myometrium vascularity on Doppler ultrasonography. A diagnosis of uterine AVM was suspected and she was prescribed high-dose oral contraceptive pills. There are reports of uterine AVMs responding to oral contraceptive pills, but this did not happen in the present case, and hence embolization was planned. Angiographic uterine artery embolization is the preferred therapy, for UAVM especially is young women who desire to preserve fertility [37]. Uterine AVMs are more common than previously thought. Clinical suspicion is essential for prompt diagnosis and treatment. Color Doppler sonography is important for proper assessment of these lesions. The size and site of lesion helps in deciding whether the patient needs medical or surgical intervention.

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