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Original Research Article

A study on diagnosis and management of arteriovenous malformation of uterus

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ABSTRACT

Background: Arteriovenous malformation is abnormal connection between an organ's arterial and venous circulation. In acquired AVM, history of uterine procedure seems inevitable. Their clinical feature is usually vaginal bleeding. It is diagnosed by 2-D ultrasonography combined with colour doppler. Most of the time they resolve spontaneously; however, if left untreated, uterine artery embolization or hysterectomy comes in hand. The purpose of this study was to evaluate the role of TVUS and colour doppler in the diagnosis and follow-up of treated cases of uterine AVM. This study also aims to evaluate different modalities to manage uterine AVM.

Methods: This was a retrospective study done at tertiary care centre from January 2018 to December 2019 to assess the presentation, treatment, and clinical pictures of patients with uterine AVM that were diagnosed with TVUS. Authors reviewed both (1) clinical data (2) ultrasound data of patients. The diagnostic criteria were "subjective" with a rich vascular network in the myometrium with the use of colour Doppler images and "objective" with a high PSV of 20 cm/sec in the vascular web.

Results: Thirteen patients met the diagnostic criteria mentioned above. Out of that 100% presented with on and off bleeding per vaginum. Recent and remote history of uterine procedures were in found in 84.6% (n=11) of cases. UAE was done in 53.8% (n=7) cases. Thirty-three (33%) (n=5) cases spontaneously resolved when closely monitored with serial imaging and serum beta- HCG levels. Hysterectomy was needed in 7.4% (n=1) of patients of AVM.

Conclusions: Uterine AVM occurred after unsuccessful pregnancies or uterine procedures. Triage of patients for expectant treatment, hormonal treatment vs intervention with uterine artery embolization based on their clinical status, which was supplemented by objective measurements of blood velocity measurement in the AVM, appears to be a good predictor of outcome.

Keywords: Arteriovenous malformation, Curettage, Uterine artery embolization, Uterus

INTRODUCTION

Arteriovenous malformation is a pathologic phenomenon describes as abnormal connections between arterial and venous end of circulation which was first described by Dubreil and Loubat in 1926 as "aneurysme cirsoide de l'uterus."¹ They are vascular lesions that may cause life threatening gynaecologic haemorrhage.^{2,3} They can occur when the thin wall of the abnormal vessels is disrupted either naturally after menstruation or artificially after instrumentation.^{4,5} AVMs are classified broadly as either

congenital or acquired.⁵ Acquired uterine AVMs are usually traumatic and result from previous uterine surgery including diagnostic or therapeutic curettage, caesarean delivery, or myomectomy.⁶⁻⁸ The clinical symptoms of uterine AVMs can appear gradually or suddenly, with patients most commonly presenting with heavy or irregular vaginal bleeding after a miscarriage, uterine surgery, or treated or untreated CSP. The natural history is variable; some cases slowly revert to normal circulation, and the condition disappears over a period of weeks to months, although some persist without

regression, which puts the patient at higher risk of haemorrhage. AVMs represent 12% of all pelvic and intraperitoneal haemorrhages; in 30% of cases, a blood transfusion is necessary.⁹⁻¹¹ With significant bleeding, treatment is of the essence, often in the form of uterine artery embolization (UAE). Importantly, curettage for patients with heavy vaginal bleeding because of an AVM may exacerbate the bleeding and may be life-threatening when the diagnosis of AVM has not been made before the intervention.¹² Angiography became the “gold standard” diagnostic method.¹¹ Most recently, transvaginal ultrasound scanning (TVUS) has emerged as an efficient, simple, and accessible diagnostic modality to detect and follow the vascular pattern of the AVM with the use of blood velocity blood flow indices.¹² The aim of this study is to review the value of TVUS in the diagnosis and treatment of suspected uterine AVMs to outline the disease’s natural history.

METHODS

This study is a retrospective review of medical records and ultrasound images to assess the presentation, diagnosis, management, treatment, and clinical outcomes of patients with uterine AVMs. All patients who presented to tertiary care centre from January 2018 to December 2019, and were diagnosed with pregnancy related uterine AVM on 2-dimensional TVUS were eligible for inclusion in the study. All patients are first examined with the grayscale mode followed by

colour/power Doppler interrogation. There were no restrictions on age or racial/ethnic origins for inclusion. Criteria for the sonographic diagnosis were (1) unusual, tubular, tortuous, anechoic structures seen by 2-dimensional grayscale ultrasound imaging on sagittal and/or transverse section of the uterus, which subjectively reveal an unusually rich vascularity with tortuous-appearing blood vessels that are concentrated in a small area of myometrium adjacent to the uterine cavity, with or without clearly visible products of conception (POC) that are detected by grayscale ultrasound imaging followed by colour or power Doppler imaging and (2) objectively a demonstration of high velocity blood flow within the vascular “web” with a peak systolic velocity (PSV) of >20 cm/sec. Authors used the highest PSV value to represent the AVM. Information to be reviewed included (1) TVUS data (images, measured dimensions, Doppler velocities, and resistive indices) and (2) clinical data (patient age, reproductive status, surgical history, clinical presentation, inciting event or procedure, clinical course, time intervals that included detection-to-resolution or detection-to-treatment, and treatment rendered). Patients selected for UAE based on clinical status or sonographic findings were referred to the department of interventional radiology. An operator performed the diagnostic angiogram using digital subtraction angiography, followed by the therapeutic intervention that consisted of embolization of the feeding vessel. The diagnostic and therapeutic intervention was always performed in bilateral uterine arteries.

Table 1: Patient demographics, clinical data and outcome of the 13 patients with EMV/AVM.

Case	Age	G/P	Clinical presentation	Surgical history\MTP pills	PSV (CM/S)	UAE	Transfusion	Clinical course	Outcome
1	22	G2P0A1L0	Caesarean scar pregnancy	D and E	90	Yes	Yes	Serial imaging+UAE	Resolution
2	24	G2P0A1L0	Incomplete abortion after MTP pills	MTP pills	88	Yes	No	Serial imaging+UAE	Resolution
3	18	G1P0A0L0	Incomplete abortion	D and E	78	No	No	Serial imaging + B-HCG+iron tablets	Resolution
4	33	G4P2A1L2	Missed abortion	D and E	90	No	No	Serial imaging +B-HCG+iron tablets+oc pills	Resolution
5	20	G2P1A0L1	Missed abortion	D and E	77	No	No	Serial imaging + B-HCG+ iron tablets	Resolution
6	26	G3P1A1L1	Incomplete abortion	D and E	90	Yes	No	Serial imaging+ UAE	Resolution
7	27	G3P0A2L0	Missed abortion	D and E	70	No	No	Serial imaging + B-HCG+ iron tablets	Resolution
8	34	G1P1A0L1	Missed abortion	D and E	86	Yes	Yes	Serial imaging + UAE	Resolution
9	25	G0P0L0	Increased menstrual bleeding PV	MTP pills	68	Yes	No	Serial imaging + B-HCG+iron tablets	Resolution
10	19	G2P0A1L0	Missed	D and E	90	Yes	No	Serial imaging	Resolution

Case	Age	G/P	Clinical presentation	Surgical history\MTP pills	PSV (CM/S)	UAE	Transfusion	Clinical course	Outcome
			abortion					+ UAE	
11	21	G2P1A1L1	Incomplete abortion	D and E	78	Yes	No	Serial imaging + UAE	Resolution
12	28	G3P2A0L2	Missed abortion	D and E	70	No	Yes	Hysterectomy	Hysterectomy
13	22	G2P0A1L0	Post D and E	D and E	80	Yes	No	Serial imaging + UAE	Resolution

RESULTS

There were 13 patients who met the diagnostic criteria of uterine AVM. Table 1 includes their mean age, gravidity, parity, and clinical diagnoses. Mean age was 24.4 years (range, 18-33 years). All patients presented with bleeding per vaginum. Overall surgical history included 11 patients (84.6%) with either recent or remote uterine surgery and 2 patients (15.4%) had history of ingestion of MTP pills. Eleven patients have undergone curettage (84.6%). Ultra sound were performed every 2 weeks or, in very few cases until resolution. Treatment was varied and included: expectant management alone with serial ultrasound scans and serial serum human chorionic gonadotropin until complete resolution (n=4) (33.3%), hormonal treatment (combined oc pills) were given in 1 patient (n=1) (7.6%), UAE (n = 7) (53.8%), hysterectomy (n=1) (7.6%). The overall episode time from diagnosis to resolution of the AVM was 2-15 weeks. Seven patients who required UAE, resolved after the procedure and required no further intervention. Embolization was able to stop the bleeding in these patients and lead to the resolution of the vascular pattern as judged by an ultrasound examination. Methotrexate was given to 1 of the 11 patients. Out of seven cases of patients who had history of D and E, 1 case was having undiagnosed CSP and had received one dose of methotrexate in private hospital. Patient presented with profuse vaginal bleeding at tertiary care centre, severe anaemia and USG showed AVM at CS site of 4×3 cm. Patient was managed with B/L UAE and serial imaging with USG. Hysterectomy was performed in 1 patient in the series where D and E was attempted and there was profuse haemorrhage requiring hysterectomy. Three patients in the series required a blood transfusion (23%): the first for intractable bleeding before hysterectomy, two were for symptomatic anaemia before undergoing UAE.

DISCUSSION

AV malformation is an abnormal connection between arterial and venous circulation system without intervening capillary bed.² Pelvic AV malformation is a rare cause of heavy and life-threatening vaginal bleeding. Mean age in the present study is 24.4 years. Yoon et al and Ilan et al in their study identified the mean age of their study population as 33.5 years and 31.8 years respectively.^{13,14} In this study 84.6% cases had history of

recent or remote uterine procedures. Similarly, Yoon et al and Ilan et al had 93% and 70% cases with such history respectively.^{13,14} UAE was modality of treatment successfully treating 53.8% of cases in this study without requiring further intervention thus having success rate of 100%. Yoon et al noted 100% cases requiring UAE out of which 61% cases didn't require any further intervention after primary UAE. Out of 39% cases remaining who failed to respond to UAE, required further intervention like repeat embolization, medical treatment with oc pills, danazol, methotrexate, occlusion of internal iliac artery and hysterectomy as a last resort.¹³ Ghai et al mentioned 93% cases being resolved with repeat UAE, only 6.6% cases were required to undergo hysterectomy due to presence of intractable bleeding after several cycles of UAE.¹⁵ Wang et al reported 83% cases being successfully treated with first attempt of embolization.¹⁶ Repeat cycles of UAE were required in 5.3% cases, proving success rate of 88% of the cases. No post-embolization complications were noted in present study. Ghai et al noted 26% cases having moderate pelvic pain post embolization being managed by analgesia and 13% cases having post-embolization fever being resolved spontaneously.¹⁵ Similarly in Wang et al 16% cases were having post-embolization pelvic pain and fever which were treated with analgesics and fever being resolved spontaneously.¹⁶ In present study 33% cases resolved spontaneously with serial imaging and serum HCG levels being closely monitored which was comparable to Elan et al (48%).¹⁴ After intractable bleeding per vaginum, hysterectomy was done in 7.4% in present study. In yoon et al 10% cases had undergone hysterectomy due to failure of repetitive UAE to resolve AVM. Blood transfusions were necessary in 23% cases in present study comparable to elan et al (11%).¹⁴

CONCLUSION

AVM, a relatively rare phenomenon, occurs as a consequence of intrauterine treatment procedures and usually has heavy vaginal bleeding. Clinicians should be familiar and be aware that the sonographic diagnosis can be based only by Doppler interrogation of the uterus. Spontaneous abortions, sharp uterine curettage and ingestion of MTP pills risks for an AVM.

Although in the past uterine artery angiography was the gold standard for diagnosis, presently transvaginal grey scale and colour Doppler ultrasound evaluation is

emerging as the simplest, best, and most cost-effective diagnostic imaging modalities. Triage of patients for either conservative follow up or treatment with UAE may be based on their clinical picture and by measurement of blood velocity in the area of AVM. Prevention of potentially life-threatening haemorrhage and preservation of fertility are some of the main advantages that embolization for AVMs has over more definitive surgical options, such as hysterectomy. Gynaecologists should be aware of the existence of AVM and rule it out before performing a curettage or aspiration for prolonged uterine bleeding.

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