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UTERINE ARTERIOVENOUS MALFORMATIONS – DIAGNOSIS AND TREATMENT IN SERIES OF CASES

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Abstract: Uterine arteriovenous malformation (UAVM) is a rare but potentially life-threatening diagnosis. Misdiagnosed and inappropriately treated Uterine arteriovenous malformation can lead to excessive hemorrhage because of specific conditions: abnormal direct multiple fistulous communication between an artery and vein without an intervening capillary bed. This makes a high-pressure gradient into venous system, enlargement of some venous vessels and multidirectional high velocity blood flow. There are different treatment options, one of which is embolization of uterine arteries.

The aim of this study is to present the etiology, clinical presentation, diagnostic options and therapeutic approaches to patients with Uterine arteriovenous malformation.

This study is a prospective follow-up of series of cases with Uterine arteriovenous malformation, which have different etiology and different uterine localization - cervical and corporal.

Our diagnostic method is ultrasonography – transvaginal, using both 2D grayscale, Color Doppler and Pulsed Wave Doppler. The machines used in this study are Samsung Hera and Medison.

We present four cases of arteriovenous malformation (one of them congenital and three others: after normal delivery, after caesarean section; dilatation and curettage were executed for all of them). All the cases resulted in hemorrhagic shock and were successfully treated later with uterine artery embolization.

Our study presents to the auditory unique cases initially misdiagnosed and mistreated with uterine curettage. They were correctly diagnosed later by ultrasonography and treated successfully through selective embolization of the feeding vessels. The research represents our personal experience in diagnosis and treatment of that life threatening condition and all cases were well illustrated with sonographic images, Color Doppler technic and embolization technics.

A good therapeutic outcome is a result of obtained collaboration between a gynecologist, an expert sonographer and an invasive cardiologist.

Uterine arteriovenous malformation can be safely and effectively treated with uterine artery embolization with even high chances of preserving women's childbearing function.

Keywords: Uterine arteriovenous malformations, embolization of uterine arteries, hemorrhagic shock, endovascular treatment.

Field: Medical sciences and Health

1. INTRODUCTION

Uterine arteriovenous malformation (UAVM) is defined as a communication between an artery and vein without an intervening capillary bed (Ruiz Labarta, Pintado Recarte, González Leyte, Arribas, Álvarez Luque, Cuñarro López, García-Montero, Fraile-Martinez, Ortega and De León-Luis, 2022). UAVM are divided into two groups – congenital and acquired or traumatic. Congenital UAVMs are the result of abnormal embryologic development of primitive vascular structures, which have multiple feeding arteries, a central nidus and numerous large draining veins (Hoang, Van, Trinh, et al. 2021). Acquired UAVM are complications of dilatation and curettage (D&C), cesarean section, gestational trophoblastic disease (GTD), endometrial cancer, infection (Nakashololo, Khan, Dunn, Snyman and Mh Ismail, 2021 May), (Lamrissi, Mabengui, Mourabbih, Jalal, Fichtali and Bouhya, 2022 May), (Hammad, Nausheen and Malik, 2022 July). They are described as arteriovenous fistulas between intramural arterial branches and the myometrial venous plexus infection (Nakashololo, Khan, Dunn, Snyman and Mh Ismail, 2021 May). UAVM is an uncommon but eventually life-threatening diagnosis (Hammad, Nausheen and Malik, 2022 July). Its clinical presentation is vaginal bleeding, which has potential to become excessive leading to hemorrhagic shock (Farias, Santi, Lima, Teixeira and De Biase, 2014 March). Therefore, UAVM should be suspected in every patient with severe metrorrhagia.

The aim of this study is to present the etiology, clinical presentation, diagnostic options and therapeutic approaches to patients with UAVM.

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2. MATERIALS AND METHODS

This study is a prospective follow-up of series of cases with UAVM, which have different etiology and localization.

Our method of diagnosis is ultrasonography – transvaginal, using both 2D grayscale, Color Doppler and Pulsed Wave Doppler or Samsung machines.

Digital subtraction angiography is a unique invasive method for both diagnostic and therapeutic purposes. Uterine artery embolization is the gold standard for UAVM treatment.

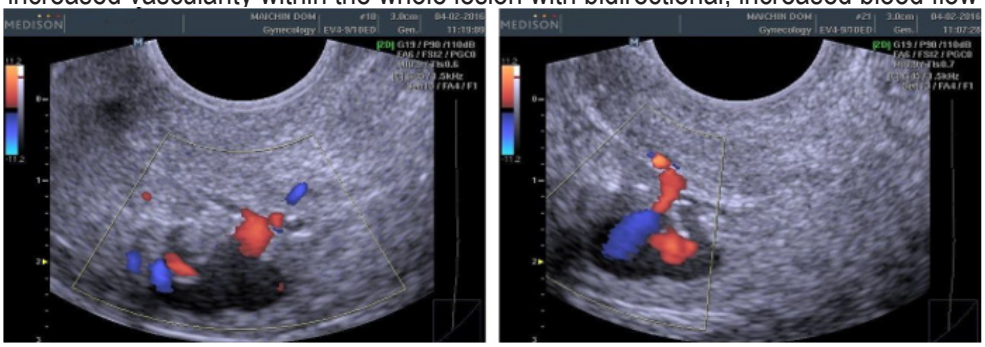
3. RESULTS

Our study presents unique cases of UAVM with different localization and successful embolization of the feeding vessels.

3.1. Case 1

A 20-year-old female, G0P0A0, presented to the emergency room with severe vaginal bleeding which has begun after intensive physical exercise. She denied any previous operative procedures incl. curettages and admission of antithrombotic medications. After admission to the hospital the patient underwent dilatation and curettage (D&C). She stayed in the reanimation unit for 5 days because of an excessive hemorrhage that led to a hemorrhagic shock and a hemotransfusion. An attempt to vaginal ligation of the uterine arteries was made without success. On the fifth day the patient was diagnosed with cervical AVM, and urgent endovascular embolization of the uterine arteries was performed.

Figure 1. TVUS Color Doppler of hypoechoic mass, localized in the uterine cervix, presenting increased vascularity within the whole lesion with bidirectional, increased blood flow



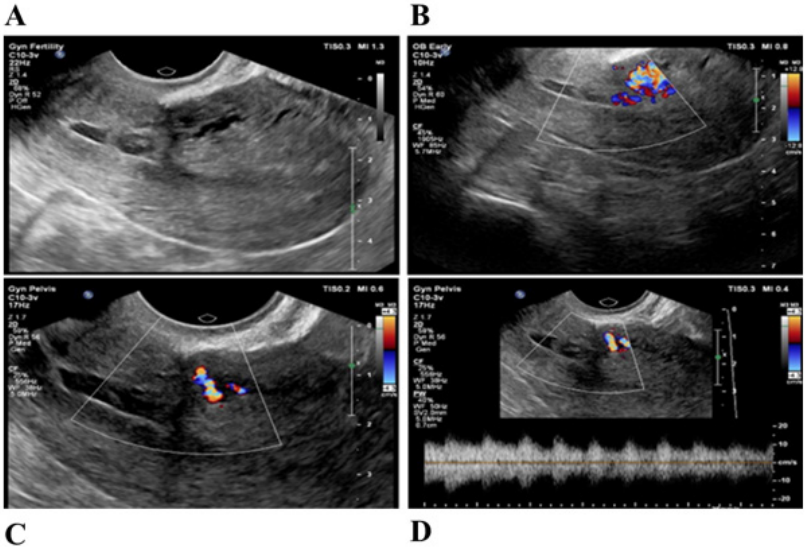
Source: Tsankova M., University hospital of obstetrics and gynecology „Maichin Dom“ Department, Medical University, Bulgaria

In Cases 2, 3 and 4 we are presenting uterine AVM in corporal part of the uterus

3.2. Case 2

A 39-year-old female, G2P1(PN)A1 was admitted to the gynecology department due to severe intermenstrual metrorrhagia. The patient reported one D&C due to spontaneous abortion in 12-th g. w. 13 years ago. There was no history of admission of any antithrombotic medications. The initial treatment was dilatation and curettage (D&C). In the postoperative period another episode of vaginal bleeding led to a second D&C which resulted in hemorrhagic shock and reanimation procedures. A month later the uterine AVM was diagnosed, and embolization of the uterine arteries was performed.

Figure 2. A. 2D – Gray scale TVUS revealing ill-defined hypoechoic mass in the myometrium of anterior uterine wall. B. and C. – Color Doppler sonography of the presented mass showing increased vascularization with aliasing, turbulent high-velocity flow. D. PW Doppler applied to the mass – revealing peak systolic flow and low resistance.

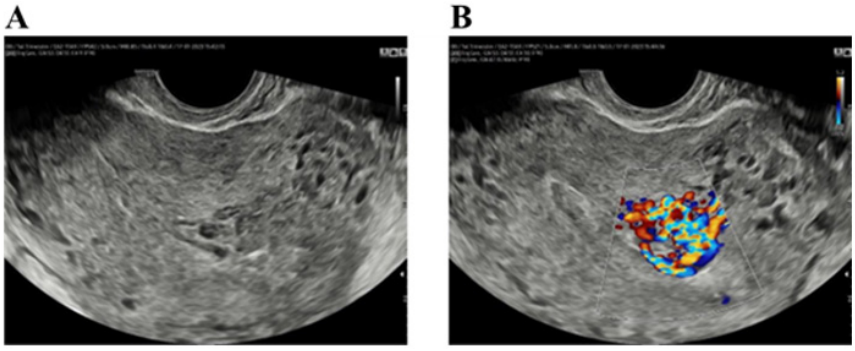


Source: Tsankova M., University hospital of obstetrics and gynecology „Maichin Dom“ Department, Medical University, Bulgaria

3.3. Case 3

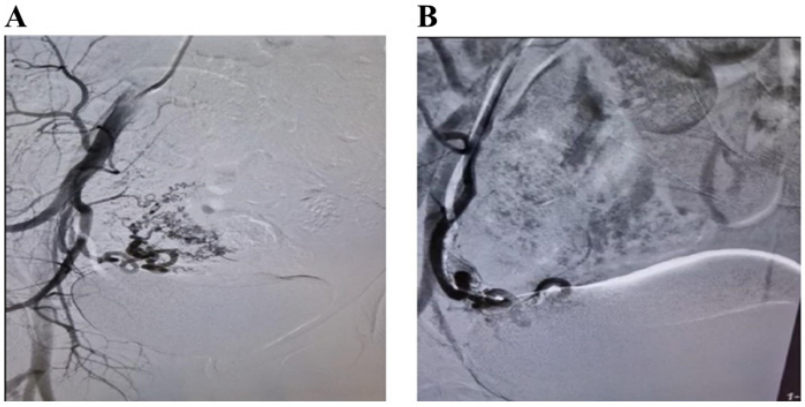
A 32-year-old female, G3P2(SC)A1 presented to the emergency with profound and continuous intermenstrual vaginal bleeding. The patient had a history of two cesarean sections and one D&C due to spontaneous abortion. The patient complained of often episodes of heavy menstruations, which have been treated with hormones (both oral contraceptives and gestagens), hemostatic medications without a result. They led to secondary anemia and impaired quality of life, including disorders of psychiatric spectrum (depression). The patient was sent to the hospital for a therapeutic uterine curettage. A transvaginal ultrasound (TVUS) was performed. On a 2D-Doppler image an enlarged uterus with no signs of intra- or extrauterine gestation was shown. An anechoic mass in the anterior wall was visualized, which after application of Color Doppler US showed increased vascularity and color aliasing phenomenon. Pulsed Wave Doppler revealed high velocity flow with low resistance. This led to diagnosing UAVM. The patient was referred to a cardiology department, where uterine artery embolization was performed. Digital subtraction angiography was performed and revealed increased vascularization in the arterial phase. After the embolization, the arterial phases show reduction in the vascularization of the UAVM.

Figure 3. A. 2D Gray scale TVUS of lesion within the myometrial layer with heterogeneous echogenicity (compared to the previous cases). B. Application of Color Doppler reveals high number of vascular structures with aliasing and multi-directional blood flow.



Source: Tsankova M., University hospital of obstetrics and gynecology „Maichin Dom“ Department, Medical University, Bulgaria

Figure 4. Fluoroscopy images obtained during digital subtraction angiogram of the right uterine artery. A. Before treatment – during the arterial phase a tortuous and dilated uterine artery is presented, suspicious for UAVM. B. Presents the obstructed blood flow of the described vessel after the embolization has been performed.



Source: Petrov, I., Cardiology, Angiology and Electrophysiology Department, Acibadem City Clinic Cardiovascular Center, Bulgaria

In our cases -2-coin catheters 3/4and 4/8 mm and 2 flacons particles PVA 100 micr. both sides.

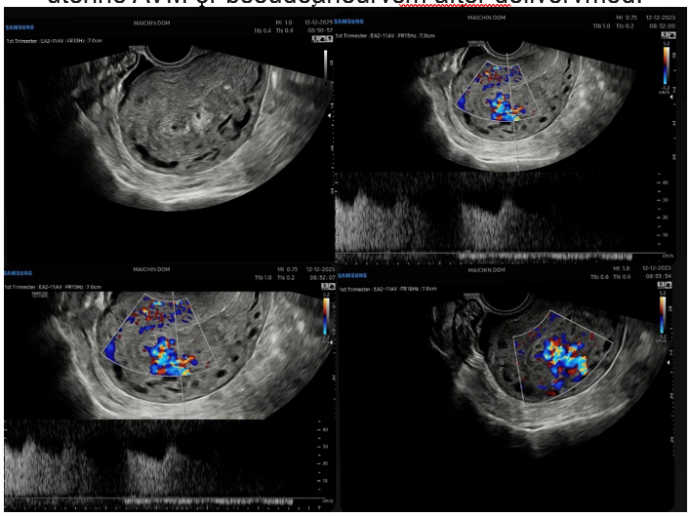
The coil embolization procedure is a catheter - based precise closure of abnormal blood in aneurysmal malformations. Once in a place, the catheter inserts different embolic agents, particles made of gelatin or plastic, that will block high velocity blood flow of uterine arteriovenous malformations and reaches directly abnormal communication between intramural arterial branches and myometrial venous plexus.

The presented cases are fully documented and therefore can be presented to the audience. Our first case was reported in 2016 and the next cases coming in 2022-2023.

3.4. Case 4

The last case of 2023 year. A patient, G1P1(PN)A0, 25 days after normal delivery. The placental period of delivery was normal. She has three episodes of heavy bleeding and uterine curettage in the postpartum period. The patient attends to the emergency department due to heavy bleeding. On Color Doppler has been diagnosed atypical low velocity blood flow of vessels in the uterus, AVM or pseudoaneurysms. An urgent embolization was performed.

Figure 5. Group of images – Gray scale sagittal scan of uterus with clearly visible uterine cavity with hypoechoic “lakes” surrounded by hyperechogenic irregular lines and color Doppler with multidirectional blood flow, aliasing effect and low resistance index of uterine branches inside. Specific characteristics of uterine AVM or pseudoaneurysm after delivervmed.



Source: Tsankova M., University hospital of obstetrics and gynecology „Maichin Dom“ Department, Medical University, Bulgaria

4. DISCUSSIONS

Uterine arteriovenous malformations were described by G.Dubreil and E.Loubat in 1926 (Dubreil and Loubat, nd). Although the incidence of this condition is considered relatively rare, a prospective study of 959 women found UAVM in 5,2% of women after D&C and in 0.22% of women after delivery. Only one of all UAVM was classified as clinically significant (Yazawa, Soeda Hiraiwa, et al. 2013). This may be since most of the studies include only symptomatic UAVM or have a retrospective model.

UAVM can be diagnosed using various imaging studies – ultrasound, computed tomography (CT), magnetic resonance imaging (MRI), digital subtraction angiography (DSA). The initial investigation is usually ultrasound, trans-abdominal or transvaginal. On 2D Gray scale ultrasound UAVM appear as heterogeneous, ill-defined mass with multiple hypoechoic structures of different size, eventually with endometrial and/or myometrial thickening. Nakasholo, Khan, et al. 2021 May), (Timmerman, Wauters, Van Calenbergh, Van Schoubroeck, Maleux, Van Den Bosch and Spitz, 2023 June), (Timor-Tritsch, Haynes, Monteagudo, Khatib and Kovács, 2016 June), (Vijayakumar, Srinivas, Chandrashekar and Vijayakumar, 2013). Typical findings on Color Doppler ultrasound are multidirectional blood flow with aliasing phenomenon, localized in different zones (in myometrium or rarely uterine cervix) with high velocity blood flow rate and low velocity indexes (Timmerman, Wauters, Van Calenbergh, Van Schoubroeck, Maleux, Van Den Bosch and Spitz, 2023 June), (Timor-Tritsch, Haynes, Monteagudo, Khatib and Kovács, 2016 June), (Vijayakumar, Srinivas, Chandrashekar and Vijayakumar, 2013). Peak systolic velocity flow can be used as a sign for UAVM prognosis (Timmerman, Wauters, Van Calenbergh, Van Schoubroeck, Maleux, Van Den Bosch and Spitz, 2023 June), (Timor-Tritsch, Haynes, Monteagudo, Khatib and Kovács, 2016 June), (Vijayakumar, Srinivas, Chandrashekar and Vijayakumar, 2013). Results of a study of 30 patients with UAVM showed that UAVM with peak systolic velocity flow (PSVF) <0.39m/sec have a good possibility of spontaneous resolution, whereas UAVM with PSVF >0.83m/sec usually require conservative or surgical treatment (Timmerman, Wauters, Van Calenbergh, Van Schoubroeck, Maleux, Van Den Bosch and Spitz, 2023 June). Other imaging tools can also be used, if necessary, for example CT angiography and MRI with angiography (Masood, Rana, Khan, et al. 2022). Hysteroscopic identification of UAVM is a feasible option for diagnosis and treatment (Calzolari, Cozzolino, Castellacci, Dubini, Farruggia and Sisti, 2017 April-June). Digital subtraction angiography (DSA) is the gold standard for diagnosing UAVM. It provides detailed images and information regarding the arterial supply of the lesion, its size and venous drainage. Because of the invasiveness of the procedure, its use is limited only to cases required to be treated with endovascular embolization, transforming the procedure from diagnostic to therapeutic (Masood, Rana, Khan, et al. 2022).Histopathologically, UAVM is described as a very thick venous structure, in which the arteries have incomplete or complete absence of elastic membranes and a completely absent tunica muscularis media (Calzolari, Cozzolino, Castellacci, Dubini, Farruggia and Sisti, 2017 April-June). Treatment of UAVM depends on the clinical presentation. As earlier discussed, most of the UAVM are diagnosed only after becoming symptomatic, so a treatment plan is intended. It depends on the regularity and intensity of bleeding. Hemodynamically stable patients with mild to moderate bleeding, which has resolved spontaneously, can be treated conservatively for up to 6 months, using medicaments such as GnRH antagonists and danazol. This approach is associated with a low chance of success (Nonaka, Yahata, Kashima and Tanaka, 2011 February). All other patients, inappropriate for conservative approach, especially those presenting with recurrent and/or profuse, life-threatening bleeding, possibly leading to hemorrhagic shock (as the presented cases) should be treated interventional and/or surgically. Transcatheter uterine artery embolization and ligation of uterine/internal iliac artery are minimally invasive and fertility-preserving methods with high success rates (Kim, Shin, Kim, Yoon, Ko, Gwon, Yang and Sung, 2014 March). A systematic review of uterine artery embolization of UAVM, which includes 371 patients in 95 studies, showed global success rate of 88.4% with a low risk 1,8% of adverse outcomes (one case of disseminated intravascular coagulopathy, one uterine artery rupture, one non-flow limiting dissection of the internal iliac artery and three cases of pulmonary embolism) even in patients with later pregnancies. 15,6% of all patients experienced mild complications defined as post-embolization syndrome – pelvic/abdominal pain with or without fever, transient of permanent amenorrhea (Ruiz Labarta, et al. 2022). Hysteroscopy is another possible option, also preserving fertility (Calzolari, et al. 2017 April-June). Hysterectomy is definitive surgical treatment option in case of failure of minimally invasive methods or lack of desire for future childbearing (Ore, Lynch and Rumsey, 2015 January). There are a lot of examples of successful pregnancy after UAE or UAVM described in the literature. (Ruiz Labarta, et al. 2022), (Delplanque, Le Lous, Proisy, Joueidi, Bauville, Rozel, Beraud, Bruneau, Levêque, Lavoué and Nyangoh Timoh, 2019 January). Some of the reported obstetric complications in this specific group are spontaneous abortions, placenta previa or accreta, postpartum hemorrhage and higher rate of cesarean

section in comparison to the normal population. (Ruiz Labarta, et al. 2022), (Soeda, Kyozuka, Suzuki, Yasuda, Nomura and Fujimori, 2014).

A differential diagnosis of UAVM includes retained products of conception, gestational trophoblastic disease (GTD), uterine postpartum pseudo aneurysmal retained parties of placenta and abnormally invasive placenta (Nakasholoło, Khan, Dunn, Snyman and Mh Ismail, 2021 May). Serum levels of b-Hcg are elevated in cases of GTD and uterine pseudoaneurysm.

5. CONCLUSIONS

The presented cases clearly demonstrated typical ultrasound images of uterine AVM because of excessive uterine bleeding and related complications. Uterine artery embolization proved to be an effective in our cases life-saving procedure. The study shows different clinical situations for iteral AVM: congenital, after normal delivery, after C section and after uterine curettage.

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