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Case Report

Repeated pulmonary embolism with cardiac arrest after uterine artery embolization for uterine arteriovenous malformation: A case report and literature review

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ABSTRACT

Objective: Uterine arteriovenous malformation (AVM) is an abnormal and nonfunctional communication between uterine arteries and veins, currently managed by uterine artery embolization (UAE). Pulmonary embolism (PE) is the most severe and life-threatening complication of this procedure.
Case report: We report a case of 27 year-old woman with heavy vaginal bleeding and abdominal pain caused by AVM. UAE was performed uneventfully, but 2 h after the procedure the first attack of pulmonary embolism occurred, treated by anticoagulation therapy. Second attack happened on the third postinterventional day. Considering vaginal bleeding, continued extracorporeal membrane oxygenation (ECMO), and suspicion of embolic particles arising from uterus, a subtotal hysterectomy was done. Patient was discharged two weeks following surgery, after complete recovery.
Conclusion: Although AVM is managed by UAE, clinicians must be aware of complications. To avoid PE, we suggest only large sized microspheres for carefully selected patients.

Introduction

Uterine arteriovenous malformations (AVM) are abnormal and nonfunctional communications between uterine arteries and veins [1]. In the past treatment option for AVM was hysterectomy in cases without realized reproductive desires, and uterine artery embolization (UAE) was performed only exceptionally. With increased experience and development of techniques and equipment, embolization become a first choice of treatment for patients of all age groups. As each invasive procedure, UAE has numerous complications, and pulmonary embolism is the most severe and life-threatening one, reported to be successfully treated by therapeutic anticoagulation [2,3]. The purpose of this paper is to report a patient suffering of repeated pulmonary embolism with cardiac arrest after UAE for uterine AVM treated by hysterectomy, with prolonged hospital stay, but good outcome. According to the literature data available, it is the first case of repeated PE and cardiac arrest after UAE for uterine AVM treated by hysterectomy and anticoagulants.

Clinical report

A 27 year-old patient was referred to our Department in April 2018 for abdominal pain and heavy menstrual bleeding. These sufferings lasted for 12 months. In September 2017, she had the same complaints, and ultrasound (US) performed revealed formation 27×32 mm in diameter, which was interpreted as submucous uterine myoma. For abundant bleeding dilation and curettage (D&C) procedure was done, and histological evaluation revealed chronic endometritis and glandular fibrous polyp. From September 2017 she received specific form of combined oral contraceptive pills

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for control of abnormal uterine bleeding, throughout a period of 6 months. In March 2018, she had extensive vaginal bleeding again and was urgently hospitalized in regional hospital with hemoglobin level of only 64 g/l. Magnetic Resonance Imaging (MRI), performed immediately thereafter, revealed dilation of numerous venous vessels of pelvis, but details related to communication between vessels were unclear.

Regarding obstetric history, she is gravida 3, para 2, with one molar pregnancy in 2008. For this molar pregnancy D&C was performed, followed by blood transfusion and four courses chemotherapy according to the protocol for gestational trophoblastic diseases. Her last episode of vaginal bleeding was from March 5 to April 10, 2018. On admission, she was apyretic, adiaphoretic, anicteric, with body mass index 26.2, and her heart and respiratory rates were within normal range (78 beats/minute and 16 respirations/minute, respectively).

MRI with contrast done in our institution revealed that structure of the myometrium is not homogeneous, and the crimped vessels expanded up to 6–12 mm. In post-contrast MRI images pathological features were observed: in arterial phase enlarged arteries were identified along with an early contrasting of extremely dilated veins; a moderate amplification of the signal from the myometrium and venous vessels was noticed, which overall confirmed the presence of uterine AVM.

Treatment options were discussed with the patient, and she has chosen UAE. UAE was performed on the next day. The right femoral artery was punctured under local anesthesia and the right and left iliac arteries were selected. Obtained angiograms visualized pronounced crimp, expanded branches of the left and right uterine arteries in the form of an extensive pathological mesh-like feature of vessels (Fig. 1A). At the level of proximal portion of uterine arteries, the catheter was filled with 500–700, 700–900, 900–1200 μm occlusive particles, and special MREY spiral-shaped items in diameter of 8 × 8 mm and 10 × 8 mm were introduced into uterine arteries for occlusion purposes. After that, angiography confirmed stagnant flow (Fig. 1B–D).

Two hours after the procedure, the patient presented with sudden dull chest pain, shortness of breath, and pale skin, accompanied by sweating, acrocyanosis, hypotonia of 60/30 mmHg, and tachypnea (35 respirations/minute). After immediate resuscitation, she was transferred to the intensive care unit (ICU), with suspect pulmonary thromboembolism. She had cardiac arrest without palpable pulses, and got cardiopulmonary resuscitation (CPR) for 4 min. After achievement of normal cardiac activity, she was hemodynamically unstable. The subsequent echocardiography (ECG) revealed right atrium and ventricle enlargement, moderate tricuspid insufficiency and inferior vena cava dilatation with elements of spontaneous echo contrasting. Systolic pressure in the pulmonary artery was 55 mmHg. Extracorporeal membrane oxygenation was initiated and the patient was transferred to the ICU of cardiac surgery center. Computer tomography (CT) showed extensive filling defects in the left and right trunk of pulmonary arteries, lower arteries of the left pulmonary lobe, and right atrioventricular enlargement. In ICU patient received anticoagulants, antibiotics, beta-receptor blockers, diuretics, gastroprotective drugs, cardiotonic agents, blood components transfusion, along with the support of all vital functions as well as careful monitoring of vital parameters. Three days after UAE, vaginal bleeding started and the first 2 days was found to be normal menstrual bleeding. On the third day, bleeding became heavy with blood clots, followed by abdominal pain and extensive dry coughing. The second CT was performed, revealing both pulmonary arteries thromboembolism, right atrioventricular enlargement, and bilateral exudative pleuritis. CT of pelvis organs detected hectically traced vessels in diameter of 0.5 cm, forming a conglomerate of coils, while details of communication between vessels were unclear. Considering the actual clinical presentation (extensive vaginal bleeding, continued ECMO, condition after UAE and suspicion of thromboembolism with embolus arising from uterus) we decided to perform a subtotal hysterectomy. Intraoperative findings confirmed CT interpretations: uterus in diameter of 6.0 × 7.0 × 4.5 cm with dilated both arteries and venous vessels, was sent to histological examination (Fig. 1E and F). The patient stayed in the ICU for 5 days, until systemic and hemodynamic stabilization. After that, she was transferred to gynecological department on the 11th day, where she recovered completely.

Discussion

Uterine AVM, didactically classified as congenital or acquired (traumatic), is not frequent in clinical practice, and our knowledge is limited to isolated case reports and small case series [4–6]. Congenital AVMs are the consequence of abnormal vascular differentiation arising from arrested vascular embryologic development and resulting in anomalous differentiation and communication between arteries and veins [5]. Acquired AVM is usually related to previous interventions on uterine tissue (myomectomy, cesarean section, curettage), retained products of conception, infection, gestational trophoblastic diseases, or cervical and endometrial carcinoma [1,7,8].

Regarding contemporary diagnostic tools, uterine AVM can be diagnosed by ultrasound scan, CT, MRI and angiography. Grey-scale ultrasound scan can reveal multiple anechoic structures with serpentine contours in myometrium. Color Doppler scan improves sensitivity and reliability, enabling visualization of vascular tangles of tortuous vessels with high-velocity and low-resistance flow [9]. In our patient grey-scale ultrasound scan, performed in the regional center, identified formation within posterior uterine wall, and misinterpreted it as a submucous uterine myoma in diameter of 27 × 32 mm. CT and MRI are more accurate in diagnosis of AVM. Both diagnostic tools very precisely define the localization, diameter, vascularity, as well as eventual involvement of adjacent structures [10]. In our Clinic, we performed MRI with contrast and precisely identified uterine AVM without the presence of uterine myoma.

Angiography was confirmed to be very reliable for diagnosis of AVM, with the additional advantage of immediate treatment by embolization [11]. Therefore, this procedure was introduced into clinical practice, and was found to be the gold standard for precise determination of AVM [12]. In our case angiography showed pronounced crimp, hypertrophy of both uterine arteries feeding a tortuous, hypertrophic arterial mass with large accessory feeding vessels, and with presented early venous contrast, revealing that this vascular anomaly is AVM. From technical point, an initial pelvic arteriogram was obtained to delineate the dominant arterial supply to the malformation. After that, selective catheterization of the dominant uterine artery was performed and a coaxial system and microcatheter was used for embolization. We used embolic materials Embosphere, biocompatible and non-absorbable, with cellular-adhesive properties enabling complete and prolonged mechanical occlusion. The Embosphere microspheres are made from the molecules of trisacryl, bound with gelatin. In our patient, we used particles sized 500–700 μm, 700–900 μm and 900–1200 μm. The contralateral uterine artery was catheterized to allow embolization of any persistent vascular supply to the malformation. Continuous fluoroscopic monitoring during embolization was performed to avoid shunting of embolic agents to the pulmonary vasculature, particularly in high-flow AVMs. Although the procedure went well, only 2 h after procedure in our patient was the first attack of pulmonary embolism.

In the past, treatment options for AVM was hysterectomy in cases without realized reproductive desires, while UAE was

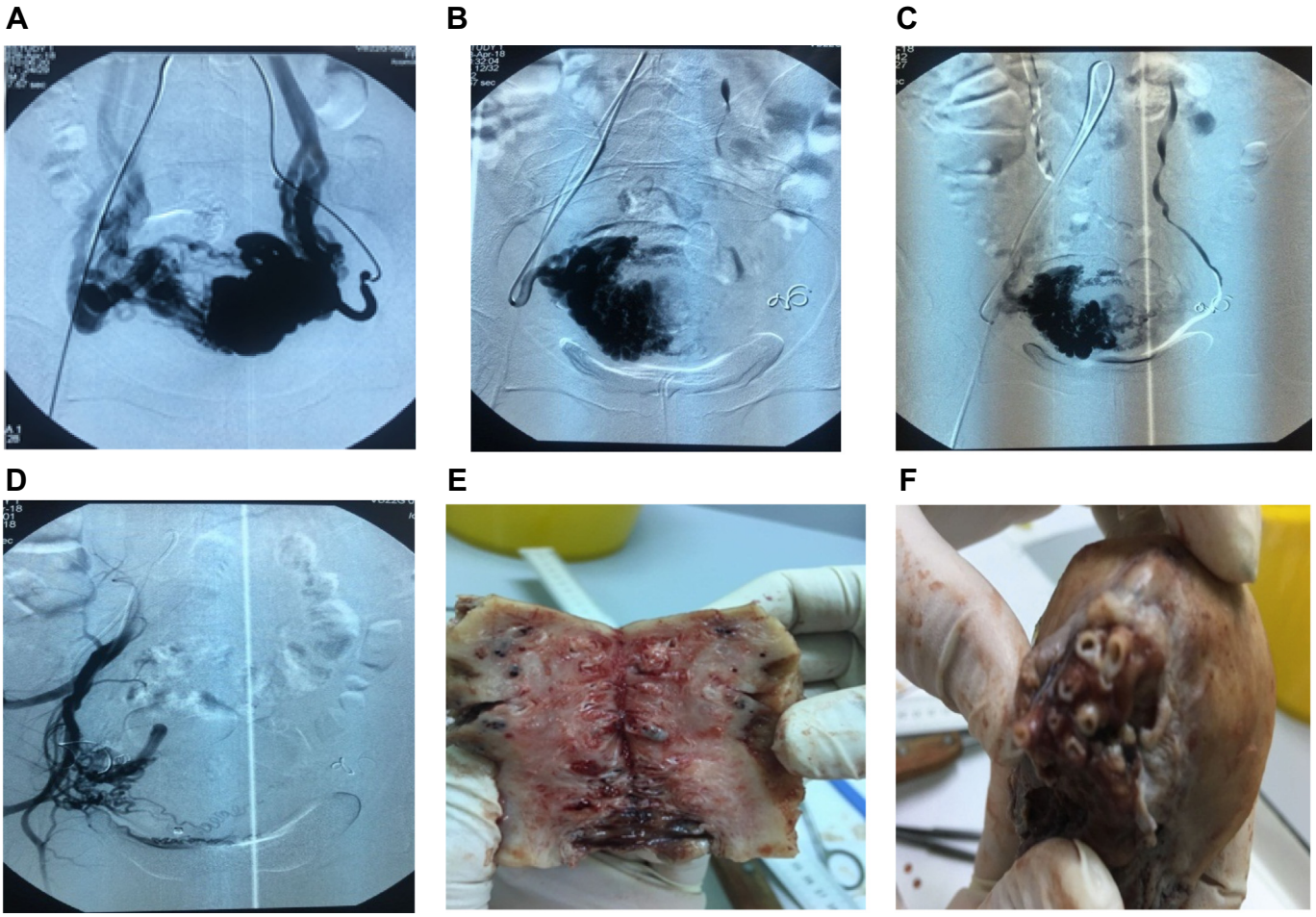

Fig. 1. Angiogram before UAE - extensive pathological mesh-like feature of vessels (A). Angiograms after UAE – revealed stagnant flow (B, C, D). Intraoperative findings: sagittal section of uterus with dilated vessels (E, F).

performed only exceptionally. With increased experience and development of techniques and equipment, embolization become the first choice of treatment for patients of all age groups. But, as every invasive procedure, UAE has very well known complications [13–15].

Acute pulmonary embolism is the most severe and life threatening complication of UAE. It requires early recognition and immediate treatment with intravenous heparin. In some cases, repeated catheterization and vascular filters were found to reduce mortality. Although pulmonary embolism is very rare after UAE, its' prevention is of utmost importance. In order to decrease the occurrence of this medical emergency several preventive measures and procedures are advised: 1. Risk for pulmonary embolism assessment before UAE, 2. Careful blood vessel puncture without disruption of the endothelial cells, and 3. Decreasing the time of sandbag compression and limb immobilization after arterial embolization. In most cases, pulmonary embolism is successfully treated with therapeutic anticoagulation. Unfortunately, our patient had two episodes of pulmonary embolism. The first one was treated by intensive anticoagulation therapy, while after the second one hysterectomy was done in order to eliminate the source producing embolic particles.

Uterine AVM is currently treated by UAE, but clinicians must be aware of complications that can appear following the procedure. Very rare complications might happen and be severe and life threatening; so far, every particular case of uterine AVM must be evaluated very carefully and treatment approach should be chosen individually. When embolizing AVMs we recommend the use of only large sized particles (700–900 μm or 900–1200 μm) in the carefully selected patients in order to prevent such life-threatening complications.

Conflict of interest

The authors have no conflicts of interest relevant to this article.

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