



Case report

Acquired uterine arterio-venous malformation post molar pregnancy suction-curettage: 2 case reports

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ABSTRACT

Introduction: Acquired uterine AVM is an abnormal and non-functional connection between uterine arteries and veins. It is reported as a consequence of uterine trauma as curettage procedures, pelvic surgery.

Cases presentation: The authors report 2 cases of uterine AVM post suction-curettage procedure on molar pregnancy incidentally discovered in ultrasound screening in the usual follow-up of this disease, diagnosed in the Department of Maternity of Mother and Child University Hospital Abderrahim Harouchi of Casablanca.

Clinical discussion: Uterine AVMs are extremely rare and should be considered in cases of heavy and persistent uterine bleeding. Uterine AVMs are either acquired or, more rarely, congenital, due to abnormal differentiation of the primary vascular structures during embryogenesis. The invasive technique allows confirmation of the diagnosis and identification of the major feeding vessels when embolization may be indicated as a treatment option.

Conclusion: Acquired uterine arteriovenous malformation is the result of uterine trauma; it presents as uterine bleeding through the vagina that can be life threatening, the rarity of the condition makes it difficult to diagnose, however the evolution of investigative techniques helps in the diagnostic process.

1. Introduction

Arteriovenous malformation (AVM), also called arteriovenous fistula, is a short circuit between the arterial and venous circulation of an organ [1]. It is a potentially rare and fatal source of uterine hemorrhage due to abnormal vascular connections [2]. Acquired arteriovenous malformations are often the result of uterine trauma [3] such as curettage, cesarean section, or pelvic surgery. They are also associated with infection, retained product of conception, gestational trophoblastic disease (GTP). Because of its potentially dangerous condition, a prompt diagnosis of uterine AVM is necessary to avoid bad consequences. The work has been reported with respect to the SCARE 2020 criteria [4].

2. Cases reports

2.1. Case 1

25-year-old patient, first procedure, followed for a molar pregnancy with an initial B-HCG level of 500,092 IU. The initial screening

ultrasound before aspiration did not show any abnormality of the vascularization of the uterine myotrium. She was admitted to the clinic for aspiration with an obvious decrease in B-HCG level. The histopathology report shows a hydatidiform molar pregnancy without malignancy. Two weeks after aspiration-curettage, an ultrasound was performed and showed multiple anechoic intrauterine images, confusing a diagnosis of gestational trophoblastic disease or choriocarcinoma. The patient was then referred to our third level maternity ward for further treatment. The B-HCG level at the first check-up in our facility was 100 IU/L. The decrease in B-HCG level was significant and not consistent with the ultrasound diagnosis. We performed advanced pelvic screening and transvaginal gray-scale ultrasound, which objectified an enlarged uterus with a heterogeneous myometrium with multiple anechogenic images around taking up the entire myometrium, with a thin-line uterine cavity (Fig. A, A1, A2). The most suspicious feature was a rhythmic rocking of these images, which made us think of an acquired AVM and rule out the diagnosis of GTD.

To confirm the diagnosis, color Doppler was applied, which objectified anechoic structures in the myometrium with a high-velocity flow

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pattern (Fig. A, A3, 4). Pelvic MRI was also performed to map the AVM, which showed in sagittal view a hypointense T2 lesion on the posterior wall of the myometrium measuring 5 cm (Fig. B, B1) with T2 hyper-signal nodules, associated with diffuse tubular hypointense images (Fig. B, B1). The contribution of MRI in the diagnosis of AVM was very low.

Despite the fact that it was asymmetric, embolization was proposed to our patient to avoid any future obstetrical complication or severe hemorrhage, in particular the AVM was diffuse to the whole wall of the uterine myotrium and the risk of hysterectomy may be higher in this case. The patient did not adhere to our proposal and was lost to follow-up during the COVID-19 pandemic.

2.2. Case 2

A 48-year-old female patient, 3rd pare, without any contraception, was followed 3 months earlier for a molar pregnancy with an initial B-HCG level of 657,320 IU. Hemoglobin level was 10 g/dl.

Pelvic and transvaginal ultrasound showed a diffuse vesicular appearance in the uterine cavity consistent with a molar pregnancy.

The patient underwent an ultrasound-guided aspiration-curettage procedure, with an empty uterine cavity with no myometrial abnormality on immediate post-suction ultrasound. The histopathology report showed a complete hydatidiform pregnancy. Weekly biological monitoring showed encouraging results with a decrease in the B-HCG level which was negative in 10 weeks.

3 months after aspiration-curettage, she presented to the emergency room with heavy bleeding and a blood pressure of 100/50 mm Hg, PR: 109 beats/min. The B-HCG level was negative. Hemoglobin level was 7.3 g/dl.

A transvaginal ultrasound was performed after the patient was hemodynamically stabilized. The ultrasound showed a cluster of vascular channels taking up the uterus (Fig. C). These vascular channels presented a turbulent colored flow when we applied the color Doppler (Fig. D); one of these vascular channels presented a huge fundal dilatation with a turbulent flow evoking the formation of an acquired aneurysm.

Vascular embolization was suggested to stop the bleeding but could not be done urgently, and because of the amount of bleeding a hysterectomy with preservation of the adnexa was performed after the patient's consent. The histopathological results confirmed the diagnosis of AVM and showed no evidence of malignancy.

3. Discussion

Uterine AVMs are extremely rare and should be considered in cases of heavy and persistent uterine bleeding [5]. Uterine AVMs are either acquired or, more rarely, congenital, due to abnormal differentiation of the primary vascular structures during embryogenesis [2,6].

Acquired arteriovenous malformations result from poor blood shunting between the intramural arterial branches and the venous plexus following uterine trauma. The most common iatrogenic causes

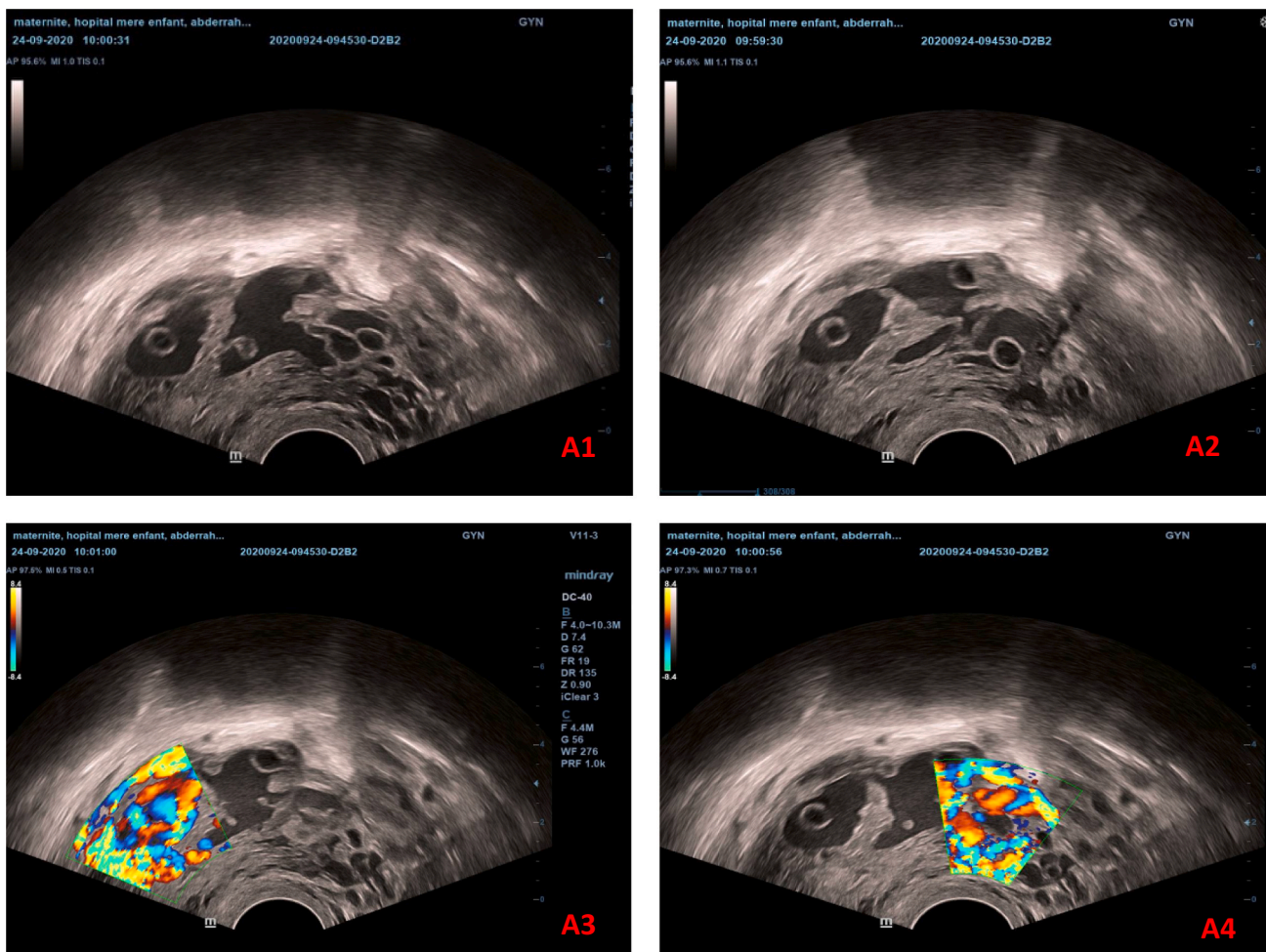


Fig. A. Transvaginal ultrasound, sagittal cut of the uterus. A1–A2: gray scale screening show a big size uterus with tubular anechoic images diffuse to all the myometrium. The cavity are empty. A3–A4: color Doppler apply show a high velocity flow pattern that confirm the diagnostic of AVM.

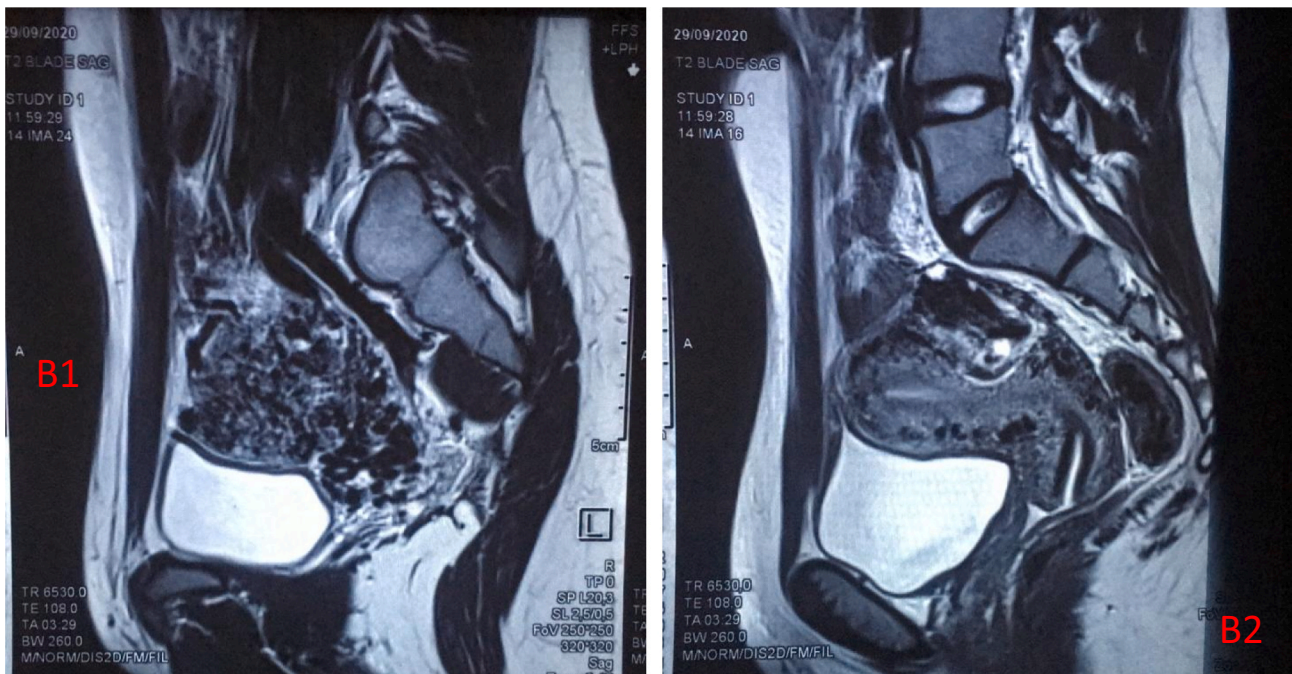


Fig. B. Sagittal view of the uterus in pelvic T2 MRI.
 B1: tubular hypo intense lesions diffuse to all the myometrium.
 B2: hypointense lesion in the posterior wall of the uterus that showing inside 2 nodular hyperintense lesion. The cavity line is free and de junction zone is normal.

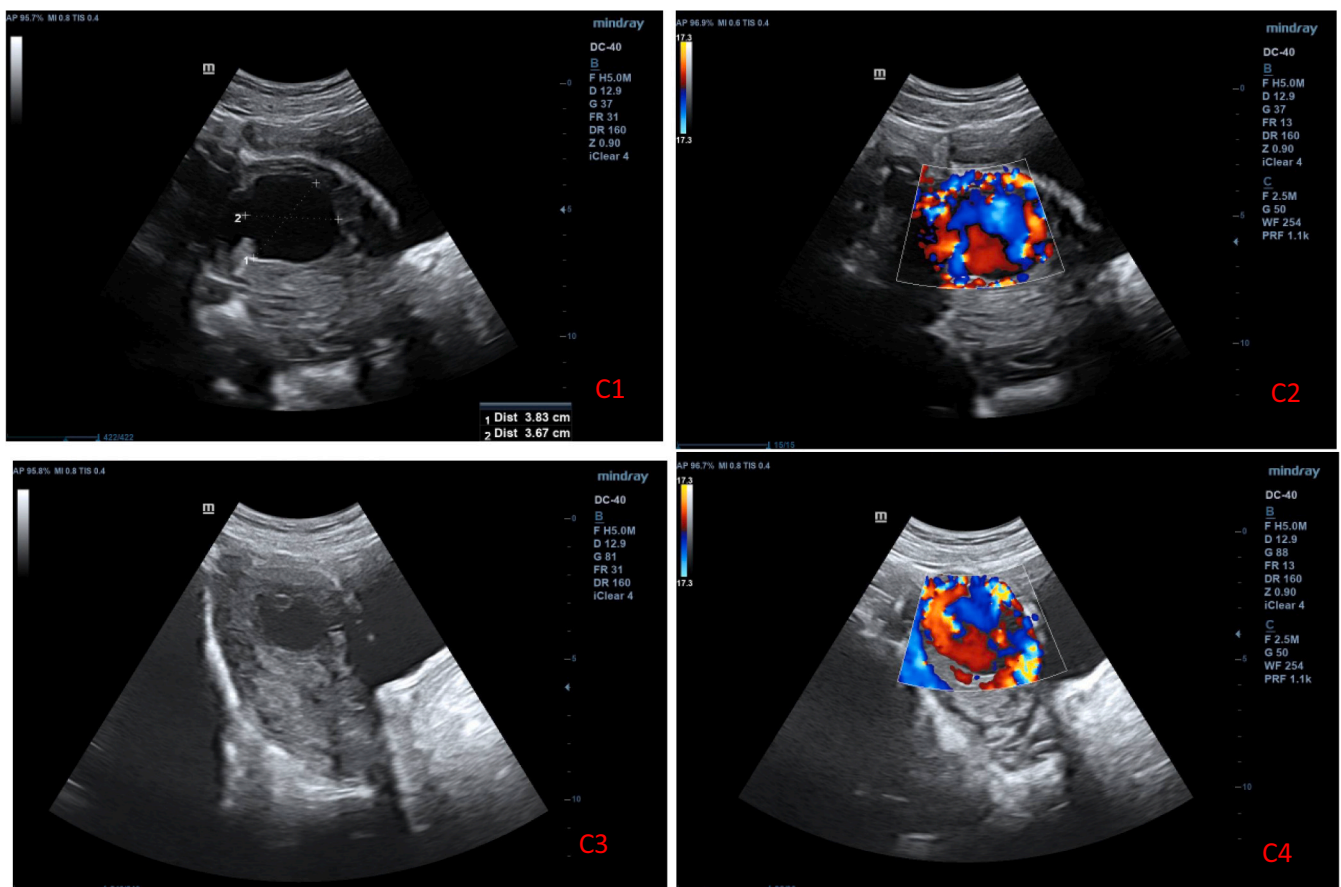


Fig. C. Pelvic ultrasound, axial (C1) and sagittal (C3) cut of the uterus that show a huge anechoic image in the fundus uterine wall pretting confusion with a gestational sac. The Doppler color make the diagnostic of AVM and showed a turbulent flow (C2, C4).

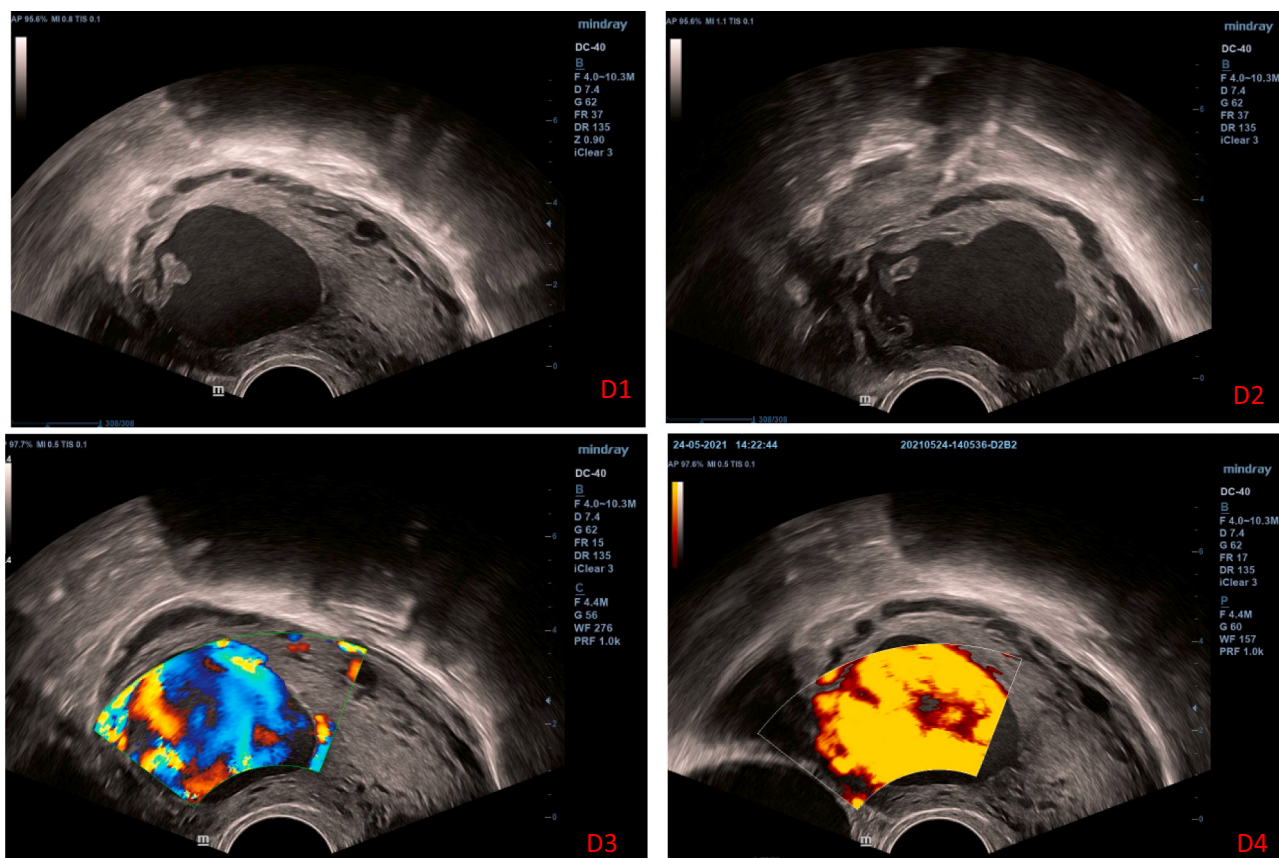


Fig. D. Endovaginal ultrasound, gray scale screening of the uterus showed an irregular anechoic image in the myometrium in sagittal cut (D1) and axial cut (D2). The cavity is empty with small hematometry. The application of color Doppler (D3) and energy Doppler (D4) object if a huge aneurism with a turbulent flow.

are surgical procedures on the uterus, such as cesarean section, myomectomy, or normal vaginal surgery. Less commonly, they may arise from gestational trophoblastic disease, endometrial malignancies, intrauterine devices, infections or fibroids. In our case, the AVM occurred after aspiration-curettage of a molar pregnancy, as no Doppler abnormality had been previously diagnosed. Dubreuil and Loubat reported the first case of uterine AVM in 1926. AVMs tend to occur in women of childbearing age, but very rarely in women who have never given birth.

The most common presentation of uterine AVMs is hemorrhage of unknown cause [7], including menorrhagia or menstrual extension and heavy bleeding during curettage and irregular vaginal bleeding. The first patient was completely asymptomatic and the finding was made as part of the usual follow-up of the molar pregnancy. Some patients even present with shock due to sudden or massive bleeding and curettage does not stop the bleeding or may even exacerbate this condition. Like our second patient, the hemorrhage was significant and an emergency hysterectomy was indicated to stop the bleeding due to lack of embolization.

Diagnosis of uterine AVMs can be made using noninvasive methods such as transvaginal ultrasound (TVS) and color Doppler [6,7]. However, TVS findings may be nonspecific and therefore this examination cannot be used to differentiate and exclude other causes of PPH, such as retained product of conception and gastrointestinal tract dysfunction. Color Doppler increases the accuracy of TVS examination in the diagnosis of uterine AVM [1]. The most common findings on color Doppler imaging are hypervascularization within the lesion, turbulent flow, and multiple, tortuous feeding vessels [3,8,9]. Digital subtraction angiography is the gold standard technique for the diagnosis of uterine AVMs. Angiographic analysis can reveal bilateral uterine artery hypertrophy feeding a tortuous, hypertrophic arterial mass with large accessory feeding vessels and early drainage into enlarged, hypertrophic veins

during the arterial phase. The invasive technique allows confirmation of the diagnosis and identification of the major feeding vessels when embolization may be indicated as a treatment option. Magnetic resonance allows for accurate diagnosis of AVMs through improved soft tissue characterization. Multiplanar imaging capabilities and the use of gadolinium-based contrast media. MRI with gadolinium-based contrast shows hypervascular dominant arterial flow and allows accurate assessment of vascular supply drainage. The management of AVM depends on many factors including clinical findings, hemodynamic status of the patient, age, desire for fertility. In early reports, hysterectomy and ligation of the internal iliac arteries or uterine arteries was the preferred treatment [7,10]. Since the first uterine artery embolization (UAE) procedure reported by Forsman et al., UAE has gradually become widely accepted because of its fertility preservation [6,10]. This procedure is highly effective, safe, and has a low risk of complications.

4. Conclusion

Acquired uterine arteriovenous malformation is the result of uterine trauma; it presents as uterine bleeding through the vagina that can be life threatening, the rarity of the condition makes it difficult to diagnose, however the evolution of investigative techniques helps in the diagnostic process. Uterine artery embolization remains the treatment of choice for AVMs for patients who wish to preserve their fertility.

Consent

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient.

Ethical approval

I declare on my honor that the ethical approval has been exempted by my establishment.

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CRedit authorship contribution statement

Amine Lamrissi: Corresponding author surgeon operator and writing the paper.

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Jalal Mohamed: Surgeon operator and correction of the.

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Declaration of competing interest

The authors declare having no conflicts of interest for this article.

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