Imaging and hysteroscopic findings in a patient with postpartum hemorrhage caused by acquired uterine arteriovenous fistulas

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Running head: acquired uterine arteriovenous fistulas

Case

A 24-year-old woman (gravida 2, para 1, aborta 1) gave birth to a healthy 3.11 kg full-term female baby, with Apgar scores of 10 and 10 at 1 and 5 min, respectively, via spontaneous vaginal delivery at our obstetrics inpatient department. A lateral episiotomy was repaired, and B-ultrasonography revealed that the placenta was lodged in the anterior wall of the uterus, about 31 mm thick, grade III maturity (Figure 1a). Eventually, the entire placenta ejected itself without complications 9 min after the delivery of the baby. The woman's immediate postpartum course was unremarkable, and bleeding was minimal; she lost about 400 ml of blood during delivery and the post-delivery procedure. However, her hemoglobin levels and platelet counts suddenly dropped from 12.0% prenatal to 7.1% postnatal and from 143 prenatal to 107 postnatal, respectively. Because she had had an arrested intrauterine pregnancy a year before this incidence, she underwent dilation and evacuation (D&E) and was discharged from the hospital on postpartum day 2.

However, the patient returned on postpartum day 12 and complained of a hemorrhage rapidly from the uterus. Her prenatal course had been uneventful, without underlying coagulation defects, medical diseases, or drug exposures. Only one therapeutic arrested intrauterine pregnancy had been documented. Two massive vaginal bleedings had occurred 3 days before her admission and resulted in a total blood loss of about 500 ml. A physical examination had revealed she was drowsy. Hemorrhaging aside, the patient had reported no abdominal pain or anything else. Because she very strongly wished to preserve her fertility, she was transferred to the Reproductive Center of our hospital.

She was hemodynamically stable when she arrived. Her physical examination showed a normal heart rate (90 b.p.m.) and ortho-arteriotomy (126/80 mmHg), but she harbored signs of cyanosis in her extremities and denied experiencing abdominal pain and dizziness. There were also no signs or symptoms of high output heart failure, except her heart rate for 90 b.p.m. at rest. We noted disseminated intravenous coagulopathy, as laboratory data showed severe anaemia (haemoglobin: 6.5 g/dL) and abnormal coagulation tests. As a result, the patient was supplemented aggressively with intravenous fluids and massive blood components (3 units of packed red blood cells).

Subsequently, she underwent conventional ultrasonography(US), which revealed that the hypoechoic area of the uterus anterior wall myometrium close to the endometrium was 2.99 × 1.67 cm with ill-defined edges (Figure 1b). Color Doppler flow imaging (CDFI) exposed a distorted and expanded blood flow signal. Dilated, tortuous vessels were visible on the anterior and left sides of the uterus. Pulsed wave (PW) ultrasound showed a turbulent spectrum and venous blood flow at the peak of systole. Uterine and pelvic blood flow exhibited a high velocity and low resistance, with a peak systolic velocity (PSV) of 78.5 cm/s and a resistance index (RI) of 0.29 (Figure 1c). The initial diagnosis using conventional US was suspicion of a uterine arteriovenous fistula (UAVF).

During hospitalization, postpartum females with uterine atony should be given oxytocin or synthetic prostaglandins to control hemorrhage. Our patient was hemodynamically stable during the first week in the hospital. She communicated her appreciation for the conservative management she received. She was administered oral ferrous sulfate, which normalized her hematocrit and haemoglobin levels; her hemoglobin rose to 8.7 gm%. Her platelet count and coagulation profile were normal.

Yet, a week after her initial complaint, the patient reported sudden "gushes" of bright red blood per vagina on postpartum day 22. Her estimated blood loss was 400 mL. The vaginal bleeding gradually ceased eventually, and she was transfused with 2 units of packed red blood cells and 270 ml of fresh frozen plasma. However, she suffered from an abrupt and profuse vaginal hemorrhage in the uterine cavity again two days later. Her estimated blood loss this time was 800 mL, and her hemoglobin levels dropped from 10.9 to 6.0 g/dl. She was hemodynamically unstable, and a physical examination showed she was drowsy, tachycardic (98 b.p.m.), and hypotensive (88/56 mmHg), with signs of cyanosis in her extremities. She denied having abdominal pain but reported dizziness. We noted disseminated intravenous coagulopathy again, as laboratory data showed severe anaemia (haemoglobin: 6.5 g/dL) and abnormal coagulation tests. The patient was given oxytocin or synthetic prostaglandins to control hemorrhage in postpartum uterine atony, but she failed to respond to treatment. Subsequently, she was supplemented aggressively with intravenous fluids and massive blood components (6 units of packed red blood cells and 6 units of fresh frozen plasma).

Her family requested that a uterine-preserving procedure be couducted if possible. Hence, Pelvic Digital Subtraction Angiography (DSA) was promptly performed, revealing a large AVF over the left uterine artery with active bleeding. The arterial phase during arteriography showed massive dilatation of uterine arteries to accommodate the high-volume shunting through the uterus. Transarterial embolization (TAE) of the bilateral uterine arteries was immediately carried out with microspheres for embolization, occluding the UAVF and active bleeders, as well as the bilateral uterine artery and its branches. Eventually, the vaginal bleeding ceased gradually(Figure S2). The patient was discharged uneventfully four days after the TAE procedure.

She went home on post-procedure day number 1 in stable condition. She was prescribed for ferrous sulfate and instructed to show up for follow-up within 2 weeks.

At the follow-up visit 2 weeks later, the patient had no more episodes of vaginal bleeding.

Hysteroscopic removal of the mass has been planned for day 47 after postpartum, but on hysteroscopic inspection of the uterine cavity, an abnormal cavity 2.99 × 1.67 cm mass consistent with a UAVF was discovered in the upper right uterine cavity (Figure S3).

Discussion

postpartum hemorrhage (PPH) is considered to be the leading cause (25%) of maternal death, especially in

developing countries,[1] that requires multidisciplinary management, including gynecologists, anesthesiologists, and interventional radiologists. Knowledge of the obstetrical and surgical history of patients with PPH is required to anticipate the most probable cause of PPH. In fact, visual estimation of blood loss (VEBL) was described as the most common and practical way.[2] Some researchers noted that in spite of the relative accuracy of the weighing method, it could not prevent the process of severe PPH. The key actions to successful management of PPH are early recognition and prompt treatment.[3]

AVF results from an abnormal connection between an artery and a vein have been described in the pelvic vasculature and, more rarely, in the uterus.[4] A UAVF is a rare cause of congenital or acquired-in-nature PPHs that represent 1% to 2% of all genital and intraperitoneal hemorrhages and, as such, should be considered in any postpartum female complaining of vaginal bleeding, particularly if the patient has undergone instrumentation of the uterus.[5-8] Acquired UAVF has been attributed to various causes, including malignancies, pregnancy-related, previous uterine trauma from repeated D&E, surgery, infection, and diethylstilbestrol exposure.[5,6,9-14] Most reported cases are acquired secondary to D&E but rarely to vaginal delivery.[15] A UAVF is most commonly identified when it causes complications during pregnancy, typically in women between 20 and 40 years old, suggesting that hormonal changes during pregnancy and the menstrual period may play a role in its pathogenesis.[16] The primary clinical manifestation of a UAVF is paroxysmal massive vaginal bleeding that it potentially life-threatening. The characteristics of vaginal bleeding in our case were a massive gush of vaginal blood that suddenly stopped, possibly due to the opening and closing of the blood sinus. The amount of vaginal bleeding was difficult to estimate, but it was enough to cause severe anemia, syncope, and unstable vital signs. This bleeding requires blood transfusion in 30% of cases.[17] While the true incidence of a UAVF is unknown, and fewer than 150 cases have been reported.[18] O'Brien et al.[19]proposed a rough incidence of 4.5%, which would make its diagnosis an even more critical issue for women with unexplained vaginal bleeding.

A UAVF is easily diagnosed now using color-Doppler ultrasonography (CDUS).[20] Other imaging modalities of importance include pelvic magnetic resonance imaging (MRI) , hysteroscopy, and DSA.[21] While DSA is considered the gold standard modality for diagnosing a UAVF,[5] many authors have found transvaginal ultrasound (TVUS) and CDUS more preferable diagnostic methods in the last two decades because they are less invasive.[22] Grey scale imaging can reveal subtle myometrial heterogeneities or anechoic spaces.[19] CDUS provides a more specific image and presents a color mosaic with thickened vessels and flow reversals. Color-Doppler allows for the identification and localization of increased vascularity, whereas spectral flow Doppler generates a waveform from which systolic and diastolic velocities may be measured. The spectral analysis of AVF lesions reveals a tangle of vessels with a high-velocity flow; spectral Doppler shows high-velocity and low-resistance flow, with low RI values ranging from 0.25 to 0.55 and high PSV values in the range of 40-100 cm/s.[19] In our case, a 78.5 cm/s PSV was recorded in the mid-range, and the patient's continued symptomatic status necessitated a more aggressive approach.

The differential diagnosis of a UAVF comprises several conditions, including hemangiomata, sarcoma of the uterus, trophoblastic disease, and pelvic varicose veins. In the latter condition, the vessels do not pulsate, and they are situated in the outer half of the myometrium.[21,22] In contrast to a real AVF with a fistula, a non-AVF should be considered subinvolution of the placental bed, which is defined as failure to obliterate the placental bed vessels in the absence of retained placental tissues after cessation of pregnancy or after abortion.[22] A correct sonographic diagnosis is, thus, very crucal.[23] However, relying only on Doppler measurements could result in the overdiagnosis of an AVF, as increased vascularity in the endometrium, which resolves spontaneously in 1-2 weeks, can also be noted immediately post D&C.[24] With DSA, hypertrophied uterine arteries contributing to a large area of hypervascularity and rapid outflow into pelvic venous channels indicate the presence of a UAVF.[25] Although contrast medium-enhanced DSA has been the conventional criterion for standard diagnostic tests, its current use is rare; it is now predominantly used during embolization therapy. Failure to recognize a UAVF could lead to an improper treatment, a life-threatening hemorrhage, and hysterectomy procedures. However while a rapid, prompt, and precise recognition of a UAVF as the cause of bleeding is critical because fistulas are life-threatening and uterine instrumentation may aggravate the condition, the entity in the postpartum must not be overdiagnosed, for many so-called UAVFs have

spontaneously resolved at follow-up imaging.[15,23]

A UAVF treatment is individualized based on clinical manifestations and fertility requirements. Five main factors must be considered in the planning and treatment of patients with a UAVF: these include hemodynamic state, size and location of the lesions, degree of bleeding, age, and the desire for future fertility.[15]Intervention options from conservative management to definitive surgical hysterectomy are available to patients.[8,11,26-28] DSA is the gold standard for diagnosing an AVF and also an interventional treatment technique. Because retaining fertility function and relieving clinical symptoms are most important for these women,[29] bilateral UAE is regarded as a method that effectively provides adequate symptomatic relief and retains fertility with minimal side effects, lower complication rates, and major surgical risks.[12] Selective uterine artery embolization (UAE) , which has replaced surgery as the optimal treatment modality for symptomatic UAVFs, has advantages that include a >95% success rate, with a 4% complication rate in retrospective review articles.[30,31] UAE complications include post-embolization syndrome in the form of severe pelvic pain and radiation exposure, infection, embolization of nontarget organs, impairment of ovarian function, intrauterine adhesions, and rebleeding after blood recanalization. Several successful intrauterine pregnancies after the UAE of UAVFs have been reported including a successful twin pregnancy, which suggests that adequate collateral blood supply can develop to support a full-term pregnancy.[32] Peitsidis et al. reported a 27 % pregnancy rate following bilateral UAE.[13] Women who become pregnant after UAE are at risk of malpresentation, cesarean delivery, preterm birth, and PPH.[33] While the impact of UAE on future fertility and pregnancy outcomes has been studied extensively, the subject remains somewhat controversial.

Herein, we present a woman who suffered from secondary PPH following vaginal delivery; her condition was diagnosed using DSA. This report stresses the fact that the clinical suspicion of an acquired UAVF is crucial to promptly diagnosing and treating secondary PPH. Still, diagnosing and treating this condition is remains challenging for physicians. By sharing this case report, we hope our experience will add to what data exist already on UAVFs.

Conflict of interest

The authors declare no potential conflict of interest.

Author contributions

Conception and design: Sijie Yi and Yuanhuan Xiong.

Acquisition of data: Sijie Yi, Xia Jin and Xiuxiu Peng.

Analasis and Intepretation of data: Sijie Yi, Xia Jin and Yuanhuan Xiong.

Drafting of the manuscript: Sijie Yi and Yuanhuan Xiong.

Critical revision of the manuscript of important intellectual content: Yuanhuan Xiong.

Details of ethics approval

The patient has provided written informed consent for publication, available upon request. The case study was performed in agreement with principles of the Declaration of Helsinki.

Data availability statement

The datasets generated during and/or analysed during the current study are available from the corresponding author

on reasonable request.

Supporting Information

Additional supporting information may be found online in the Supporting Information section at the end of the article

Figure S1. (a) B-ultrasonography revealed that the placenta was lodged in the anterior wall of the uterus.(red arrow)

(b) Transvaginal sonography Sagittal grayscale image of the uterus showed a low-echoic cystic lesions in the anterior uterine body (red arrow) measuring 2.99 cm × 1.67 cm in the anterior wall of the myometrium of the uterus.

(c) Color-flow Doppler revealed a mosaic pattern of blood flow within these cystic spaces, Pulsed Doppler analysis revealed an elevation of the PSV (78.5 cm/s) while RI (0.29) was low , illustrating a typical high flow, low resistance, blood flow pattern.(red arrow).

Figure S2. The dilated uterine artery and tortuous arteriovenous net seen during the UAE. Angiography before embolization of the left (A) and right (B) uterine arteries. (C) and (D) show the angiography study following successful embolization of right and left uterine arteries, respectively.

Figure S3. Hysteroscopic image of the uterine cavity. The green arrows point prominent abnormal cavity from the UAVF.

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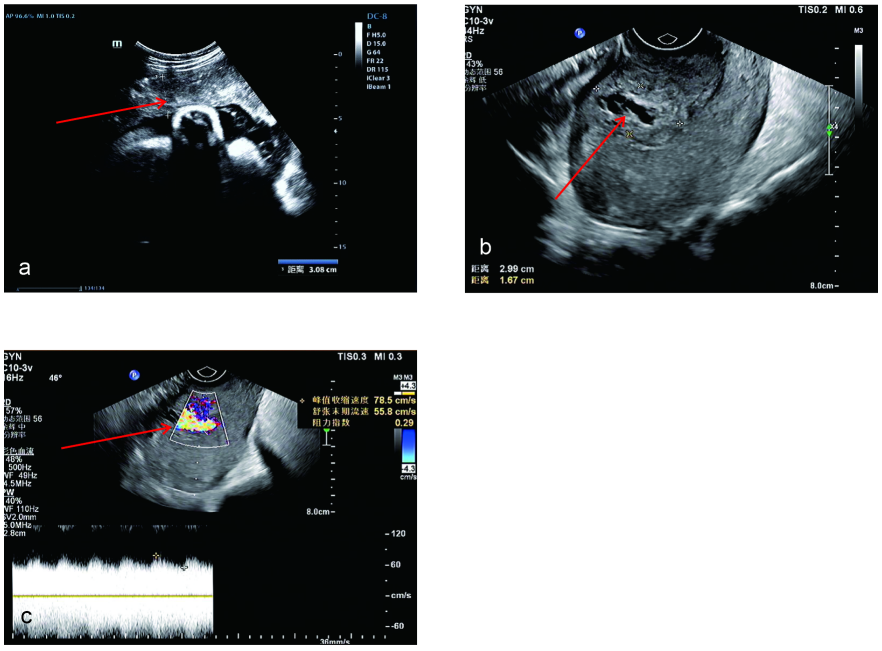

Figure S1

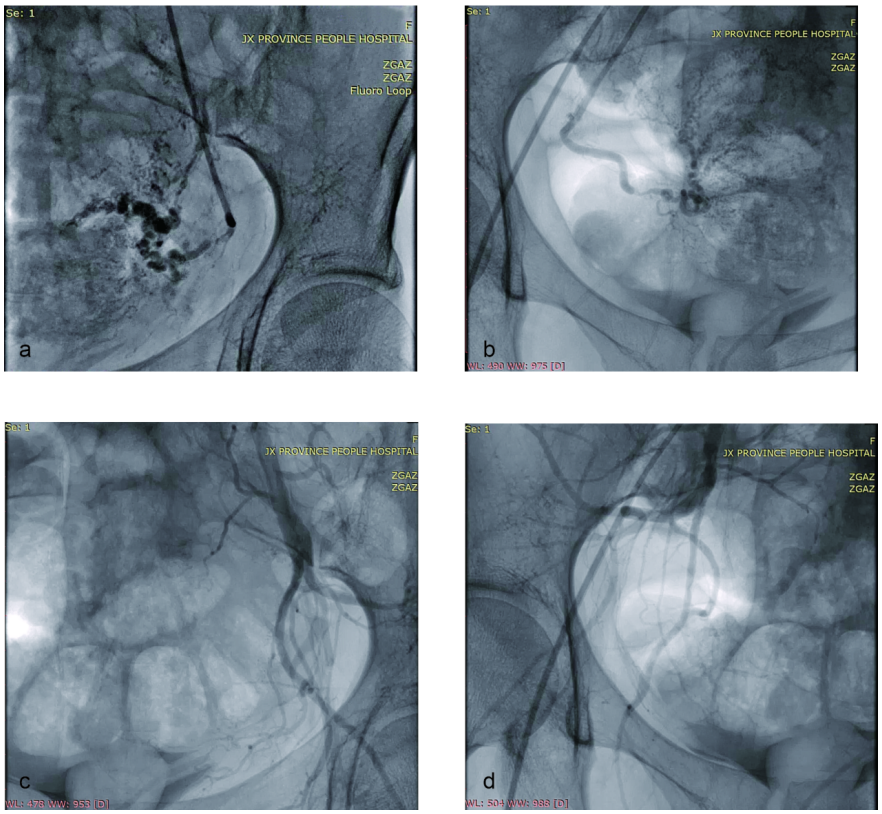

Figure S2

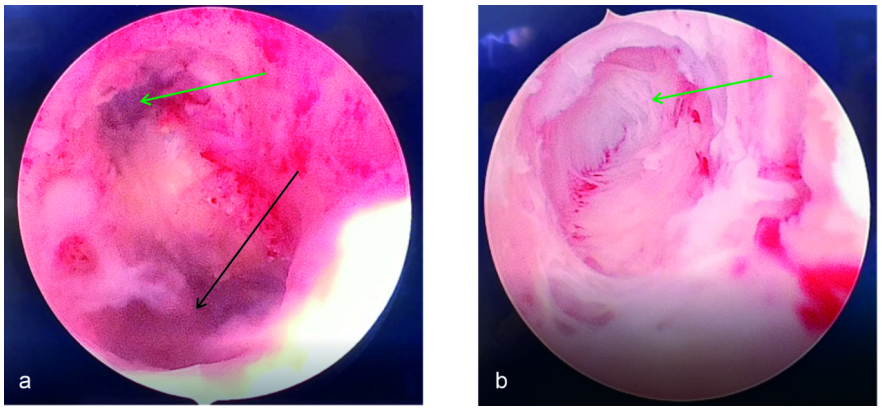

Figure S3