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Clinical vignette

Uterine artery embolization for arteriovenous malformation of the cervix

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INTRODUCTION

In the reproductive age, vaginal bleeding is a frequent presentation; however, its association with serious underlying pathologies cannot be underestimated. One such pathology is the arteriovenous malformation of the cervix (AVM). Defined by the anomalous interconnection of arterial and venous systems within the cervical region, AVM poses significant clinical ramifications, including severe hemorrhage, resultant anemia, hemodynamic compromise, and in critical cases, lethal outcomes.

According to the classification delineated by the International Society for the Study of Vascular Diseases (ISSVA) in 2018, vascular malformations are broadly categorized into simple and mixed varieties. The former is further segregated into distinct subtypes, namely capillary, venous, lymphatic, and arteriovenous malformations [1]. Although these vascular irregularities can present in a plethora of anatomical sites, their incidence in the pelvic anatomy is particularly uncommon. This assertion is evidenced by a limited number of documented cases, with less than 150 reported instances of uterine arteriovenous malformations spanning from 1926 to 2023 [2]. Moreover, the etiological differentiation of

uterine AVMs is pivotal, bifurcating them into congenital or acquired origins. Notably, the acquisition of AVMs is concomitantly associated with several gynecological interventions and conditions, encompassing events such as spontaneous vaginal delivery, cesarean section, gestational trophoblastic disease, diagnostic curettage, placement of intrauterine devices, and specific infections [2, 3].

In this article, we will present a case of a 27-year-old woman who suffered from recurrent heavy vaginal bleeding due to AVM. However, the case reported herein did not have any risk factors before the first presentation of massive vaginal bleeding, and the lesion was located in the cervix. Considering the patient's medical history, symptoms, signs, and laboratory and imaging findings, a congenital arteriovenous malformation of the uterus may have caused vaginal bleeding. We will describe how she was diagnosed and treated with a novel technique called uterine artery embolization (UAE), which successfully stopped the bleeding and preserved her fertility. We will also discuss the causes, symptoms, diagnosis, and treatment options for AVM, as well as the advantages and disadvantages of UAE compared to other methods.

CASE PRESENTATION

Basic information

A 27-year-old woman, with no prior history of abortion, delivery, or pelvic surgical procedures, was recurrently admitted to our medical facility due to episodes of "severe vaginal hemorrhage" (as depicted in Table 1). Despite presenting with intense vaginal bleeding accompanied by symptoms of shock, the ultrasound evaluations consistently failed to detect any anomalous blood flow patterns. By the time of her fourth bleeding episode, a congenital vascular malformation was posited as a potential diagnosis. Intriguingly, an episode of vaginal bleeding transpired during her preparations for oocyte cryopreservation, necessitating an emergent uterine arteriogram. The arteriogram revealed that both uterine arteries exhibited protracted, gestation-reminiscent tortuous formations (illustrated in Figure 3A). Consequently, an immediate bilateral uterine artery embolization was executed.

Treatment

The patient underwent UAE under local anesthesia and sedation. The procedure involved inserting a catheter into the femoral artery and advancing it to the uterine arteries on both sides. Then, small metal coils were deployed into the uterine arteries to block the blood supply to the AVM. The procedure was successful, and no complications occurred.

Prognosis and follow-up

The patient showed no signs of abnormal vaginal bleeding and was subsequently discharged nine days after the procedure. Post-discharge, her oocytes were cryopreserved at the Reproductive Unit to safeguard her fertility. Encouragingly, during subsequent follow-ups, the patient reported no instances of abnormal uterine bleeding and, in a delightful turn of events, has successfully conceived.

DISCUSSION AND CONCLUSION

The patient was admitted to our institution following her third incident of vaginal bleeding. Urgent pelvic Doppler ultrasonography was conducted, which astonishingly did not demonstrate any aberrant blood flow patterns. With suspicions leaning towards abnormal uterine bleeding, potentially due to anovulation or a localized endometrial irregularity, an expedited curettage was undertaken, effectively achieving hemostasis. Nonetheless, in a perplexing turn of events, the patient manifested another bleeding episode, underscoring the severity and intricacy of her condition. Intriguingly, both her coagulation function and pathological evaluations returned within standard parameters. Moreover, the inefficacy of the GnRH-a treatment, coupled with an absence of a familial history of analogous conditions, intensified our investigative endeavors. After diligently ruling out uterine organic lesions, ovulatory disturbances, and systemic coagulation-associated disorders, our diagnostic focus shifted towards the possibility of cervical vascular malformations.

Vascular malformations present a diagnostic conundrum. They seldom resolve autonomously and frequently exhibit progressive enlargement over time. In severe scenarios, they can precipitate profuse, life-imperiling hemorrhage. Stratifying vascular anomalies are pivotal, as therapeutic strategies diverge based on the anomaly's classification. Within the ambit of the PALM-COIN etiological taxonomy of abnormal uterine bleeding, uterine

arteriovenous malformations are demarcated as AUB-N [4]. Emblematic clinical hallmarks encompass vaginal bleeding, pelvic discomfort, menorrhagia, and abrupt inception and cessation of copious bleeding. The symptomatic tableau of AUB-N is inherently ambiguous, often mandating an array of diagnostic modalities to pinpoint the underlying etiology. Color Doppler ultrasonography stands as the diagnostic modality of choice for delineating uterine arteriovenous malformations, owing to its prowess in vividly capturing the multifarious blood flow dynamics within the anomalous echogenic region. For a more panoramic diagnostic vista, MRI and CT imaging are valuable adjuncts [5].

Treatment selection is contingent upon the patient's age, reproductive aspirations, and the gravity of their ailment. Therapeutic modalities encompass medications, UAE, and hysterectomy. Distinctively, UAE is a less invasive approach, efficacious in halting bleeding while concurrently preserving uterine functionality. This technique offers benefits such as diminished complication risks, abbreviated hospitalization duration, and a swifter convalescence in comparison to surgical procedures, notably hysterectomy. A multitude of case studies have documented successful conceptions following UAE [6]. Scholarly research indicates that UAE does not detrimentally affect ovarian reserves, rendering it a viable treatment option in this context [7].

This case accentuates the importance of factoring in pelvic vascular malformations when assessing differential diagnoses for recurrent and sudden vaginal hemorrhage, especially given the paramount significance of preserving a woman's ability to conceive. Congenital malformations of the cervical artery are seldom discussed in medical literature. Veteran obstetricians and gynecologists, with their wealth of experience, are optimally equipped to recognize and diagnose this condition with precision. This case was a profound lesson for our team. Beyond the immediate health implications, it highlighted the intrinsic value of ensuring a woman's reproductive potential. Even in the absence of traditional risk factors like miscarriage or previous pregnancies, persistent and sudden vaginal bleeding necessitates specialized diagnostic techniques, such as DSA, to pave the way for timely interventions that not only save lives but also safeguard future motherhood possibilities.

Article information and declarations

Ethics statement

The studies involving human participants were reviewed and approved by Affiliated Hospital of Guangdong Medical University, Zhanjiang, China. The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individual for the publication of any potentially identifiable images or data included in this article.

Author contributions

Supervision: Ying Zhang; writing–original draft; Shangao Huang: writing–review&editing: Yueling Wu.

Conflict of interest

The authors declare that they have no competing interests.

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Supplementary material

Table 1, Figures 1–3.

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Table 1. Admission summary for recurrent vaginal bleeding

Number of times (Date)	Symptoms	Auxiliary examinations	Processing	Prognosis
First time (Apr 20th,2021)	Heavy vaginal bleeding with dizziness and weakness in a quiet state of sleep	Ultrasound suggested a cystic mass (10 cm^2) in the left anterior aspect of the uterus.	Laparoscopic left ovarian cyst debulking and pelvic adhesion release	Self-stopping
Second time (Apr 27th,2021)	Heavy vaginal bleeding, abdominal pain, dizziness, and weakness	HGB: 65 g/L	GnRH-a injection therapy	Self-stopping
Third (May 5th,2021)	Heavy vaginal bleeding with lower abdominal pain,	Ultrasound suggested slightly strong echogenicity from	Hemostatic and uterotonic agents, hematopoietic	Bleeding stopped after treatment

	dizziness, weakness, and inability to call for help	the cervical canal to the vagina. (Fig. 1)	treatment, diagnostic curettage, no abnormal postoperative pathological findings	
Fourth (May 21st, 2021)	Heavy vaginal bleeding, unrelieved abdominal pain, profuse sweating with dizziness, headache, general weakness	HGB: 62 g/L CTA suggested a tortuous vessel on the left side of the cervix attached to the endocervical lining (Fig. 2 and 3)	Uterine contraction agents, GnRHa injection therapy	Self-stopping
Fifth (May 31st, 2021)	Sudden and heavy vaginal bleeding	DSA suggested prolonged pregnancy-like tortuous malformation of both uterine arteries	Uterine artery spring embolization	Bleeding stopped after treatment

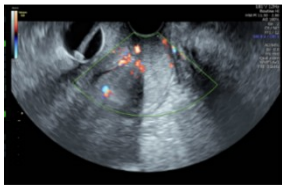

Figure. 1. Gynecologic Doppler ultrasound showing slightly strong echogenicity from the cervical canal to the vagina

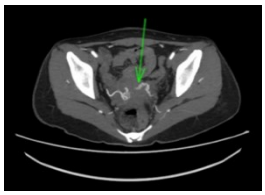

Figure. 2. CTA showing a tortuous vessel on the left side of the cervix attached to the endocervical lining (indicated by the green arrow)

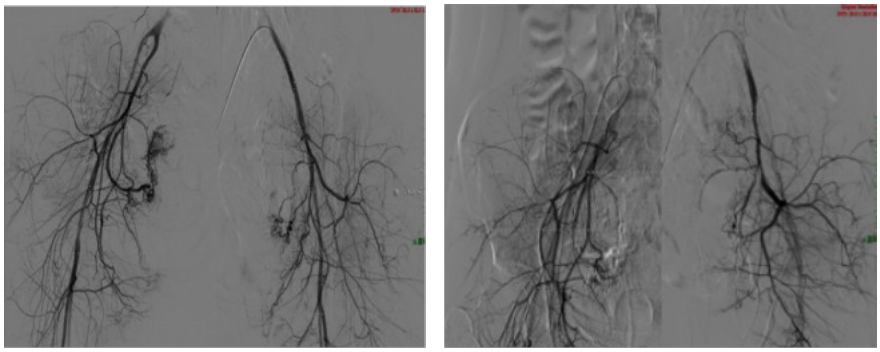

Figure. 3. Before DSA treatment, (A) bilateral uterine artery gestational-like tortuous prolongation. After DSA treatment, (B) bilateral uterine artery flow arrest, no distal visualization