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Case report

Modified compression sutures for treatment of asymptomatic uterine arteriovenous malformation in a low-resource setting: A case report

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ABSTRACT

Introduction and importance: Uterine arteriovenous malformation (UAVM) can be present at birth or acquired later, often after trauma like cesarean delivery. It can cause severe vaginal bleeding but may have no symptoms. What makes our case special, other than being a rare condition, is the surgical technique used.

Case presentation: A 24-year-old woman came in with abdominal pain at 38 weeks pregnant. She had a cesarean delivery 13 months before. She had an uncomplicated repeat cesarean but bled heavily after from uterine atony. A 5 × 7 cm asymptomatic uterine AVM was found incidentally in the right uterine horn. After the transfusion, B-Lynch sutures were used to treat the atony and AVM. The patient recovered well after the sutures. Follow-up ultrasound showed the AVM got much smaller and no more bleeding.

Clinical discussion: While conventional approaches advocate hysterectomy or uterine artery embolization (UAE), our case, situated in a low-income setting, necessitated innovative strategies. With embolization unavailable, and surgery carrying inherent risks, the B-lynch Procedure emerged as a pragmatic choice.

Conclusion: Uterine AVM with no symptoms can happen after cesarean delivery. In low-resource settings, modified compression sutures can effectively treat heavy bleeding after delivery and shrink AVM size, avoiding hysterectomy.

1. Introduction

Arteriovenous malformation (AVM) is a condition characterized by anomalous connections between arteries and veins, without the presence of intervening capillary vessels, resulting in direct blood flow from the arterial to the venous system with high flow and pressure gradients [1]. Uterine AVM can give rise to significant vaginal bleeding and potentially fatal hemorrhage in severe cases or may be asymptomatic. AVM has two forms: acquired, which is more prevalent and may be associated with cesarean section, gestational trophoblastic neoplasia, curettage, cervical or endometrial neoplasia, and congenital, which arises from embryonic differentiation failure [2–4]. Due to underdiagnosis or misdiagnosis, the true and accurate incidence rate of AVM is difficult to ascertain [2].

So, what makes our case special, other than being a rare condition, is the surgical technique used, and how we managed it despite the limitation of sources in our country.

The work has been reported in line with the SCARE criteria [5].

2. Presentation

A 24-year-old gravid woman presented to our medical institution complaining of intense abdominal pain and symptoms of impending labor. Reviewing her medical history revealed no noteworthy personal or familial medical conditions. However, it was noted that she had undergone a cesarean section approximately 13 months before her visit due to breech presentation. Physical examination did not reveal any observable abnormalities. The patient's blood pressure was recorded as 120/70 mmHg and her heart rate was 84 beats per minute at the time of presentation. An ultrasound examination was performed to evaluate the progress of her pregnancy, estimating a gestational age of 38 weeks based on biparietal diameter (BPD) and femur length (FL) measurements, with the placenta positioned posteriorly and a breech presentation. Therefore, due to the current fetus being in a breech presentation in

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addition to a history of prior cesarean section, the patient was admitted to the surgical department and underwent a Lower-Segment Transverse Cesarean Section under spinal anesthesia, successfully delivering the fetus and placenta. However, postpartum hemorrhage was encountered immediately following delivery due to uterine atony, necessitating the initiation of management strategies, including Oxytocin (Pitocin), Methylergonovine (Methergine), Misoprostol (Cytotec), and a single unit of blood transfusion. During this procedure, the presence of an asymptomatic 5*7 cm arteriovenous malformation (AVM) was incidentally detected, accompanied by hypervascularity and myometrial weakness, in the right horn of the uterus [Fig. 1], which had previously gone undiagnosed. The AVM was visually confirmed during the surgical procedure, which is particularly interesting given the absence of prior uterine surgeries or interventions in the affected area. Regarding perfusion, the malformation relied on the occurrence of an anastomotic connection linking the uterine artery and the ovarian artery. Following the blood transfusion, a second surgical intervention was initiated. While hysterectomy was a possible option, it was decided to employ the B-Lynch procedure, utilizing modified compression sutures [Fig. 2]. After the removal of the Vicryl thread's needle, the thread is securely ligated to a nylon thread using a straight needle. This technique enables the nylon straight needle to effectively traverse the entire thickness of the uterus. Subsequently, utilizing the Vicryl thread exclusively, we proceeded to create vertical and transverse sutures, shaping them into a square configuration that exerted mechanical pressure on the uterus, leading to a reduction in its dimensions and facilitating hemostasis. In the postoperative period, two units of blood and two units of plasma were administered as needed within the first 48 h. Uterotonic medications were also administered to address uterine atony, and prophylactic antibiotics were given as routine care to prevent infection. Hemorrhaging was successfully controlled, and the patient's vital signs remained within normal limits. Consequently, she was discharged from the hospital on the second day after the surgery. Regular outpatient follow-up visits have been scheduled for the patient. A subsequent ultrasound examination performed after 12 days demonstrated a significant reduction in the size of the arteriovenous malformation, with no evidence of ongoing hemorrhage, indicating the effectiveness of the compressive sutures procedure and the successful avoidance of unnecessary hysterectomy.

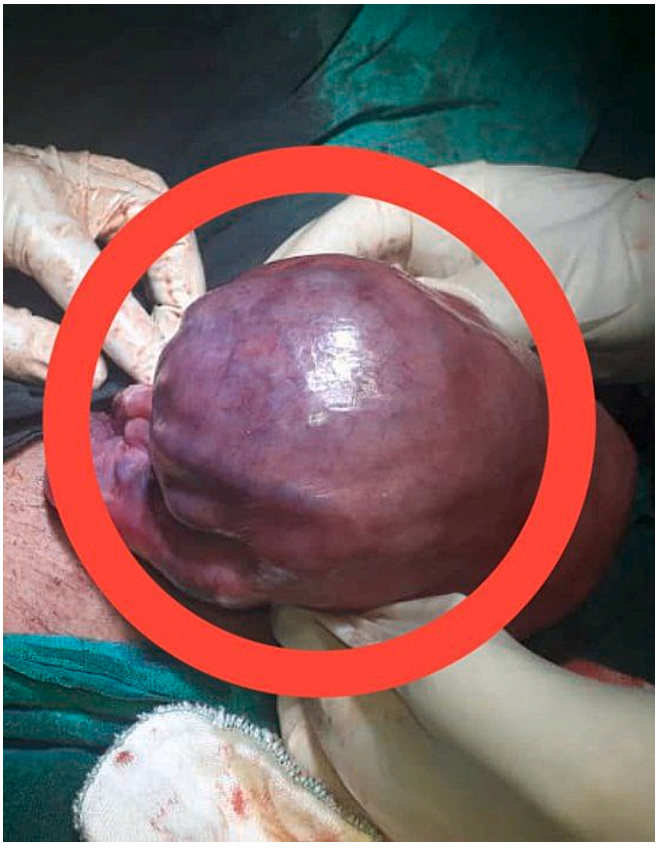

Fig. 1. 5*7 cm arteriovenous malformation in the right horn of the uterus.

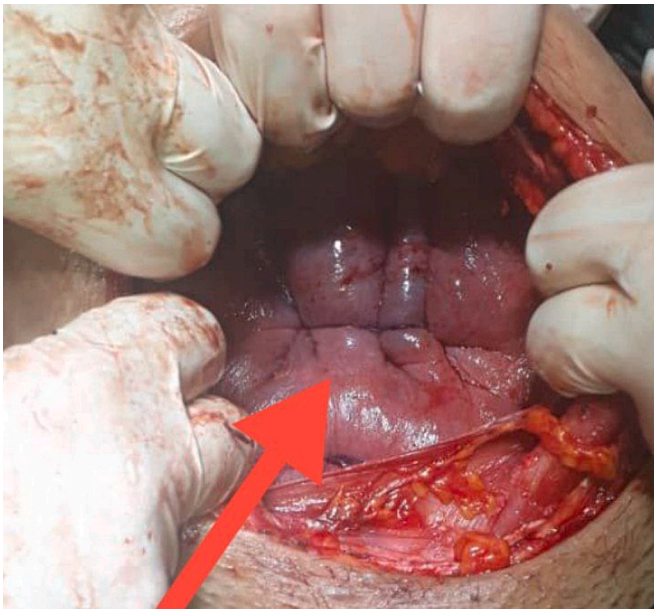

Fig. 2. The uterus after applying modified compression sutures.

3. Discussion

UAVM is a rare life-threatening disease that can be divided into two sections which are congenital and acquired. Congenital UAVM is usually caused by the arteries and veins being abnormally connected as a result of abnormal fetal angiogenesis [6]. Whereas acquired UAVM is usually caused by trauma on the uterine which can occur in surgeries, cesarian sections, and curettages or can be caused as a part of other disorders such as neoplastic disorders – which include gestational trophoblastic disease (GTD) and endometrial adenocarcinoma. Notably, the patient did not undergo any procedures or exhibit symptoms suggestive of these neoplastic disorders during her previous cesarean section, which was a lower cesarean section and not considered a risk factor and does not match the site of the AVM discovered; All of the previous information suggests that the AVM in our case could be congenital [7,8].

This disease is considered rare and its prevalence is unknown as there are only about 150 cases that have been reported between the years 1926 and 2021, although an increase can be considered due to the increasing availability of imaging tools [1].

Still, many cases that have been treated medically could have not been reported. However, UAVM could be considered to take up to 1 to 2 % of all genital and intraperitoneal bleedings [1].

The most common symptom is vaginal bleeding which varies from intermediate to severe and can be accommodated with other symptoms that could include postpartum hemorrhage and menorrhagia. Medical history is of the utmost importance in helping with the diagnosis, this history could include severe uterine bleeding after curettage along with findings during the physical examination that include palpitations of dilated vessels in the genital tract, a pulsing mass, and a large throbbing uterus [1].

Diagnosing AVM can be challenging, and a variety of methods can be used to diagnose the condition accurately.

One of the most common ways to diagnose AVM is through the use of

2-dimensional grayscale ultrasound scanning. This method can reveal irregular tortuous tubular hypoechoic lesions, showing increased vascularity. Transvaginal ultrasound scanning can now be used successfully in diagnosing and following up after treatment. Color Doppler shows turbulent high-velocity blood flow, which can determine the severity of the case depending on PSV (peak systolic velocity) values in the range of 40–100 cm/s and the resistance index of the vessel ranging from 0.25 to 0.55.

In the past, uterine angiography was the gold standard diagnostic method for AVM. However, it is rarely used today for diagnosis unless it is used in treatment [9,10].

CT and MRI can also be used to diagnose AVM. These methods can provide a detailed view of the affected area and help determine the severity of the condition.

In this particular case, the AVM was asymptomatic and went unnoticed, making it an unexpected finding. We did not diagnose the AVM through any screening method; it was only visible to the naked eye. We have also observed hypervascularity accompanied by myometrial weakness in the right horn of the uterus.

The management of uterine AVM depends on the patient's age, desire for future fertility, and the severity of symptoms. Traditionally, hysterectomy has been the treatment of choice for symptomatic uterine AVM in women who no longer desire future fertility [6]. However, with the advancement of embolization techniques and equipment, uterine artery embolization (UAE) has become an effective alternative to hysterectomy. According to a study, embolization should be the initial therapeutic option for all women with UAVM, regardless of their pregnancy desire [11].

Uterine artery embolization (UAE) is a widely used option for managing postpartum hemorrhage (PPH). A systematic review, included 54 patients, reported a primary success rate of 61 % with transcatheter embolization (TCE), which increased to 91 % with repeat procedures. In cases of recurrence of bleeding, there are no established guidelines for treatment, and patients may receive pharmacological, surgical, or re-embolization interventions, which may involve embolization of the contralateral or ipsilateral side. However, clinical factors and the patient's overall condition are important considerations when making treatment decisions [7].

Another interventional method is the uterine balloon tamponade. This procedure has a high success rate for treating severe postpartum hemorrhage and appears to be safe.

Prolonged medical treatment has also been delineated and could be appropriate for individuals without a past of profuse hemorrhaging. Notably, efficacious utilization of the combined oral contraceptive pill has been documented in a patient, wherein the ultrasound examination conducted at the end of 3 months manifested a reduction of the lesion [12].

Some authors have reported that the administration of intramuscular methylergonovine maleate followed by oral therapy has been linked to the amelioration of an ultrasound-detected lesion. However, they emphasize that noninvasive treatment should only be initiated in patients who can undergo rigorous clinical monitoring [13].

Due to the potential risk of spontaneous rupture of subserosal uterine arteriovenous malformation (AVM), some authors propose surgical excision as the preferred treatment for this type of lesion [14].

In our case, although the patient was asymptomatic, management of the postpartum hemorrhage is necessary to prevent potential recurrent bleeding in the future. However she couldn't conduct embolization or uterine balloon tamponade because we are in a low-income country, so we don't have these techniques available. Another reason is that embolization can't be used in the Ovarian artery as in our patient, because it may lead to ovary ischemia. Also because of the patient's desire to stay fertile, we can't risk having another surgery on the uterine. So as it's already an open surgery, we've decided to do the B-lynch Procedure, utilizing modified compression sutures to manage Uterine Atony and the Arteriovenous Malformation. Moreover, due to the presence of severe hemorrhage and marked hypervascularity in the right horn, the triple ligation procedure was deemed unfeasible before the implementation of the B-Lynch technique. Thus, to mitigate the risk of hemorrhage, we proceeded with the B-Lynch technique.

4. Conclusion

AVM is a rare condition but being in the uterine makes it crucial and ticklish. As doctors, we must always pay attention to it especially in women with history of multiple cesarean sections, which has the same importance as early discovery of such disease considering its risk on the patient and her fertility. A modified compression sutures is a practical method to treat Asymptotic Uterine AVM in low-income countries.

Consent for publication

Written informed consent was obtained from the patient's parents for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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HK is the first author, contributed to drafting, editing, reviewing, bibliography and corresponding.
GAA contributed to drafting, editing, reviewing, and bibliography.
AA contributed to drafting, editing, reviewing, and bibliography.
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DA contributed to Supervision.
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Conflict of interest statement

The authors declare that they have no competing interests.

Data availability

Not applicable.

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