

Late Postpartum Hemorrhage due to Vascular Malformations of the Uterus with Feeding from the Femoral Artery: A Rare Case

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Abstract

Introduction: Pseudoaneurysm of the uterine arteries is a rare cause of late postpartum hemorrhage (PPH) but could potentially be a life threatening complication.

Case Presentation: We report a case of a patient with pseudoaneurysm of the uterine arteries following a cesarean section, who presented with late PPH. Bilateral uterine artery ligation was done to conserve the uterus, however, the hemorrhage did not cease. In order to control the bleeding, embolization was suggested. During angiography, a pseudoaneurysm of the feeding femoral artery was noticed and embolization was performed to save the patient.

Discussion: Uterine artery pseudoaneurysm is one of the uncommon causes of secondary postpartum hemorrhage and could lead to a life-threatening situation. Management should be multidisciplinary and the gold standard for treatment and diagnosis is through interventional radiology.

Keywords: Late Postpartum Hemorrhage, Uterine Artery Embolization, Uterine Artery Pseudoaneurysm

1. Introduction

Vascular malformations of the uterus are rare causes of delayed postpartum hemorrhage (PPH). In this case report, we aim to present a delayed intermittent heavy postpartum hemorrhage that initially was managed by bilateral ligation of both internal iliac arteries and secondly by insertion of an intra-uterine balloon catheter. As a result of ongoing bleeding with a suspicion of arteriovenous malformation (AVM) and pseudoaneurysm, embolization was performed to control the bleeding and to avoid hysterectomy.

2. Case Presentation

A 22-year-old woman (gravid 1, para 1) was referred to our hospital with vaginal bleeding 17 days after delivery at term. A cesarean section was performed due to full arrest and fetal distress. During surgery, the uterine angle on the right side was extended through to the uterine artery bed which was subsequently repaired and terminated by a successful operation.

On postoperative day 0, the patient's hemodynamic signs were stable and continued to be stable during the fol-

lowing 2 days, after which the patient was discharged from the hospital.

In the early morning of postoperative day 13, the patient was faced with heavy vaginal bleeding during urination. While visiting another hospital, the clinical examination revealed a shock situation with a systolic blood pressure of 50 mmHg and an undetectable diastolic blood pressure. The laboratory results revealed a hemoglobin level of 4.5 g/dL. As a result, she received 10 units of packed cells and 10 units of fresh-frozen plasma (FFP), which yielded no response during a 2-hour observation period. Consequently, she was transferred to the operating room and underwent laparotomy. The bilateral internal iliac arteries were ligated and vaginal bleeding was controlled. After the surgery, the patient was monitored for 2 more days and other than vaginal spotting, no further complications were observed at time of discharge.

After three days, the patient experienced her second episode of vaginal bleeding. Her vital signs were stable: BP: 100/60, PR: 104, T: 36.5°, and the Hb was 10.4 mg/dL. Vaginal examination was normal and no signs or symptoms of endometritis were detected.

With a transvaginal ultrasonography, the uterus was measured to have a diameter of 140 × 46 mm, which was

considered to be in the normal range for postpartum. At the incision site, a subfascial hematoma with a diameter of 114X90 mm and a volume of 152 cc was observed. The lower segment of the uterus was normal. There were no retained products of conception. A massive intravaginal blood clot with vaginal expansion was also noted. Furthermore, the sonography was not followed by a Doppler study. In the report, the cervix and the lower segment of the uterus were suggested as the origin of bleeding.

However, a third episode of bleeding occurred after about 24 hours. It was heavy and the patient underwent uterine balloon tamponade with a volume of 400 cc, after which the bleeding was controlled. Nevertheless, her haemoglobin (Hb) was decreasing, almost from 10.4 mg/dL to 7.5 mg/dL. She was then transferred to the ICU and received 2 units of packed cells and intravenous liquid therapy. Her vital signs, the bag and line of balloon were carefully checked. After 5 - 6 hours we noticed blood flow in her line, revealing that the uterine bleeding was still persisting.

By means of intervention consultation, we referred her for embolization. The angiography displayed a saccular aneurysm with a diameter of 14.8 mm with an irregular margin almost in left site of the uterus. The aneurysm was fed by one of the accessory branches of the left common femoral artery, which was selectively cauterized with a microcatheter and micro guidewire. Subsequently, one vial of Onyx was used for embolization (Figure 1A - 1C).

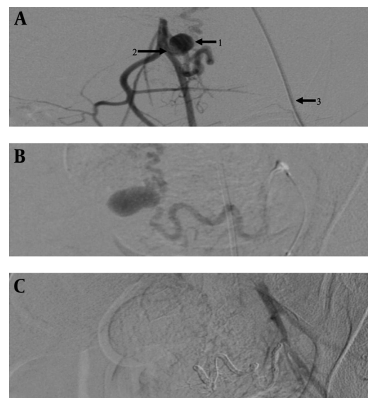


Figure 1. A, Pre-Embolization: 1) Pseudoaneurysm Sac, 2) Pseudoaneurysm Neck, 3) Catheter; B, During Embolization with Microcatheter; C, Post-Embolization

3. Discussion

Postpartum hemorrhage (PPH) is a major challenge for obstetricians. Of all maternal mortalities, 11% is due to obstetric hemorrhage, which is still one of the most important causes of maternal mortality (1). Excessive vaginal

bleeding from 24h after delivery up to 6 weeks is defined as late postpartum hemorrhage.

There is no obvious definition for quantity of blood loss and this can vary from increased lochia to massive hemorrhage; the diagnosis is therefore subjective. The overall incidence of secondary postpartum hemorrhage is 0.47% - 1.44% (2).

There are different etiologies for secondary postpartum hemorrhages and management depends on the cause. In spite of the association with minor morbidities, there are still requirements of readmission to hospital, use of antibiotics, and surgical intervention. In rare cases, major hysterectomy, arterial ligation or radiological intervention is possible and despite the use of all available interventions, maternal death may still result from massive secondary postpartum hemorrhage (1). All common causes of delayed PPH, with exclusion of endometritis and retained product of conception, were ruled out and bilateral uterine arteries were ligated. Hysterectomy was not an option for a young fertile woman.

Since pseudoaneurysm is not prevalent and in some cases small in size, the diagnosis is only possible if sufficient clinical suspicion exists. Good quality transvaginal ultrasonography with Doppler is usually the first key to determine the diagnosis, but angiography is the gold standard (3).

Abu Ghazza et al. reviewed 16 cases of PPH with pseudoaneurysm and showed that transvaginal sonography was successful in diagnosing 12 of these cases. A characteristic vascular appearance with a "to and from" sign can be seen in Doppler results of pseudoaneurysms that could differentiate it from other possible diagnoses such as fibroid, hematoma or an abscess (3).

3.1. Vascular Surgery Recommendation

Uterine arteriovenous malformation leading to postpartum hemorrhage is rare and has an unknown definite incidence. AVM can be congenital but is commonly acquired when it is presented with secondary postpartum hemorrhage, because of the fact that sufficient time is necessary for a pseudoaneurysm to initiate (4). The most common cause of PPH is endometritis or retained products of conception, which could be managed with antibiotics or through an evacuation.

AVMs are abnormal arteriovenous connections that can occur anywhere in the body. Abnormal development of primitive vessels that form connections between pelvic arteries and veins in the uterus can be a way in which they are produced.

They are characterized by several feeding and draining vessels with an interconnecting nidus with turbulent flow.

AVMs are rare and are associated with PPH in less than 1% of cases.

Acquired vascular malformation results from iatrogenic or traumatic injury to the uterine artery vascular bed (5).

Abu-Ghazza also showed that out of 16 patients in his study, 10 underwent a cesarean section, 3 of them had a uterine evacuation, and another 3 had a normal vaginal delivery without any complications (3).

In terms of etiology, a cesarean section at advanced dilatation with uterine angle extension is the most common antecedent event, where direct trauma and suturing around the uterine artery bed cause abnormal vascular connections. Failure to completely suture causes the bleeding vessels at the apex angle to tear and leads to leakage into the surrounding tissues (6). Recent curettage, particularly in situations in which it is difficult or the placental tissue is very adherent, may be also a direct cause of vascular trauma. When these abnormalities present after an uncomplicated vaginal delivery, it is proposed that the vascular bed of the myometrium is disrupted by the mechanism of delivery or, in fact, more likely that the malformation pre-existed and only presented after delivery (6).

3.2. Interventionist Aspect

Embolization of the pelvic vascular system is well established for PPH in general. It is also minimally invasive. If the site of hemorrhage is accurately identified, selective embolization can preserve the normal vascular supply of the uterus. Abu-Ghazza describes a high degree of success in managing vascular malformations with embolization (3). Embolization success rates were reported as 85% - 95% (7, 8). As flow through the vessels returns over time, it preserves both the uterus and fertility (9).

Embolization is performed through percutaneous catheterization of the femoral artery. Typically, a temporary agent such as a gelatin sponge is used to decrease the perfusion pressure and to stop hemorrhage in PPH (9).

Proximal and non-selective embolization present a higher risk for the pelvic blood supply (9). In our case, which was an emergency situation with unstable hemodynamic conditions and no suspicion of AVM, ligation of the bilateral internal iliac arteries was primarily performed. AVM with feeding from the femoral artery branch is very rare and has not previously been reported. Of course, it is to mention that angiography could detect it.

Pregnancies following embolization are reported to have included a series of known AVMs, although long-term reproductive sequelae are not yet clear. However, the embolization process itself carries potential morbidity including infection, neurological damage, and bladder

necrosis (10). Hysterectomy is mentioned as the only treatment if bleeding is intractable despite the available managements, as mentioned above, and hemodynamic instability also makes interventional radiology unsafe.

One of the uncommon reasons of secondary postpartum hemorrhage is uterine artery pseudoaneurysm, which can cause a life-threatening situation. Management should be multidisciplinary and the gold standard for treatment and diagnosis is through interventional radiology.

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Footnote

Conflict of Interest: The authors declare that they have no conflict of interest.

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