Case Report

Recurrent Postpartum Hemorrhage: A Case of Uterine Artery Pseudoaneurysm Probably Induced by Anticoagulants

Sangam Jha*, Sharda Jha, Pammy Praveena, Jafeesha Brightsingh
Department of Obstetrics and Gynecology, AIIMS, Patna, Bihar, India

Abstract

Pseudoaneurysm formation often occurs when there is inadequate sealing at an arterial puncture site. We present the case of a 27-year-old primigravida with rheumatic heart disease and a history of mitral valve replacement on anticoagulants who experienced recurrent episodes of postpartum hemorrhage (PPH). Despite conservative management and adjustments to anticoagulant therapy, the bleeding persisted. Further investigations revealed a small pseudoaneurysm originating from the left uterine artery. Bilateral uterine artery embolization (UAE) using polyvinyl alcohol particles was successfully performed. The patient's condition improved, and she was discharged on a carefully regulated medication regimen. This case highlights the importance of considering rare causes of PPH in high-risk patients, such as uterine artery pseudoaneurysm. Anticoagulants could be a potential contributor of its spontaneous rupture. Prompt diagnosis and appropriate intervention, such as UAE, can effectively manage PPH and prevent adverse outcomes.

Keywords: Anticoagulants, pseudoaneurysm, secondary postpartum hemorrhage, uterine artery, uterine artery embolization

Introduction

Secondary postpartum hemorrhage (PPH) refers to bleeding from the genital tract occurring between 24 h and 12 weeks after delivery.[1] It can be categorized as minor (500–1000 ml) or major (>1000 ml) depending on the amount of blood loss. The World Health Organization's PPH summit in 2022 highlighted that PPH is the leading cause of maternal mortality worldwide.[2] The primary causes of secondary PPH include infection, retained products of conception, tears in the vagina or cervix, coagulation disorders, and rare conditions, such as arteriovenous malformation, uterine artery pseudoaneurysm (UAP), uterine artery erosion, or arteriovenous fistula, as well as cesarean scar dehiscence. In certain high-risk patients, like those with heart disease, coagulation abnormalities can overshadow other causes of PPH, particularly in individuals taking anticoagulants due to valve replacement. UAP can be treated with uterine artery embolization (UAE), a minimally invasive procedure that has a high success rate and typically preserves fertility. Failure to recognize and treat UAP promptly can have severe consequences, potentially requiring a hysterectomy or leading to fatal outcomes. Here, we present the case of a patient with rheumatic heart disease (RHD) who experienced recurrent episodes of PPH following mitral valve replacement (MVR), with UAP identified as the underlying cause. The presence of coagulopathy masked the UAP, but successful management was achieved using UAE.

Case Report

A 27-year-old primigravida at 39 weeks of gestation, with a history of RHD and prior MVR, presented with leaking per vaginum. She had severe mitral stenosis, moderate mitral regurgitation, mild tricuspid regurgitation, atrial fibrillation, premature ventricular contractions, and severe pulmonary arterial hypertension (PAH). She was on warfarin 3 mg OD

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Address for correspondence: Dr. Sangam Jha, Department of Obstetrics and Gynecology, AIIMS, Patna, Bihar, India.
E-mail: sangam.jha78@gmail.com

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and metoprolol 25 mg OD. An emergency cesarean section was performed due to the severity of PAH, resulting in the delivery of a healthy baby boy. However, the patient experienced postoperative complications, including difficulty maintaining blood oxygen saturation, requiring ventilatory support for 24 h. She was started on anticoagulants, digoxin, and metoprolol from the 3rd postoperative day.

On postoperative day 8, the patient developed persistent fever, which was initially attributed to an *Acinetobacter* infection detected through urine culture. Despite appropriate antibiotic treatment, the fever persisted. On postoperative day 12, she developed wound complications, including mild induration, wound discharge, and wound gape. The wound culture was sterile, and management involved daily debridement and dressing.

On postoperative day 15, the patient experienced heavy vaginal bleeding, losing approximately 1000 cc of blood. She was managed conservatively with uterotonics and blood transfusion. As ultrasound (USG) pelvis and Doppler were grossly normal, coagulopathy was suspected due to an elevated prothrombin time-international normalized ratio (PT-INR) of 4.33. Anticoagulants were stopped. Ultrasonography showed no abnormal findings. The bleeding was controlled, and anticoagulation was resumed after 72 h. However, on postoperative day 21, she had another severe episode of bleeding, losing approximately 1–1.5 l of blood, despite a normal uterine examination. As this time too, her PT-INR was 4.11, so cause was attributed to coagulopathy Her hemoglobin dropped to 6.3 g/dl. Conservative management with fluid resuscitation and blood products was initiated, but her condition deteriorated, requiring transfer to the high-dependency unit and administration of noradrenaline to stabilize blood pressure. The repeat sonography with Doppler study did not reveal any pathology this time too.

Given all the common causes of postpartum hemorrhage had been ruled out, the patient underwent diagnostic hysteroscopy and laparoscopy, which did not reveal any pathology. Further evaluation through computerized tomography (CT) angiography detected a small mass, which showed a dappled contrast filling on the arterial phase arising from the distal end of the left uterine artery [Figure 1a and b]. Bilateral UAE using polyvinyl alcohol particles was performed [Figure 2a]. Postembolization digital subtraction angiography demonstrated complete occlusion of Uterine Pseudoaneurysm [Figure 2b]. The patient showed improvement, and wound healing progressed well.

Following the procedure, the patient's condition was closely monitored for 2 weeks, during which the wound gape healed successfully. Once the target INR was achieved, she was discharged on a medication regimen consisting of warfarin 4 mg OD, metoprolol 25 mg OD, digoxin 0.25 mg OD, and Ecosprin 75 mg OD. On 2-month follow-up, she was completely symptom free.

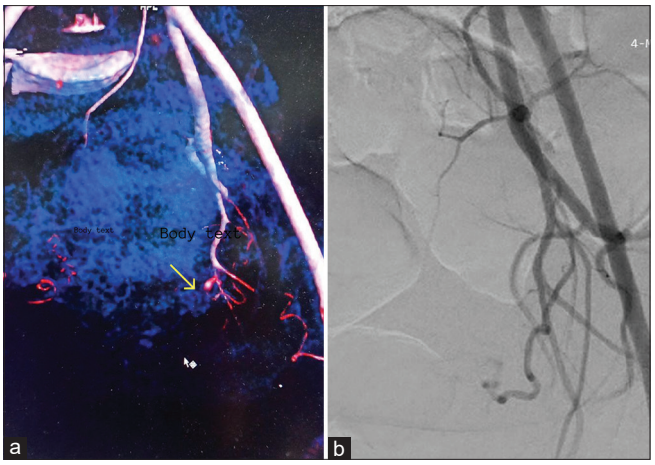

Figure 1: (a) Yellow arrowhead showing uterine artery with pseudoaneurysm in 3D image of computerized tomography (CT) angiography, (b) CT angiography showing aneurysm of left uterine artery

Discussion

Pseudoaneurysms are a rare cause of PPH, with an incidence rate of 3–6/1000 deliveries.[3] Histologically, a pseudoaneurysm is characterized by a single layer of connective tissue, making it more prone to rupture and bleeding. Vascular injury resulting from trauma, surgery, or infection allows blood to enter the periarterial tissue, leading to the development of a pseudoaneurysm.[4] Anticoagulation therapy has been identified in several reports as a potential factor contributing to the rupture of uterine pseudoaneurysms.[5] The spontaneous healing of pseudoaneurysms typically occurs through thrombosis. However, any disruption in normal hemostasis can impede this process. In our case, we attributed the use of anticoagulants, as potential factors interfering with the pseudoaneurysm's natural resolution. They typically present with intermittent massive bleeding, although they can also remain asymptomatic or manifest with abdominal pain or fever. Case reports indicate a variable interval between surgery and symptom onset, ranging from 6 to 140 days.[6,7] In our index case, the patient experienced recurrent PPH, with the first episode occurring after a 15-day interval.

Diagnosis of UAP can be achieved through color Doppler USG, CT angiography, and magnetic resonance imaging.[8] Although color Doppler USG is the initial diagnostic modality for UAP, it may fail to reveal abnormalities, especially in cases with small-sized pseudoaneurysms, as happened in our case.[9] CT angiography is a superior diagnostic tool as it provides clear images and allows for precise localization of the aneurysm. Therefore, CT angiography should be utilized to rule out other causes of delayed postpartum bleeding and determine the exact location of the UAP.

Several treatment options have been suggested for managing UAP, including selective transcatheter arterial embolization,

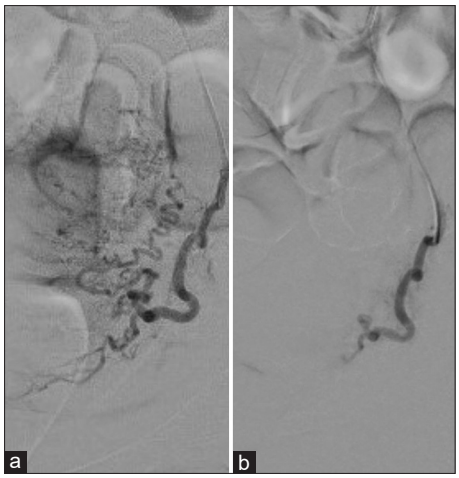

Figure 2: (a) Digital subtraction angiography image showing left uterine artery pseudoaneurysm (UAP), (b) Complete occlusion of the left UAP by embolization

ligation of the internal iliac artery, USG-guided thrombin injection, and hysterectomy.[8,10] Angiographic arterial embolization has been demonstrated as the most effective approach in managing symptomatic UAP.[11] UAE is a noninvasive and safe technique for managing postpartum bleeding, preserving reproductive potential.

Surgical intervention is suitable in cases of acute heavy bleeding where there is insufficient time or lack of facilities for embolization. Hysterectomy is an option for acute massive hemorrhage when fertility preservation is not a concern. Ligation of either the uterine or internal iliac arteries is another surgical option, although it carries the risk of repeat dissection, anesthesia requirement, and difficulty in identifying tissue planes and arteries due to inflammation and hematoma. However, the effectiveness of surgical ligation is limited to <50% due to the presence of uterine collaterals. Uterine balloon tamponade and laparoscopic surgeries have been mentioned in some studies, but they were not successful in our case.

Conclusion

This case report highlights the anticoagulants as a potential cause for spontaneous rupture of UPA leading to recurrent PPH. The diagnosis of this rare condition can be challenging, requiring advanced imaging techniques such as CT angiography. Treatment options include UAE, which offers effective control of bleeding while preserving fertility. Early diagnosis and a multidisciplinary approach are crucial for successful management of this condition.

Ethical approval

Ethical approval has been obtained for the case report from the institute (AIIMS/Pat/IEC/2023/81).

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initial will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

Authors' contribution

Data collection and analysis was done by JB and ShJ. Clinical work by SJ and PP. Concept, designing, and writing of the manuscript were done by SJ. All the authors have read and approved the final version of the manuscript.

Availability of data and material

Data will be available from authors on reasonable request.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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