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Schmidt, Luke
Serna-Gallegos, Tasha
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Publication Date

2020-04-01

DOI

10.1016/j.crwh.2020.e00184

Case Reports in Women's Health 26 (2020) e00184

Contents lists available at ScienceDirect

Case Reports in Women's Health

journal homepage: www.elsevier.com/locate/crwh

A ruptured vulvar labial artery pseudoaneurysm causes a secondary postpartum hemorrhage: A case report

Marie-Claire Leaf [a,*], Luke Schmidt [a], Tasha Serna-Gallegos [b], Felicia Lane [c]

[a] Department of Obstetrics and Gynecology, University of California, Irvine-Medical Center, Orange, CA, USA
[b] Division of Female Pelvic Medicine and Reconstructive Surgery, University of New Mexico, Albuquerque, NM, USA
[c] Division of Female Pelvic Medicine and Reconstructive Surgery, University of California, Irvine-Medical Center, Orange, CA, USA

ARTICLE INFO

Article history:
Received 11 February 2020
Received in revised form 18 February 2020
Accepted 19 February 2020

Keywords:
Pseudoaneurysm
Postpartum hemorrhage
Obstetric care
Case report

ABSTRACT

Background: Postpartum hemorrhage is the most common cause of maternal morbidity in the United States. However, secondary postpartum hemorrhage is rare and includes pseudoaneurysms, which represent only 3.3% of all cases of secondary postpartum hemorrhage. Vulvar labial artery pseudoaneurysm had never been reported in the literature.
Case: This is a case of ruptured vulvar labial pseudoaneurysm leading to secondary postpartum hemorrhage. Computerized tomography angiography showed it to be located in a distal branch of the vulvar labial artery. This location is unique, although there are reported cases of pseudoaneurysms in the uterine artery. The patient was successfully treated with arterial embolization.
Conclusion: Recognition of a ruptured pseudoaneurysm as the cause of postpartum hemorrhage allows for its proper management by arterial embolization.

1. Introduction

Postpartum hemorrhage is the most common cause of maternal morbidity in the United States [1]. Primary postpartum hemorrhage encompasses the majority of hemorrhage cases and is defined as over 1000 mL blood loss within 24 h after delivery. Causes of primary postpartum hemorrhage include uterine atony, lacerations, retained placenta, or defects of coagulation [1]. In contrast, secondary postpartum hemorrhage occurs more than 24 h after delivery and up to 12 weeks postpartum and is rare, with an incidence reported to be 0.23% to 1% of all pregnancies [1,2]. The most common causes of secondary postpartum hemorrhage include sub-involution of the placental bed, retained products of conception, or infection such as endometritis. These can generally be treated with uterotonics, dilation and curettage, or antibiotics. Less frequently, a pseudoaneurysm can cause a secondary postpartum hemorrhage, which is the focus of this case study. A pseudoaneurysm is a collection of blood in the outer layer of the artery that remains contained. Pseudoaneurysms account for an estimated 3.3% of all secondary postpartum hemorrhages [2].

The majority of case reports describe pseudoaneurysms of the uterine artery leading to significant uterine bleeding. Although data are limited, one retrospective study showed that 75% of pseudoaneurysms were located within the uterine arteries [3]. In documented cases of ruptured pseudoaneurysm, whether after a cesarean section or a vaginal delivery, arterial embolization has resulted in cessation of uterine bleeding in all patients [3–5]. The goal of this case report is to describe a case of a ruptured vulvar labial artery pseudoaneurysm leading to secondary postpartum hemorrhage. This is an extremely rare anatomical location for a pseudoaneurysm after obstetrical deliveries; it has not hitherto been described in the literature (a detailed review of PubMed revealed no published reports). Recognition of a pseudoaneurysm as the etiology of secondary postpartum hemorrhage is an important addition to the literature. Early diagnosis is facilitated by pelvic computerized tomography (CT) angiography and embolization is an efficacious treatment of this complication.

2. Case

A 28-year-old woman, gravida 1 para 1, who underwent a normal spontaneous vaginal delivery at term, was transferred to a tertiary hospital on postpartum day one for a higher level of care in the setting of a worsening secondary postpartum hemorrhage. The etiology was described by the referral hospital as significant vaginal lacerations requiring extensive vaginal and perineal repair. Her condition was complicated by disseminated intravascular coagulopathy (DIC) diagnosed by hypofibrinogenemia (fibrinogen = 31 mg/dL). The referral hospital had an initial estimated blood loss of 3.5 L , which, in combination with her DIC, warranted administration of 14 units of packed red blood cells (pRBC), 3 units of cryoprecipitate, 4 units of fresh frozen

* Corresponding author at: 333 City Blvd W, Suite 1400, Orange, CA 92868, USA.
E-mail address: marieclm@hs.uci.edu (M.-C. Leaf).

plasma (FFP), and 1 unit of platelets. Once she arrived at the tertiary hospital, she was transferred to the intensive care unit (ICU) and intubated. The laboratory findings were notable for a hemoglobin of 9.0 g/dL and a fibrinogen of 372 mg/dL, which had normalized compared with the reported fibrinogen from the referral hospital. The pelvic exam was remarkable for moderate swelling of the perineum, and a small vaginal genital hiatus with many sutures palpated along the posterior vaginal wall that were intact. There was scant vaginal bleeding. After observation for approximately 24 h at the hospital (postpartum day two) with normal postpartum lochia, the patient was stable and successfully extubated. She remained stable with a hemoglobin of 8.5 g/dL, a fibrinogen of 462 mg/dL, and coagulation panel within normal limits (prothrombin time 14.6 s, international normalized ratio of 1.1, and partial thromboplastin time 31.2 s), and she was discharged home on hospital day four (postpartum day four).

The patient presented to the emergency department on the following day (postpartum day five) with profuse vaginal bleeding, which had started one hour prior to arrival, concerning for another secondary postpartum hemorrhage. Her vital signs were remarkable for tachycardia (heart rate = 97–112 beats per minute) and hypotension (blood pressures = 60–98 / 31–61 mmHg). Her pelvic exam was notable for edematous, non-ecchymotic labia and a large palpable defect proximal to the hymen and to the intact suture line, along the posterior wall of the vagina, filled with dark red blood clots concerning for a ruptured rectovaginal hematoma. Furthermore, the rectovaginal septum was intact. Vaginal packing was placed and the blood transfusion protocol was initiated. A bedside trans-abdominal ultrasound showed a thin endometrial stripe at 0.8 cm and a small amount of blood clots in the vagina. Given ultrasound findings that did not correlate with the degree of vaginal bleeding and maternal instability, CT angiography of the abdomen and pelvis was completed. It demonstrated active extravasation of contrast into the right labia that appeared to be originating from the anterior division of the internal iliac artery (Figs. 1 and 2). Interventional radiology (IR) was consulted for possible uterine artery embolization. The IR team suggested against a minimally invasive approach such as embolization because of concern of labial and/or vaginal necrosis given that the pseudoaneurysm was located in a distal branch. A multidisciplinary meeting with Vascular Surgery and Female Pelvic Medicine and Reconstructive Surgery was called and the consensus was that an open approach to a vaginal or labial aneurysm would ultimately make it difficult to isolate such a distal branch of the internal iliac artery.

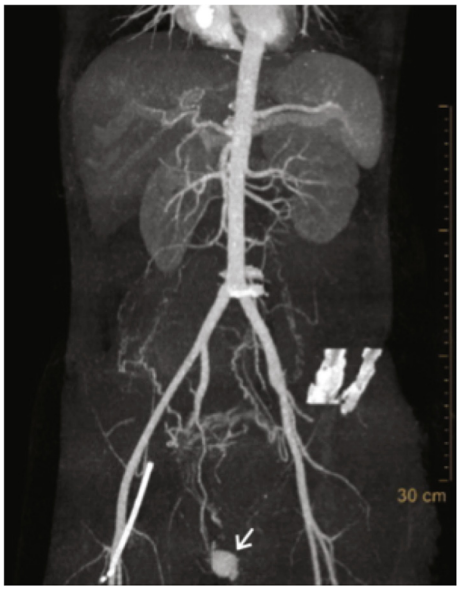

Fig. 2. Initial CT angiography demonstrating active extravasation of contrast into the right labia that appeared to be originating from the anterior division of the internal iliac artery.

Additionally, the perineal blood supply contains numerous arterial branches and collateral vasculature from both the internal and the external iliac arteries, making labial necrosis unlikely by the embolization of one distal vessel [6]. However, given the IR team's reluctance to recommend embolization, the decision was made to observe the patient's vaginal bleeding, resuscitate her with blood products and repeat a CT angiography promptly if bleeding continued.

After two days of observation (postpartum day six), her vaginal bleeding slowly continued, despite daily vaginal packing changes. A repeat CT angiography of the pelvis demonstrated a persistent 2.4 cm pseudoaneurysm in the right labia originating from a branch of the anterior division of the right internal iliac artery, with evidence of active extravasation from the pseudoaneurysm posteriorly (Fig. 3). Given the bleeding had not resolved with conservative measures, IR was amenable to embolization. On hospital day three (postpartum day seven), the patient underwent a pelvic angiogram with percutaneous

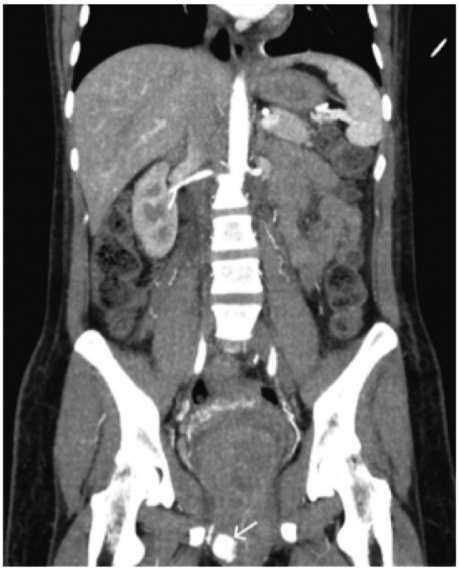

Fig. 1. Initial CT of the pelvis and abdomen demonstrating active extravasation of contrast into the right labia.

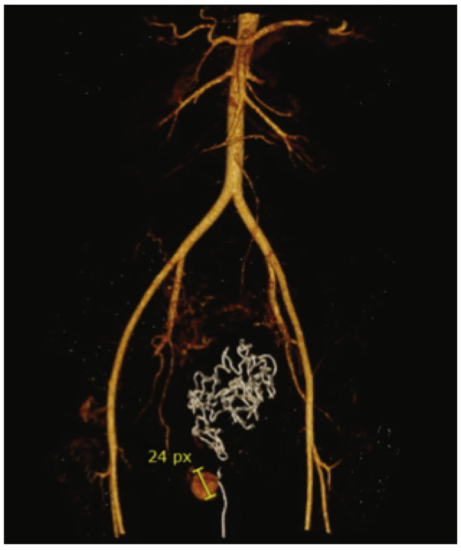

Fig. 3. Repeat CT angiography demonstrating a pseudoaneurysm on a branch of the anterior division of the right internal iliac artery.

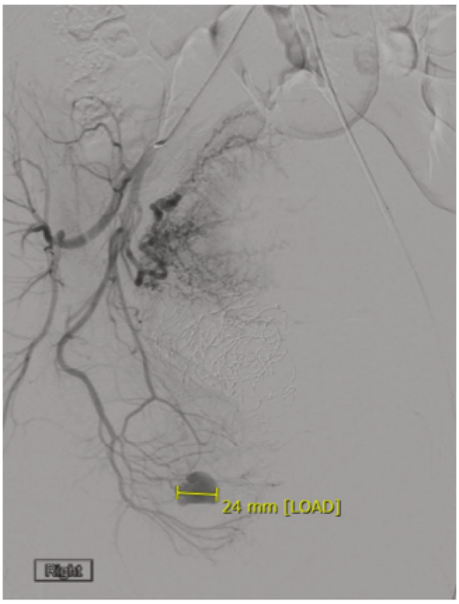

Fig. 4. Right pelvic angiogram with embolization by interventional radiology.

ultrasound-guided thrombin injection and N-butyl cyanoacrylate (NCBA) embolization of the right labial artery by the IR team (Fig. 4). Following this procedure, the patient's bleeding decreased significantly. She was discharged home on hospital day six (postpartum day ten). On postpartum day 13, she returned for a short-interval follow-up visit. Her bleeding had ceased and she was meeting all early postpartum milestones. On a brief pelvic exam, there was no evidence of vaginal or labial necrosis. The plan was for her to return to clinic for a 6-week postpartum visit, but she was lost to follow-up despite multiple attempts to contact her.

3. Discussion

This case brings to light a rare, yet life-threatening cause of secondary postpartum hemorrhage. To date, the literature has predominantly described the uterine arteries as the origin of postpartum pseudoaneurysms, with only two other reports found describing it in another location. One of the two cases found it originating from the left internal pudendal artery and the other in the vaginal artery. In both cases, controlled bleeding was successfully achieved with arterial embolization [7,8]. This case report is the first to describe a suspected vulvar labial pseudoaneurysm as the etiology of a severe secondary postpartum hemorrhage.

Early identification of a pseudoaneurysm can lead to decreased maternal morbidity from blood transfusions and rapid recovery by arterial embolization. The best diagnostic imaging has yet to be established in the literature, but based on this case report, pelvic CT angiography should be considered. It allows for a detailed anatomical survey of the pelvic vasculature, facilitating a more accurate target for IR treatment, when compared with another modality such as ultrasound.

In conclusion, a ruptured pseudoaneurysm should be considered when a patient presents with a secondary postpartum hemorrhage, unresponsive to conservative measures and after more common etiologies have been ruled out by clinical presentation and ultrasound imaging. Pelvic CT angiography should be the imaging of choice to detect a pseudoaneurysm. The IR team should be consulted for embolization quickly, as conservative measures are unlikely to control the bleeding from an aneurysm. Even in the event of a pseudoaneurysm in a distal branch such as in this case with the vulvar labial artery, embolization can successfully control the bleeding, with no evidence of long-term side-effects such as labial or vaginal necrosis.

Contributors

Marie-Claire Leaf was the primary author of the manuscript.
Luke Schmidt was a secondary author of the manuscript.
Tasha Serna-Gallegos was a secondary author of the manuscript.
Felicia Lane was the final editor of the manuscript.

Conflict of Interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

Funding

No funding from an external source supported the publication of this case report.

Patient Consent

Obtained.

Provenance and Peer Review

This case report was peer reviewed.

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