Case Reports in Women's Health 44 (2024) e00667

Contents lists available at ScienceDirect

Case Reports in Women's Health

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

Successful management of intraoperative cesarean section bleeding due to cervical arteriovenous malformation: A case report

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ARTICLE INFO

Keywords:
Arteriovenous malformations
Obstetric labor complications
Obstetric surgical procedure
Uterine bleeding

ABSTRACT

Cervical arteriovenous malformation is an exceedingly rare condition that can lead to antepartum hemorrhage, posing risks for both maternal and perinatal morbidity. We report the case of a 30-year-old primigravida, at a gestational age of 31^{+2} weeks, who presented to hospital with antepartum hemorrhage. A speculum examination revealed a 500 mL blood clot. Despite a thorough examination, the cause of the antepartum hemorrhage remained elusive. An emergency cesarean section was done due to hypovolemic shock and a fetal NICHD category III assessment. Following the delivery of the fetus, a pulsatile tubular structure was identified at the endocervix and biopsied. Suture ligation followed by insertion of a Bakri balloon, effectively controlled the bleeding with blood loss of 1200 mL. Histopathology confirmed the diagnosis of arteriovenous malformation. This case underscores the importance of recognizing cervical arteriovenous malformation and demonstrates the effectiveness of suture ligation and balloon tamponade in managing associated hemorrhage, offering insights for similar cases.

1. Introduction

Antepartum hemorrhage is a leading global cause of perinatal and maternal morbidity and mortality. While placenta previa and placental abruption are significant causes of this condition [1], unexplained antepartum hemorrhage, or antepartum hemorrhage of unknown origin, accounts for approximately half of all such cases [2]. Antepartum hemorrhage has been linked to preterm delivery [3].

Cervical arteriovenous malformation (AVM) during pregnancy is an exceedingly rare disorder. It is capable of causing severe antepartum and intrapartum hemorrhage, thereby jeopardizing both the mother and fetus. Only one published report could be found of cervical AVM during pregnancy, which originated from Korea [4]. The diagnosis was based on Doppler ultrasonography and magnetic resonance imaging during the antenatal period without pathological confirmation. The pregnancy concluded with favorable outcomes.

Our report presents a further case of cervical AVM in pregnancy, with the diagnosis based on intraoperative findings. Tissue samples were submitted for pathological analysis to validate the diagnosis. Moreover, we describe an alternative technique to arrest the hemorrhage induced by this condition while preserving fertility.

2. Case Presentation

A 30-year-old primigravida presented at 31^{+2} weeks of gestation with an initial episode of third-trimester antepartum vaginal bleeding. Her prior antenatal care was unremarkable, second-trimester ultrasonography revealed no fetal anomalies, and the placenta was positioned posteriorly in the mid-section with no noted cervix involvement. Upon admission, her vital signs were stable, with a blood pressure of 119/78 mmHg and a heart rate of 109 bpm. A speculum examination revealed approximately 500 mL of fresh blood in the vagina. The cervical os appeared closed without abnormal lesions. Transabdominal ultrasonography indicated a single viable fetus weighing 1429 g in a vertex presentation. The placenta was low-posteriorly located with adequate amniotic fluid. Doppler ultrasonography was not conducted.

Thirty minutes after being transferred to the labor room, the patient exhibited continuous vaginal bleeding, amounting to 300 mL, with deteriorating vital signs (blood pressure had dropped to 60/30 mmHg). External fetal monitoring indicated a National Institute of Child Health and Human Development (NICHD) category III status. An emergency cesarean section was initiated due to the antepartum hemorrhage and the NICHD category.

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https://doi.org/10.1016/j.crwh.2024.e00667
Received 26 October 2024; Received in revised form 12 November 2024; Accepted 13 November 2024
Available online 19 November 2024

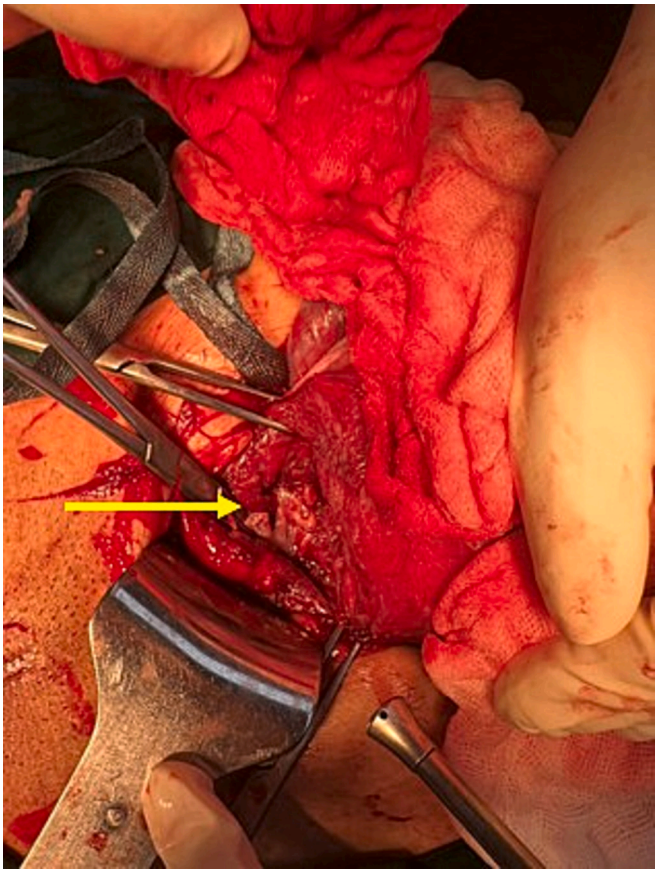

Fig. 1. Intraoperative photograph. Whitish tubular-structured tissue measuring 3 × 3 cm in the endocervical region exhibiting pulsatile bleeding (indicated by the arrow).

A low midline skin incision was made. No vascular abnormalities were observed in the lower uterine segment. After a low transverse uterine incision, the newborn was safely delivered (body weight 1310 g; APGAR scores 1 and 3 at 1 and 5 min, respectively). The placenta was fully extracted from its posterior position. Notably, a whitish, tubular structure measuring 3 × 3 cm with pulsatile bleeding was identified in the endocervical region (Fig. 1). Despite attempts to halt the bleeding with figure-of-eight suture ligation, the blood flow persisted, albeit slower. A Bakri balloon was inserted via the uterine incision and inflated with 150 mL of water. The bleeding was finally controlled. The patient lost an estimated 1200 mL of blood but experienced no other intraoperative complications. She was transfused with 4 units of packed red blood cells. The Bakri balloon remained in place for 24 h. After its deflation, no rebleeding occurred, and the patient was discharged on postoperative day 3.

For diagnostic clarification, a biopsy was taken before the Bakri balloon insertion. Histologically, it displayed multiple interconnected vascular channels of varying diameters and wall thicknesses (Fig. 2). Higher magnification revealed a sporadic smooth muscle layer and an endothelial lining without dysplastic characteristics. The surrounding stromal tissue exhibited decidual changes, with no lymphatic channels and minimal thrombosis. Additional staining and immunohistochemical tests revealed an irregular, smooth muscle layer with heterogeneous diameters (Fig. 3). These attributes were consistent with AVM.

The newborn initially presented with respiratory distress syndrome but recovered and was discharged on day 49. He is currently in good health with normal neurodevelopment.

At follow-up on postoperative day 10, the mother's transvaginal ultrasonography displayed an 8 × 7 mm hypodense lesion with significant vascular flow in the endocervical region. By postoperative day 43, this had diminished, as shown in Fig. 4.

3. Discussion

The patient in the case reported here had an undetected cervical AVM that induced severe hemorrhage during the third trimester of pregnancy. A Bakri balloon tamponade via a uterine incision successfully managed the bleeding.

Arteriovenous malformations are uncommon and can be either congenital or acquired. Typically manifesting as profuse vaginal bleeding, AVMs can threaten life. The initial diagnosis often involves Doppler ultrasound, but computed tomography angiography remains the gold standard. However, the latter is not always feasible in emergency settings. Most uterine AVMs are acquired following uterine curettage, cesarean sections, pelvic infections, gestational trophoblastic diseases, or exposure to diethylstilbestrol [5]. Conversely, congenital AVMs are exceedingly rare and are postulated to arise from anomalies in primitive vascular structures [6,7].

Most gynecologic AVMs localize to the myometrial regions, deriving their supply from uterine vessels. Notably, only two cervical AVM cases seem to have been published. In a report by Kim et al., Doppler ultrasonography at 32 weeks of gestation displayed numerous rapidly flowing vessels. Speculum examination revealed engorged vessels on the surface of the cervix. Magnetic resonance imaging confirmed the lesion as a cervical AVM. Despite an emergency cesarean section 2 weeks later due to massive bleeding, the patient had an uncomplicated postpartum period. Bleeding was halted via suture ligation of the cervical AVM and bilateral uterine artery ligation [4]. In the second report, Val-Bernal and colleagues discovered a cervical AVM posthysterectomy in a nonpregnant patient with menorrhagia. The AVM had preoperatively been misdiagnosed as having a sizable uterine leiomyoma [8].

The present case resembled the two published cases insofar as the patient had no history of pelvic surgery, including curettage. However, the case diverged from these studies in that the AVM remained undiagnosed until delivery, when it presented as an emergency. The urgency of the patient's condition precluded a comprehensive evaluation for a definitive diagnosis before the cesarean section. Although placenta previa, vasa previa, and placental abruption are primary considerations for third-trimester antepartum hemorrhages, the pulsatile bleeding pointed to a potential cervical AVM.

This is the first report of a cervical AVM supported by pathological findings. Among the differential diagnoses were pregnancy-associated vascular dilation, cavernous hemangioma, and lymphangioma. As uniformly sized vascular channels characterize these alternatives, none corresponded with the patient's pathological presentation of irregular vascular channels. Thus, an AVM diagnosis was the most plausible.

Given the scarcity of documented cervical AVM-induced hemorrhages, we advocate prioritizing hemodynamic stabilization. Several interventions are available to arrest the bleeding, depending on the

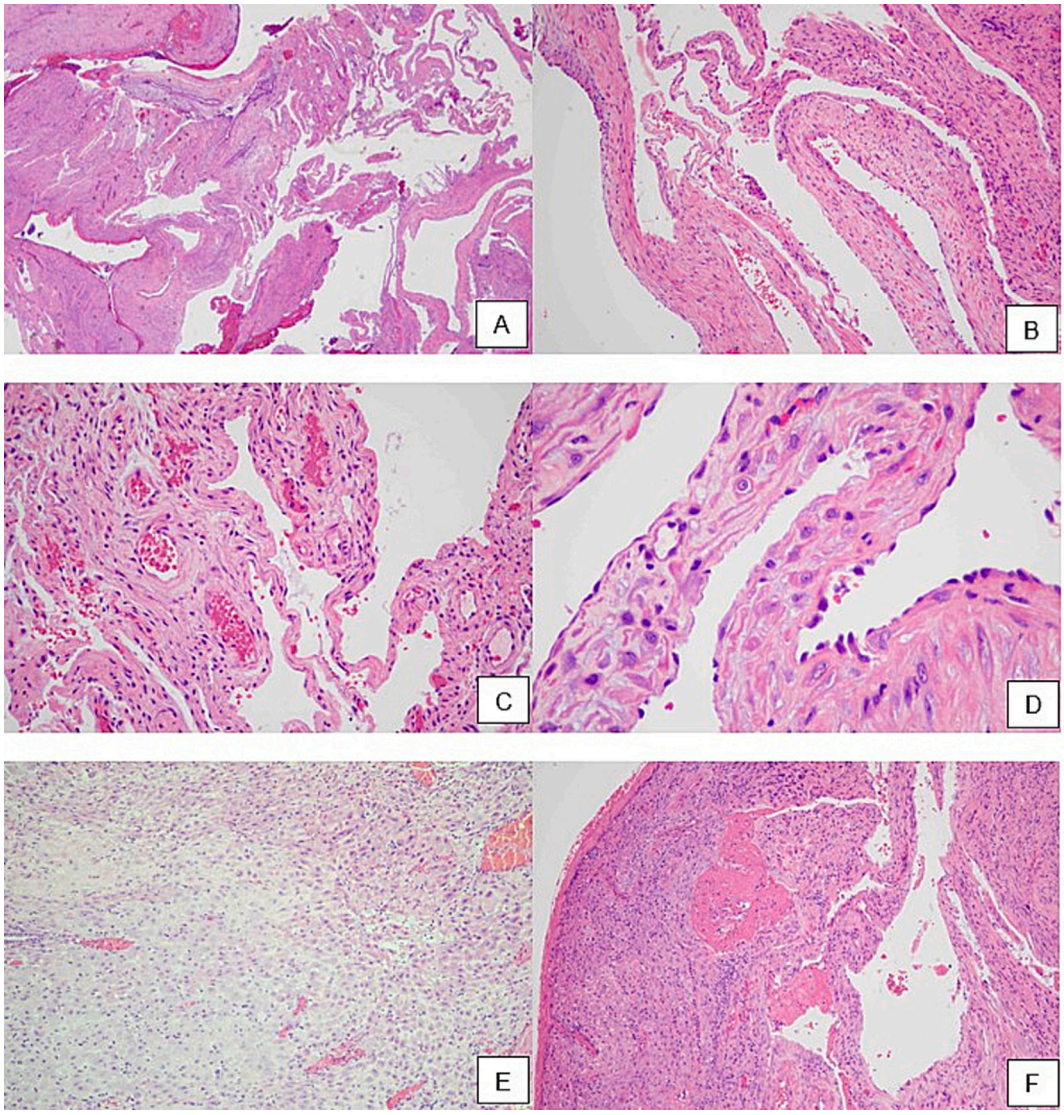

Fig. 2. Microscopic findings of the lesion. A: The tissue displays a mix of large thick-walled and thin-walled vascular channels (HE; 20×). B: Interconnecting channels span between large and small vessels (HE; 100×). C: Small vessel filled with blood (HE; 200×). D: Endothelium cells present focal plumping but are negative for dysplasia (HE; 400×). E: The residual tissue illustrates decidualized stroma (HE; 100×). F: Vascular thrombi are discernible (HE; 100×).

hemorrhage's severity and the patient's reproductive aspirations. Options include suture ligation, Bakri balloon tamponade, uterine artery ligation, and endovascular procedures, with hysterectomy a last resort if bleeding persists. After the initial suture ligation slowed the hemorrhage in the present case, the bleeding was successfully stemmed with a Bakri balloon. By 6 weeks postoperatively, the lesion had receded. This outcome paralleled the observations of Kim et al. and reinforced the supposition that the cervical AVM might have emerged due to the hyperdynamic state during pregnancy [4].

In conclusion, cervical AVM is an exceedingly rare and life-threatening disorder. Should third-trimester antepartum hemorrhage manifest without an obvious etiology, clinicians must maintain a high suspicion for this condition. Combining suture ligation with a Bakri balloon tamponade offers a viable strategy to halt AVM-induced bleeding.

Contributors

Vitcha Poonyakanok contributed to patient care, conception of the case report, and drafting the manuscript.

Kridtin Jarutatsanangkoon contributed to acquiring and interpreting the data, and revised and submitted the figures.

Pattarawalai Talungchit contributed to patient care, undertaking the literature review, revising the article critically for important intellectual content, and drafting the manuscript.

All authors approved the final submitted manuscript.

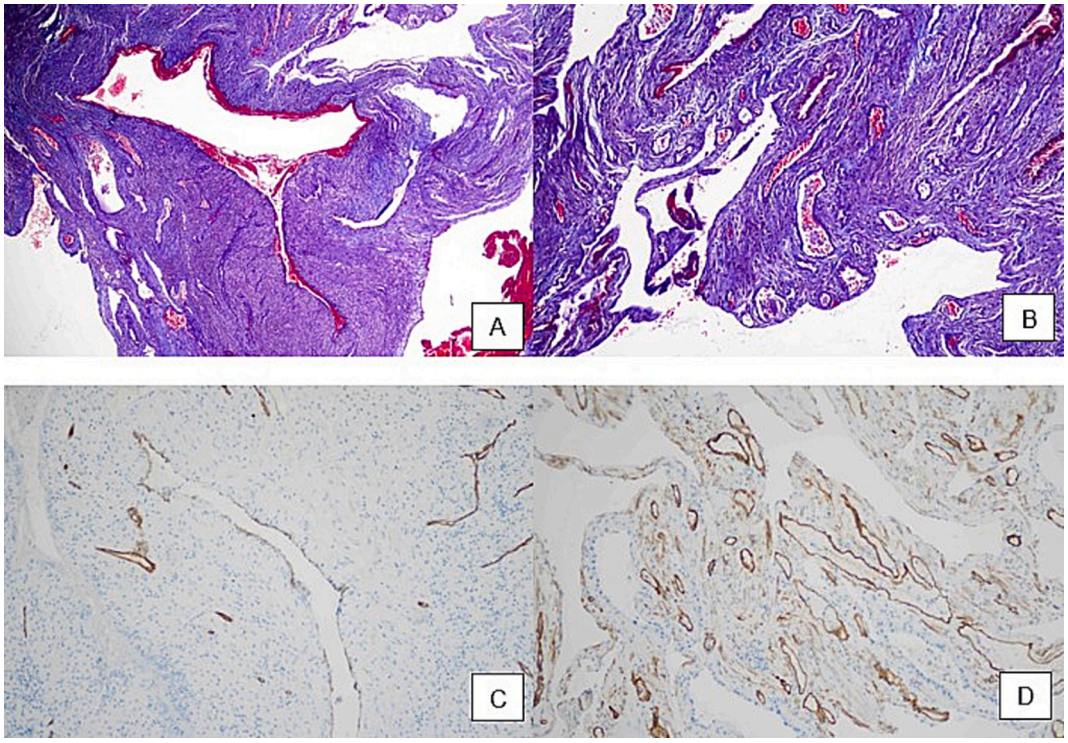

Fig. 3. Specialized stains from the lesion. A and B: Masson-trichrome stain reveals sparsely distributed disorganized smooth muscle fibers (MT stain; 40× and 100×). C and D: Immunostaining for CD34 confirms the presence of endothelial lining (CD34, 100×).

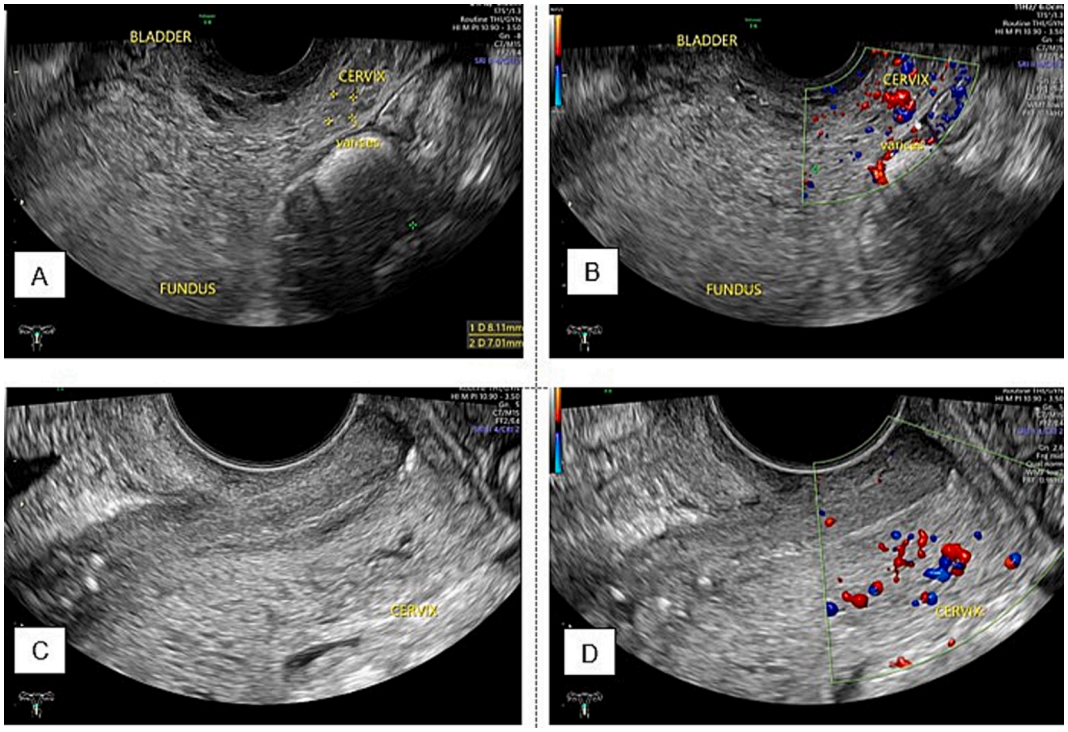

Fig. 4. A and B: Transvaginal ultrasonography of the cervical varix on postoperative day 10. C and D: Transvaginal ultrasonography of the cervical varix on postoperative day 43.

Funding

This work did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Patient consent

Written informed consent was obtained from the patient for publication of this case report and the accompanying images.

Provenance and peer review

This article was not commissioned and was peer reviewed.

Acknowledgments

The authors thank the patient, who agreed to allow us to publish the clinical data.

Conflict of interest statement

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

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