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Trans-arterial embolization for intractable primary postpartum hemorrhage caused by arterial aneurysms with arteriovenous fistulas in the lower vagina bilaterally: a case report

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Abstract

Failure of conservative management for controlling postpartum hemorrhage (PPH) is not uncommon, particularly when PPH is caused by vascular lesions. Awareness of this possibility and initiating timely trans-arterial embolization (TAE) are essential for improving the outcome. Herein, we describe the case of a 34-year-old woman presenting with arterial aneurysms with arteriovenous fistulas in the lower vagina bilaterally, which caused intractable PPH. Conservative management failed to resolve the PPH; however, TAE successfully controlled the bleeding, and the patient recovered smoothly. Knowledge of this possible etiology for intractable PPH is crucial for timely TAE. This case report aims to highlight the pivotal role of TAE in detecting and treating this unusual cause of PPH.

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Introduction

Postpartum hemorrhage (PPH) is a major cause of maternal morbidity and mortality worldwide.[1] PPH is usually clinically assessed by caregivers in emergent situations.[2] The most common etiologies of PPH are genital tract laceration, uterine atony, uterine rupture, placental retention, and coagulopathy.[3–5] Rarely, accompanying vascular anomalies, such as aneurysms, arteriovenous fistulas (AVFs), and arteriovenous malformations may cause aggressive bleeding.[6] Trans-arterial embolization (TAE) is an effective and minimally invasive procedure for managing PPH.[4,5] Here, we present a case of lower vaginal arterial aneurysms with AVFs that caused intractable PPH, which was successfully controlled by TAE after failure of conservative management.

Case report

All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional and/or national research committee, and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. Informed verbal and written consent for trans-arterial embolization and signed consent to publish this case report were obtained from the patient. This case report obtained institutional review board approval from the Chang Gung Medical Foundation Institutional Review Board, No: 202001292B0. The reporting of this study conforms to the CARE guidelines.[7]

A 34-year-old woman with an obstetric history of G3P0AA2 was 38+ weeks pregnant and otherwise healthy. She was admitted for vaginal delivery after spontaneous rupture of the amniotic membranes. She sustained a second-degree perineal laceration during the delivery and underwent primary repair. PPH occurred after delivery, and initial medical management was immediately initiated but was ineffective. After blood transfusion and vaginal packing with surgical pads, she temporarily returned to normotensive status. As vaginal bleeding recurred, angiography was performed, and arterial aneurysms with AVFs (Figure 1) fed by the internal pudendal arteries (Figure 2) were identified in the lower

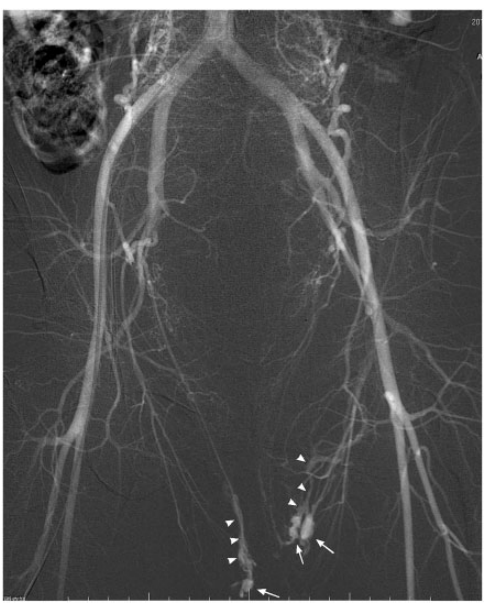

Figure 1. Pelvic angiography showing arterial aneurysms (arrows) with arteriovenous shunting, with early draining veins (arrowheads) in the lower vagina bilaterally.

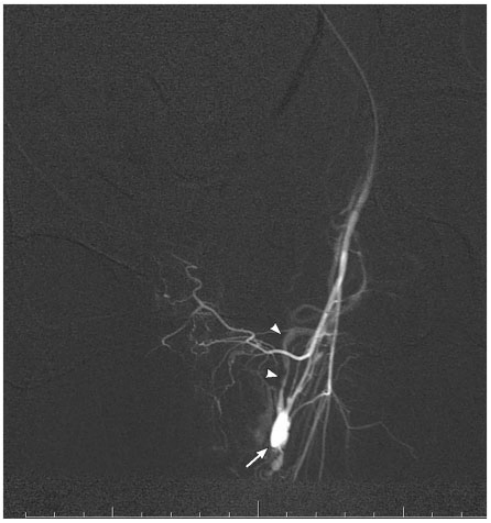

Figure 2. Left internal iliac arteriography clearly showing an arterial aneurysm (arrow) with early draining veins (arrowheads) arising from the internal pudendal artery. Trans-arterial embolization with coils (VortX^{TM}-18, 3 mm $\times$ 2.5 mm; Boston Scientific, Marlborough, MA, USA) was performed at both sides of the vagina by super-selective catheterization of the feeding vessels.

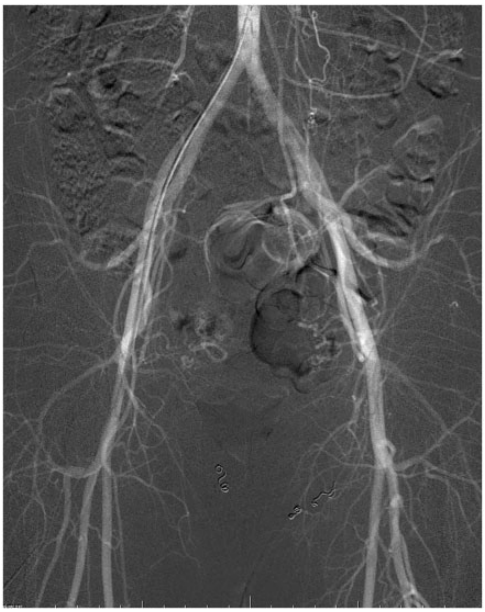

Figure 3. Follow-up angiography performed 3 months after trans-arterial embolization showing complete obliteration of the previous vascular lesions.

vagina, bilaterally. Hemostasis was then achieved by TAE of the aneurysms with metallic coils placed in the distal internal pudendal arteries. After 5 days of hospitalization, the patient recovered well and was discharged smoothly, with no specific complaints at the follow-up examination. Repeat angiography 3 months later revealed obliteration of the previous vascular lesions (Figure 3).

Discussion

The most common etiologies of PPH are genital tract laceration, uterine atony, uterine rupture, placental retention, and coagulopathy.[3–5] Routine conservative management for PPH comprises resuscitation, blood transfusion, and uterine control, such as with the administration of uterotonic drugs, uterine compression, and intrauterine balloon tamponade. If conservative management fails, TAE or surgical management should be initiated without delay.[6]

As reported, angiographic active extravasation is a factor in TAE failure; ruptured arterial vascular lesions typically results in vigorous bleeding that may render conservative treatment ineffective. Vaginal arterial aneurysms with AVFs as a cause of PPH have not been previously reported in the English literature. In the current patient, the rare condition of arterial aneurysms with early draining veins in the lower vagina bilaterally was clearly shown by angiography, and the aneurysms were managed with TAE.

There is no established theory explaining the association of vaginal vascular anomalies with gestation. Angiographic studies have demonstrated that arteriovenous communication might exist in normal uterine pregnancies (0.9%), with higher rates in

women who experience miscarriages, and with missed abortions (20%) and ectopic pregnancies (39%).[8] Retained products of conception (RPOC) from miscarriage or delivery may stimulate the development of hypervascular placental polypoid masses and neo-vascular lesions arising from the villi over time.[9] AVFs may develop over time as the villi undergo necrosis, which explains why AVFs develop in late gestation.[9] Most vascular lesions associated with RPOC are located in the uterus or ectopic gestational tissue near the uterus,[9] unlike in our case, in which the lesions were located in the vagina. However, there was a lack of tissue specimens in our case to confirm the existence of RPOC.

Anatomically, there are several arteries that supply the vagina. The anterior and lateral surfaces of the vagina are fed by the vaginal artery, the middle portion is fed by the inferior vesicular artery, the lower portion is fed by the internal pudendal artery, and the posterior surface is fed by the middle rectal artery.[4] Familiarity with this vascular anatomy is key to facilitate and complete TAE in the emergent situation of PPH.

TAE is effective for controlling PPH, with a high success rate, and this method is even recommended as first-line therapy.[10] Although successful TAE could technically preserve the uterus, patients should be counseled that fertility might be affected.[11] The most commonly used embolic material in TAE for PPH is gelatin sponge particles, with metallic coils or N-butyl cyanoacrylate as bail-out materials.[3–5] With the uncommon condition of vaginal arterial aneurysms with AVFs causing PPH, small particles and liquid embolizers should be used carefully in TAE as they may pass through the AVF and embolize the inferior vena cava and pulmonary arteries.

The vascular anomaly of arterial aneurysms with AVFs in the lower vagina bilaterally contributing to intractable PPH is rare and renders conservative treatment ineffective. Obstetricians should be aware of this unusual lesion to facilitate timely TAE. Angiography is especially pivotal and should be performed preoperatively to detect and treat this uncommon condition.

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Author Contributions

CHW and PWW drafted the manuscript. HYC performed the surgery. CHW, YCW, and HYC collected and analyzed the clinical and imaging data. CHW critically revised the manuscript for important intellectual content. CHW and PWW edited the manuscript. CHW and PWW reviewed the draft. All authors read and approved the final manuscript.

Declaration of conflicting interests

The authors declare that there is no conflict of interest.

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