

Postpartum hemorrhage secondary to pseudoaneurysm of the uterine artery - A case report*

BY VANESSA MARIE TY LIM, MD AND THELMA MARIE AVENDANO ELIO MD, FPOGS

Department of Obstetrics and Gynecology, St. Luke's Medical Center - Quezon City

ABSTRACT

Obstetric hemorrhage is the most important cause of maternal mortality worldwide approximately 27.1%. Pseudoaneurysm of the uterine artery is rare but a potentially life-threatening complication reported to occur. In the Philippines, experience with these kinds of vascular abnormalities is limited. This is a case of a 26-year old Gravida 2 Para 1 (1011) eight weeks post-cesarean section with profuse vaginal bleeding. Transvaginal ultrasonography with doppler revealed arterio-venous malformation of the cervix with high arterial pulsations. Pelvic angiography revealed brisk contrast extravasation and pooling at the fundal branch of the left uterine artery consistent with pseudoaneurysm for which she successfully underwent superselective coil embolization. Repeat ultrasonography after one week showed absence of arterial pulsations on the cervical echogenicity. Despite a recent proliferation of reports, uterine artery pseudoaneurysm still remains to be a rare cause of unexplained postpartum hemorrhage, which requires a high index of clinical suspicion for diagnosis.

Keywords: Cesarean section, postpartum hemorrhage, pseudoaneurysm, uterine artery embolization

INTRODUCTION

Obstetric hemorrhage is the single most important cause of maternal mortality worldwide accounting for 27.1 percent of all direct maternal deaths.¹ It is defined as blood loss above 500 milliliters for vaginal delivery and 1000 milliliters for cesarean section; or blood loss greater than 500 milliliters in the first 24 hours following delivery; or a 10 percent decrease in the hemoglobin or hematocrit level; or a need for blood transfusion.² Postpartum hemorrhage accounts for 19.7 percent of all obstetrical hemorrhage.¹ This can be classified as primary or secondary. Primary postpartum hemorrhage can be due to uterine rupture, uterine atony and retained placenta, as well as, serious vaginal trauma, uterine inversion, and extra-uterine pregnancy. Secondary hemorrhage (or late/delayed hemorrhage) is excessive vaginal bleeding that occurs between 24 hours and six weeks following delivery² or even up to 12 weeks postpartum.³ Etiology of secondary hemorrhage include subinvolution of the placental site, retained products of conception, and endometritis. Other causes of delayed hemorrhage may be von Willebrand disease or other inherited coagulopathies.³ These usually present within 7-14 days post-delivery. Such bleeding when occurring later in the puerperium may be confused with the first menstrual period which is often the result of an anovulatory bleed and thus, may be heavy, prolonged and

painful. In these cases, prompt recognition and treatment is critical, although definitive management will vary depending on the cause of the bleeding. Once patient has been resuscitated and stabilized, initial treatment includes use of uterotonic agents and antibiotics. A transvaginal or pelvic ultrasound is often the first diagnostic procedure done, though it may not reliably differentiate between retained placental fragments and blood clots within the uterine cavity.

A pseudoaneurysm of the uterine artery is a rare but potentially life-threatening complication which has been reported to occur, not only following cesarean section, but even after uncomplicated vaginal delivery.^{4,5} A pseudoaneurysm is an extra-luminal collection of blood with turbulent flow that communicates with the parent vessel through a defect in the arterial wall which may result from local trauma to the vessel following surgery, neoplasm or infection leading to perivascular hematoma. Following hematoma formation, there is central liquefaction leaving the cavity with persistent communication between the parent artery and the hematoma. The absence of a three-layered arterial wall lining differentiates it from a true aneurysm which is less common.^{6,7} Typically, a pseudoaneurysm contains a single layer of loose connective tissue which enlarges due to extraluminal turbulent blood flow and subsequently susceptible to rupture.⁸

Review of Pubmed/Medline, and EBSCO for the past five years using the keywords Pseudoaneurysm + Postpartum hemorrhage + Cesarean Section + Embolization yielded 17 articles all with full access available. All of the

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reported cases of pseudoaneurysm of the uterine artery presented with active vaginal bleeding except in one case report wherein the patient was asymptomatic.⁹

In the Philippines, there is only one reported case of delayed postpartum hemorrhage due to ruptured uterine artery pseudoaneurysm. Thus, experience with these kinds of vascular abnormalities is limited. Here is one recent case of ruptured uterine artery pseudoaneurysm.

CASE REPORT

This is a case of a 26-year old Gravida 2 Para 1 (1011) Filipino woman status post Cesarean Section who was admitted for the first time for profuse vaginal bleeding.

Antenatal history and review of past medical history revealed a diagnosis of gestational diabetes mellitus at six months age of gestation. She was seen and managed by an endocrinologist with diet and capillary blood glucose monitoring only. No other medications were taken. She has no known drug allergies. Her first pregnancy, one year prior to the current pregnancy, was a non-induced, non-septic, spontaneous abortion at eight weeks for which she underwent completion curettage. Post-operative course was unremarkable. Follow-up transvaginal ultrasound done eight months after the curettage, revealed a normal sized anteverted uterus, with normal cervix, proliferative endometrium and bilateral polycystic ovaries. Her family medical history was unremarkable. The patient is a non-smoker, not an alcoholic beverage drinker and has been regularly menstruating monthly since 12-years old consuming three to four pads per day lasting for three days. She has a monogamous relationship with one sexual partner and has no history of any oral contraceptive use.

For this pregnancy, first trimester ultrasound at 9 weeks age of gestation revealed a single, live, intrauterine pregnancy at 8 weeks and 2 days age of gestation by crown rump length with a yolk sac and cardiac activity at 178 beats per minute. There was no subchorionic hemorrhage and the cervix was described to be unremarkable, measuring 3.6 x 3.8 x 3.3 cm with normal ovaries and corpus luteum on the right. The rest of her prenatal course and subsequent sonologic evaluations were unremarkable.

The patient delivered at 39 weeks age of gestation by low transverse Cesarean section for malpresentation (transverse lie – fetal back up, fetal head maternal right, left scapula anterior) to a 3,546 grams baby girl, Apgar score of 9 and 9 with a Ballard score of 38 weeks appropriate for age. Intraoperative course was unremarkable. There was a gravid uterus term in size with a well-formed lower uterine segment. The estimated blood loss was 800 cc. The patient was discharged after the third post-operative day with minimal vaginal bleeding that ceased after three weeks. However, vaginal spotting, consuming one panty

liner per day with no other associated symptoms, recurred after one week. On consult with her attending physician, internal examination was unremarkable. Patient was prescribed Methylergonovine maleate 125 micrograms per tablet, one tablet every 12 hours for three days, which eventually ceased the bleeding until this episode. Patient was advised to have a transvaginal ultrasound done if there will be any recurrence of bleeding. Four hours prior to admission, the patient developed profuse vaginal bleeding, approximately one liter associated with occasional dizziness and weakness but with no abdominal pain, hence, she was admitted.

During admission, review of systems was unremarkable. On physical examination, the vital signs on presentation were: blood pressure of 130/80 mmHg; pulse rate of 84 beats per minute; respiratory rate of 20 breaths per minute; and temperature 37.5 degrees Celsius. Physical examination revealed pink palpebral conjunctivae, anicteric sclera, regular heart sounds, and clear lung fields. The abdomen was soft, non-tender with normal bowel sounds. Pelvic examination revealed a normal-sized uterus with a cervix that admits the tip of the finger. The cervix and adnexae were non-tender. Blood clots were visible in the vaginal vault and at the cervical os.

Admitting impression was a 26-year old Gravida 2 Para 1 (1011) postpartum hemorrhage rule out retained products of conception, status post cesarean section for malpresentation.

Patient was placed on nothing per orem. Intravenous access was obtained and blood was sent for a complete blood count, blood typing with RH screening, and routine coagulation profile. Laboratory tests revealed hemoglobin of 10.6 g/dL with normal coagulation profile and blood type of A+ (Appendix A). Urine pregnancy test was negative. The patient remained hemodynamically stable at the emergency room.

A transvaginal ultrasound was facilitated to rule out retained placental fragments. The transvaginal ultrasound revealed an anteverted uterus that was normal in size with a thin endometrium measuring 0.74 cm, characterized as hyperechoic and intact, with normal ovaries (Appendix B, Figure I, A – C). There was note of a distinct region at the left lateral cervical stroma measuring 2.6 x 1.9 x 2.0 cm composed of slightly echogenic borders and a central irregular cystic anechoic structure measuring 1.7 x 0.6 x 1.2 cm which on color Doppler shows bright mosaic color flow and high arterial pulsations which could be an arterio-venous malformation of the cervix (Appendix B, Figure I, D-F). Assessment at this time was delayed postpartum hemorrhage probably secondary to arterio-venous malformation of the cervix. The patient was informed that treatment options were either to perform minimally invasive pelvic angiography with possible embolization,

Appendix A: Tabulation of the Laboratory Results

2014		October 31	October 31	November 1
Complete Blood Count	Hemoglobin (11.6 - 15.5 g/dl)	10.6 g/dl	9.1 g/dl	12.2 g/dl
	Hematocrit (36.0 – 47.0 %)	32.0 %	28.0 %	35.5 %
	Red Blood Cells (4.20-5.40 mil/mm ³)	3.09 mil/mm ³	3.07 mil/mm ³	4.06 mil/mm ³
	White Blood Cells (4800-10800 mm ³)	13230/mm ³	13770/mm ³	12830/mm ³
	Neutrophils (40 – 74 %)	76 %	71 %	75 %
	Lymphocytes (19 – 48%)	18 %	22 %	18 %
	Eosinophils (0 – 7 %)	1 %	1 %	1 %
	Monocytes (3 – 9 %)	4 %	6 %	6 %
	Platelet Count (150, 000 - 400, 000/mm ³)	251, 000 /mm ³	264, 000 /mm ³	171, 000 /mm ³
Partial Thromboplastin Time (29.5 – 39,9 secs)		31.0 secs		
Prothrombin Time Control		11.1 secs		
Prothrombin Time Test (11.9 – 14.2 secs)		12.6 secs		
INR (0.9 – 1.19)		1.05		
Blood and Rh Typing		A Positive		
Serum Creatinine (0.60 – 1.20 mg/dl)		0.52 mg/dl		

or a surgical uterine artery ligation. Patient opted to undergo embolization and was immediately referred to an interventional radiologist for evaluation and possible embolization. Blood products were then prepared. Pre-operatively, hydrocortisone 100 mg per intravenous route was given one hour prior to the planned procedure to decrease the rate of hypersensitivity reaction to the contrast dye to be used in the procedure.

Patient then had another episode of profuse vaginal bleeding amounting to approximately 500cc. Physical examination revealed generalized pallor with vital signs as follows: blood pressure of 100/70 mmHg; pulse rate of 105 beats per minute; respiratory rate of 22 breaths per minute; and temperature 37.8°C. Double intravenous access was secured, 100 ml of six percent hydroxyethyl starch 130/0.4 in 0.9 percent sodium chloride injection (Voluven®) was given on fast drip followed by two units of packed Red Blood Cells. Repeat complete blood count four hours after the baseline revealed hemoglobin of 9.1 g/dL

(Appendix A). Patient became hemodynamically stable. The patient was then transferred to the Catheterization Laboratory and underwent Pelvic angiography and superselective coil embolization of the left uterine artery using Boston Scientific VortX® fibered platinum coils (two 3 x 2.5 mm size) under total intravenous anesthesia with supplemental local anesthesia without complications. Intra-operatively, there was note of brisk contrast extravasation and contrast pooling at the fundal branch of the left uterine artery consistent with pseudoaneurysm (Appendix C, Figure I, A and B). The lower abdominal aorta, the visualized bilateral common iliac, bilateral external iliac and right internal iliac vasculatures showed normal intimal outline and branching pattern with no evidence of arterio-venous malformation (Appendix C, Figure II). Post left uterine artery embolization pelvic angiographic study confirmed complete occlusion of the vessel with no note of collateral vessels (Appendix C, Figure I, C and D). Another unit of properly typed and crossmatched packed

Appendix B: Transvaginal ultrasound results

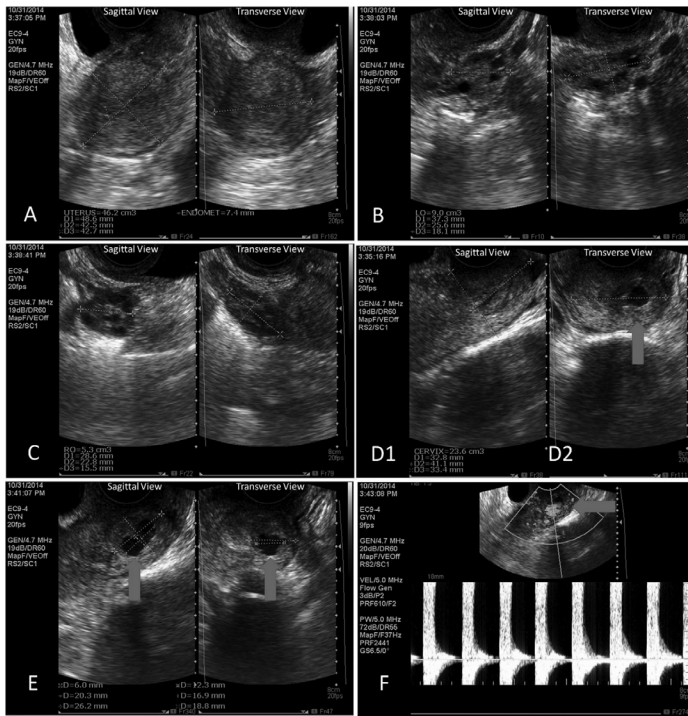


Figure I. On admission (October 31, 2014): A – Uterus; B – Left Ovary; C – Right Ovary. The uterus was normal in size, anteverted, with thin endometrium measuring 0.74 cm, hyperechoic and intact, with normal ovaries. D – Cervix, red arrow showing the Pseudoaneurysm at the left lateral cervical stroma, E – Pseudoaneurysm with anechoic center; F – Bright mosaic flow on Color Doppler.

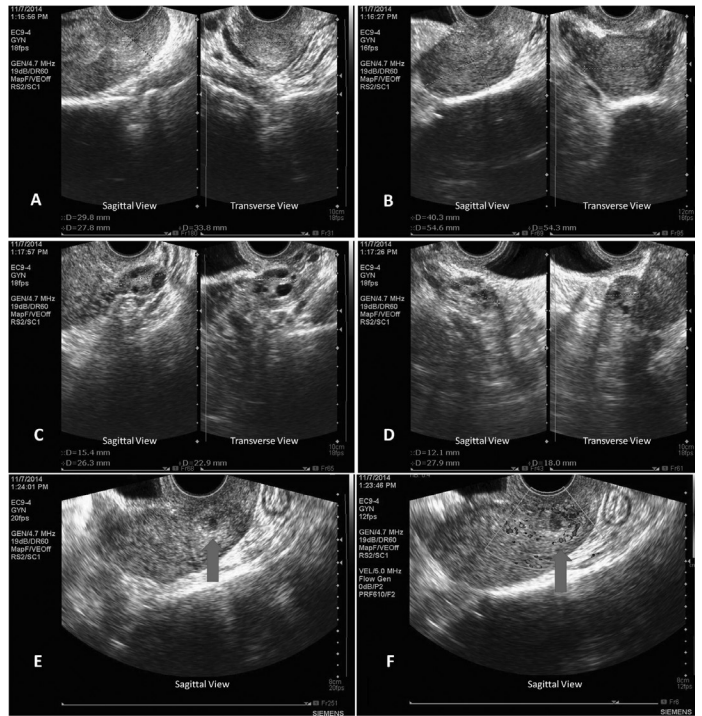


Figure II. One week after Uterine Artery Embolization (November 7, 2014): A – Cervix; B – Uterus; C – Left Ovary; D – Right Ovary. Red arrow showing the Pseudoaneurysm: E and F – The previously noted cervical sonologic structure still has slight hyperechogenic borders and central irregular hypoechoic structure measuring 1.5 x 1.1 cm, now without arterial pulsations on Color Doppler.

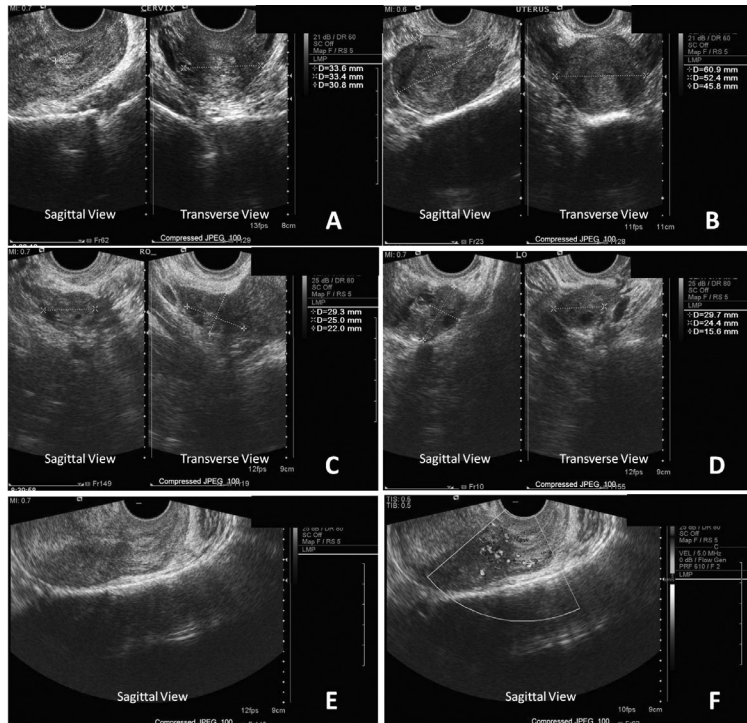


Figure III. Five weeks after Uterine Artery Embolization (December 3, 2014 - Day 3 of menses): A – Cervix; B – Uterus; C – Left Ovary; D – Right Ovary. The previously noted hypoechoic structure is no longer appreciated (E), but still with moderate flow in the said site on Color Doppler (F).

Red Blood Cells was transfused. Repeat complete blood count revealed hemoglobin of 12.2 g/dL. Post-operatively, the patient had occasional vaginal spotting of dark red to brownish discharge with no other symptoms. On the third post-operative day, speculum examination was done to evaluate the brownish discharge. The cervix was pink and smooth with no lesions, and there was no active bleeding per os. The patient was then discharged stable and improved.

The final assessment was delayed postpartum hemorrhage secondary to a ruptured pseudoaneurysm of the left uterine artery status post superselective coil embolization, status post primary cesarean section for malpresentation. Repeat transvaginal ultrasound one week after embolization revealed a normal sized anteverted uterus with thin endometrium and normal ovaries (Appendix B, Figure II, A – D). The previously noted cervical sonologic structure still has slight hyperechogenic borders and central irregular hypoechoic structure measuring 1.5 x 1.1 cm, now without arterial pulsations (Appendix B, Figure II, E and F). Regular menstrual flow with no dysmenorrhea resumed after five weeks. Follow-

up transvaginal ultrasound five weeks after embolization on the third day of menses revealed a normal sized anteverted uterus with thickened endometrium 1.77 cm, intact, and normal ovaries (Appendix B, Figure III, A – D). The cervix is normal in size with the previously noted hypoechoic structure no longer appreciated, but still with moderate flow in the said site (Appendix B, Figure III, E and F). Repeat transvaginal ultrasound nine months after embolization revealed a normal sized anteverted uterus with thin endometrium 0.60 centimeters, intact, and normal ovaries. The cervix is normal in size with intact canal and no lesion. Patient is currently asymptomatic with regular menstrual flow.

CASE DISCUSSION

A pseudoaneurysm of the uterine artery is a rare but potentially life-threatening complication.^{4,5} However, the actual incidence of ruptured pseudoaneurysm in patients presenting with postpartum hemorrhage is unknown. Many single case reports and series are found in literature. Gyoung et al (2013) reviewed medical

Appendix C: Pelvic Angiography and Embolization

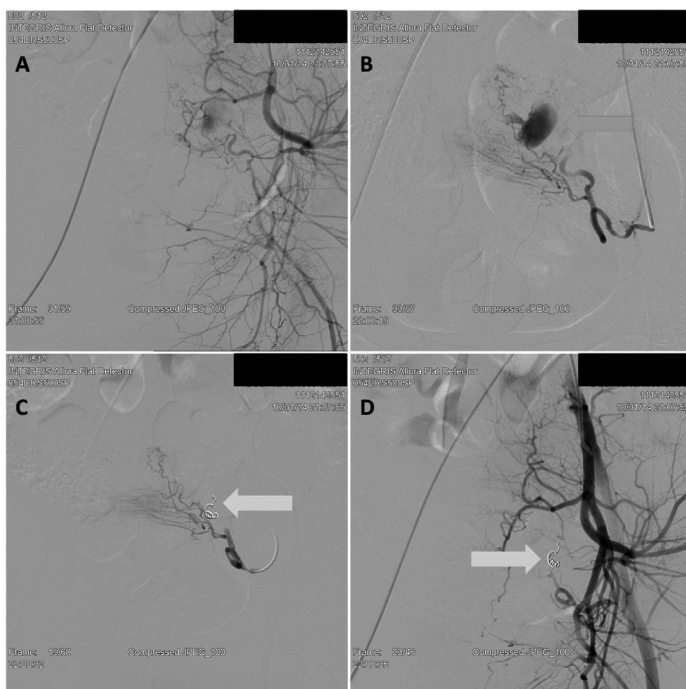


Figure I. Pelvic Angiography of the Left Uterine Artery and Embolization: A and B – There was noted brisk contrast extravasation and contrast pooling at the fundal branch of the left uterine artery consistent with pseudoaneurysm (red arrow); C – Superselective Coil Embolization using Boston Scientific VortX fibered platinum coils (blue arrow); D – Post-Embolization Pelvic Angiography.

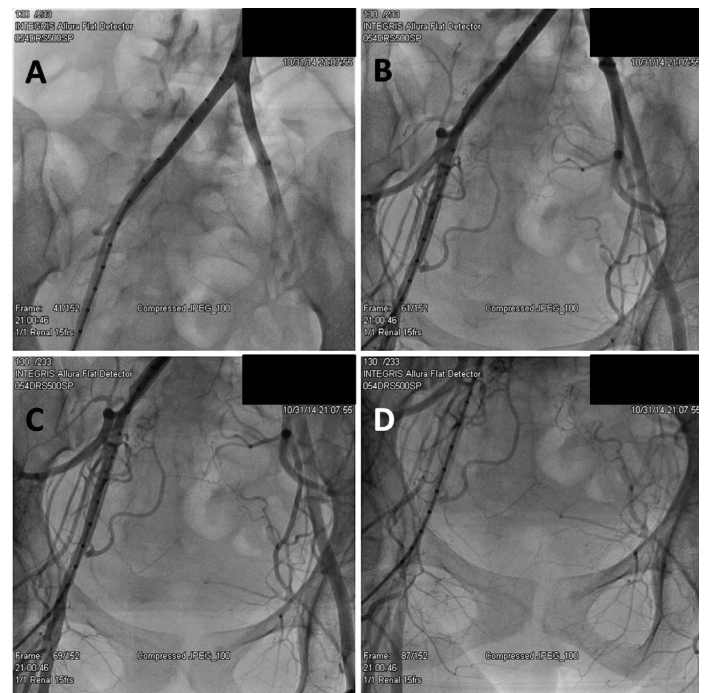


Figure II. Pelvic Angiography: A to D – The lower abdominal aorta, the visualized bilateral common iliac, bilateral external iliac and right internal iliac vasculatures showed normal intimal outline and branching pattern with no evidence of arteriovenous malformation.

records from the period of March 2004 to December 2010 in Seoul, South Korea which included 250 patients who underwent emergency transarterial embolization for massive postpartum hemorrhage. A total of 35 ruptured pseudoaneurysms were identified on angiograms in 33 patients, of which 17 were from the uterine artery with an incidence rate of 6.8%, 31.4% from the left uterine artery and 17.1% from the right uterine artery.¹⁰ In the study by Dohan et al (2013), a single-center in France reported 588 patients with postpartum hemorrhage during the period of July 1994 to December 2012, which were successfully treated with pelvic artery embolization, 18 of which were found to have arterial pseudoaneurysm with an incidence rate of 3.06% which confirms that the condition is rare but not a negligible cause of postpartum hemorrhage.¹¹

There is one reported case of uterine artery pseudoaneurysm in the Philippines by Beltran et al published by the Philippine Journal of Cardiology in 2007 at the Philippine Heart Center of the Philippines entitled "Bloody Mary: A Case of Uterine Artery Pseudoaneurysm" in a 26-year old primiparous patient who underwent cesarean section for failed induction.¹² Patient had an unremarkable past medical and family medical history. She had a two-week history of recurrent vaginal bleeding approximately two weeks after her Cesarean section. Transvaginal ultrasound revealed an intramural uterine mass with an arterial waveform on Doppler studies. Computed Tomography of the lower abdomen confirmed the presence of a left uterine artery pseudoaneurysm for which selective embolization was successfully done.

Yi and Lee (2012) retrospectively reviewed medical records in Gangneung Asian Hospital during the period of October 2006 to June 2010.¹³ Eight women had delayed postpartum hemorrhage due to uterine pseudoaneurysm leakage. The mean age of women was 31.0 ± 4.4 years (range of 27 – 38 years), similar to the study of Gyong et al (2013), the mean age of which was 31.6 (range of 20 – 37 years), and Dohan et al (2013), the mean age of which was 32.2 ± 3.1 years (range of 28 – 39 years).^{10,11} Hence, postpartum hemorrhage due to ruptured uterine pseudoaneurysm more likely occurs in the reproductive age group. The index patient is 26 years old.

The patient's obstetrical history is a Gravida 2 Para 1 (1011), as with the other studies, mean parity was 2.0 ± 1.1 (range of 1-4) with 87.5% having a history of cesarean section, and in the study of Gyoung et al (2013) 54.5% were multiparous with 66.7% having a history of spontaneous vaginal delivery and 33.3% having a history of cesarean section.^{10,13} On the other hand, Dohan et al (2013) noted that 67% were multiparous with 11.1% having a history of dilatation and curettage, 61.1% having a history of cesarean section, 16.67% having a history of operative vaginal delivery, and 22.2% spontaneous vaginal

delivery.¹¹ In the analysis of cases, Isono et al (2010) reported that the most frequent cause of uterine artery pseudoaneurysm was cesarean section accounting for 47.4% of all cases.¹⁴

Review of current literature shows that Cesarean section is the major cause of pseudoaneurysm of the uterine artery although several studies have shown that it may also be secondary to vaginal delivery, pelvic surgeries (adenomyomectomy, myomectomy), and dilatation and curettage. The condition becomes symptomatic within one to two months from the surgical procedure. These studies conclusively indicate that trauma to the uterus predisposes the patient to the development of pseudoaneurysm. As in the case presented, the patient had both a history of dilatation and curettage as well as a more recent cesarean section. The sentinel event prior to the development of the pseudoaneurysm was the cesarean section rather than the previous dilatation and curettage. However, development of the pseudoaneurysm may also occur in patients with no history of such surgery. In the case report by Matsubara et al (2014), a primiparous woman with an uncomplicated vaginal delivery had persistent vaginal bleeding 47 days post-operatively and had further profuse vaginal bleeding after evacuating a questionable hematoma seen on ultrasound.¹⁵ A pelvic angiography revealed a pseudoaneurysm of the left uterine artery for which bilateral uterine artery embolization was done. McGonegle et al (2006) also reported a delayed postpartum hemorrhage six weeks after a normal uncomplicated spontaneous vaginal delivery wherein there was a two centimeter pseudoaneurysm supplied by both uterine arteries, which was embolized successfully.⁵

Pseudoaneurysms may likewise develop after pelvic surgeries. In the study by Matsubara et al (2013), a woman underwent extensive adenomyomectomy 16 months prior to an in vitro fertilization pregnancy.¹⁶ This ended in an abortion for which she underwent a dilatation and curettage at nine weeks for vaginal bleeding. Post-operatively she had profuse vaginal bleeding and on pelvic angiography, there was note of a pseudoaneurysm from the left uterine artery occupying the endometrial cavity. Bilateral uterine artery embolization was done successfully. Wan et al (2014) also reported that three out of the eight patients who underwent myomectomy developed pseudoaneurysm and extravasation of the dye into the uterine cavity.¹⁷ All of which underwent embolization. Oishi et al (2013) presented a case of a woman with massive uterine bleeding a month after laparoscopic myomectomy secondary to a 2 cm right uterine artery pseudoaneurysm treated with selective uterine artery embolization. Thus, it has been proposed that prior surgical procedures in the uterus lead to trauma to the blood vessels which progress to the development of the pseudoaneurysm.¹⁸

However, there is debate as to the integrity of the assumption that pseudoaneurysms are caused by trauma to the uterus. Since there are cases wherein the pseudoaneurysm is nowhere located near the injured site, it is postulated that there is a different underlying cause for its development. In the review of cases by Yi and Lee (2012), pseudoaneurysm of the uterine artery may develop due to trauma, such as that caused by surgical procedures in the uterus.¹³ However, a pseudoaneurysm leakage was observed in the non-traumatic area of the uterus over the fundal area, for which they presumed that the vascular lesion may have developed due to a disruption of the arterial wall by the placenta implanted at the fundus rather than trauma. As in this case presented, the placenta was likewise implanted at the fundus; however, the pseudoaneurysm is located at the cervix, a non-traumatic area which is located neither near the implantation of the placenta nor the incision site from the Cesarean section. At the time of the cesarean section, the uterus and cervix were carefully inspected and palpated to rule out any extension of the incision. Bilateral broad ligaments were noted to be intact with no lacerations, or hematomas noted. In all the literature reviewed, there was no note of a pseudoaneurysm in a similar location. Hence, the underlying cause for the formation of the pseudoaneurysm in this case is currently unclear. Thus, the absence of a traumatic event during delivery does not preclude the occurrence of a pseudoaneurysm.

All of the reported cases of pseudoaneurysm of the uterine artery present with postpartum hemorrhage or with active vaginal bleeding except in one case report wherein the patient was asymptomatic. Kuwata et al (2010) reported a case of a Japanese woman with parity of four, who had a history of two vaginal deliveries and two cesarean sections.⁹ As dictated by institutional policies, six days post-operatively, the patient underwent a routine ultrasound which revealed the finding of a left pelvic mass, adjacent to the uterus, and Magnetic Resonance Imaging that was consistent with a pseudoaneurysm, pelvic angiography was done and revealed a left uterine artery pseudoaneurysm. Hence, a selective uterine artery embolization was done. Pseudoaneurysms are susceptible to rupture due to extraluminal turbulent blood flow that causes it to enlarge.¹⁹ However, while some pseudoaneurysms rupture and induce profound hemorrhage, others may rupture but be resealed by the surrounding tissues, causing intermittent bleeding.⁹ As in this case presented, patient initially had episodes of intermittent vaginal spotting for one week followed by profuse vaginal bleeding thereafter.

In the study by Yi and Lee (2012), there was predominance of delayed postpartum hemorrhage with range of five to 40 days (mean \pm SD 20.3 \pm 12.0 days)

from delivery, as it was in the case of the patient.¹³ In the study of Gyuong et al (2013), most women had primary postpartum hemorrhage 87.9% as compared to secondary postpartum hemorrhage of only 12.1%.¹⁰ This is also evident in the study by Dohan et al (2013), 83.3% of women presented with primary postpartum hemorrhage, whereas only 16.67% presented as secondary postpartum hemorrhage wherein vaginal bleeding occurred 10-18 days after vaginal delivery and five weeks after cesarean delivery.¹¹ Hence, ruptured pseudoaneurysm must be considered as a cause of vaginal bleeding after delivery, since it is not limited to patients presenting with primary postpartum hemorrhage but should likewise be considered for those with secondary postpartum hemorrhage.

If the pseudoaneurysm communicates with the uterine cavity, postpartum hemorrhage may occur as stated above. However, if the pseudoaneurysm has no communication, hemorrhage may be confined to the abdominal cavity, leading to abdominal pain and subsequent acute abdomen.⁹ Kulkarni et al (2013) reported a case of a 30 year old, gravida 2 para 2, who presented with giddiness, lower abdominal pain and persistent vaginal bleeding for 20 days after a term Cesarean delivery.¹⁹ Patient underwent curettage but there was still persistence of bleeding. On ultrasound Doppler of the pelvis, there was note of an anechoic focus which on bilateral internal iliac angiograms revealed focal pseudoaneurysm of the left uterine artery for which she underwent selective arterial embolization. Fortunately in the case presented, the patient presented only with profuse vaginal bleeding which indicates that the pseudoaneurysm had a communication with the cavity. This was likewise seen on the pelvic angiography, where the extravasation of the dye was draining into the vaginal canal.

In a literature review of 17 cases, definitive diagnosis of pseudoaneurysm is established using various imaging tests such as pelvic angiography (41.2%), computed tomography (29.4%), or color Doppler ultrasonography (29.4%).¹⁴ Ultrasound findings usually reveal echogenic foci or mass with a swirling flow (high velocity), or yin-yang pattern on color Doppler either in the uterine cavity, myometrium, or pelvic cavity.^{5,9,14,20,21} This diagnostic tool is non-invasive and may be an initial screening test in an emergency setting with a 95% diagnostic sensitivity.¹⁹ As in the case presented, a transvaginal ultrasound was done as the initial screening test which revealed a distinct region at the left lateral cervical stroma measuring 2.6 x 1.9 x 2.0 cm composed of slightly echogenic borders and a central irregular cystic anechoic structure measuring 1.7 x 0.6 x 1.2 cm which on color Doppler showed bright mosaic color flow and high arterial pulsations (Figure II). However, Doppler findings may be similar for both

arterio-venous malformations and pseudoaneurysm. In the case presented, the transvaginal ultrasound indicated presence of a possible arterio-venous malformation of the cervix. Although computed tomography angiography may be informative in specific cases to help localize, identify feeding vessels, and perfusion sac owing to its three-dimensional reformatted images, it should be restricted to hemodynamically stable patients as it induces a delay in the treatment due to pre-procedural preparations.^{11,20}

The reference standard for the diagnosis of pseudoaneurysm still remains to be pelvic angiography for the main reason that all patients presented were actively bleeding in the setting of postpartum hemorrhage which needed rapid and effective control with preservation of fertility.^{11,20,21} In this case, patient is a 26-year old gravida 2 para 1 who is desirous of future pregnancies with active vaginal bleeding but hemodynamically stable – a good candidate for pelvic angiography. Pelvic angiography in this case revealed brisk contrast extravasation and contrast pooling at the fundal branch of the left uterine artery consistent with pseudoaneurysm. The lower abdominal aorta, the visualized bilateral common iliac, bilateral external iliac and right internal iliac vasculatures showed normal intimal outline and branching pattern with no evidence of arterio-venous malformation. Hence, the definitive diagnosis of the pseudoaneurysm was only seen once the pelvic angiography was done.

A pseudoaneurysm can result in life-threatening profuse postpartum hemorrhage when untreated or treated inadequately.²¹ Once a bleeding pseudoaneurysm is diagnosed, the conservative management initiated is frequently unsuccessful in controlling the bleeding.¹³ In the literature reviewed, treatment methods for pseudoaneurysm include hysterectomy and uterine artery ligation but emergency pelvic arterial embolization is a well established therapeutic option for controlling either primary or secondary postpartum hemorrhage, and it is currently accepted as the treatment of choice in specific cases of pseudoaneurysm in patients with the advantage of preserving fertility by selectively blocking the blood supply to the diseased blood vessels.

The patient in this case report consented to the emergency pelvic angiography with arterial embolization after all available treatment options were presented. Pelvic arterial embolization also has advantages such as easy identification of bleeding sites with decreased re-bleeding from collaterals as more distal occlusion of bleeding vessels are carried out, and shorter hospital stays.^{19,22} If profuse hemorrhage occurs while waiting for embolization, hemorrhage may be decreased by uterine packing using a balloon catheter or gauze, which is a method used to treat atonic bleeding.³ This technique can compress the bleeding site and decrease hemorrhage; however, no trial

of this procedure for pseudoaneurysm has been reported as the insertion may lead to further injury to the vessel, and may induce profuse hemorrhage.¹⁵ In the study of Dohan et al (2013) involving 18 patients, pseudoaneurysm is well treated by pelvic arterial embolization with a primary success rate of 94%, and a secondary success rate of 100%.¹¹ The secondary success rate was due to only one patient requiring repeat embolization. No hysterectomy or arterial ligation was needed in any case.

In congruence with the previous study, Yi and Lee (2012) reported all eight patients were treated with bilateral uterine artery embolization with no additional embolization sessions or hysterectomies needed.¹³ In a literature review involving 17 cases, Isono et al (2010) reported a success rate of 94.1% for pelvic arterial embolization in patients with pelvic arterial pseudoaneurysm.¹⁴ Technical success of pelvic artery embolization was achieved in a literature review of 33 patients, 17 patients of whom were pseudoaneurysm of the uterine arteries, clinically 93.9%, however, one patient had persistent vaginal bleeding despite bilateral uterine artery embolization for which she underwent emergency hysterectomy, revealing a uterine rupture. Another patient had recurrence of hemorrhage 13 days after unilateral uterine artery embolization for which she underwent emergency hysterectomy, confirming that bleeding was secondary to remnant placenta and not due to the persistence pseudoaneurysm.⁹

Almost all literature reviewed reported bilateral uterine artery embolization with some including embolization of other feeding vessels with exceptional success rates. However in this case after the pelvic angiography, only a unilateral uterine artery embolization was performed since the feeding vessel that was supplying the pseudoaneurysm was only from the left uterine artery. Repeat pelvic angiography after the superselective coil embolization revealed no contrast extravasation with no other feeding vessels from the contralateral side. In literature search, there have also been reports of cases where only selective unilateral uterine artery embolization was done with a similar success rate and no subsequent complications, as in the cases reported by Kuwata et al (2010), Koo et al (2013), and Kulkarni et al (2013).^{9,19,22}

In the case report by Matsubara et al (2014), hemorrhage immediately ceased after the embolization of the pseudoaneurysm, and repeat ultrasound confirmed the absence of flow within the mass.¹⁵ Two months after the embolization, ultrasound showed no mass within the uterine cavity, and menses resumed without dysmenorrhea after another month. As in the case presented herein of a gravida 2 para 1 (1011) diagnosed with ruptured pseudoaneurysm of the left uterine artery, vaginal bleeding immediately stopped after the

embolization. Repeat transvaginal ultrasound one week after the procedure revealed absence of pulsations within the mass, and after five weeks, complete absence of the mass in the cervix with resumption of menses with regular flow and no dysmenorrhea.

There were no major embolization-related complications in all the literature reviewed. Though Choi et al (2011), Singhal et al (2014), and Yun et al (2011) reported that patients may have low-grade fever, hematoma at the site of catheter placement, and contrast-related side effects after the procedure and transient ischemic phenomena.^{8,21,23} In this case, the patient had no short-term complications as stated above, with an unremarkable post-operative course. As in the study of Erickson et al (2007), uterine artery embolization was performed in 20 patients with postpartum hemorrhage, including three patients with pseudoaneurysm.²⁴ All achieved complete hemostasis, and no major short- or long-term complications were observed. Four women subsequently became pregnant and gave birth to seven healthy infants. As in the literature review of Kuwata et al (2010), eight of 12 patients received embolization, with all eight achieving complete hemostasis without short- or long-term complications.⁹ The other four either had hysterectomy or Thrombin injection. Recently, instead of arterial embolization, thrombin injection directly into the pseudoaneurysm under ultrasound guidance has been reported in a study by Kovo et al (2007) and Kulkarni et al (2013); however, its indication and effectiveness have not yet been determined. Choi et al (2011) reported a case of a 3.8 centimeter pseudoaneurysm in the myometrium supplied by the left uterine artery.^{6,19,23} Initially, an ultrasonography-guided thrombin injection was attempted but failed. They attributed the failure to the high blood flow velocity and the large cavity of the

pseudoaneurysm. Hence, embolization was performed instead. However, surgery may be more suitable in cases of acute and massive bleeding in which there is no time for embolization.^{18,19}

CONCLUSION

Postpartum hemorrhage is an obstetrical emergency, following a vaginal or cesarean section delivery, wherein it is important to promptly determine the diagnosis and provide management to prevent maternal sequelae. Despite a recent proliferation of reports, uterine artery pseudoaneurysm still remains to be a rare cause of unexplained postpartum hemorrhage, which requires a high index of clinical suspicion for diagnosis. Ruptured pseudoaneurysm is not limited to patients presenting with primary postpartum hemorrhage but should likewise be considered for those with secondary postpartum hemorrhage. Prior surgical procedures in the uterus predispose a patient to the development of a uterine artery pseudoaneurysm but uncomplicated vaginal deliveries may also lead to such. In the case presented since the pseudoaneurysm is located in the cervix, which is a non-traumatic area, the cause for the development of the pseudoaneurysm still remains to be unclear but the history of a prior cesarean delivery may have predisposed the patient to such an event. Pelvic angiography and subsequent left uterine artery pseudoaneurysm coil embolization was performed successfully in the case presented, with no short-term complications and with regular menses resuming after five weeks. Hence, the diagnostic and therapeutic treatment of choice is still pelvic angiography with embolization. However, in hemodynamically unstable patients, a life-saving hysterectomy should be done.

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