


**Case Report**

## Clinical Complications Induced by Placental Site Subinvolution: Secondary-Type Hemorrhage after Delivery and Pseudo Aneurysms after Non-Traumatic Delivery may be Caused by Atony of the Uteroplacental Artery

Yuji SHIINA<sup>1\*</sup>, Masahiro YAMAMOTO<sup>2</sup>, Shiori SANO<sup>1</sup>, Yuta GONAI<sup>3</sup>, Kazuyoshi SAKAI<sup>1</sup>

### Abstract

**Background:** Persistence of extravillous trophoblasts causes hemorrhage and many other problems.

**Methods:** Three cases of critical hemorrhage are presented. Two cases of secondary-type postpartum hemorrhages that occurred after discharge and one case of pseudoaneurysms that occurred after miscarriage.

**Results:** In postpartum hemorrhage cases, placental site subinvolution was diagnosed. Microscopic examination of the resected uterus showed subinvolved arteries containing large thrombi. Immunohistochemical analysis showed that extravillous trophoblasts were present within the walls of subinvolved vessels. Failure of involution on the placental site sometimes occurs following miscarriages as well. In post-miscarriage pseudoaneurysms case, the distinctive hemorrhage in this patient consisted of bleeding from various sites in the myometrium after the placental chorion had been expelled without leaving a remnant. The bleeding pattern was extremely similar to the postpartum hemorrhage described above. Though, we could not confirm extravillous trophoblasts directly because of the success of conservative treatment, these postpartum and post-miscarriage hemorrhage were thought to have the same pathophysiology.

**Conclusion:** The cause of both hemorrhages may have been atony of the uteroplacental artery and can be explained by subinvolution.

**Keywords:** Doppler-Diagnostic; Gestational Trophoblastic Disease; Placental Pathology; Post Partum Hemorrhage; Vascular Biology

### Introduction

Two cases of secondary-type critical postpartum hemorrhages that occurred after discharge are presented. In both cases, the patient delivered a baby at full term uneventfully. Massive bleeding occurred suddenly more than ten days after delivery. Since the patients were in critical condition, hysterectomy was performed immediately to save the lives of the patients. Microscopic examination of the resected uterus showed the possibility of placental site subinvolution. The correlation of atony of the uteroplacental artery and secondary-type postpartum hemorrhage was discussed. One case of patient who had a spontaneous miscarriage at 11 weeks' gestation and delivered the gestational sac (chorionic tissue) almost entirely, with no remnant observed, who then suffered a development of a pseudoaneurysm and critical hemorrhage, and the uterus was eventually saved by means of two uterine artery embolizations, is presented. Success of the conservative treatment failed us to do pathological diagnosis directly, but the close similarity

### Affiliation:

<sup>1</sup>Department of Obstetrics and Gynecology, Yamagata Prefectural Shinjo Hospital, Japan

<sup>2</sup>Department of Morphological and Physiological Science, Faculty of Life Sciences, Kumamoto University, Japan

<sup>3</sup>Department of Obstetrics and Gynecology, Faculty of Medicine, Yamagata University, Japan

### \*Corresponding Author

Yuji SHIINA, Department of Obstetrics and Gynecology, Yamagata Prefectural Shinjo Hospital, Japan

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in appearance to secondary-type postpartum hemorrhages suggested the same pathophysiology of post-miscarriage hemorrhage. The correlation of atony of the uteroplacental artery and such hemorrhage was also discussed. Subinvolution is believed to constitute the underlying pathophysiology of pseudoaneurysms.

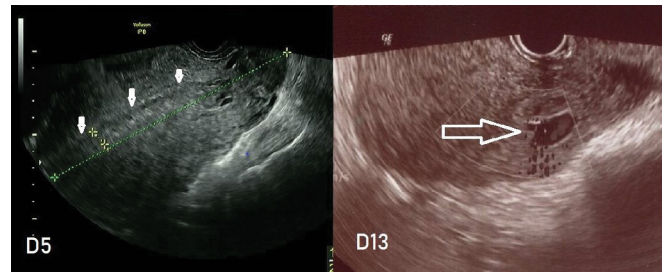
## Case presentation

### Case 1

This case involved a 31-year-old, gravida 3 para 2 woman whose pregnancy progressed without any particular complications. On day 281 of pregnancy, the uneventful spontaneous delivery of a 3,550-gm boy occurred after a normal labor of 9.5 hours. The placenta was expelled and showed no apparent signs of a defect 3 minutes after delivery. The amount of bleeding was 270 gm until placental delivery. Massive bleeding occurred suddenly 1 hour after delivery, with blood loss of 836 gm. Transabdominal ultrasonography showed an intrauterine coagula mass measuring 50 mm across at the cervical portion, raising suspicion of cervical atony. The cervical coagula mass was extruded, with blood loss of 431 gm. Total blood loss since delivery was 1537 g. The administration of a uterotonic in combination with intravaginal gauze packing successfully stopped the bleeding. Transvaginal ultrasonography showed the absence of any remnants in the uterus on day 5 (Figure 1 D5). She was discharged on day 6 according to schedule. Massive bleeding occurred suddenly 13 days after delivery, with blood loss of about 880 gm. It happened without warning. Transvaginal ultrasonography showed pulsatile blood flow from the myometrium to the uterine cavity (Figure 1 D13). These phenomena were seen at several points of the myometrium to the uterine cavity. The patient was in critical condition, so hysterectomy, with blood loss of 575 gm, was performed immediately to save the patient's life. Though total blood loss during this event was 1455 gm, there was much bleeding that was not counted. Blood transfusion of 4,000 ml was finally required.

### Case 2

This case involved a 32-year-old, gravida 2 para 1 woman whose pregnancy progressed without any particular complications. On day 284 of pregnancy, the uneventful spontaneous delivery of a 3,315-gm girl occurred. The placenta was expelled and showed no apparent signs of a defect, with blood loss since delivery totaling 191 gm. She was discharged on day 6 according to schedule. Massive bleeding occurred suddenly 11 days after delivery, with blood loss of 275 gm and over. It happened without warning. Enhanced computed tomography showed vivid blood flow toward the uterine cavity, and the patient was in critical condition. Hysterectomy, with blood loss of 813 gm, was performed



**Figure 1:** Grayscale imaging and pulsed-wave Doppler sonography in Case 1.

On Day 5 (D5), the absence of any remnant was confirmed in a pre-discharge examination (small arrows). On Day 13 (D13), pulsatile blood flow from the myometrium to the uterine cavity was shown (big arrow).

immediately to save the patient's life. Though total blood loss counted during this event was 1088 gm, there was much bleeding that was not counted. Finally, blood transfusion of 2,400 ml was required.

### Case 3

A 33-year-old, gravida 2, para 1 woman had become pregnant by frozen-thawed embryo transfer with luteal phase support. However, the pregnancy did not proceed uneventfully, and although a fetal heartbeat was finally observed at 8 weeks 5 days, it was lost at 9 weeks 5 days. Intrauterine fetal death was diagnosed, at which point the Crown-Rump Length (CRL) was 10 mm, and the Gestational Sac (GS) measured 26 mm. Spontaneous miscarriage occurred at 11 weeks 0 days, with the uterine contents expelled as a single unit. Blood loss when this expulsion occurred was 325 g. The patient subsequently continued to bleed despite uterotonic administration. Because the bleeding failed to stop, transvaginal ultrasound was performed, and although no large remnant was observed within the uterus, 289 g of blood was lost during the examination. Gauze packing and additional uterotonic administration gradually brought the bleeding under control, but by this time, a further 180 g of blood had been lost. Total blood loss due to the miscarriage was 794 g.

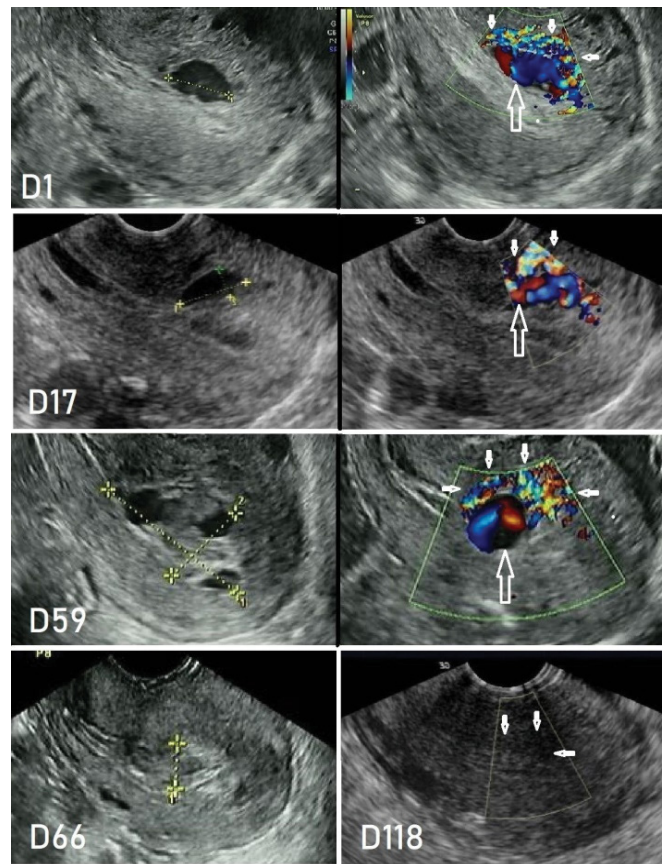
On Day 1, although the hemorrhage had ceased, a pseudoaneurysm appeared (Figure 2 D1). On grayscale imaging, there was an 18-mm, cystic lesion within the uterus, and on close examination, a space was opening up between the organic tissue, including decidua that was filling the uterine cavity, and the myometrium. Pulsed-wave Doppler sonography demonstrated abundant perfusion in the myometrium, and this blood was flowing into the space and causing the appearance of a whirlpool yin-yang sign. On Day 2, uterine artery embolization was performed. On Day 10, this cystic lesion disappeared temporarily. Abundant perfusion

was still evident in the myometrium, but the significant flow of blood from the myometrium to the uterine cavity was no longer evident. On Day 17, however, a cystic lesion separated by organic tissue again appeared between the myometrium and the uterine cavity (Figure 2 D17). As before, a large amount of blood was flowing into it from the myometrium, causing the appearance of a whirlpool yin-yang sign. On Day 23, uterine artery embolization was again performed. On Day 31, the only signs of perfusion were within the myometrium, and the flow of blood from there to the uterine cavity had disappeared.

After embolization, only a brownish vaginal discharge was apparent, but on Day 41, this changed to fresher blood. On Day 45, reappearance of the cystic lesion was confirmed. In view of the potential complications of frequent embolization, it was decided to adopt a policy of watchful waiting at this point. The bleeding gradually diminished each day, but on Day 55, the patient suffered a large hemorrhage, although fortunately it had abated by the time she reached the hospital. On Day 59, she again came to the hospital with major bleeding, and this time it also decreased with conservative therapy. The cystic lesion with abundant perfusion had reappeared within the uterus (Figure 2 D59). Until this point, this cystic lesion had appeared to be covered with organic tissue, but on this occasion, the organic tissue occupied most of the uterine cavity, and it now appeared to be the main component, with a few cystic lesions present within it. On Day 66, the patient again came to the hospital because of major hemorrhage, but the bleeding had stopped by the time she arrived. Ultrasound showed only organic tissue in the uterine cavity, with no cystic lesion (Figure 2 D66). During this time, a GnRh antagonist was administered from Day 23 to Day 103 to prevent bleeding due to the resumption of menstruation. After Day 100, genital bleeding suddenly stopped. On Day 118, there was no longer any organic tissue in the uterus, and the endometrium had recovered its normal state during the follicular phase. All signs of perfusion in the myometrium had also disappeared (Figure 2 D118).

### Case summaries

Cases 1 and 2 were discharged according to the usual schedule and did not exhibit any major abnormalities until Day 13 and Day 11, respectively. In Case 1, a primary-type hemorrhage due to uterine atony during delivery occurred, and this immediately diminished. No other trauma or coagulopathy occurred during delivery that might have caused a major hemorrhage, and neither a remnant nor infection was present in the puerperal period. These two cases were secondary-type critical hemorrhages that occurred post-discharge. The onset was sudden, with no clear trigger, but the symptoms were acute. In both cases, the patients hemorrhaged large amounts of fresh blood, and these were extremely dangerous



**Figure 2:** Grayscale imaging and pulsed-wave Doppler sonography.

On Day 1 (D1), a pseudoaneurysm appeared. There was an 18-mm, cystic lesion within the uterus. A space was opening out between the organic tissue, that was filling the uterine cavity, and the myometrium. Pulsed-wave Doppler sonography demonstrated abundant perfusion in the myometrium (small arrows), and this blood was flowing into the space and causing the appearance of a whirlpool yin-yang sign (big arrow). On Day 17 (D17), a cystic lesion separated by organic tissue again appeared between the myometrium and the uterine cavity. A large amount of blood was flowing into it from abundant perfusion in the myometrium (small arrows), causing the appearance of a whirlpool yin-yang sign (big arrow). On Day 59 (D59), the cystic lesion (big arrow) with abundant perfusion (small arrows) had reappeared within the uterus. Until this point, this cystic lesion had appeared to be covered with organic tissue, but on this occasion, the organic tissue occupied most of the uterine cavity, and it now appeared to be the main component, with a few cystic lesions present within it. On Day 66 (D66), only organic tissue in the uterine cavity, with no cystic lesion. On Day 118 (D118), there was no longer any organic tissue in the uterus, and the endometrium had recovered its normal state during the follicular phase. All signs of perfusion in the myometrium had also disappeared (short arrows).

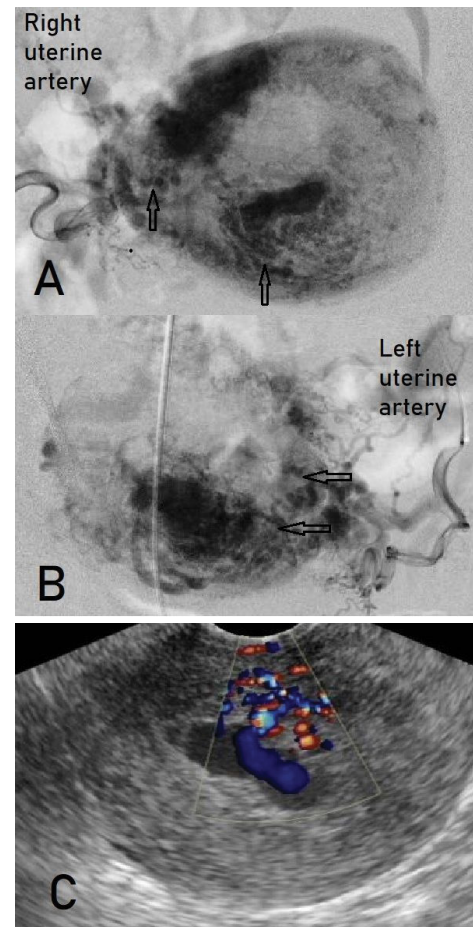
hemorrhages that might well have resulted in a fatal outcome before the patient reached hospital. The hemorrhages in Cases 1 and 3 appeared extremely similar on pulsed-wave Doppler sonography. In Case 1, the absence of any remnant

was confirmed in a pre-discharge examination on Day 5. When she came to hospital with massive bleeding, pulsatile blood flow in the direction of the empty uterine cavity was observed. Because of the patient's poor general state, only a single page of records was preserved, but during the period in which she was observed, similar pulsatile blood flows were present not just in one site, but from several different sites. In Case 3, angiography identified a number of bleeding points in the myometrium that were hemorrhaging toward the uterine cavity. These vessels were supplied by both the left and right uterine arteries, and the bleeding points extended over a wide area (Figure 3A, 3B). Ultrasound also confirmed major bleeding into the uterine cavity from abundant perfusion in the myometrium (Figure 3C). In this spontaneous miscarriage, the gestational sac (chorionic tissue) was expelled as a single unit, and the abnormal bleeding was not due to a remnant or atony. No intrauterine procedure had been performed, and trauma and iatrogenic causes were therefore excluded. Despite the difference between full gestation and miscarriage, in both cases, the pathology consisted of repeated alternating hemorrhages into the uterine cavity from different points in the myometrium following expulsion with no remaining placental or chorionic tissue.

### Pathology

On macroscopic examination of the resected uterus from Case 1, an area with a hard, rough, irregular surface had spread inside the uterus, and it was conjectured that this was where the hemorrhage had occurred (Figure 4). Microscopic examination of this area showed large, patent, and dilated superficial subinvolved arteries containing large thrombi adjacent to normally involuted vessels with occlusive fibrointimal thickening (Figure 5A, 5B). Immunohistochemical analysis for cytokeratin showed that Extravillous Trophoblasts (EVTs) were persistently present within the wall of subinvolved vessels (Figure 5C), while fewer EVT were within involuted vessels (Figure 5D). No retained placenta or placenta accreta was noted. The figure shows a site with a mixture of normal involuted vessels and subinvolved vessels, but most of the vessels were subinvolved. The resected uterus from Case 2 showed very similar findings. In both Cases 1 and 2, the possibility of placental site subinvolution was judged to be the most likely diagnosis.

(A, B) Low and high magnification of dilated subinvolved arteries filled with thrombi (arrows) adjacent to involuted arteries with slit-like lumens (arrowheads). (C, D) Immunohistochemistry for cytokeratin highlights extravillous trophoblasts, which are abundant in a subinvolved artery (C) and are few in an involuted artery (D).



**Figure 3:** Angiography and pulsed-wave Doppler sonography.

Angiography identified a number of bleeding points (arrows) in the myometrium that were hemorrhaging toward the uterine cavity. These vessels were supplied by both the left and right uterine arteries (A, B). Ultrasound also confirmed major bleeding into the uterine cavity from abundant perfusion in the myometrium (C).

### Discussion

Various different terms for placental site subinvolution are used in the literature, including “Subinvolution of the Placental Implantation Site (SPIS),” “placental site Vessel Subinvolution (VSI),” and “Subinvolution of the Placental Site (SPS);” in this study, “placental site subinvolution” is used. At term, half a liter of blood per minute must be delivered to the placenta to serve the physiological needs of the fetus[1]. In order to maintain adequate uteroplacental flow, vascular remodeling plays an important role during pregnancy. In the early weeks of pregnancy, the decidual segment of the spiral arteries at the site of placentation begins to show morphological changes. The wall has lost its normal structure, and then it is invaded by infiltrating trophoblasts[2]. Throughout the second and third trimesters, uteroplacental arteries increase the blood supply, facilitated by the loss of musculoelastic tissue from their walls[3] and

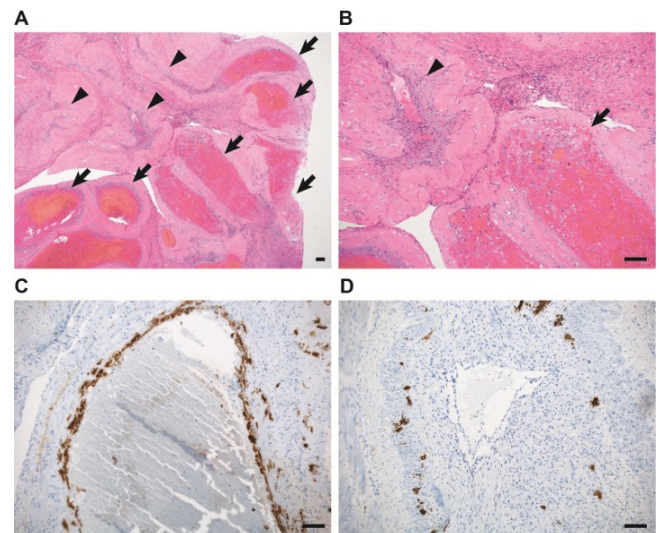


**Figure 4:** Macroscopic examination of the resected uterus in Case 1. An area with a hard, rough, irregular surface had spread inside the uterus.

the reduction of peripheral vascular resistance in the placental bed by physiological changes[4]. In the third trimester, the physiological mechanism of involution of uteroplacental vessels begins, and endovascular cytotrophoblasts are replaced by maternal-derived endothelial cells. There is reendothelialization of the uteroplacental vessels with a disappearance of the trophoblasts and regeneration of the elastic media[5]. Regeneration of the internal elastic lamina causes an occlusive form of intima layer thickening via its fibrotic transformation, local endarteritis, and thrombotization[6]. After delivery, there is necrosis and sloughing of the decidua, which completes the normal involution of the uterus. However, in the subinvolution process at the site of placental implantation, the normal remodeling of the uteroplacental arteries during the third trimester is either delayed or inadequate, resulting in the persistence of low-resistance vessels with increased flow[7]. Subinvolved placental bed vessels in the superficial myometrium are large, patent, and dilated with intravascular thrombosis. Their walls are distorted and thickened due to deposition of hyaline with partial absence of the endothelial lining[8], which can lead to severe intermittent vaginal bleeding after delivery[9].

In Case 1, the identification of Extravillous Trophoblasts (EVTs) by microscopic examination of pathological specimens (Figure 5C) demonstrated that the areas of surface irregularity observed on macroscopic examination were the sites of placentation. The sites of continued remodeling during pregnancy were the hemorrhage sites. Incomplete involution at these sites was also observed (Figure 5A, 5B), suggesting that the cause of these hemorrhages may have been atony of the uteroplacental artery. The bleeding in Cases 1 and 2 was due to placental site subinvolution. Pseudoaneurysms were

initially thought to be related to intrauterine procedures. They can occur after a traumatic delivery or traumatic pregnancy termination, such as cesarean section or dilatation and curettage[10]. A recent analysis showed that half of the pseudoaneurysms occurred after non-traumatic deliveries[11]. In these cases, the reasons for bleeding were vague until now. If the sites of hemorrhage through which the blood is spurting were physically covered with some substance, this would cause the bleeding to slacken or stop. We suggest that, in these cases, it was the pooling of the blood within this covering itself that caused the appearance of pseudoaneurysms. The covering may have been the surrounding uterine wall, residual tissue such as decidua, fibrin or other coagulatory substances, or organized inflammatory substances. Whether the covering stops the bleeding may depend purely on whether the force of the hemorrhage or the covering action is greater. The fact that total hysterectomy has been required in Case 1 and 2 following full-term delivery suggests that bleeding may be forceful at full term [9]. At the miscarriage stage, e.g. Case 3, the bleeding is therefore gentler than at full term, to the extent that it was possible to preserve the patient's uterus with conservative treatment. In Case 3, the recurrent pseudoaneurysms appeared to be around the same size each time, but in fact, the force of the bleeding must have weakened with each uterine artery embolization. The tissue covering gradually started to take over, with outbreaks of bleeding sometimes occurring in the interim (Figure 1 D1 to D17 to D59). Some authors have attempted to assess hemorrhagic force objectively in terms of Peak Systolic



**Figure 5:** Microscopic examination in Case 1. Scale bar means 100  $\mu$ m.

(A, B) Low and high magnification of dilated subinvolved arteries filled with thrombi (arrows) adjacent to involuted arteries with slit-like lumens (arrowheads). (C, D) Immunohistochemistry for cytokeratin highlights extravillous trophoblasts, which are abundant in a subinvolved artery (C) and are few in an involuted artery (D).

Velocity (PSV) and establish an index for the feasibility of conservative management. Timmerman et al. reported that patients with  $PSV > 0.83$  m/s have a substantial threat of hemorrhage, whereas those with  $PSV < 0.39$  m/s may undergo conservative management safely in the absence of symptoms [12]. However, because there has been insufficient evaluation of stopping this bleeding, this index is not generally used. Other studies have shown that some lesions with elevated PSVs resolve spontaneously with conservative management [13]. The sole difference is that vessels that are bleeding will stop bleeding if they are physically covered strongly enough, and they form pseudoaneurysms if they are covered weakly.

Clinical symptoms are expressed on the balance power between push and cover. The mechanism of push, i.e. Cause of hemorrhage, in Case 3 are discussed. The distinctive hemorrhage in this patient consisted of bleeding from various sites in the myometrium after the placental chorion had been expelled without leaving a remnant. The walls of the myometrium contained abundant perfusion seeking an exit, and blood spurted out from atonic vessels (Figure 3C). The abundant perfusion evident on pulse-wave Doppler sonography (Figure 2 D1, D17, and D59) showed that the tissue was in a special state, and when this tissue reverted to its non-pregnant state, this abundant perfusion was no longer visualized (Figure 2 D118). These special states of the myometrium spreaded widely where the placental chorion had been expelled. There was no remnant, local vascularization, trauma, or any hemolytic disease. Incomplete contraction could only explain temporary hemorrhage, but in this case, the event continued for more than a hundred days. There must be a structural change in this portion. We suggest the possibility that this might be atonic vessels on the surface of the myometrium. In some abortion cases, atony of the uteroplacental artery was also thought to be the cause of bleeding [14,15], and it might show a close similarity in appearance to Case 1 and 2 hemorrhages. Case 1 and 2 were presented as placental site subinvolution. Given its close similarity in appearance to these hemorrhages, it was thought that this special state of myometrium was placental site subinvolution. There is no direct evidence, but the curetted endometrium of post-abortion endometritis often shows features suggestive of the persistence of extravillous trophoblasts. The residuum of the pregnancy is represented by hyaline material, particularly related to the endometrial spiral arteries, in which effete cells, almost certainly trophoblasts, are embedded. This finding represents a failure of involution of the placental bed and of the vascular physiological changes following an abortion [16]. In the first and second trimesters, the uteroplacental arteries in the placental bed undergo morphological changes [5,6], and endovascular cytotrophoblasts and endothelial cells co-exist. If uteroplacental arteries in this site are disproportionate,

and an excess amount of endovascular cytotrophoblasts is present, the situation might be the same as subinvolution and could cause severe hemorrhage after miscarriage due to atony of the uteroplacental artery. Many issues surrounding the persistence of extravillous trophoblasts have recently emerged, so subinvolution might be an appropriate area of research. According to some reports, focal increased color vascularity after miscarriage appears in the second trimester [17]. When trophoblastic tissue was confirmed in women who underwent uterine re-evacuation due to suspected RPOC, the rate of a new infertility problem was significantly higher than when there were negative pathologic findings [18]. A relationship between chronic endometritis and infertility-related conditions such as repeated implantation failure and recurrent miscarriage has been reported [19]. These conditions seem to be produced by subinvolution. Close investigation of uteroplacental arteries in the myometrium at the site of placental implantation will clarify the pathophysiology of the remodeling process during the first and second trimesters. The status of endovascular cytotrophoblasts and endothelial cells may explain the clinical state.

### Availability of Data and Materials

Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

### Authors contributions

Y.S. and M.Y. designed the research study. Y.S., S.S., Y.G. and K.S. performed the research. They were attending physicians of these patients. M.Y. provided help and advice on pathological findings. Y.S. wrote the manuscript. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

### Ethical Approval and Consent Participate

That the research was conducted in accordance with the ethical standards established in the Declaration of Helsinki of 1947. We got written informed consent from all patients, allowing to use medical examination and treatment information for a case report.

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### Conflict of interest

The authors have no conflicts of interest to declare.

## Supplement

We have continued to report neovascular lesions after delivery or miscarriage. These comprise following pathophysiologies;

1. Retained Products of Conception (RPOC) with vascularization.
2. Acquired Arteriovenous Malformations (AVMs).

AVMs are present as vessel lesions from the start of placentation irrespective of the chorion, and these two constitute completely different pathological conditions[20,21].

3. Placental site subinvolution, which includes pseudoaneurysms.

In this paper, we presented the third condition. Subinvolution and physical covering can explain the cause of pseudoaneurysms after non-traumatic delivery reasonably. Subinvolution believed to constitute the pathophysiology of pseudoaneurysms.

The addition of the following two widely known pathophysiologies to the three described above enables the full classification of neovascular lesions after delivery or miscarriage.

4. Placenta Accreta Spectrum (PAS), which includes Cesarean Scar Pregnancy (CSP).

Kim et al, suggested that a vascular lesion may be caused by the erosive property of the syncytiotrophoblastic tissue and chorionic villi during placentogenesis in early pregnancy and CSP[22]

5. villous-derived malignancies (e.g., invasive moles and placental site trophoblastic tumors (PSTTs)). Invasive moles and PSTTs may be differentially diagnosed by abnormally high levels of Human Chorionic Gonadotropin (hCG) and of Human Placental Lactogen (hPL)[23], respectively.

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