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# [Case report]



# Introduction and utility of resuscitative endovascular balloon occlusion of the aorta for cases with a potential high risk of postpartum hemorrhage: A single tertiary care center experience of two cases

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#### Abstract

Postpartum hemorrhage is an important obstetric complication and the leading cause of maternal mortality worldwide. Occasionally, we encounter unexpected massive postpartum hemorrhage diagnosed for the first time after delivery. Therefore, it is essential to pay attention to patients with a high risk of postpartum hemorrhage. The authors report two cases of patients at high risk of postpartum hemorrhage that were successfully managed by resuscitative endovascular balloon occlusion of the aorta before cesarean section. Case 1: A 32-year-old woman with a history of cesarean section and who conceived using assisted reproductive technology was diagnosed with partial placenta previa at 25 weeks of gestation. Because of tocolysis failure, emergent cesarean section with resuscitative endovascular balloon occlusion of the aorta was performed at 36 weeks of gestation. Natural placental resorption was observed. She was discharged at 5 days after delivery without significant hemorrhage. Case 2: A 41-year-old woman with suspected placenta accreta spectrum due to a cesarean scar pregnancy was referred to our hospital at 33 weeks of gestation. A planned cesarean section with resuscitative endovascular balloon occlusion of the aorta was conducted at 37 weeks of gestation. There was no visual evidence of abnormal placental invasion of the myometrium, and natural placental resorption was observed. She was discharged at 5 days after delivery without significant hemorrhage.

**Key words**: Intra-aortic balloon occlusion, pregnancy, postpartum hemorrhage, preventive medicine, resuscitative endovascular balloon occlusion of the aorta

## Introduction

Postpartum hemorrhage (PPH) is an important obstetric complication that can lead to life-threatening conditions. Massive PPH is the most common cause of maternal death worldwide<sup>1,2)</sup>. Life-threatening massive PPH occurs in one of every 300 cases<sup>3)</sup>. Placenta accreta spectrum (PAS) is thought to

be caused by the adherence of the placenta to the defective site of the decidua and is a well-known obstetric complication that can cause life-threatening massive PPH<sup>4</sup>). A history of cesarean section with the presence of placenta previa, which completely covers the internal uterine ostium, is a risk factor for PAS<sup>4,5</sup>). In such patients, careful diagnosis and multidisciplinary management strategies are re-

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quired prior to cesarean section to reduce the risk of maternal mortality<sup>6</sup>). Unfortunately, in cases where there is no placenta previa, PAS is sometimes diagnosed for the first time as an unexpected PAS in women with a cesarean scar pregnancy (CSP) or those where the patient conceived using assisted reproductive technology (ART) <sup>4,6-8</sup>, leading to a failure to manually remove the placenta after delivery. When the adhesion area is limited, manual removal may be possible; however, when the whole placenta is adhered, manual removal can cause life-threatening PPH<sup>9</sup>).

Resuscitative endovascular balloon occlusion of the aorta (REBOA) was initially intended for the surgical treatment of abdominal aortic aneurysms in 1953 by Edwards *et al.*<sup>10)</sup> and was later applied to traumatic hemorrhagic shock<sup>11)</sup>. Recently, there have been several reports on the use of REBOA for temporary aortic occlusion to reduce massive PPH during cesarean hysterectomy for PAS<sup>12-14)</sup>.

Located in central Fukushima, Ohta Nishinouchi Hospital is a leading tertiary care center for severe maternal-fetal conditions. It has maternal-fetal units and altitude emergency and critical care medical centers and has experienced several severe maternal conditions<sup>5,8,15-17)</sup>. We introduced REBOA as a maternal rescue for cases with massive PPH in 2016. Furthermore, we have also introduced this novel technique from 2021 for cases with high suspicion of severe PPH to prevent maternal morbidity and mortality based on our experience with suspicion of unexpected PAS. Although several cases have been reported on the utility and validity of REBOA for the control of PAS, the efficacy of REBOA for unexpected PAS is limited.

Here, we report two patients at high risk of PPH who were successfully managed with REBOA before cesarean section.

# Case Report

Case 1

A 32-year-old, gravida 3, para 1, Japanese woman with a history of one previous cesarean section was transferred to our hospital because of abnormal genital bleeding at 25 weeks of gestation. She conceived using ART because of secondary sterility. At the time of transfer, transvaginal ultrasound examination showed a significantly shortened cervical length and the presence of a placenta covering the intra uterine ostium. Magnetic resonance imaging (MRI) at 30 weeks showed evidence of partial

placenta previa, which completely covered the intra uterine ostium and was mainly located on the postwall side of the myometrium (Figure 1). Although it was not a conventional suspect lesion for PAS, such as placenta previa, mainly located in the anterior wall of the myometrium, we suspected that the patient had abnormal placentation, due to the abnormal placenta location, one previous cesarean section and history of ART in the present pregnancy, which are risk factors for PPH.

We decided to use REBOA before cesarean section and occluded the aorta in case of severe PPH or cesarean hysterectomy due to unexpected PAS. At 36 weeks of gestation, an emergent cesarean section with REBOA was performed because of the onset of labor. At the time of cesarean section, because natural placental resorption and significant uterine contractions were observed, balloon occlusion was not required during the cesarean section. The REBOA and sheath were removed soon after cesarean section. The estimated total blood loss during the cesarean section was 270 g, and the patient was discharged on postpartum day 5 without any complications.



Fig. 1. MRI of the pelvis at 30 weeks of gestation showed evidence of partial placenta previa in which the intra-os was covered completely. It was mainly located on the posterior wall side of the myometrium (arrow).

## Case 2

A 41-year-old woman with a suspected CSP and risk of PAS attended our hospital at 33 weeks of gestation. Although there was no evidence of placenta previa, transabdominal ultrasound examination at a previous hospital showed that the placenta appeared to be located at the uterine scar of a previous cesarean section. MRI at 35 weeks showed no evidence of placenta previa; however, the risk of CSP was difficult to exclude because the enlarged uterine scar of the previous cesarean section appeared to be covered by the placental margin (Figure 2). Based on these data, the patient was suspected to have a CSP.

A planned cesarean section with REBOA was performed at 38 weeks gestation. During laparotomy, we observed no medusa head infiltration of the uterine surface by the placenta, along with bulging of the uterine wall, indicating the presence of abnormal placentation and suggestive of PAS. The placenta resorbed naturally after delivery, and significant uterine contractions were observed. We did



Fig. 2. MRI of the pelvis at 30 weeks of gestation showed no evidence of placenta previa; arrow (1) indicates that the placenta is located on the myometrium anterior wall. Arrow (2) shows that the enlarged thin uterine myometrium seems covered by the placental margin, suggesting that the placenta is located on the cesarean scar.

not use occlusion during the course of the cesarean section, and the REBOA and sheath were removed immediately after the surgery. The estimated total blood loss during the cesarean section was 500 g, and the patient was discharged on postpartum day 5 without any complications.

#### REBOA procedure

After spinal anesthesia, a 7 Fr sheath was placed via the right femoral artery under the guidance of surface ultrasound (Figure 3a). REBOA was then placed above the celiac artery under X-ray guidance (Figure 3b). Finally, the effect of REBOA was confirmed by filling the exploratory balloon with 10 mL of saline water just before the cesarean section. At the time of filling and expansion of the exploratory balloon, maternal blood pressure at the right radial artery increased from 137/85 mmHg to 172/90 mmHg and from 99/51 mmHg to 132/65 mmHg, while maternal heart rate decreased from 74 beats/min to 45 beats/min and from 90 beats/min to 50 beats/min in Case 1 and Case 2, respectively. Based on these maternal physiological changes, we confirmed that the distal aortic flow was occluded by REBOA.

# **Discussion**

In the field of trauma, compared with resuscitative thoracotomy, REBOA is a relatively less invasive intervention for life-threatening hemorrhage. Recently, REBOA has also emerged as a novel procedure for preventing significant obstetric hemorrhage<sup>13)</sup>. REBOA involves the insertion of a balloon catheter device that can be placed at the bedside. Conventionally, REBOA is most frequently used in trauma, but there is growing evidence supporting its use in non-traumatic hemorrhagic events, including gastrointestinal bleeding, ruptured viscera, aneurysms, and obstetric bleeding<sup>18)</sup>. The first report on the use of REBOA in the field of obstetrics was in 1995, in a case of PAS<sup>19)</sup>. Since then, over 1,000 cases of aortic balloon occlusion have been reported in obstetrics, mainly for cases with PAS to control obstetric hemorrhage during cesarean hysterectomv<sup>20)</sup>.

In addition to the typical PAS characterized by placenta previa and previous cesarean section history, the concept of unexpected PAS, which is defined as the occurrence of severe obstetric hemorrhage for the first time at the time of delivery, is growing<sup>4,6-8)</sup>. Unexpected PAS is usually defined as unsuspected PAS at the time of delivery due to the absence of placenta previa, which is the typical form of



Fig. 3a. After spinal anesthesia, a 7 Fr sheath was placed via right femoral artery under the guide of surface ultrasound (arrow indicates right femoral artery).

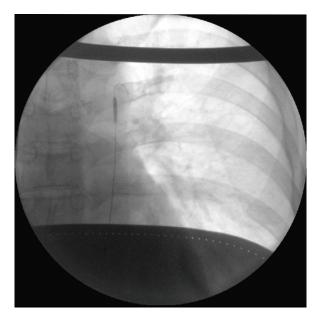


Fig. 3b. REBOA was then placed above the celiac artery under the guidance of x-ray. After the x-ray, a balloon was moved toward a more distal zone and set just upper the celiac artery.

conventional PAS<sup>4,6-8)</sup>. Of note, unexpected PAS is an important concern because it is less likely to be managed in a PAS center with a multidisciplinary team, where specialized care may lower maternal morbidity and mortality<sup>21)</sup>. As a result, despite the

lesser degree of placental invasion, worse obstetric outcomes could occur in "unexpected PAS" compared to "expected PAS"6). Therefore, in a clinical setting, paying careful attention to pregnant women at risk of unexpected PAS is essential<sup>4</sup>). Kyozuka et al. reported that repeated cesarean sections in ART pregnancies are strongly associated with unexpected PAS<sup>4</sup>. Therefore, we introduced REBOA in Case 1, where the patient had a suspected CSP, to control obstetric bleeding. CSP, which is defined as a gestational sac implant at a previous myometrium scar due to cesarean section, is a rare form of ectopic pregnancy, accounting for 6.1% of ectopic pregnancies and 0.15% of pregnancies in women who have previously undergone cesarean section<sup>22,23)</sup>. In most cases, CSP is diagnosed between 6 and 8 weeks of gestation when the gestational sac is clearly visible on ultrasound<sup>24)</sup>. Because CSP is rare, accurate diagnosis of CSP after the second trimester is challenging if there is no evidence of placenta previa<sup>7)</sup>. CSP without placenta previa sometimes presents as an unexpected PAS at the time of laparotomy. Suzuki et al. reported a case of unexpected PAS in a patient with CSP that required emergent cesarean hysterectomy by a multidisciplinary emergent team because of severe abnormal placental implantation. However, Case 2 showed no evidence of placenta previa on transvaginal ultrasound during perinatal care. CSPs are divided into two types based on imaging presentation and progression<sup>25)</sup>. In type 1 (endogenic CSP), the gestational sac grows toward the uterine cavity, while in type 2 (exogenic CSP), the gestational sac grows toward the bladder. In a previous study, the diagnosis of type 1 CSP was delayed, and emergent cesarean hysterectomy by a multidisciplinary emergent team was needed because of unexpected PAS, which usually necessitates an immediate cesarean hysterectomy<sup>7)</sup>. In contrast, type 2 CSP could develop as placenta previa and be diagnosed as conventional PAS, which allows enough time to set up a multidisciplinary medical team. As shown in Case 2, once the obstetric care provider suspects type 1 CSP, immediate transfer to a multidisciplinary medical center is reasonable due to the high risk of massive PPH or emergent cesarean hysterectomy<sup>7)</sup>.

# Future challenges

REBOA increases proximal blood pressure through aortic occlusion. However, because this occlusion induces distal ischemia of the visceral organs and lower extremities, which causes inflammatory sequelae and may be life-threatening or limbthreatening<sup>26)</sup>, the attending physician should take the benefits and risks of REBOA into consideration. In addition, REBOA may lead to complications of organs proximal to the occlusion due to increased blood flow. Uchino *et al.* reported the case of an 86-year-old woman with fatal massive intracranial hemorrhage because of increased proximal blood flow using REBOA during the management of a severe pelvic fracture<sup>27)</sup>. Cognizant of these physiological changes during the REBOA procedure, our institution excluded patients with hypertensive disorders of pregnancy, such as new onset hypertension after 20 weeks of gestation, from REBOA due to the potential risk of brain hemorrhage<sup>28)</sup> due to the increased blood flow resulting from REBOA.

Furthermore, because the terms "REBOA" and "Unexpected PAS" are still not commonly encountered among obstetricians, we thought that multidisciplinary simulation and prophylactic application of REBOA to cases with unexpected PPH in addition to conventional PAS may be essential for gaining expertise in REBOA.

In conclusion, we have introduced REBOA as a multidisciplinary medical procedure for cases with suspected unexpected PAS. Future collaborative studies should be performed in centers with multidisciplinary management protocols for highly suspect to unexpected placenta PAS to ensure better understanding of the characteristics and outcomes of the disease.

## Conflict of interest disclosure

The authors have no conflicts of interest to declare.

# **Contributors**

All authors were involved in the clinical care of the patients and contributed to the conception, drafting, review, and revision of the manuscript. All authors have read and approved the final version of the manuscript and take full responsibility for the work.

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# Patient consent

Informed consent was obtained from the patients for the publication of this work.

# Provenance and peer review

This case report was peer reviewed.

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