Retained Products of Conception Fed by the Inferior Mesenteric Artery: A Case Report

Yumiko GOTO[*1, 2], Yasuhira KANNO[*1], Masaru HAYASHI[*1], Kanako MITSUZUKA[*1], Tatsuya SEKIGUCHI[*3], Takahiro SUZUKI[*1] and Hitoshi ISHIMOTO[*1]

[*1]*Department of Obstetrics and Gynecology, Tokai University School of Medicine*
[*2]*Department of Clinical Genetics, Tokai University Hospital*
[*3]*Department of Diagnostic Radiology, Tokai University School of Medicine*

(Received May 25, 2020; Accepted June 22, 2020)

Retained products of conception (RPOC) refer to the persistence of placental or fetal tissue in the uterus following delivery or miscarriage. RPOC may cause massive postpartum or post-abortion hemorrhage. Arterial embolization (AE) is an effective choice of management for postpartum hemorrhage including RPOC. We report a case of hemorrhagic RPOC, in which uterine artery embolization with transcervical resection did not achieve hemostasis, and laparotomy with uterine compression sutures was subsequently required. The RPOC was apparently fed by an aberrant branch derived from the inferior mesenteric artery (IMA). AE of IMA was not performed because of possible necrosis of the descending colon and rectum. A physician should be aware that AE is not an all-encompassing hemostatic technique for postpartum bleeding, such as with RPOC, and should keep alternatives in mind.

Key words: retained products of conception, uterine artery embolization, transcervical resection, aberrant branch

INTRODUCTION

Retained products of conception (RPOC) refer to the persistence of placental and/or fetal tissue in the uterus following delivery or miscarriage. The incidence of RPOC after the first trimester, second trimester, and normal delivery are reported to be 17%, 40%, and 3–5%, respectively [1, 2]. RPOC may cause massive postpartum or post-abortion hemorrhage and could necessitate a hysterectomy. Several reports have shown that uterine artery embolization (UAE), in combination with transcervical resection (TCR), is an effective choice of management for RPOC, which can reduce the amount of bleeding [3, 4]. We report a case of hemorrhagic RPOC, in which UAE with TCR did not achieve hemostasis, and laparotomy with uterine compression sutures was subsequently required. The RPOC was apparently fed by an aberrant branch derived from the inferior mesenteric artery (IMA). Arterial embolization (AE) of IMA was not performed because of possible intestinal necrosis.

CASE REPORT

A 28-year-old woman, 0G0P, vaginally delivered a 3400-g female infant at 39 weeks of gestation at another clinic. At delivery, the blood loss was 1500 mL and a partial defect of the placenta was noted. The patient was conservatively managed with a hope of spontaneous passage of the retained placental tissue. On day 14 post-delivery, an intrauterine mass with abundant blood flow was recognized by ultrasound at the same clinic. On day 19 post-delivery, she was referred to our institution, and at the first examination, she suddenly presented with active genital bleeding with hypotension. Transvaginal sonography revealed an intrauterine mass of 4 cm in diameter that was located near the internal cervical os with abundant arterial blood flow (Fig. 1). Computed tomography (Fig. 2) and magnetic resonance imaging (Fig. 3) also indicated an intrauterine mass with blood flow. A diagnosis of RPOC was made. UAE was immediately performed (Fig.4), and initial hemostasis was satisfactory with a remarkable decrease in human chorionic gonadotropin titers from 419.6 IU/L to 53.7 IU/L within 1 week after UAE (Fig. 5).

TCR was planned and performed on day 28 post-delivery. At the TCR, several minutes after the removal of the mass, a sudden uncontrollable hemorrhage occurred. The RPOC was located mainly at the posterior uterine cervix and the resected tissue was fragile. After resecting the mass, the sudden bleeding occurred from the 9 o'clock direction of the uterine os. Sutures and uterotonic agents were ineffective. The total amount of blood lost was 2026 mL.

Angiography demonstrated spontaneous recanalization of the right uterine artery (Fig. 6a); the vessel was embolized, but hemostasis was not achieved. Detailed pelvic angiography revealed an aberrant branch derived from the IMA that apparently fed the uterus (Fig. 6b, c). Because embolizing the vessel would damage the

Yumiko GOTO, Department of Clinical Genetics Tokai University Hospital, 143 Shimokasuya, Isehara, Kanagawa 259-1193, Japan
Tel: +81-463-93-1121 Fax: +81-463-92-2600 E-mail: gotoyumi@is.icc.u-tokai.ac.jp

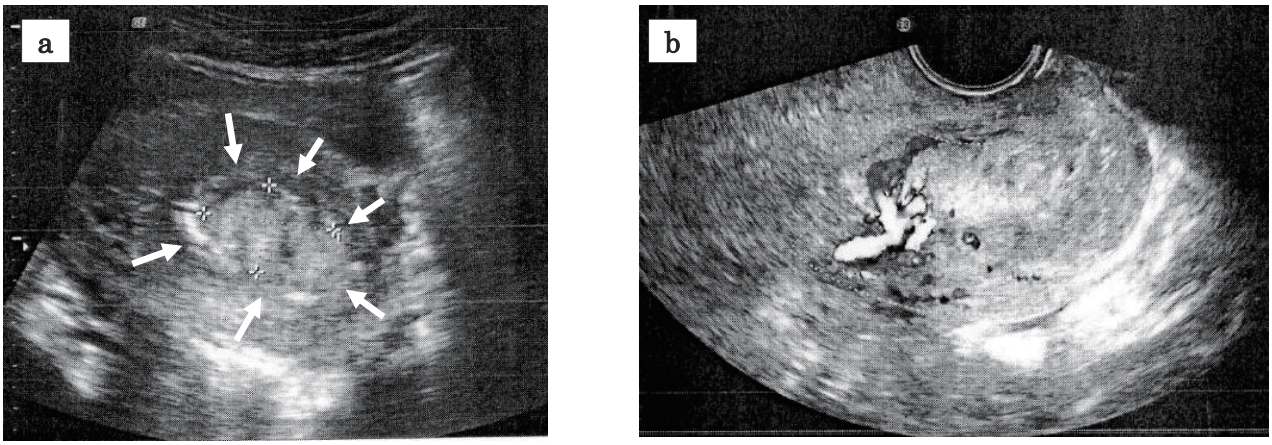

Fig. 1 Ultrasonographic images at our hospital.
A 4-cm sized mass was detected near the internal uterine os indicated by arrows (a). The mass was diagnosed as a retained product of conception with hypervascularity (b).

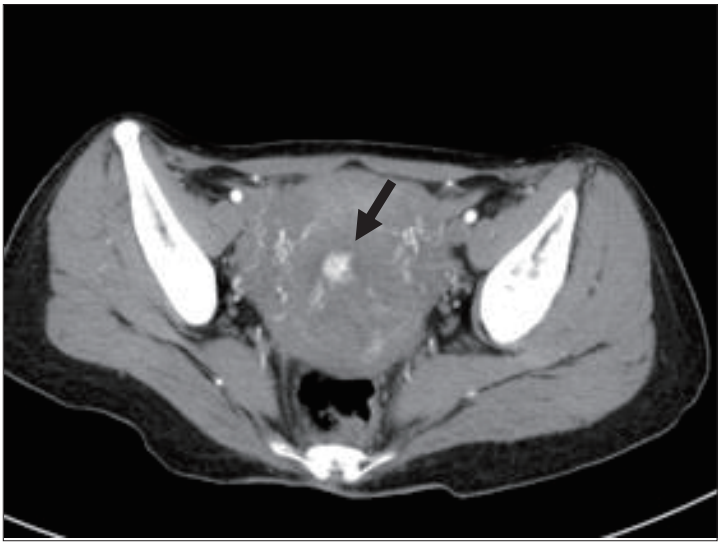

Fig. 2 Computed tomography (CT) images.
Dynamic enhanced CT during the arterial phase showed a hypervascular lesion (arrow) in the uterine cavity corresponding to a retained product of conception.

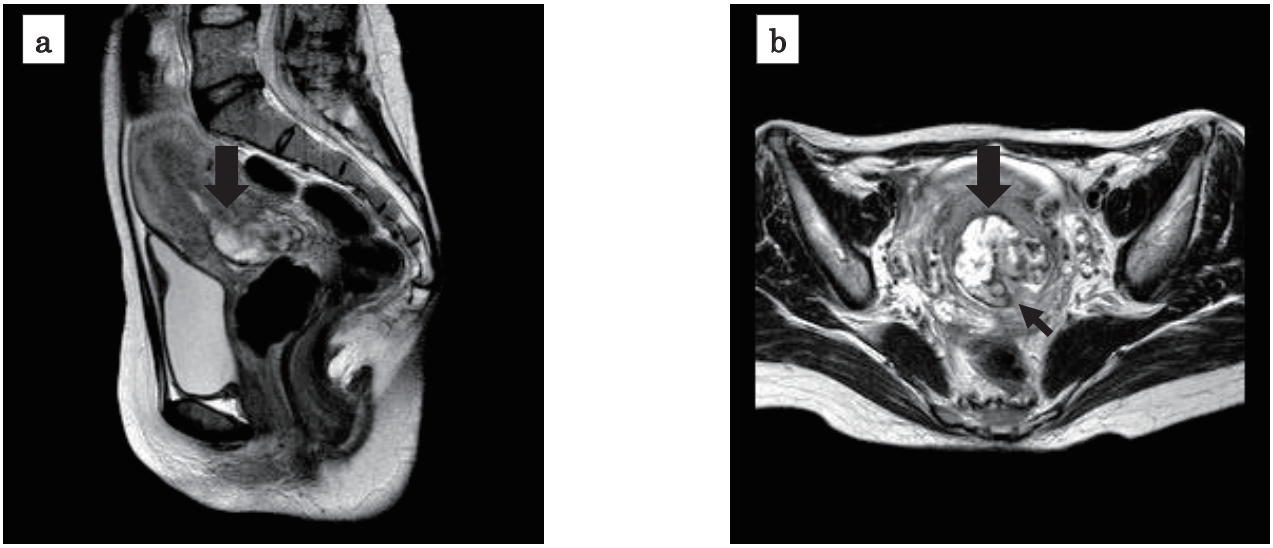

Fig. 3 T2-weighted sagittal (a) and axial (b) magnetic resonance images.
Magnetic resonance imaging showed a heterogeneous mass (arrow) with both high- and low-signal intensity on the T2-weighted image. The mass was located in the lower uterine cavity and cervical canal and protruded into the cavity from the posterior uterine wall (small arrow).

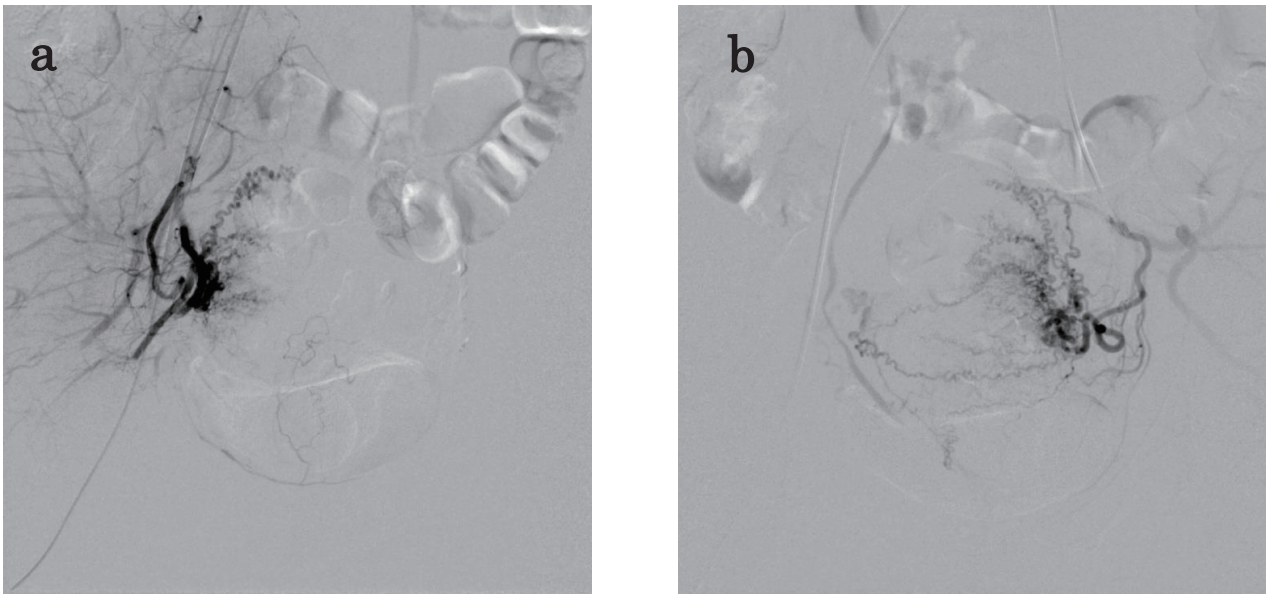

Fig. 4 Angiographic images for the first uterine artery embolization.
Angiography revealed leakage from the right (a) and left (b) uterine arteries.

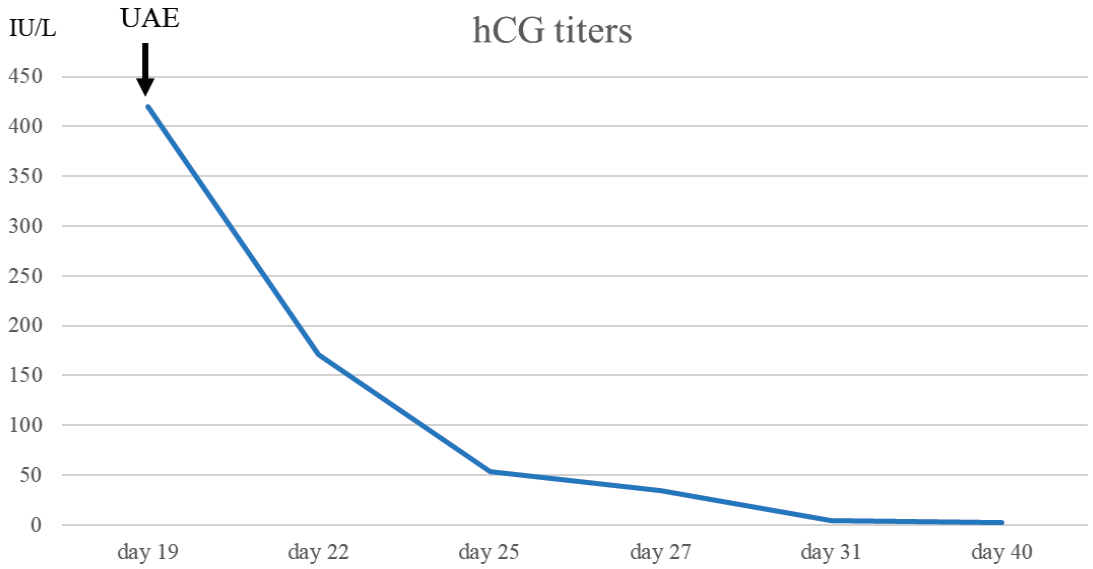

Fig. 5 Human chorionic gonadotropin (hCG) titers.
After the first uterine artery embolization was performed on day 19 post-delivery, hCG titers substantially decreased.

intestine and colon, laparotomy with vertical compression sutures to the uterus was alternatively performed after the TCR had finished. There was no intraperitoneal bleeding and no apparent damage to the intestine and colon. The bleeding was only from the inside of the uterus. Superficial endometriotic lesions were observed at the back of the uterus. Compression sutures were placed at the lower part of the uterus from the anterior to the posterior wall with absorbable suture (1 vicryl, Ethicon, NJ, USA) and the knots were tied tightly to compress the lower part of the uterine cavity. The total blood loss was 904 mL. Blood transfusion was performed with 8 units of red cell concentrates, 6 units of fresh frozen plasma, and 10 units of platelet concentrates.

Pathological examination demonstrated that the removed lesion consisted of distal villi, some of which were degenerated. Accumulation of neutrophils was partially seen. These findings were compatible with the diagnosis of RPOC. She was discharged from our hospital on postoperative day 8 without complications and subsequently underwent Kaufmann treatment.

DISCUSSION

RPOC appear in 17%, 40%, and 3–5% of cases after delivery in the first, second, and third trimesters, respectively [1, 2]. The cause of RPOC is considered to be due to the degeneration of retained placenta with fibrin deposition and vitrification, and developing marked vascularity. Therefore, the risk factor for RPOC is retained placental fragments, which is common in placental abruption by hand after delivery,

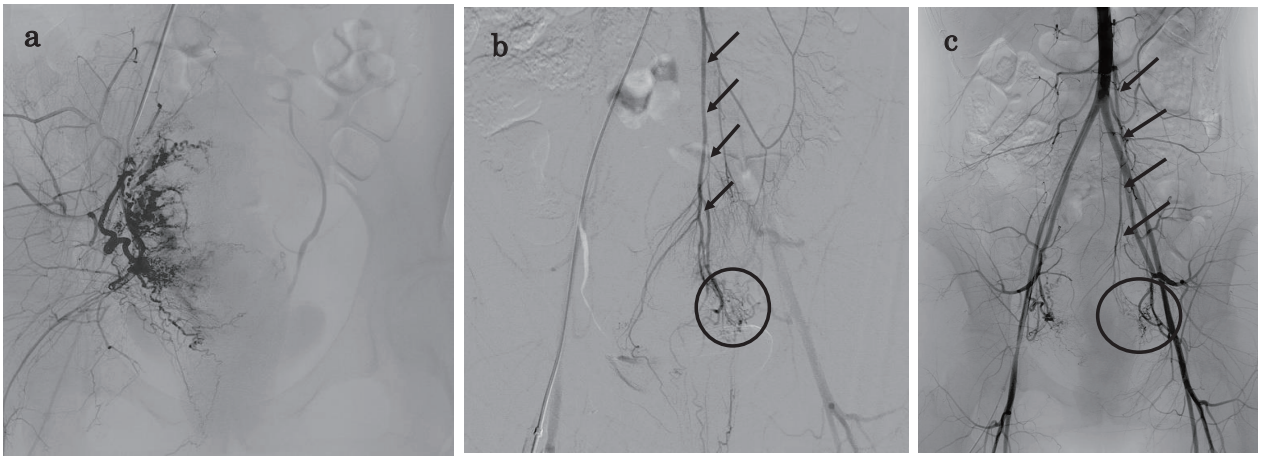

Fig. 6 Angiographic images after the transcervical resection.
The right uterine artery showed massive blood flow (a).
The aberrant branch derived from the inferior mesenteric artery (arrows) revealed a stain (circle) corresponding to the uterine cavity (b, c).

placenta accreta, and accessory placenta. Early recognition of clinical symptoms such as abdominal pain and vaginal bleeding, and more frequent use of ultrasonography may lead to early and accurate diagnosis of RPOC [5]. The pathological diagnosis is made by the existence of chorionic villi, which represents persistent placenta or trophoblast [2].

Though RPOC may cause massive postpartum or post-miscarriage hemorrhage and even maternal death, the treatment of RPOC is controversial. Postpartum hemorrhage causes 25% of all maternal deaths [6]. RPOC treatment options are careful observation [7, 8], dilation and curettage [9], UAE [10], TCR [11], use of uterotonic medications such as prostaglandin E1 analogs [12], and hysterectomy.

In the present case, we performed UAE with TCR as the first-line treatment for RPOC. However, the procedure was not effective due to an aberrant collateral branch from the IMA, which was observed at the second UAE.

When a UAE is not successful, one can opt for a secondary UAE, hysterectomy, and vertical compression suture. Though the rate of hysterectomy is 4–12% [13, 14], we prevented hysterectomy by performing a compression suture. This may contribute to preserve fertility.

It is known that there are collateral blood flows to the uterus derived from the vaginal artery, external pudendal artery, external iliac artery, and the IMA. McLucas (2009) first reported the collateral supply to the uterus from the IMA during a UAE for fibroid treatment [15]. Smoger (2010) also showed collateral blood supply to the uterus from the left ovarian artery fed by the IMA during a UAE for adenomyosis treatment [16]. Further, Chang (2013) examined 559 cases, which underwent UAE for fibroid or adenomyosis treatment. It was determined that 1.3% of cases (n = 7/559) had collaterals from the IMA to the uterus and that patients with adenomyosis had collaterals from the IMA more frequently than patients with fibroids only [17]. Shin (2015) reported a case with collaterals from the IMA found by angiography for postpartum hemorrhage [18]. The only case in which collaterals from the IMA existed after UAE was shown by Dixon (2012) [19]. The collaterals were found 15 years later in another UAE.

The present case is the first report to show an RPOC lesion fed not only by the uterine artery but also by the IMA. The presence of an aberrant arterial collateral blood feed is possible. Placing uterine compression sutures may be a hemostatic method of choice in such a situation. Although IMA embolization has been performed in several studies, we did not perform it to avoid intestinal necrosis.

The reason why the collateral circulation derived from the IMA emerged in the present case remains unknown. However, as pelvic endometriotic lesions are frequently fed by branches from the IMA [20], the endometriotic lesions in our case may possibly have been fed by a small and narrow branch of the IMA, which may have led to the apparent collateral blood flow seen at the second angiography.

In conclusion, RPOC may have collateral vessels that cause massive hemorrhage. Though UAE may be a mainstream choice for management of RPOC, alternatives should be kept in mind for such cases, particularly when the RPOC is fed by an artery that is not embolizable. Obstetricians should be aware that a UAE is not an all-encompassing hemostatic technique for postpartum bleeding, such as with RPOC.

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