Case Reports in Women's Health 42 (2024) e00630

Contents lists available at ScienceDirect

Case Reports in Women's Health

journal homepage: www.elsevier.com/locate/crwh

Large uterine arteriovenous malformation successfully treated with combined endovascular treatment and supracervical hysterectomy: A case report

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ARTICLE INFO

Keywords:
Uterine arteriovenous malformation
Supracervical hysterectomy
Embolization
Balloon occlusion
Paradoxical embolization

ABSTRACT

Uterine arteriovenous malformations (AVMs) are rare conditions that cause life-threatening bleeding. Endovascular treatment or total hysterectomy is performed to safely treat most AVMs. This case report describes a 54-year-old female patient with a large uterine AVM, uterine bleeding, and cardiac overload that was difficult to manage but successfully treated. Total hysterectomy poses a high risk of hemorrhage due to significant uterine and internal iliac vein dilation; thus, embolization of feeding arteries was performed with N-butyl cyanoacrylate. However, a postembolization computed tomography scan detected paradoxical embolization of the liver, kidneys, and spleen. Therefore, supracervical hysterectomy was performed with preoperative coil embolization and intraoperative balloon occlusion of the feeding arteries. In this case, supracervical, not total, hysterectomy needed to be performed as the shunts were determined to be in the uterine corpus.

1. Introduction

Arteriovenous malformations (AVMs) are anomalous anastomoses of arteriovenous vessels without capillaries. They are high-flow vascular lesions with one or more arteriovenous shunts and abnormally dilated and tortuous vessels. AVMs develop in various body organs [1], but uterine AVMs are rare and occur in approximately 0.63% of cases after childbirth or abortion [2]. The main symptom includes abnormal uterine bleeding, occasionally life-threatening [3]. Heart failure can result from large AVMs due to increased arteriovenous volume [1]. Treatment of uterine AVM includes total hysterectomy or embolization [3]. Transcatheter arterial embolization aims to minimize invasiveness and preserve fertility, whereas hysterectomy is performed in cases of failed embolization or when the patient has no desire to preserve fertility [2]. There are only a few case reports of uterine AVM being surgically treated with preoperative embolization [4,5] or intraoperative arterial balloon occlusion [6–9].

This report describes a case of a large uterine AVM with cardiac overload that was successfully managed with endovascular treatment and supracervical hysterectomy.

2. Case Presentation

A 54-year-old woman, gravida 8 para 3, attended hospital with complaints of excessive uterine bleeding for the first time after menopause. She reported a surgical history of tubal pregnancy, one vaginal delivery, two cesarean sections, and two miscarriages that required intrauterine curettage in early pregnancy and two in mid-term. She had experienced menorrhagia and occasionally repeated abnormal uterine bleeding after her last cesarean section. Subsequently, she underwent intermittent gonadotropin-releasing hormone agonist therapy, although the cause of her uterine bleeding remained unknown. She had heavy uterine bleeding and required a blood transfusion at 49 years of age.

A transvaginal ultrasound examination detected abnormally developed and dilated blood vessels within the myometrium and around the uterus. The patient was referred for a thorough assessment and

Abbreviations: AVM, arteriovenous malformations; NBCA, N-butyl cyanoacrylate.

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https://doi.org/10.1016/j.crwh.2024.e00630
Received 9 May 2024; Received in revised form 10 June 2024; Accepted 11 June 2024
Available online 13 June 2024

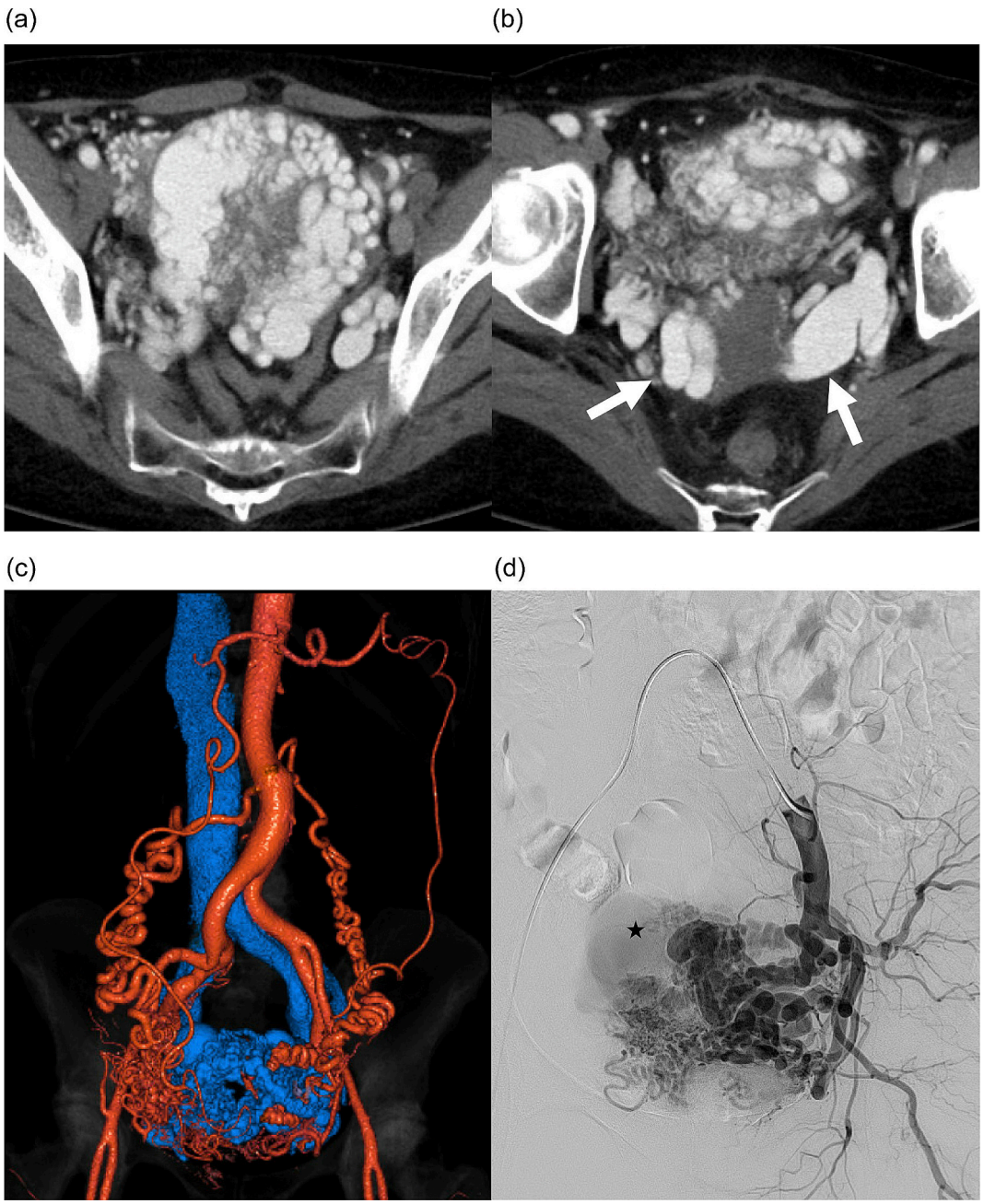

Fig. 1. Computed tomography and angiography images before embolization.
(a, b) Pelvic axial contrast-enhanced CT images showing a large uterine AVM. (a) Suspected shunts in the uterine corpus (arrowhead). (b) Dilated bilateral uterine and internal iliac veins as drainers (arrows). (c) Three-dimensional reconstructed images of contrast-enhanced CT of uterine AVM. (d) Left internal iliac artery angiography. A dilated venous space [14] is detected at the right side of the uterine corpus, immediately after tortuous feeding artery enhancement. The shunts of the AVM were considered present at the venous space periphery. AVM: arteriovenous malformations; CT: computed tomography.

treatment of a suspected uterine AVM.

Contrast-enhanced computed tomography (CT) revealed a large uterine AVM with dilated blood vessels occupying almost the entire uterus, along with markedly enlarged surrounding vessels (Fig. 1a). The bilateral uterine and internal iliac veins were dilated as the primary drainers, and the left internal iliac vein exhibited extensive dilation (Fig. 1b). The following feeders were observed: bilateral internal iliac artery branches, including uterine arteries, right external iliac artery, and the bilateral ovarian arteries, and gastroepiploic arteries due to greater omentum adhesion to the vesicouterine pouch (Fig. 1c). This uterine AVM was suspected to be the cause of the uterine bleeding. Chest radiography revealed cardiomegaly, and echocardiography detected diastolic left ventricular flattening and increased cardiac index (4.3 L/min/m^2), indicating right cardiac overload due to an AVM.

The surgical approach may have posed a high risk of life-threatening hemorrhage due to extensively dilated uterine and internal iliac veins and tortuous arteries in the adherent vesicouterine pouch. Therefore, the primary treatment involved transcatheter embolization of the feeding arteries.

Embolization using coils is considered inappropriate as it causes proximal embolization and AVM recurrence [10]. CT or transthoracic echocardiography identified neither pulmonary AVMs nor intracardiac shunts. Thus, paradoxical embolization was deemed to be unlikely, and N-butyl cyanoacrylate (NBCA) was administered as a liquid embolic

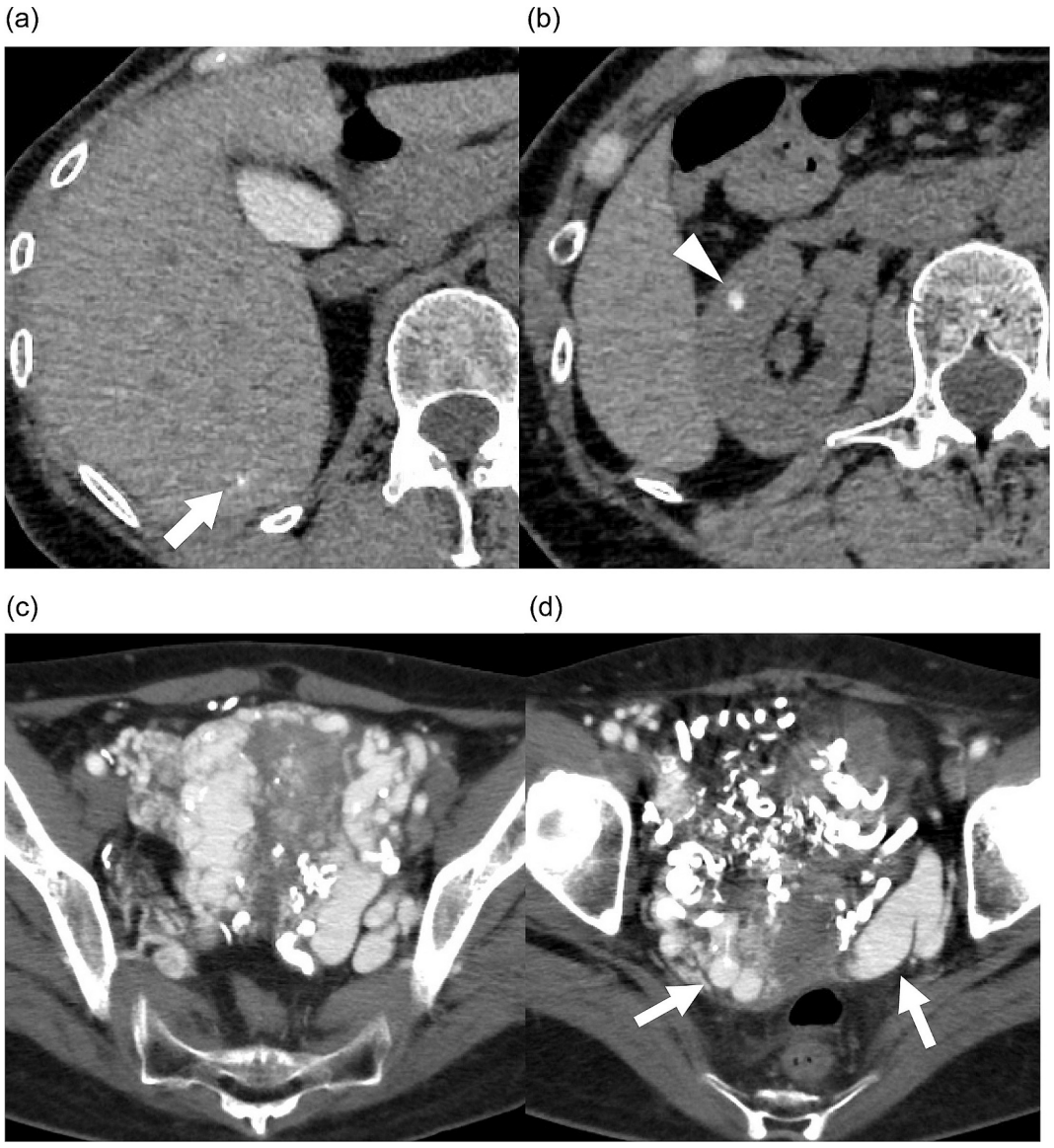

Fig. 2. CT images after NBCA embolization of a uterine AVM.
(a, b) Axial CT images at the upper abdomen. Hyper-attenuated nodules in the posterior lobe of the liver (arrow) and the right kidney (arrowhead), suspected to be paradoxical embolization of lipiodol. (c, d) Axial contrast-enhanced CT images at the pelvis. (c) The uterine AVM is smaller compared with the pretreatment image in Fig. 1 (a). (d) Remaining extensive dilation of the bilateral uterine to internal iliac veins, especially on the left (arrows). AVM: arteriovenous malformations; CT: computed tomography; NBCA: N-butyl cyanoacrylate.

agent. Pre-embolization aortography revealed a dilated venous space on the right side of the uterine corpus after tortuous feeding artery enhancement (Fig. 1d). The AVM shunts were considered to be located near the dilated venous space in the uterine corpus. Three embolization sessions were performed under local anesthesia and moderate sedation. NBCA mixed with iodized oil at ratios of 1:2–1:5 was used to embolize multiple feeding arteries that branch from the bilateral internal iliac arteries and omental branches of the bilateral gastroepiploic arteries. Postembolization CT revealed acceptable amounts of pulmonary emboli caused by passage of the embolic agent through the uterine AVM. However, multiple small foci of iodized oil were detected in the liver, spleen, and kidney, consistent with paradoxical embolism (Fig. 2a, b). The passage of the embolic agents through a patent foramen ovale was suspected. The patient demonstrated no clinical or imaging signs of cerebral embolism, but further embolization using NBCA was discontinued (Fig. 2c).

Total hysterectomy was considered difficult as significant uterine and internal iliac vein dilation remained despite blood flow reduction in the AVM after NBCA embolization (Fig. 2d). Therefore, supracervical hysterectomy was decided upon, as the shunts were found in the uterine corpus. This procedure could be performed without accessing the abnormally dilated veins and with minimal vesicouterine pouch dissection, thereby minimizing the risk of excessive bleeding. Moreover, an intraoperative balloon occlusion of the bilateral internal iliac arteries was planned after preoperative coil embolization of the feeding arteries that were unblockable by balloon obstruction.

Surgery was performed in a hybrid operating room 4 days after coil embolization. Two 4-F guiding sheaths were cannulated in the left femoral artery before laparotomy. A balloon catheter (TMP PED balloon catheter; Tokai Medical Products, Aichi, Japan) from each sheath was cannulated into the bilateral internal iliac arteries. Additionally, a 3-F sheath was inserted via the right femoral artery, allowing a quick transition to a larger sheath for aortic balloon occlusion in case of heavy bleeding. Supracervical hysterectomy, bilateral oophorectomy, and partial omentectomy were performed under balloon catheter inflation (Fig. 3a). The uterus was soft, probably due to blood vessel

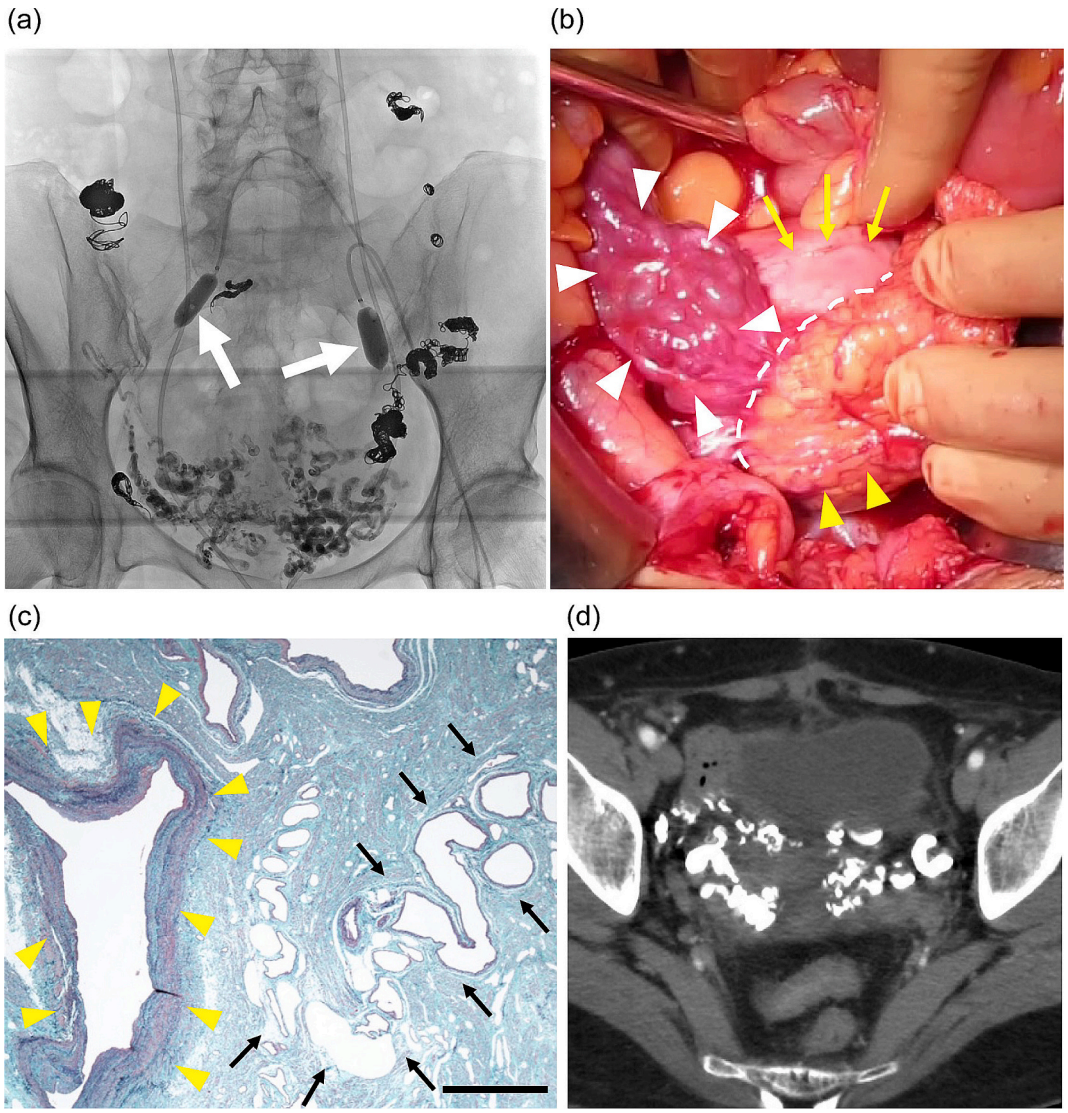

Fig. 3. Intra- and postoperative images.
(a) Intraoperative pelvic radiograph. Two balloon catheters were cannulated from the left femoral artery. The balloons (arrows) were inflated in the bilateral internal iliac artery. NBCA and lipiodol casts were observed in the uterine AVM, and metallic coils in the bilateral ovarian, right round ligament, and right lateral sacral arteries. (b) Photograph at supracervical hysterectomy initiation. Yellow arrows indicate the uterus. The greater omentum was adherent to the vesicouterine pouch (white dotted line). The arteries of the omentum and ovary remained distended (white and yellow arrowheads, respectively). (c) Histopathology image of Elastica-Masson staining. The vascular structures with heterogeneous wall thickness and irregular morphology were observed (yellow arrowheads). Several dilated and tortuous vessels were observed (black arrows). Scale bar: 1000 μm. (d) Axial contrast-enhanced CT image at the pelvis six months postoperatively showing no uterine AVM. NBCA and lipiodol casts were observed in the uterine cervix and cardinal ligaments. AVM: arteriovenous malformation; CT: computed tomography; NBCA: N-butyl cyanoacrylate.

development, and the greater omentum was adherent to the vesicouterine pouch. The omentum and ovarian arteries remained distended despite preoperative embolization (Fig. 3b). The greater omentum was first divided and bilateral ovarian vessels were cut. Vesicouterine pouch adhesions were then carefully dissected, allowing shunt resection. Finally, the lateral uterine vessels were ligated and cut at a higher level than the internal orifice, and the uterine corpus was resected. The operation lasted 3 h and 13 min, and the total blood loss was 180 mL. No complications were observed intra- or postoperatively. Histopathological examination revealed vascular structures with heterogeneous wall thickness and irregular morphology, compatible with uterine AVM (Fig. 3c). CT after 6 months detected no pelvic AVM (Fig. 3d). The patient was asymptomatic and progressing well, and echocardiography indicated recovery from right cardiac overload.

3. Discussion

A supracervical hysterectomy was successfully performed with preoperative embolization and intraoperative arterial balloon occlusion for the treatment of a large uterine AVM. Uterine AVMs are primarily associated with endometrial curettage, cesarean section, and chorionic disease [1]. The patient in this case had repeated abnormal uterine bleeding after her last cesarean delivery, indicating that cesarean section possibly caused her AVM, exacerbated by greater omentum adhesion to the vesicouterine pouch. After a long period the AVM had progressed to a critical condition with cardiac overload.

The goal of AVM treatment is complete shunt resection or obstruction [11]. Feeding artery proximal embolization prevents complete shunt closure, causing the recurrence of AVM due to collateral artery development. Therefore, proximal embolization is suitable only if surgical resection is immediately performed.

In this case, uterine AVM involved many feeding arteries, and the draining veins were extensively dilated. Therefore, the safety of surgery was unclear even after embolization. NBCA embolization was performed as the primary treatment and as close to the shunts as possible.

The surgical procedure was reconsidered when NBCA was unavailable. It was assumed that a total hysterectomy was required for shunt removal before initiating treatment. However, supracervical hysterectomy was considered a viable option as the shunts were located in the uterine corpus. Supracervical hysterectomy is safe and useful, despite being rarely reported for uterine AVMs with highly dilated uterine veins, as these make access to the cervical level risky, or for uterine AVMs with vesicouterine pouch adhesions and vascularization after cesarean sections. In the present case, the combination of preoperative coil embolization and intraoperative internal iliac artery balloon was effective in reducing blood flow and ensuring safe surgery.

Paradoxical embolization is a life-threatening complication of tumor embolization [12,13]. In the present case, NBCA and lipiodol fragments were speculated to pass through the uterine AVM and the patent foramen ovale. Liquid embolic agents may predominantly pass through arterial venous connections of AVM rather than through those of tumors. A close examination of the patent foramen ovale by performing microbubble tests may be considered before using liquid embolic agents for a large uterine AVM.

Here, a case of uterine AVM with marked vascular dilation throughout the pelvic region is reported. The large uterine AVM was safely treated by blood flow reduction with endovascular treatment and appropriate surgical procedures for shunt removal.

Contributors

Fumika Hamaguchi contributed to patient care, manuscript drafting, and literature review.

Yasuyuki Onishi contributed to patient care, case report conception, manuscript drafting, and literature review.

Yusuke Sagae contributed to patient care, case report conception, manuscript drafting, literature review and critical article revision for important intellectual content.

Koji Yamanoi contributed to patient care, case report conception and literature review.

Hironori Shimizu contributed to patient care, literature review, and critical article revision for important intellectual content.

Masaki Mandai contributed to patient care and critical article revision for important intellectual content.

All authors approved the final submitted manuscript.

Declaration of generative AI and AI-assisted technologies in the writing process

Generative AI and AI-assisted techniques were not used in writing this case report.

Funding

This work did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Patient consent

Obtained.

Provenance and peer review

This article was not commissioned and was peer reviewed.

Acknowledgments

We are grateful to Dr. Keigo Osuga, Professor of Diagnostic Radiology at Osaka Medical and Pharmaceutical University, for his helpful advice on the treatment plan and embolization technique.

Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

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