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Case report

Successful transvenous embbolization for type II uterine arteriovenous malformation: A case report ☆,☆☆

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ABSTRACT

A 40-year-old female (gravida 3 para 1) presented with menstrual, urinary, and anal pain. Computed tomography revealed type II acquired uterine arteriovenous malformation, a common dilated venous sac with bilateral uterine arteries, and multiple branches of iliac arteries draining to the bilateral ovarian veins. Venous sac transvenous embolization via the left ovarian vein of dominant outflow was planned, since complete arteriovenous malformation occlusion was difficult with super-selective transarterial embolization of multiple feeders. Therefore, transarterial embolization of the minor feeder was performed before completing transvenous embolization using coils and 50% glue under left iliac artery flow control. Immediately thereafter, angiography confirmed the complete disappearance of the uterine arteriovenous malformation, and all pain symptoms remitted. In conclusion, transvenous embolization combined with adjunctive transarterial embolization can be an effective and radical treatment for type II uterine arteriovenous malformations.

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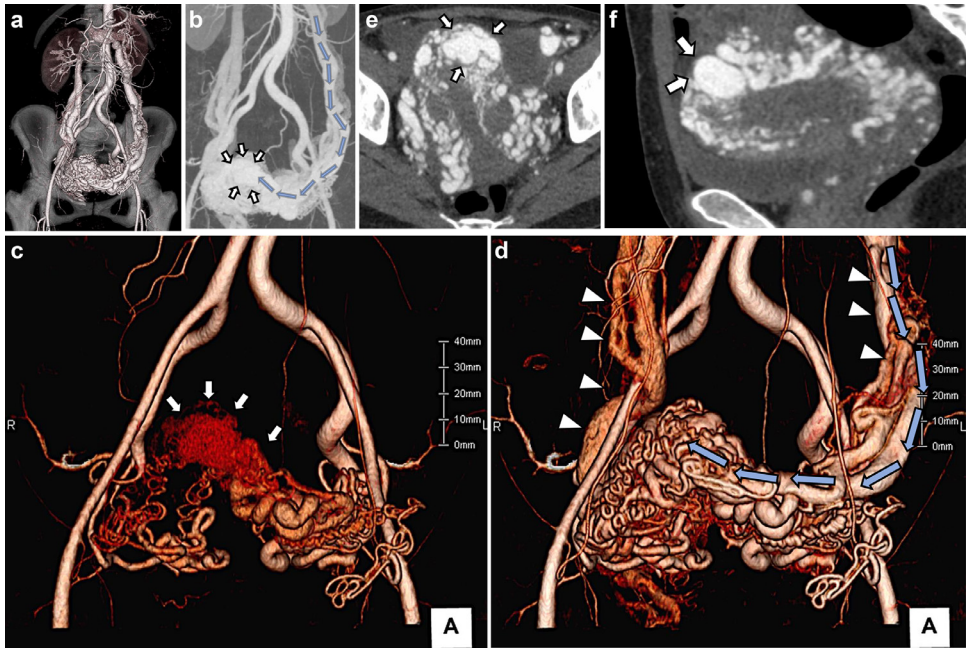

Fig. 1 – Preoperative computed tomography images.
Volume rendering (A) and maximum intensity projection (B) of CT angiography showing a huge U-AVM with early venous return of bilateral ovarian veins. Four-dimensional (4D) CT early phase (C) showing a huge U-AVM primarily fed by the dilated left uterine artery, right uterine artery, and multiple non-uterine arteries (left round ligament artery, bilateral internal pudendal artery, left obturator artery) and numerous feeding arteries continuous with a dilated venous sac. Four-dimensional CT late phase (D) showing draining bilateral ovarian veins via the myometrial venous plexus and dilated uterine veins around the uterus. The route of retrograde approach to the venous sac was continuous to the venous sac from the dilated left ovarian vein of dominant outflow. Contrast-enhanced CT arterial phase axial (E) and coronal (F) images showing a U-AVM venous sac in the myometrium at the base of the uterus and dilated uterine veins around the uterus. White arrows, venous sac; blue arrows, route of retrograde approach to the venous sac; white arrowheads, draining vein of bilateral ovarian veins). CT, computed tomography; U-AVM, uterine arteriovenous malformation.

Introduction

An acquired uterine arteriovenous malformation (U-AVM) is a rare condition causing uterine bleeding, menorrhagia, menometrorrhagia, lower abdominal pain, dyspareunia, or heart failure [1]. U-AVM often develops due to previous uterine trauma caused by dilatation and curettage (D&C), cesarean section, or pelvic surgery. D&C is the most frequent cause and is responsible for approximately 62% of cases [2,3].

Several treatments, including transarterial embolization (TAE), have been reported for U-AVMs [1]. TAE is usually chosen as the first-line treatment due to its low invasiveness. However, eliminating the vascular nidus (arteriole-venous connections without interposition of the normal capillary bed) of a huge or deeply involved U-AVM is difficult with TAE alone because the embolic material cannot sufficiently reach the nidus [3,4]. In cases where TAE results are inadequate, surgical treatment such as hysterectomy is preferred [1]. On the other hand, based on the anatomical morphology of AVMs in the body and extremities, treatment approaches other than TAE, such as nidus occlusion by transvenous or direct embolization, are proposed for complete occlusion [5,6].

Among the various AVM subtypes, transvenous embolization (TVE) is recommended for type II AVM based on Cho's classification; this type of AVM is characterized by a dilated common venous sac (VS) with multiple arteries [5,7]. However, there are no reports of TVE of the VS in a U-AVM in the literature. This report describes a case of type II U-AVM successfully treated with TVE in combination with TAE.

Case report

A 40-year-old female (gravida 3 para 1) presented with gradual worsening of severe menstrual, urinary, and anal pain. Computed tomography (CT) revealed a large U-AVM, and she was referred to our department. 15 years earlier, she had undergone therapeutic abortion followed by D&C for a hydatidiform mole. On external examination, blood samples were normal with no findings suggestive of anemia due to uterine bleeding. Since she wished to preserve her reproductive capacity and because surgical options have a potential risk of massive intraoperative blood loss, an interventional approach was selected.

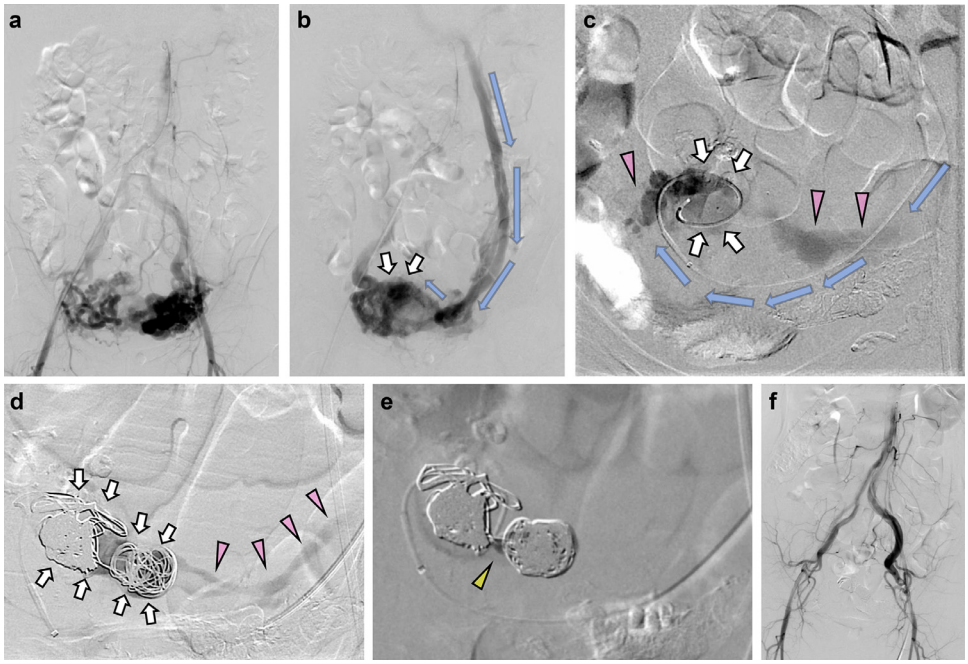

Fig. 2 – Transvenous embolization digital subtraction angiography.
Aortography before treatment demonstrating enlarged and tortuous feeding arteries (A) with enlarged bilateral ovarian veins: left ovarian vein dominant (B). A large microcatheter tip inserted into the venous sac via the left ovarian vein, with DSA of a venous sac showing the right part of the venous sac retrogradely and dilated myometrial venous plexus (C). The blue arrow shows the route of retrograde approach to the venous sac. Venous sac embolization by coiling from right to left was performed, and DSA during embolization of the left venous sac side (D) shows a decrease in visualization into the uterine vein. Following venous sac embolization using detachable microcoils to stop the flow completely, 50% glue embolization from the most distal coil through a small microcatheter tip was performed (E). Aortography after TVE demonstrating complete U-AVM disappearance (F). White arrows, venous sac; blue arrows, route of retrograde approach to the venous sac; pink arrowheads, dilated myometrial venous plexus and uterine vein; yellow arrowheads, 50% glue. DSA, digital subtraction angiography; TVE, transvenous embolization; U-AVM, uterine arteriovenous malformation.

CT images showed a huge U-AVM fed by the bilateral uterine arteries, multiple branches of iliac arteries, and arteries connecting to a dilated common VS adjacent to the uterine base. The left ovarian vein, as the dominant outflow via VS, and the right ovarian vein as the drainer, were involved via the myometrial venous plexus (Fig. 1).

Based on CT findings, the patient was diagnosed with a high-flow type II U-AVM. TVE was planned because superselective TAE of each feeder seemed not only time-consuming and difficult but also could possibly result in insufficient proximal embolization. Instead of TAE alone, we planned a transvenous approach including balloon-occluded retrograde obliteration (B-RTO) with sclerosant or coil-based TVE as a treatment option for VS embolization. Additionally, TAE combined with a transvenous approach was also planned to reduce the U-AVM shunt flow as U-AVM rupture could occur due to outflow block by unsuccessful VS occlusion via a transvenous approach alone. Therefore, we decided to treat it in two phases. We first performed TAE alone to reduce the shunt volume; thereafter, the planned radical TVE was conducted.

Pre-treatment aortography showed enlarged tortuous arteries feeding the VS and draining enlarged bilateral ovarian veins (dominant left ovarian vein) (Fig. 2A and B). The left round ligament artery and the right uterine artery were embolized as-distal-as-possible to reduce the shunt flow with a 14-17% n-butyl-2 cyanoacrylate (NBCA; Histoacryl Blue; B. Braun, Melsungen, Germany) and iodized oil (Lipiodol; Guerbet, Aulnay-Sous-Bois, France) mixture. TAE was technically successful, and a reduction in shunt volume was obtained.

Seven days after TAE, a transvenous approach via the right subclavian vein was performed. After introducing a 10-Fr sheath and a 9-Fr guiding balloon catheter, a 5-Fr balloon catheter (CANDIS: coaxial and double interruption system, Medikit, Tokyo, Japan) was inserted co-axially into the most distal portion of the main trunk of the left ovarian vein. Balloon-occluded retrograde venography (B-RTV) demonstrated no contrast stagnation in the VS and showed contrast flowing to multiple drainage veins, despite additional temporal flow control of the left internal iliac artery with a balloon catheter. Thus, B-RTO was abandoned for superselective catheterization to VS to perform coil-based TVE. A microcatheter (2.7/2.8 Fr BISOP HF, Piolax Medical Devices Inc., Kanagawa, Japan) was successfully inserted into the VS via a 5-Fr balloon catheter placed in the left ovarian vein. The VS was embolized with 10 detachable coils (8-20 mm diameter, 30-60 cm length). Since complete flow stasis was not obtained, a small microcatheter (1.9/1.9-Fr Carnelian Marvel, Tokai Medical Products, Inc., Aichi, Japan) was advanced to the distal coil

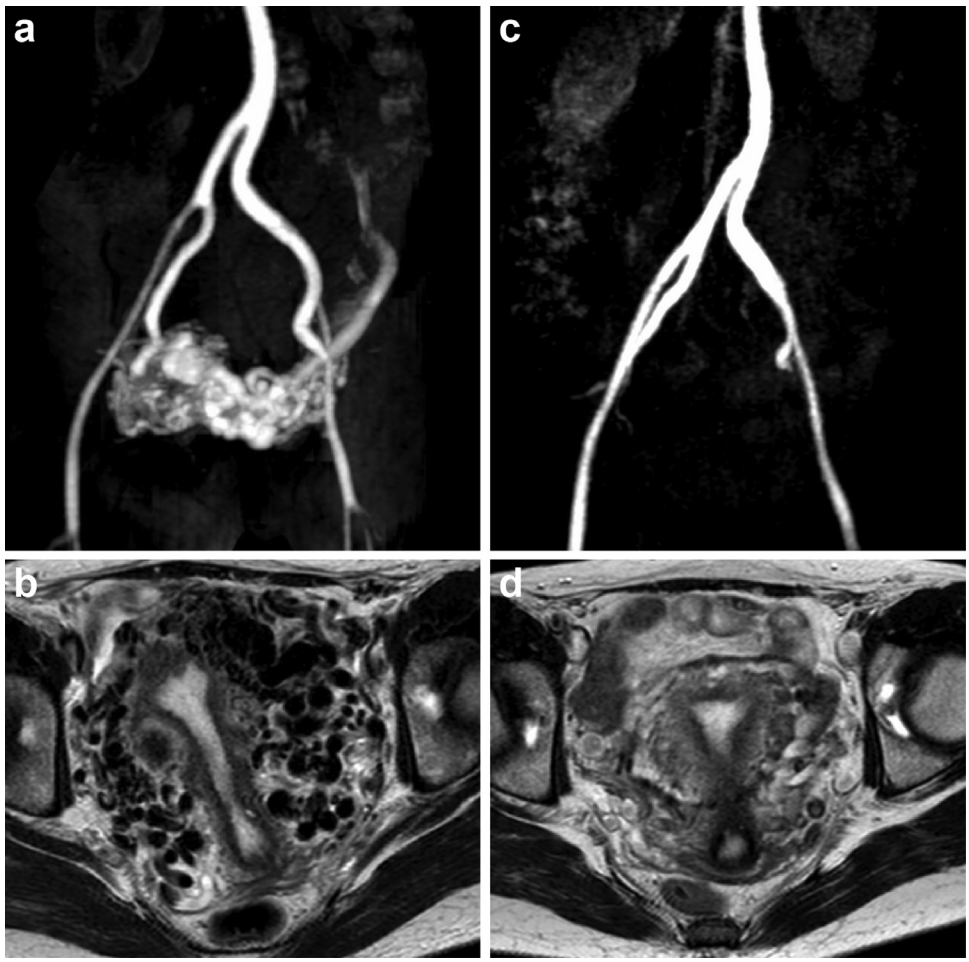

Fig. 3 – Contrast media in time-resolved magnetic resonance angiography and T2-weighted imaging. MRA (A) and T2WI (B) before treatment show a huge U-AVM with early venous return, and multiple dilated blood vessels around the uterus. MRA (C) and T2WI (D) 9 months after treatment show disappearance of U-AVM and multiple dilated blood vessels surrounding the uterus. MRA, magnetic resonance angiography; T2WI, T2-weighted image; U-AVM, uterine arteriovenous malformation.

nest (upstream of VS) via a first microcatheter (triple coaxial technique). Additional injection of a 50% NBCA-Lipiodol mixture (0.6 mL) into the coil nest through a small microcatheter (Fig. 2C-E) led to complete flow stasis. Aortography immediately after TVE demonstrated the disappearance of the U-AVM and early vein visualization (Fig. 2F).

As the 17-day inpatient postoperative course was uneventful and clinical symptoms completely disappeared, the patient was discharged.

Postoperative time-resolved magnetic resonance angiography and T2-weighted images 9 months after treatment showed complete resolution of the U-AVM and shrinkage of arteries and veins surrounding the uterus (Fig. 3).

Discussion

Acquired U-AVMs typically involve multiple small arteriovenous fistulas between small intramural uterine arteries and the myometrial venous plexus [3,8], while VS formation in U-AVM is extremely rare. In fact, it has not been reported so far. However, the mechanism of VS formation in this case is unknown.

On analysis of therapeutic outcomes and approaches according to the anatomical morphology of AVMs in the body and extremities [4,5,7,9,10], embolization for VS accessed through a direct puncture or transvenous approach is recommended for type II AVM lesions with a single VS with multiple feeders, as in the present case [5].

Percutaneous direct VS puncture is the standard approach for type II AVMs. A successful case with pelvic type II AVM, not U-AVM, has been previously reported [10]. However, direct VS puncture was not considered in the present case because percutaneous transabdominal puncture of this deep-seated and complicated vasculature lesion seemed difficult, and a puncture failure could result in severe hemorrhage.

As a treatment option other than a direct puncture, coil-based TVE has been reported to be effective for type II AVMs [5,7,9]. Complete occlusion of a renal AVM was achieved in 88%

of patients by transvenous coil embolization of VS alone [7]. In the present case, the VS was embolized by multiple coils; however, we could not achieve complete flow stasis despite additional flow reduction by combined TAE due to excessive shunt flow. Therefore, NBCA was added for successful occlusion of the VS.

As another method of transvenous approach, a case report showed the utility of B-RTO with sclerosant for a U-AVM [6]. B-RTO was initially prioritized as it is commonly used for treating gastric varices. However, the use of B-RTO was abandoned because successful B-RTO was predictably difficult. A reported case of U-AVM had a single draining vein; thus, sclerosant stagnation under balloon occlusion could be achieved. However, in the present case, blood flow stagnation could not be achieved in the VS because the U-AVM had multiple drainage veins.

TAE is usually performed as the first-line treatment for a U-AVM. Yoon et al reported a primary success rate of 61% with TAE and a secondary success rate of 91% after repeated embolization [3]. However, TAE sometimes results in inadequate outcomes due to incomplete U-AVM occlusion by insufficient embolic reach to the nidus [3,4]. Adjunctive TAE combined with TVE was reported to be useful for type II AVMs, similar to the present case, for safe and complete occlusion of AVM [9,10].

Precise image interpretation of the anatomical morphology of a U-AVM is essential to select an appropriate treatment approach other than TAE alone to avoid unnecessary surgical intervention.

TVE combined with adjunctive TAE can be an effective and radical treatment for type II U-AVMs.

Statement of Human and Animal Rights

This article does not contain any studies with human or animal subjects.

Patient Consent Statement

Informed Consent: Written informed consent was obtained from the patient for publication of this case report and any accompanying image.

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