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Case Report

Transvenous embolization for a huge pelvic arteriovenous malformation associated with prominent outflow veins

Atsushi Ugajin, MD[a], *Hiroyuki Fujii, MD,PhD*[a,*], *Akifumi Fujita, MD,PhD*[a], *Hiroyasu Nakamura, MD*[a], *Akira Fujisaki, MD*[b], *Hideharu Sugimoto, MD,PhD*[a]

[a] *Department of Radiology, Jichi Medical University, School of Medicine, 3311-1 Yakushiji, Shimotsuke, Tochigi 329-0498, Japan*

[b] *Department of Urology, Jichi Medical University, School of Medicine, Shimotsuke, Tochigi, Japan*

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ABSTRACT

We describe the case of an adult female with a huge pelvic arteriovenous malformation (AVM) measuring approx. 8 × 10 × 13 cm, treated via the transvenous approach alone. Management of huge pelvic AVMs is challenging; there is no consensus on a standardized treatment strategy. In our patient, multiple arterioles shunted to prominently dilated outflow veins. Selective occlusion of prominent outflow veins as close to the nidus as possible achieved significant flow reduction through the nidus. Our treatment may be a safe and acceptable option to control hematuria for huge pelvic AVMs associated with prominent outflow veins, especially when both transarterial embolization and direct puncture are difficult.

Introduction

An arteriovenous malformation (AVM) is an abnormal communication between arteries and veins with a nidus, which infrequently occurs in the pelvis. Given the high recurrence rate and morbidity associated with the surgical resection of AVMs, transcatheter embolization has emerged as the preferred treatment for AVMs [1]. Peripheral AVMs are classified into 4 types based on their angiographic morphologies, with implications for therapeutic approach and outcomes: type I, no more than 3 separate arteries shunt to the initial part of a single venous component; type II, multiple arterioles shunt to the initial part of a single venous component, in which the arterial components show a plexiform appearance on angiography; type IIIa, fine multiple shunts are present between the arterioles and venules and appear as a blush or fine striation on angiography; and type IIIb, multiple shunts are present between the arterioles and venules and appear as a complex vascular network on angiography [2].

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* Corresponding author.
E-mail address: hiroyuki.fujii@jichi.ac.jp (H. Fujii).

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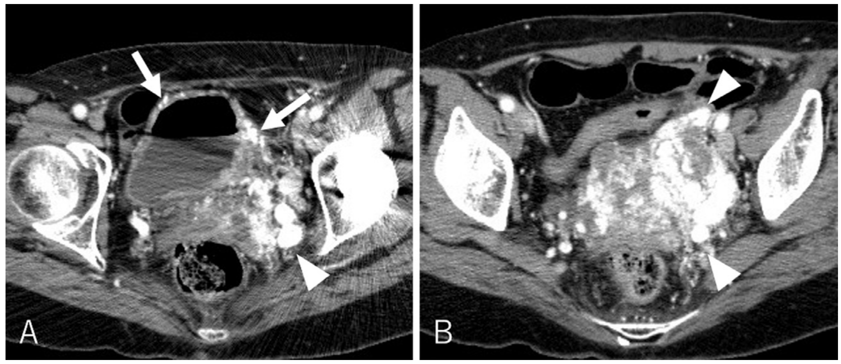

Fig. 1 – Axial contrast-enhanced CT images of the original lesion. There was a huge hypervascular lesion in the retroperitoneum and the pelvis, which involved the wall of the urinary bladder (A, *arrows*). There was no bone involvement. Note the dilated LIIV and LOV (A, B, *arrowheads*).

Fig. 2 – Coronal and axial contrast-enhanced CT images of the recurrent lesion. The infiltrating vessels in the wall of the urinary bladder were more dilated than the original lesion (A, *arrow*). An initial part of the LIIV (B, *arrow*) forms a venous pouch (B, *arrowhead*). Apart from the venous pouch, the arterioles also drained into the LOV (C, *arrow*).

In general, the goal of transcatheter embolization for AVMs is the obliteration of the nidus. Either a transarterial approach or a direct puncture has been preferentially used to embolize the nidus itself. The transvenous approach is also considered effective in some case, especially in type II AVMs. A single dilated outflow vein in type II AVMs has been referred to as a dominant outflow vein (DOV). The embolization of a DOV by the transvenous approach has achieved good therapeutic outcomes [2,3]. However, it is difficult to categorize huge AVMs with complex angiographic morphologies into any of the above-mentioned types, and there is no consensus on a standardized treatment strategy. Herein, we present the case of a patient with a huge pelvic AVM that was associated with prominent outflow veins. The AVM was successfully treated via the transvenous approach alone.

Case report

A 54-year-old woman was admitted to our hospital with gross hematuria. After the work-up for hematuria, she was initially diagnosed as having a huge pelvic AVM, measuring 8 × 10 × 13 cm. Contrast-enhanced computed tomography (CT) showed the AVM spread in the retroperitoneum and the pelvis, which involved the wall of the urinary bladder (Fig. 1). There was no bone involvement. She underwent transarterial embolization using n-butyl-2-cyanoacrylate (NBCA; Histoacryl, Braun, Melsungen, Germany) and subsequently remained stable without further hematuria. However, hematuria recurred several months after the treatment and unfortunately, the patient was lost to follow-up.

At 5 years after the initial treatment, the patient was readmitted to our hospital with recurrent gross hematuria. Contrast-enhanced CT showed the recurrent lesion in the same location. Although there was no remarkable change in the overall size (8 × 10 × 13 cm), the arteries and veins in the AVM were more dilated compared to the patient's initial admission before the previous transarterial embolization, especially the vessels in the wall of the urinary bladder. The arterioles were draining into the dilated left internal iliac vein (LIIV) which formed a large venous pouch and the dilated left ovarian vein (LOV; Fig. 2). Cystoscopy showed an intravesical, pulsatile mass arising from the wall of the urinary bladder, confirming the diagnosis of a residual pelvic AVM (Fig. 3).

Pretreatment vascular mapping angiography showed numerous, tortuous arterioles arising from bilateral internal iliac, external iliac, and inferior mesenteric arteries returning to 3 outflow veins: the LIIV, LOV, and right internal iliac vein (RIIV; Fig. 4). Among them, the LIIV with the venous pouch and the LOV were prominently dilated. We made the diagnosis of a complex pelvic AVM which could not be classified as any of

Fig. 3 – Pretreatment cystoscopy. An intravesical, pulsatile mass is arising from the wall of the urinary bladder.

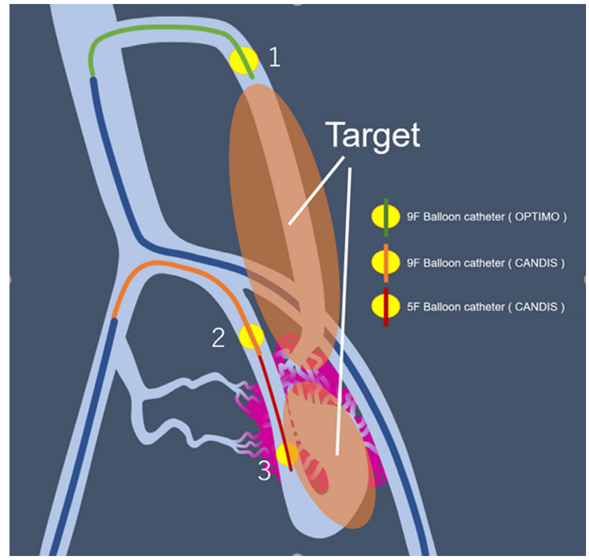

Fig. 5 – Schematic diagram of the treatment plan in the present case. To embolize the venous pouch of the LIIV and the LOV, transvenous embolization under flow control is attempted.

the 4 above-described types based on the angiographic findings; the lesion had multiple dilated arteriovenous shunts as could be seen in type IIIb AVMs, but the outflow veins were extremely dilated like a DOV in type II AVMs

Treatment

Since the initial transarterial embolization with NBCA failed to control the patent's symptoms, we planned to embolize the enlarged venous pouch of the LIIV and the LOV to achieve sufficient flow reduction through the nidus. The bilateral femoral veins were punctured. A 9-Fr balloon catheter (Optimo, Tokai Medical Products, Aichi, Japan) was advanced to the LOV via the left femoral vein. A 9-Fr/5-Fr double coaxial balloon catheter system (Candis, Medikit, Tokyo, Japan) was advanced to the LIIV via the right femoral vein (Fig. 5). The left internal iliac artery was also catheterized via the right femoral artery using a 5-Fr balloon catheter (Selecon MP, Terumo, Tokyo, Japan) to control the arterial inflow during the procedure.

We first attempted to embolize the LOV. A 5-Fr catheter (Imager, Boston Scientific, Marlborough, MA) and a 2.85-Fr microcatheter (Carry Leon, UTM, Aichi, Japan) were inserted in parallel through the 9-Fr balloon catheter. A 2.2-Fr microcatheter

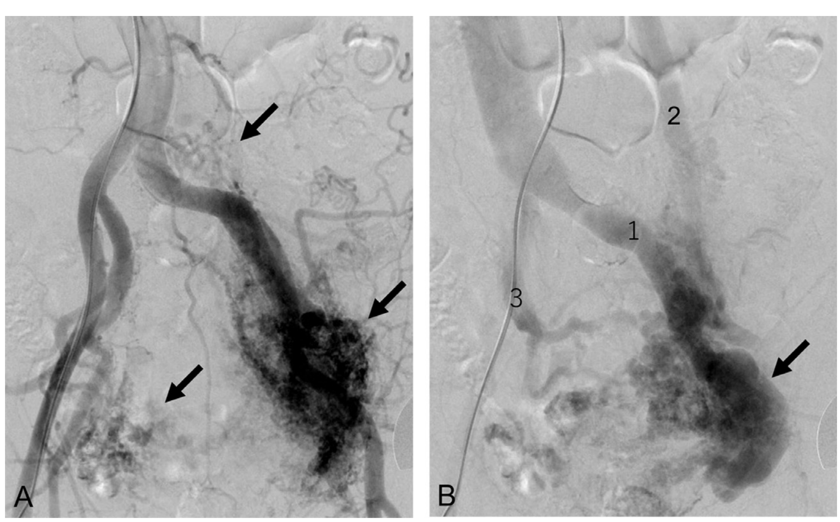

Fig. 4 – Pretreatment vascular mapping angiography. The early phase shows numerous tortuous arterioles (A, *arrows*). The delayed phase shows the three outflow veins (B: 1, LIIV; 2, LOV; 3, RIIV) and the venous pouch of the LIIV (B, *arrow*).

(Progreat β3, Terumo) was inserted through the 5-Fr catheter. Eight 0.035-inch fibered coils (Interlock-35, Boston Scientific), seven 0.020-inch bare coils (Ruby coil, Penumbra, Alameda, CA), and 5 fibered 0.018-inch coils (Interlock-18, Boston Scientific) were deployed into the LOV. Subsequently, 2 mL of a mixture of NBCA and iodized oil (Lipiodol, Guerbet, Aulnay-Sous-Bois, France; NBCA:lipiodol = 1:3) was injected between 2 coil meshes, and 1 vascular plug (Amplatzer Vascular Plug II, Abbott Vascular, Santa Clara, CA) was deployed into the proximal part of the LOV to achieve complete occlusion and reduce the number of coils used.

We then attempted to embolize the venous pouch. A 5-Fr balloon catheter (Candis) was catheterized into the venous pouch via the right femoral vein. A 4-Fr guiding catheter (Cerulean G, Medikit) through a 6-Fr guiding catheter (Envoy, Johnson & Johnson, Miami, FL) was also catheterized into the venous pouch via the left femoral vein. A 2.6-Fr microcatheter (PX SLIM, Penumbra) and a 2.2-Fr microcatheter (Progreat β3) were inserted through the 5-Fr balloon catheter or the 4-Fr guiding catheter, respectively. Twelve 0.035-inch fibered coils (Interlock-35), seventeen 0.020-inch bare coils (Ruby coil), twelve 0.018-inch fibered coils (Nester, Cook Medical, Bloomington, IN), and two 0.018-inch hydrogel-coated coils (AZUR, Terumo) were deployed into the venous pouch (Fig. 6).

At that time point, since post-treatment angiography confirmed significant flow reduction through the nidus (Fig. 7) and because the RIIV was so narrow on CT that selective catheterization close to the nidus seemed difficult, we decided not to embolize the RIIV. There were no adverse effects or complications related to transcatheter embolization. At 10 months after the treatment, cystoscopy confirmed complete disappearance of the lesion with no obvious hematuria (Fig. 8).

Discussion

We have described the case of a huge pelvic AVM associated with prominent outflow veins that was treated via the transvenous approach alone. To our knowledge, there have been no previous reports of the attempted embolization of the prominent outflow veins for a huge pelvic AVM via the transvenous approach alone.

In our patient's case, transarterial embolization seemed difficult because feeding arteries were numerous and tortuous, which could result in proximal embolization. We speculate that this was why the previous transarterial embolization resulted in a poorer outcome. Although a direct puncture might be another viable option, we expected that it would be difficult to puncture the pelvic AVM due to its deep location and the potential risk of pelvic bleeding. In addition to these reasons, because there were 2 prominent outflow veins similar to a DOV in our patient's case, we planned to embolize the AVM via the transvenous approach in accordance with the strategy for type II AVMs.

The obliteration of a single dilated outflow vein referred to as a DOV is the practical goal for the treatment of a type II AVM. However, a DOV itself is not a nidus, because the nidus exists between the DOV and the feeding arteries. Several studies have thus reported the effectiveness of transvenous embolization for a DOV using liquid materials such as ethanol or Onyx (Medtronic, Dublin, Ireland) combined with coils to penetrate the nidus retrogradely [2–6]. However, in cases of huge AVMs, occluding all outflow veins is technically difficult. In the present case, residual shunts through the RIIV could exist.

Nevertheless, we accomplished sufficient flow reduction through the nidus because prominent outflow veins (the LIIV and LOV) were occluded as close to the nidus as possible and residual shunts through the RIIV were insignificant. If we had treated the patient by embolization of the outflow veins distant to the nidus, it may have resulted in only a change in the route of the draining flow (Fig. 9).

Huge AVMs pose a higher risk of nontarget embolization and require larger amounts of liquid materials, which can cause serious complications such as pulmonary embolism, acute pulmonary hypertension, and subsequent cardiopulmonary collapse [7,8]. In the present patient's case, we therefore used mainly coils and a vascular plug. Among the various embolic materials that are available, coils may be the most feasible embolic material for high-flow outflow veins. Some detachable coils can be placed densely within the outflow veins just distal to a nidus, with a low risk of migration under flow control.

The use of NBCA after the placement of some coils helps achieve the complete occlusion of outflow veins, and it can reduce the total number of coils required. Although NBCA is a liquid embolic material, the appropriate concentration of an NBCA-iodized oil mixture can be used to shorten or lengthen the polymerization time. Moreover, a small amount of injected NBCA between coil meshes enables embolization with a lower risk of migration. Vascular plugs are also effective and more cost-effective than coils, and they present a low risk of migration. However, drawbacks of the use of a vascular plug are the need for a larger catheter tip, a relatively stiff delivery wire, and a relatively straight landing zone. Moreover, incomplete occlusion after the placement of a vascular plug makes further embolization difficult within a short segment of target vessel.

As previous reports indicated, if our strategy results in incomplete devascularization of the nidus, subsequent transarterial embolization or direct puncture should be considered [3,9]. Additional transvenous embolization can also be required in the long term when a residual shunt develops through the remaining outflow veins. Performing the embolization of prominent outflow veins first can be expected to be effective for shunt reduction and for minimizing the need for a subsequent embolization procedure, even though there is a theoretical risk of nidus rupture as a result of elevated intravenous pressure.

In conclusion, we treated a huge pelvic AVM associated with prominent outflow veins via the transvenous approach alone. Since complete devascularization of the nidus was not accomplished and the long-term effectiveness of our approach in this case has not yet been confirmed, our approach may not be a radical treatment. However, transvenous embolization of prominent outflow veins may be a safe and acceptable treatment option for the management of hematuria for huge pelvic AVMs.

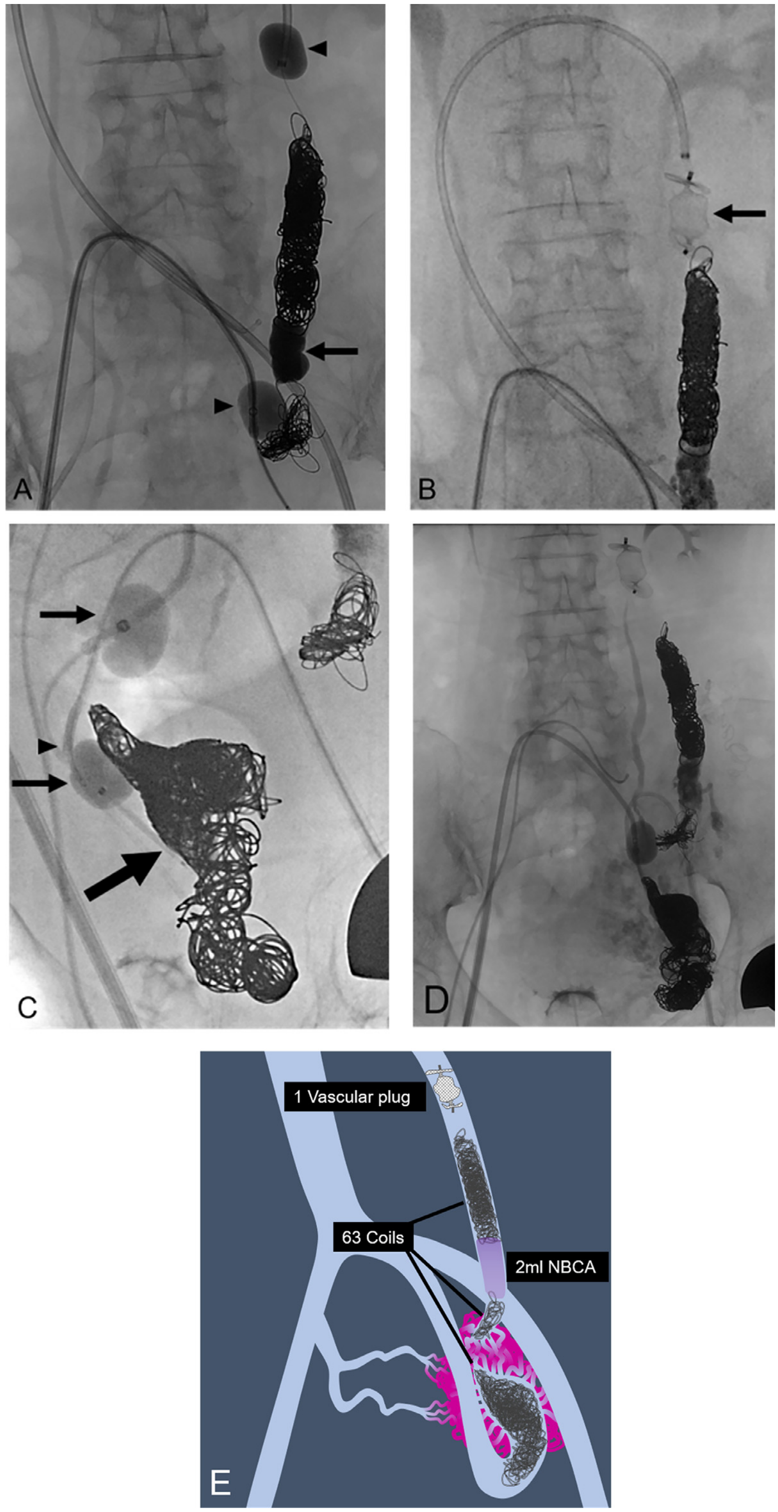

Fig. 6 – Embolization of 2 prominent outflow veins. Initially, the LOV is embolized by coils, a mixture of NBCA and iodized oil, and a vascular plug. After the placement of the coils, the mixture of NBCA and iodized oil (A, *arrow*) is injected between 2 coil meshes under venous flow control using balloon catheters (A, *arrowheads*), and the vascular plug is deployed into the proximal part of the LOV (B, *arrow*). Next, the venous pouch is embolized by coils (C, *arrow*). A 9-Fr/5-Fr double coaxial balloon catheter system (C, *thin arrows*) and a 6-Fr guiding catheter (C, *arrowhead*) are catheterized into the LIIV. Consequently, 63 coils, 2 ml of the mixture of NBCA and iodized oil, and a vascular plug are required to perform the procedure (D, E).

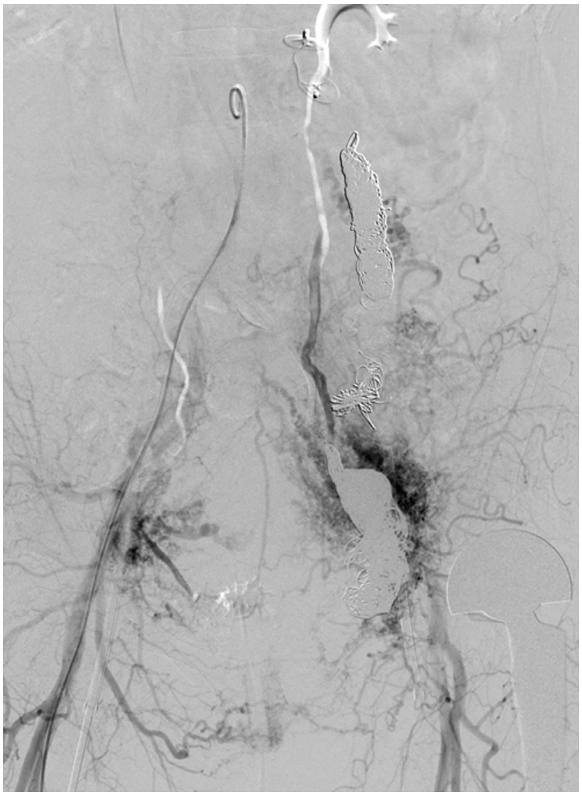

Fig. 7 – Post-treatment angiography showing the disappearance of early venous return through the LIIV, LOV, and RIIV despite the incomplete devascularization of the nidus.

Fig. 8 – Post-treatment cystoscopy. There are no abnormal signs of residual pelvic AVM.

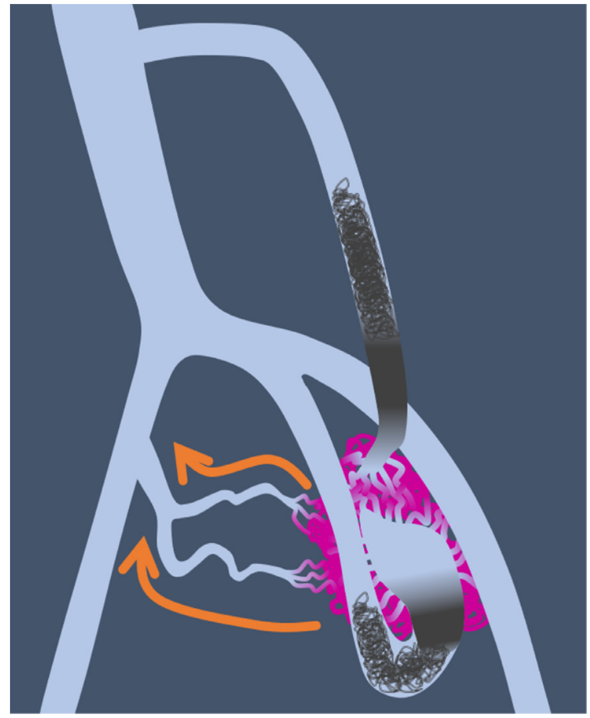

Fig. 9 – Schematic diagrams of the approaches to pelvic AVM. The embolization of the outflow veins distant to the nidus cannot lead to thrombosis in the nidus retrogradely, changing only the route of the draining flow (*arrows*).

Data availability

No data were used to support this study.

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