INTERVENTIONAL RADIOLOGY
Official journal of the Japanese Society of Interventional Radiology and Asia Pacific Society of Cardiovascular and Interventional Radiology

CASE REPORT

Double Microballoon-occluded Ethanol Embolization for Pelvic Arteriovenous Malformation: A Case Report

Keigo Osuga[1)], Naoki Yokota[1)], Kazuhiro Yamamoto[1)], Hiroki Matsutani[1)], Kiyohito Yamamoto[1)], Hiroshi Juri[1)], Hideki Ozawa[2)] and Takahiro Katsumata[2)]

1) Department of Diagnostic Radiology, Osaka Medical and Pharmaceutical University, Japan
2) Department of Thoracic and Cardiovascular Surgery, Osaka Medical and Pharmaceutical University, Japan

Abstract:
A 40-year-old man was incidentally found to have right-sided pelvic arteriovenous malformation (AVM) with an aneurysmal dominant outflow vein (DOV). The AVM had two main feeding arteries forming a cluster of fine vessels shunt to the DOV. As transvenous approach was impossible due to anatomical difficulty, transarterial ethanol embolization was performed under simultaneous double microballoon occlusion of the two feeding arteries in combination with protective coil embolization of the prostatic branches. Ethanol (13 mL) was intermittently injected from both microballoon catheters until the AV shunt was completely occluded. At 1-year follow-up, contrast-enhanced CT revealed shrinkage of the thrombosed DOV without any symptom. Our case demonstrated the usefulness of simultaneous double microballoon-occluded ethanol embolization for treating a localized pelvic AVM with a few feeding arteries.

Keywords:
pelvic arteriovenous malformations, embolization, ethanol, microballoon

Interventional Radiology 2024; 9(1): 26-30
https://doi.org/10.22575/interventionalradiology.2023-0021
https://ir-journal.jp/

Introduction

Arteriovenous malformation (AVM) is caused by abnormal connection between the arteries and veins. It rarely occurs in the pelvis. Pelvic AVMs may present as asymptomatic, abnormally dilated vessels on radiologic imaging. Symptoms include pelvic pain, dysuria, urinary frequency, hematuria, dyspnea on exertion, and high-output cardiac failure [1, 2]. Given the high recurrence rate and morbidity of the surgical treatment, endovascular treatment has emerged as the less-invasive treatment option [3]. However, there is no consensus on the standardized method regarding the choice of embolization access routes and embolic agents. We herein report a case of pelvic AVM with an aneurysmal outflow vein that is difficult to treat with transvenous embolization (TVE). The AVM was successfully treated with a single session of transarterial ethanol embolization under simultaneous microballoon occlusion of the two main feeding arteries.

Case Report

A 40-year-old man presented with headache, vomiting, and diarrhea to the emergency room of another hospital. Plain and contrast-enhanced CT (CECT) incidentally revealed abnormally dilated vessels along the right pelvic wall, indicating pelvic AVM (**Fig. 1**). His initial symptoms were relieved conservatively and considered unrelated to the pelvic AVM. After 3 months, he was referred to our department for further examination and endovascular treatment of the pelvic AVM. At the first clinic visit, he was asymptomatic and had a normal blood pressure of 110/74 mmHg and pulse rate of 63 bpm. He had a history of medication for dyslipidemia but no previous history of trauma, neoplasm, or surgery in the pelvis.

3D-CTA showed two main feeding arteries arising from the common trunk with inferior gluteal artery forming a cluster of fine vessels shunt to the inferior wall of the aneurysmal dominant outflow vein (DOV) (40 × 34 × 30 mm). The right obturator artery also gave off small branches to

Corresponding author: Keigo Osuga, osuga@ompu.ac.jp
Received: May 14, 2023, Accepted: July 18, 2023, Advance Publication by J-STAGE: February 8, 2024

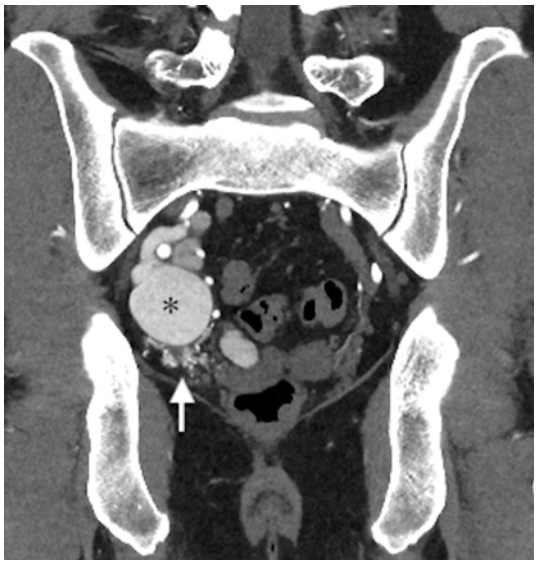

Figure 1.
Coronal MPR image of contrast-enhanced CT shows aneurysmal dilatation of the DOV (asterisk) and a cluster of fine vessels along the inferior wall of the DOV (arrow).

the shunt. Furthermore, these shunt vessels were connected with the normal branches to the prostate. There were two dilated tortuous draining veins branched from the DOV, one superiorly and the other inferiorly, with the inferior vein compressing the urinary bladder. These two veins were confluent just before entering into the right internal iliac vein. We thoroughly discussed with the patient and his family about the potential risks of massive hematuria, intrapelvic hemorrhage, and high-output cardiac failure in the future as well as the procedural risks, including serious ischemic and thromboembolic complications. We decided to perform endovascular treatment after the patient provided written informed consent for the treatment and publication.

Diagnostic angiography was first performed using the left femoral approach to evaluate the detailed vascular anatomy and means of possible access to the AV shunt. Aorto-pelvic arteriogram revealed the whole appearance of AVM in the territory of right iliac artery with the rapid outflow into the DOV and the two continuous dilated tortuous veins draining into the right internal iliac vein. The selective right internal iliac arteriogram showed the two main feeding arteries and an accessory feeding branch from the obturator artery sharing the cluster of abnormal fine vessels shunt to the DOV. Thus, it was considered as type II AVM based on Cho's classification [4]. Normal prostatic branches were also observed from the fine vessels as expected by the 3D-CTA. The balloon-occluded right internal iliac arteriogram using a 5 Fr 9-mm balloon catheter (Selecon MP Catheter II, Terumo, Tokyo, Japan) showed sluggish antegrade flow into the shunt and prostatic branches. Transvenous catheterization to the draining veins was attempted from the left femoral vein; however, it was impossible due to the acute angle and narrowing of the orifice from the right internal iliac vein.

Therapeutic embolization of the AVM was separately performed using the bilateral femoral approach under general anesthesia. A 4 Fr pig-tail catheter (SZ1350, Medikit, Tokyo, Japan) was placed in the aorta from the right femoral artery to repeat pelvic angiogram during the procedure (**Fig. 2a, b**). Another 6 Fr guiding sheath (ParentPlus60; Medikit, Tokyo, Japan) was placed in the main right internal iliac artery from the left femoral artery. Two microballoon catheters (Logos GrandMaster; PIOLAX, Kanagawa, Japan) were coaxially advanced from the same guiding catheter into the distal portion of two main feeding arteries just before the abnormal shunt vessels. Simultaneous microballoon-occluded angiography from both feeding arteries revealed blood flow stagnation of the shunt (**Fig. 2c**). The prostatic branches were not visualized due to the reversal flow by microballoon occlusion. Absolute ethanol (0.5-1 mL) was intermittently and alternately injected from each feeding artery at 5- to 10-min intervals under microballoon occlusion. The so-called sandwich technique was used for ethanol injection, in which pure radiolucent ethanol was injected after test contrast injection and the residual ethanol in the microballoon catheter was carefully flushed with a small amount of contrast media under fluoroscopy. Before each next ethanol injection, test injection of contrast media (0.5-1 mL) was performed to assess the degree of blood flow reduction or determine whether any normal branch supplying pelvic organs became visualized. Indeed, as the blood flow to the shunt decreased, the prostatic branches began to appear. Thus, one of the microballoon catheters was deflated to advance into these prostatic branches, and then protective embolization was performed using 10 detachable microcoils (Target 360 Ultra; Stryker Japan K.K., Tokyo, Japan). A total of 13 mL (0.20 mL/kg) of absolute ethanol was used until complete flow cessation of the shunt. The final pelvic arteriogram confirmed the complete disappearance of the AVM without collateral arteries from other territories (**Fig. 2 d**).

After the procedure, the patient did not complain of pelvic or buttock pain, numbness in the lower extremities, or respiratory distress. CECT performed 3 days after embolization revealed complete thrombosis of the DOV and the two contiguous draining veins with size reduction of the feeding arteries. At 1-year follow-up visit, the patient remained asymptomatic. Furthermore, the early-phase images of CECT showed shrinkage of the thrombosed DOV without early venous return (**Fig. 3a**), whereas the late-phase images showed inhomogeneous wall enhancement of the DOV (**Fig. 3b**). Further follow-up will be continued.

Discussion

Embolization of pelvic AVMs is technically challenging because the multiplicity, complexity, large size, and fast blood flow of the affected vessels often hamper safe and effective delivery of embolic agents. As pelvic AVMs may also involve arteries supplying the pelvic organs such as the bladder, uterus, intestinal tracts, and gluteal muscles, embolization carries a risk of ischemic organ or nerve damage due to migration or reflux of embolic agents. Thus, the por-

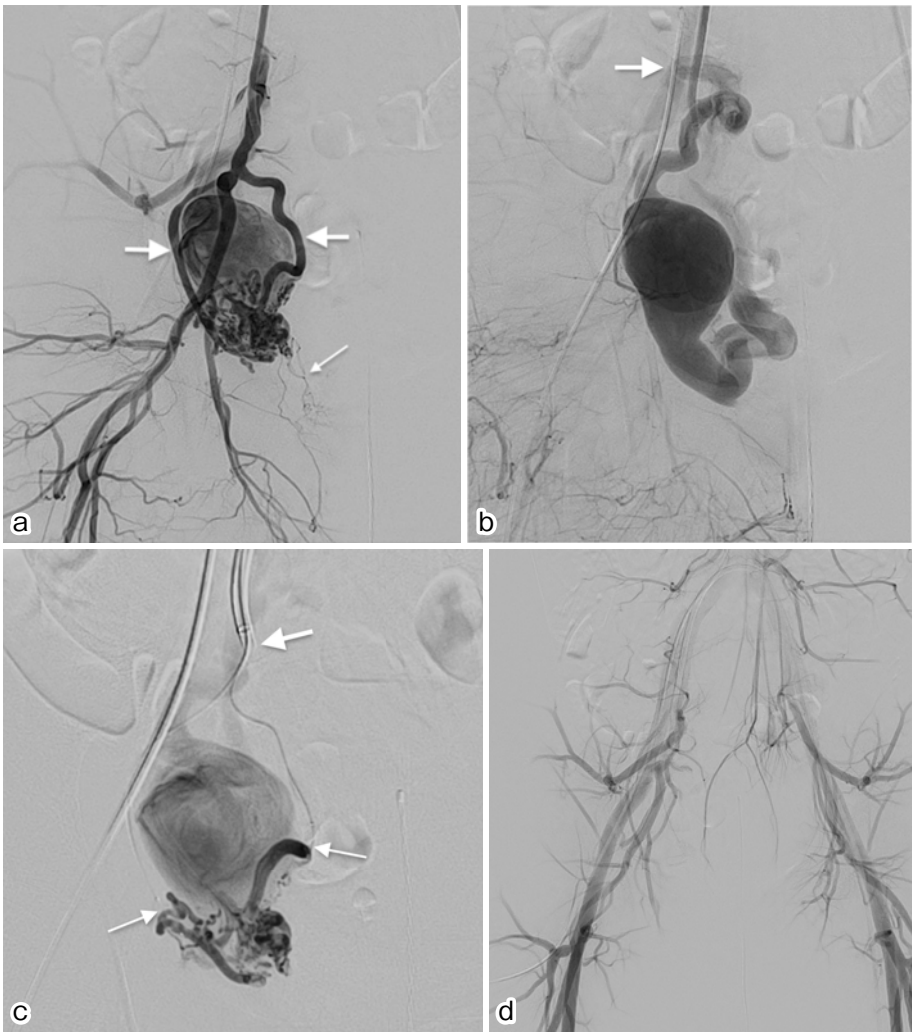

Figure 2.

a. The arterial phase of the right internal iliac arteriogram shows the two main feeding arteries (large arrows) and the cluster of fine vessels shunt to the inferior wall of the DOV. Normal prostatic branches also arise from those fine vessels (small arrow).

b. The venous phase shows the DOV and contiguous tortuous draining veins toward the right internal iliac vein, and the acute angle and narrowing of the orifice are observed (arrow).

c. Simultaneous microballoon-occluded angiography of the two feeding arteries shows blood flow stagnation of the shunt. The prostatic branches are not visualized due to the flow reversal. Arrows indicate the tips of the guiding sheath (large arrow) and each microballoon catheter (small arrows).

d. Post-embolization pelvic angiogram shows complete disappearance of the AVM without any collateral artery.

tion of the AV shunts should be precisely elucidated and selectively embolized, although the angiographic anatomy is often confusing due to overlapping of multiple affected arteries and veins.

In the early reports, transarterial embolization (TAE) was mainly performed for symptom palliation or preoperative devascularization of pelvic AVMs [2, 3, 5]. In the report by Jacobowitz et al., multiple treatment sessions (range, 1 to 11; mean, 2.4) were needed in TAE, but the AVM tended to recur due to collateral development [2]. When many feeding arteries are present, pelvic AVMs are considered difficult to treat with TAE alone.

Since the angiographic classification of AVMs was proposed by Cho et al., embolization techniques have been more tailored based on the vascular anatomy of the AV shunt [4]. Do et al. reported the treatment outcomes of pelvic AVMs, mostly type II AVMs consisting of multiple feeding arteries shunt to a single DOV [1]. The DOV was embolized with an average total of 45 coils using the transvenous or direct puncture approach and an average total of 28 mL of ethanol to eradicate the residual shunt. After the mean 1.9 sessions (range, 1-4) of treatment, 10 of 12 cases were cured and 2 had partial responses. However, TVE may be difficult when angulation or meandering of the draining

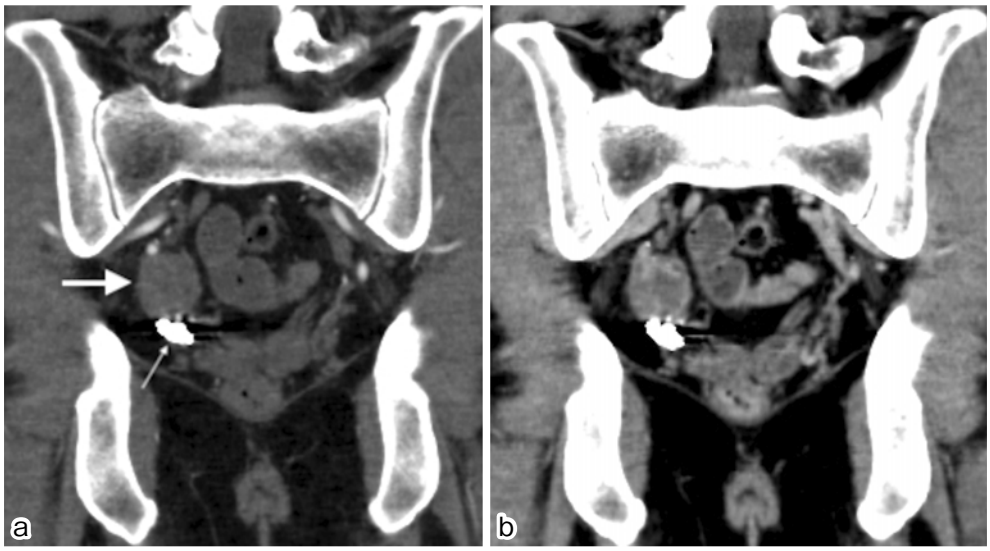

Figure 3.
a. Early-phase coronal MPR image of CECT at 1-year follow-up shows complete thrombosis and shrinkage of the DOV (large arrow). Protective coils of prostatic branches are seen inferior to the DOV (small arrow).
b. The late-phase image shows inhomogeneous wall enhancement of the DOV.

veins is significant. Direct puncture embolization may also carry bleeding risk after needle removal unless shunt occlusion is completed.

Murakami et al. reported a case of Cho's type II pelvic AVM treated only with a single session of TAE [6]. After coil embolization of the superior gluteal artery, inferior gluteal artery, and iliolumbar artery to decrease the number of feeding arteries, a 1:1 mixture of n-BCA and ethiodized oil (50% n-BCA) was injected from the main feeding artery under proximal balloon occlusion. However, as other smaller feeding arteries still remained, more diluted 12.5%-25% n-BCA was additionally injected without proximal balloon occlusion. Koganemaru et al. also reported a case of Cho's type II pelvic AVM treated with a single session of combination of TVE and TAE [7]. As TVE of the DOV using 30% n-BCA was incomplete, TAE using PVA and 20% n-BCA was added to achieve complete shunt occlusion. As demonstrated in these case reports, precise adjustment of concentration and injection control of n-BCA seem difficult, but the microcatheter should be immediately removed after each injection regardless of the completeness of the shunt occlusion. Ethanol embolization has also drawbacks of unpredictability for vessel occlusion and the risk of nontarget embolization due to reflux or migration of less radiopaque ethanol, although there is no need to remove the microcatheter immediately. Therefore, a limited dose of ethanol should be intermittently injected while carefully monitoring the process of blood flow reduction or appearance of normal branches by repeating control angiography. The recommended maximum ethanol dose was 0.14 mL/kg per bolus injection or 0.5-1.0 mL/kg per session to prevent toxicity and hemodynamic collapse [8].

The treatment indication for an asymptomatic AVM like our case remains controversial, and the Japanese clinical practice guidelines for vascular anomalies recommend judging an appropriate timing of treatment for AVMs individually depending on the symptom stage and lesion extent in consideration of the complication risks [9]. As discussed in the guideline, the response rate tends to decrease while the complication rate increases according to disease progression in symptoms or lesion size. Therefore, early intervention can be considered for localized AVMs when radical treatment is likely feasible based on the operator's skill and experience. In our case, the aim of embolization was cure rather than symptom palliation because the lesion was relatively localized without significant symptom. Because TVE was impossible due to anatomical difficulty, whereas the arterial anatomy was favored with a few feeding arteries, endovascular treatment was attempted by TAE alone. Because the two main feeding arteries shared the AV shunts before entering the DOV, only microballoon occlusion of one feeding artery would leave persistent blood flow of the AV shunts from the other feeding artery, which may allow partial escape of ethanol into the DOV. Therefore, simultaneous microballoon occlusion of the two main feeding arteries through a single guiding sheath was efficient in that a highly concentrated ethanol can maximize its sclerosing effect on the cluster of abnormal fine vessels while limiting the ethanol dose in the safe range of 0.20 mL/kg. Microballoon occlusion was also helpful in avoiding nontarget embolization as it reversed the flow direction of the normal prostatic branches. Another concern is a risk of pulmonary embolism after TAE of Cho's type II pelvic AVM without venous coil embolization, particularly when the large draining vein enters straight into the internal iliac vein. However, in our case, the risk was relatively lower considering the acute angle and narrowing of the orifice of the draining vein hampering selective catheter insertion. As a precaution, the patient remained hospital-

ized for observation until CECT was performed 3 days after the treatment. Although complete thrombosis and persistent shrinkage of the DOV was successfully achieved, delayed wall enhancement of the DOV was observed at 1-year follow-up CECT. As it was uncertain if the enhancement was the minimal *de novo* shunt or reactive change, further follow-up will be continued.

Lastly, Kishino et al. reported a case series of six patients with "paravesical space AVM" as a specific subgroup of pelvic AVM. Five cases were successfully treated with TAE using ethanol or n-BCA under venous balloon occlusion or in combination with transvenous embolo-sclerotherapy using 5% ethanolamine oleate, coils, and n-BCA [10]. They also added the literature review of similar cases, including the reports by Murakami et al. and Koganemaru et al. as stated above [6, 7]. The common features of the paravesical space AVMs include occurrence in male patients, dominance of right-side location, and Cho's type II AVM. Our case met all these features and can be categorized into this subgroup.

In conclusion, our case demonstrated the usefulness of simultaneous double microballoon-occluded ethanol embolization in combination with protective coil embolization of normal organ branches to treat a localized pelvic AVM with a few feeding arteries.

Acknowledgement: We appreciate Masanori Inoue, MD, PhD, for his valuable advise on the use of ethanol in treating pelvic AVM.

Conflict of Interest: None

Author Contribution: Guarantors and concepts of the entire case report, K.O.; clinical participation in the treatment plan, procedures, outpatient/inpatient management, and imaging data acquisition, all authors; manuscript editing and literature research, K.O., N.Y.

References

1. Do YS, Kim Y-W, Park KB, et al. Endovascular treatment combined with embolosclerotherapy for pelvic arteriovenous malformations. J Vasc Surg. 2012; 55: 465-471.

2. Jacobowitz GR, Rosen RJ, Rockman CB, et al. Transcatheter embolization of complex pelvic vascular malformations: results and long-term follow-up. J Vasc Surg. 2001; 33: 51-55.

3. Game X, Berlizot P, Hassan T, et al. Congenital pelvic arteriovenous malformation in male patients: a rare cause of urological symptoms and role of embolization. Eur Urol. 2002; 72: 1359-1361.

4. Cho SK, Do YS, Shin SW, et al. Arteriovenous malformations of the body and extremities: analysis of therapeutic outcomes and approaches according to a modified angiographic classification. J Endovasc Ther. 2006; 13: 527-538.

5. Palmaz JC, Newton TH, Reuter SR, Bookstein JJ. Particulate intraarterial embolization in pelvic arteriovenous malformations. AJR Am J Roentgenol. 1981; 137: 117-122.

6. Murakami K, Yamada T, Kumano R, Nakajima Y. Pelvic arteriovenous malformation treated by transarterial glue embolisation combining proximal balloon occlusion and devascularisation of multiple feeding arteries. BMJ Case Rep. 2014; bcr2013203492.

7. Koganemal M, Abe T, Iwamoto R, Suenaga M, Matsuoka K, Hayabuchi N. Pelvic arteriovenous malformation treated by superselective transcatheter venous and arterial embolization. Jpn J Radiol. 2012; 30: 526-529.

8. Shin BS, Do YS, Cho HS, et al. Effects of repeat bolus ethanol injections on cardiopulmonary hemodynamic changes during embolotherapy of arteriovenous malformations of the extremities. J Vasc Interv Radiol. 2010; 21: 81-89.

9. Mimura H, Akita S, Fujino A, et al. Japanese clinical practice guidelines for vascular anomalies 2017. Jpn J Radiol. 2020; 38: 287-342.

10. Kishino M, Nishida K, Kimura K, et al. Paravesical space arteriovenous malformation as a specific subgroup of pelvic vascular anomaly: a case series and review of literature. Jpn J Radiol. 2022; 38: 434-439.