Venous Rupture Following Transcatheter Arterial Embolization for Inferior Mesenteric Type II Arteriovenous Malformation

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Abstract

We treated a 64-year-old man who had an inferior mesenteric arteriovenous malformation with multiple shunts. As multiple varicosities in the draining vein became enlarged, two dilated shunts on the superior rectal and sigmoid colon arteries were coil embolized. Two days after embolization, a varicosity near the shunt (65 mm diameter) ruptured, causing intra-abdominal hemorrhage and surgical hemostasis. There were thrombi in the ruptured varicosity and its draining vein. The likely cause was a pressure increase in the incompletely thrombosed varicosity due to shunt blood flow from the remaining shunts after embolization.

Key words: arteriovenous malformation, transcatheter arterial embolization, hemorrhage, inferior mesenteric artery

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Introduction

Hemorrhage from an arteriovenous malformation (AVM) is a known complication of AVM [1]. Hemorrhage frequency depends on the characteristics of the AVM, such as lesion size. With brain AVM, hemorrhage reportedly occurs in about 2%-4% of untreated cases [2]. Endovascular embolization, microsurgery, and radiosurgery have been used to prevent hemorrhage [1, 3, 4], but no comprehensive reports have clarified the treatment for pelvic AVM. Even after embolization, rare hemorrhage has occurred in the cerebrovascular field [4-7]. However, this has not been reported in pelvic AVM.

We report a case in which massive hemorrhage occurred after transcatheter arterial embolization (TAE) for an incidentally discovered inferior mesenteric AVM.

Case Report

Our institutional review board approved this retrospective case report and waived the requirement to obtain the patient's informed consent for inclusion in this study since the patient had consented in advance.

The patient was a 64-year-old man. Computed tomography (CT) revealed a mass in the pelvis as part of a detailed examination for fever. He had similar findings on CT three years earlier, which were diagnosed as varicose veins. However, the mass had enlarged, so he underwent contrast-enhanced CT (**Fig. 1a**), which showed AVM. The inferior mesenteric artery was the feeding artery, and the marginal vein was the draining vein. The mass involved multiple varicosities in the draining vein, with 24, 65, and 46 mm sizes. Apart from the inferior mesenteric AVM, he had several AVMs in the pelvis and legs, but treatment was considered unnecessary.

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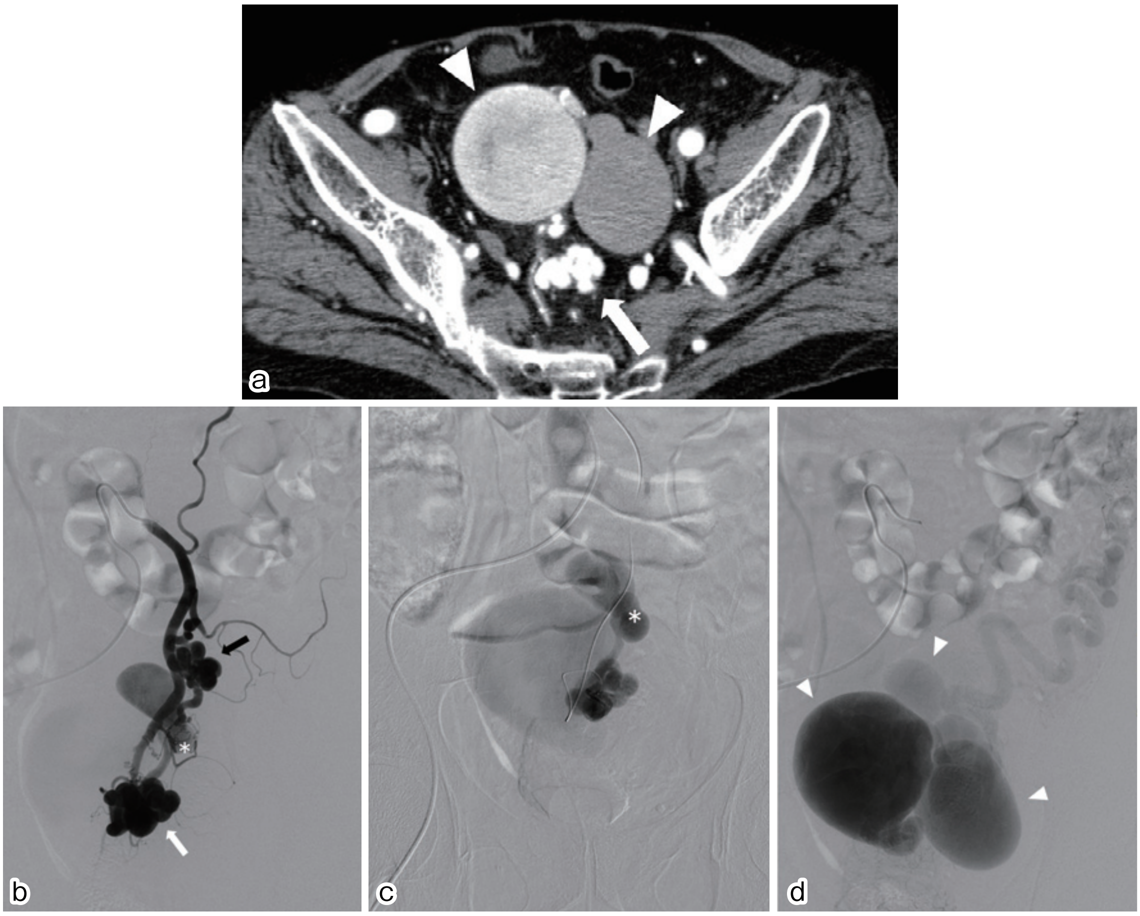

Figure 1. Computed tomography (CT) shows a pelvic mass in a 64-year-old man.
a) Arterial-phase contrast-enhanced CT. An enhanced shunt (white arrow) is visible in the pelvis. Varicosities (white arrowheads) are in the draining vein.
b) Pretreatment of inferior mesenteric arterial-phase arteriography. Many shunts are in the inferior mesenteric artery. A shunt (black arrow) continuous with the sigmoid artery and a shunt (white arrow) continuous with the superior rectal artery appear clearly enlarged. Dominant venous sac (asterisk).
c) Selective angiography. Microcatheter advanced to the feeding artery to enlarged shunt in the superior rectal artery. Dominant venous sac (asterisk).
d) Inferior mesenteric venous-phase arteriography. Multiple varicosities (white arrowheads) have formed in the draining vein.

The patient had a history of brain hemorrhage, hypertension, pulmonary embolism, deep venous thrombosis, and cellulitis.

The largest varicosity had enlarged 1.6-fold on the axial section compared with three years earlier, suggesting a risk of rupture. TAE was planned as treatment. First, angiography evaluated the blood flow. Many shunts were seen in the inferior mesenteric artery (**Fig. 1b**). These shunts were continuous to a single dominant venous sac. Of these, two shunts with the sigmoid and superior rectal arteries as feeding arteries appeared markedly enlarged and produced large shunt flow (**Fig. 1c**). The varicosities visualized on these draining veins corresponded to the varicosities identified on CT (**Fig. 1d**). According to Cho's classification, the angiographic findings showed that the AVM was diagnosed as type II [3]. Shunt blood flowed into the marginal vein along the descending colon and finally drained into the superior mesenteric vein via the middle colic vein. The inferior mesenteric vein was not visualized on CT. TAE was performed two separate times for the two enlarged shunts. In the first TAE, coil embolization was performed for the enlarged shunt in the superior rectal artery using ten coils (AZUR CX18 Detachable and Azur D-18 Helical Hydro Coil; Terumo Co., Tokyo, Japan). Two weeks later, a second TAE was performed for the enlarged shunt in the sigmoid artery using nine Hydro Coils. Shunt flow decreased after these two embolizations, and blood flow slowed in the varicosities in the draining vein (**Fig. 2a** and **Fig. 2b**). Treatment concluded with the expectation that thrombosis would occur within the varicosities and that rapid enlargement of the varicosity could also be avoided. However, small shunts in many locations were not embolized, and some degree of

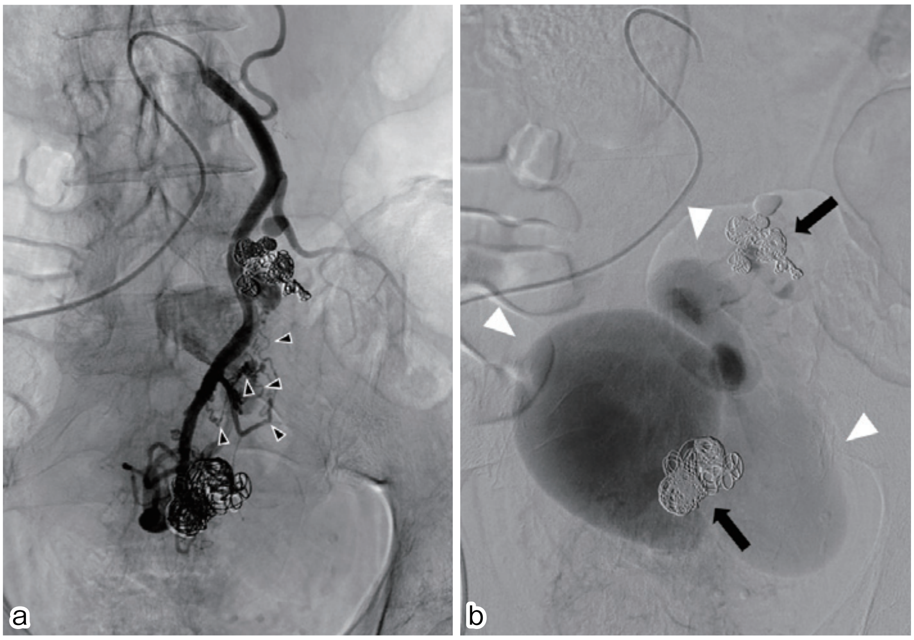

Figure 2. Inferior mesenteric arteriography after second transcatheter arterial embolization (TAE).
a) Arterial phase. The two large shunts have been embolized and blood flow is no longer evident, but remnant shunt flow is seen in small shunts (black arrowheads) that have not been embolized.
b) Venous phase. Varicosities (white arrowheads) in the draining vein via small shunts are visualized. Coils are placed in the two large shunts (black arrows).

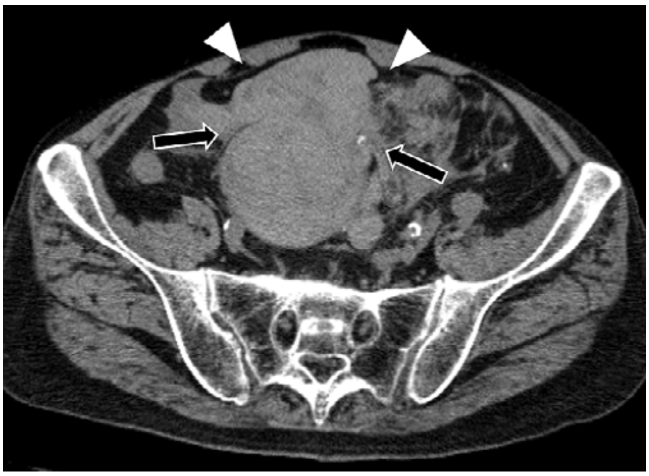

Figure 3. Computed tomography (CT) once the patient's blood pressure decreased two days after embolization.
Rupture of the largest varicosity (black arrows) is visible. A hyperdense clot continues from inside to outside varicosity (white arrowheads).

shunt flow remained.

Two days after the second TAE, the patient experienced vomiting, lower abdominal pain, and abdominal fullness, and systolic blood pressure dropped to 80～89 mmHg. CT was performed to search for the cause. Hemorrhagic ascites and a hyperdense clot within the largest varicosity continued outside the varicosities, indicating varicosity rupture (**Fig. 3**). A clot occluded varicosity situated proximal to ruptured varicosity. During emergency surgery, ruptured varicosity appeared in the draining vein of the AVM. The clot was apparent within the ruptured varicosities in the vessel distal to the varicosities. The ruptured varicosity was sutured. CT performed one month after surgery confirmed that all varicosities had thrombosed and shrunk. No recurrent bleeding occurred within 24 months after embolization. Thrombosis in the varicosities was maintained, and all varicosities had reduced in size.

Discussion

No comprehensive investigations of inferior mesenteric AVM have been reported, with descriptions limited to case reports [8, 9]. They reported performing surgical excision or embolization due to the development of symptoms of diarrhea, hematochezia, and abdominal pain. The patient had no subjective symptoms in the present case, but CT revealed enlarged varicosities. TAE was performed due to the risk of rupture. By embolizing the enlarged shunts, shunt flow decreased, mitigating the risks of the enlargement and rupture of varicosities. However, in the present case, varicosity rupture occurred. The post-rupture CT image and surgical findings showed the largest varicosity near the shunt ruptured, even though thrombosis had occurred in the varicosities and draining vein from the decreased shunt flow with the second TAE. In type II AVM, embolization of the dominant venous sac is considered a priority [3]. However, in this case, it was considered difficult to reach the dominant venous sac by transvenous approach or direct percutaneous puncture, and embolization of the inflow artery was performed. Therefore, blood flow to the dominant venous sac was reduced but remained.

The cause of rupture may have been as follows: with the closure of the enlarged shunts at two sites, the draining vein was thrombosed, but shunt flow continued from the remaining small shunts. This remaining shunt flow seemed to have caused elevated pressure in the incompletely thrombosed varicosities, which ruptured. Another possibility is that the varicosities ruptured due to a partial thrombus in the draining vein and then migrated to the hepatic side, blocking the varicosity outflow portion. In the case of brain AVM, hemorrhage reportedly occurs when, after the draining vein is blocked with clot from embolization and stasis occurs in the draining vein from the remaining shunt flow, a state develops in which venous pressure rises near the feeding artery side [5].

Since this case was a type II AVM, a rupture could have been avoided if the dominant venous sac had been embolized. Direct puncture of the dominant venous sac by an open abdominal approach, called a hybrid approach, or percutaneous puncture of the dilated outflow vein and transcatheter access to the sac may also have been possible if the sac could not be embolized. Additional embolization should also be considered when incomplete thrombosis of the draining vein and residual shunt flow appears. Administration of heparin or anticoagulant during or after embolization also needs to be considered to prevent thrombosis occurrence in the outflow vein.

In this case, after embolization of an inflow artery in a type II AVM, shunt blood flow remained, and the outflow vein ruptured.

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IRB: The study protocols for this retrospective analysis were approved by our institutional review board.

Informed Consent: Written, informed consent was obtained from the patient before publication of this case report. A copy of the written consent is available upon request.

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References

1. Feghali J, Huang J. Updates in arteriovenous malformation management: the post-ARUBA era. Stroke Vasc Neurol. 2020; 5: 34-39.
2. Stefani MA, Sgarabotto Ribeiro D, Mohr JP. Grades of brain arteriovenous malformations and risk of hemorrhage and death. Ann Clin Transl Neurol. 2019; 6: 508-514.
3. 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.
4. Lee KW, Lin YC, Lo CP, Liu CC. Intracerebral hemorrhage following endovascular embolization of brain arteriovenous malformation with a combination of Onyx and n-butyl cyanoacrylate: a case report. Clin Imaging. 2012; 36: 375-378.
5. Purdy PD, Batjer HH, Samson D. Management of hemorrhagic complications from preoperative embolization of arteriovenous malformations. J Neurosurg. 1991; 74: 205-211.
6. Jordan JA, Llibre JC, Vazquez F, Rodriguez R, Prince JA, Ugarte JC. Predictors of hemorrhagic complications from endovascular treatment of cerebral arteriovenous malformations. Interv Neuroradiol. 2014; 20: 74-82.
7. Spetzler RF, Wilson CB, Weinstein P, Mehdorn M, Townsend J, Telles D. Normal perfusion pressure breakthrough theory. Clin Neurosurg. 1978; 25: 651-672.
8. Otani Y, Okabayashi T, Shibuya Y, et al. A surgical case of inferior mesenteric arteriovenous malformation. Surg Technol Int. 2018; 33: 101-104.
9. Amir P, Nolan H, Farnoosh S, Johnathan O. Detection and intervention of clinically masquerading inferior mesenteric artery AVMs. Case Rep Radiol. 2021; 2021: 8854806.