Targeted Transarterial Embolization for Treatment of a Symptomatic Venous Varix in the Draining Vein of an Arteriovenous Malformation: Case Report and Literature Review

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Abstract

Background Venous varices in the draining vein of arteriovenous malformations (AVMs) can result in compression symptoms. This condition is extremely rare, and its treatments and long-term outcomes are unresolved.

Methods Herein, we describe the treatment of a thrombosed venous varix in a draining vein and review the relevant literature.

Results The patient presented with progressive right-sided hemiparesis and aphasia. Magnetic resonance imaging revealed flow void accumulation from the corpus callosum to the left ventricle and a 30-mm mass in the left putamen. The patient underwent targeted transarterial embolization to reduce the blood flow to the venous varix and relieve the neurologic symptoms. The patient had recovered completely from the right hemiparesis and aphasia 4 years after treatment, with a modified Rankin scale score of 0.

Conclusion Targeted transarterial embolization for symptomatic venous varix is a palliative treatment that may improve long-term functional outcomes.

Keywords
- arteriovenous malformation
- venous varix
- draining vein
- targeted transarterial embolization

Introduction

High-grade arteriovenous malformations (AVMs) have a high rupture rate and a poor prognosis.[1,2] The treatment of high-grade AVMs is challenging; multimodality treatment is often used, but the cure rate is low and the incidence of complications is high.[3] Given their high rupture rate, high-grade AVMs should be treated with caution. Targeted embolization may be performed as palliative therapy in some cases.[4]

Unruptured high-grade AVMs are symptomatic in 47.3% of cases, likely owing to the volume and location of the nidus; common symptoms include headaches, epilepsy, and focal neurologic deficits.[5,6] Symptomatic lesions must be treated according to their respective causes, and the treatment method and approach vary according to the vascular architecture of the lesion. Venous varices associated with AVMs can cause symptomatic mass effects.[7,8] Herein, we report the case of a patient with a large high-grade AVM with a

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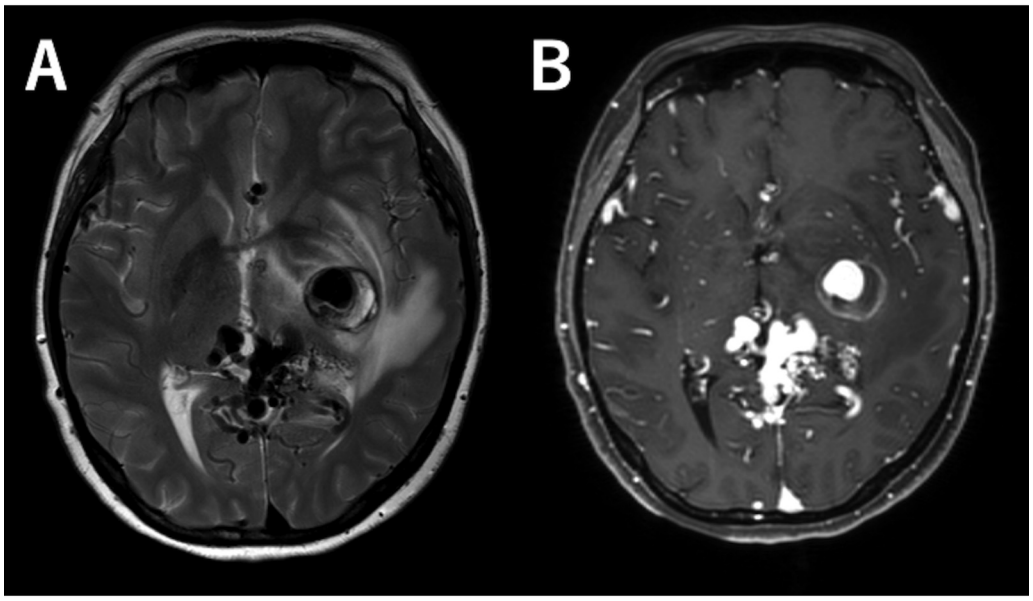

Fig. 1 Magnetic resonance image acquired at initial examination. (**A**) Preoperative contrast-enhanced T2-weighted magnetic resonance imaging of the head shows a 30-mm low-intensity area in the left basal ganglia and clusters of cerebral blood vessels showing T2 low-intensity area mainly in the left corpus callosum. In addition, a T2 high-intensity area is observed around the aneurysm. (**B**) A T1 gadolinium contrast-enhanced image shows a contrast-enhanced area within the venous varix, indicating the presence of a thrombus within the venous varix.

symptomatic thrombosed venous varix in the draining vein. The patient underwent targeted transarterial embolization (TAE) with a good long-term prognosis. AVMs with a venous varix in the draining vein are extremely rare, and previous cases are reviewed here.

Patient

A 43-year-old woman presented with progressive right hemiparesis persisting for 2 months. Axial T2-weighted magnetic resonance imaging (MRI) revealed flow void accumulation extending from the corpus callosum to the left ventricle. A 30-mm thrombosed venous varix was observed in the left basal vein, one of the draining veins. Cerebral edema was observed in the area surrounding the thrombosed venous varix, as well as in the left midbrain, pons, and superior cerebellar peduncle (►**Fig. 1**).

The patient was diagnosed with AVM and referred to our hospital for further investigation and treatment. Physical examination revealed mild motor aphasia, sensory disturbance, and paralysis of the right upper and lower limbs, with a manual muscle strength test score of 3. Cerebral angiography revealed that the bilateral pericallosal, right parieto-occipital, left medial posterior choroidal, and bilateral splenic arteries were the major feeding arteries. The nidus of the AVM was 65 mm in size. The internal cerebral vein and basilar vein of Rosenthal were markedly dilated, as was part of the left deep middle cerebral vein (22 mm; ►**Fig. 2**). The patient was diagnosed with a Spetzler–Martin grade V AVM. The thrombosed venous varix and perifocal edema were considered to be the causes of the neurologic symptoms.

Because curative treatments were not possible, palliative TAE was performed to reduce the blood flow and thereby shrink the venous varix. The first TAE targeted the feeding arteries through the bilateral posterior cerebral artery, which had the highest blood flow to the varix. Under general anesthesia, we successively inserted a 90-cm, 5-Fr FUBUKI guiding sheath (Asahi Intecc, Aichi, Japan) into the right femoral artery; the guiding sheath into the left vertebral artery using a coaxial system after heparinization; a 120-cm, 4.2-Fr FUBUKI guiding catheter (Asahi Intecc) into the basilar artery as a distal access catheter; and a Marathon microcatheter (Medtronic, Minneapolis, Minnesota, United States) into the feeding arteries. The right parieto-occipital, bilateral splenial, and left medial posterior choroidal arteries were embolized using n-butyl-2-cyanoacrylate (NBCA). Blood flow to the venous varix was reduced at the end of the first embolization, and the neurologic symptoms had lessened 1 month thereafter.

The second TAE was performed approximately 1 month after the first TAE; the left medial posterior choroidal and left splenial arteries were embolized using NBCA. Subsequently the right superior and inferior internal parietal arteries were embolized using NBCA (third TAE), and the left pericallosal artery and left superior and inferior internal parietal arteries were embolized using platinum coils and NBCA (fourth TAE).

A gradual reduction in the size of the thrombosed venous varix was observed with embolization (►**Fig. 3**). The pareses of the right upper and lower limbs and aphasia had significantly resolved after the fourth TAE. Annual MRI performed after embolization showed no enlargement of the venous varix or perifocal edema (►**Fig. 4**). The patient had recovered

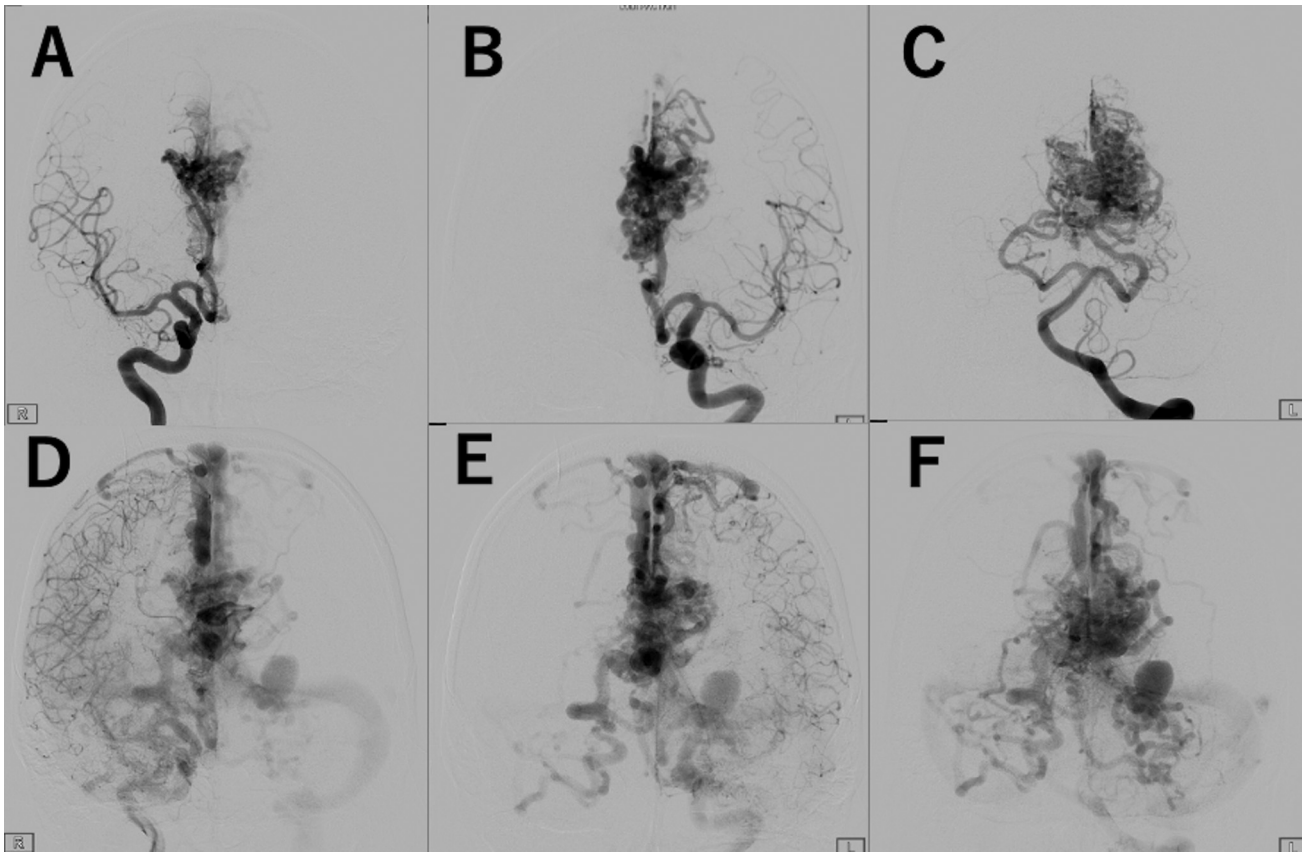

Fig. 2 Preoperative cerebral angiography. Frontal view of (**A**) left internal carotid artery angiography (ICAG), (**B**) right ICAG, and (**C**) left vertebral angiography (VAG) in the arterial phase. The venous varix appears in the venous phase of the (**D**) left ICAG, (**E**) right ICAG, and (**F**) left VAG.

completely from the right hemiparesis and aphasia 4 years after treatment, with a modified Rankin scale score of 0.

Discussion

A venous varix in the nidus of an AVM is common, whereas a symptomatic venous varix in the draining vein is extremely rare. Among the eight previously reported cases of venous varices in the draining vein, only four cases had neurologic symptoms, as did our case (►**Table 1**).[2,8–13] High-grade AVMs (Spetzler–Martin grades III–V) were detected in seven of the nine cases, including ours, and all nine patients presented with symptoms of brain tissue compression by the venous varix. Six patients had a thrombosed venous varix or perifocal edema. Hemorrhage from the venous varix in the draining vein, although usually uncommon, was observed in three patients.[7,14,15] Treatments included TAE (5 patients) and transvenous embolization (TVE; 1 patient; ►**Table 1**); in a few cases, including ours, these treatments were palliative. Palliative treatments are acceptable when complete treatment is difficult owing to the high grade of the AVM. Unlike our study, none of the previous reports assessed long-term treatment outcomes. Long-term treatment outcomes cannot be ignored, as palliative treatment of an AVM may not guarantee long-term treatment effects.

Our patient had perifocal edema in the left putamen due to a thrombosed venous varix in the draining vein of a high-grade AVM and presented with progressive neurologic deficits. Because a complete cure was difficult, palliative TAE was administered. TAE quickly decreases blood flow to a venous varix and, therefore, would likely shrink the thrombosed venous varix and alleviate the symptoms. The AVM in our case had multiple feeding arteries, and the feeding arteries with the highest blood flow to the venous varix were embolized first. The size of the venous varix and volume of the perifocal edema decreased with treatment, and the neurologic symptoms gradually resolved. Owing to the embolization of multiple feeding arteries, no cerebral hemorrhage or deterioration of the neurologic symptoms occurred during the 4 years after treatment; nevertheless, further follow-up is mandatory.

TAE uses liquid embolic agents to decrease the blood flow to the nidus, which is expected to shrink the venous varix. However, owing to the risk of liquid embolic agents migrating to normal vessels, the possibility of embolic complications increases when multiple feeding arteries are targeted. To prevent migration, selective perinidal or intranidal introduction of a microcatheter is important.[16] Additionally, intracranial hemorrhage after TAE is possible and may alter the hemodynamics of the nidus.[4] Because complete prevention of post-TAE hemorrhage is difficult, intranidal aneurysms, which may be the cause of the hemorrhage, need to be embolized. TVE of AVMs and venous varices has been reported.[8,17,18] Although TVE is

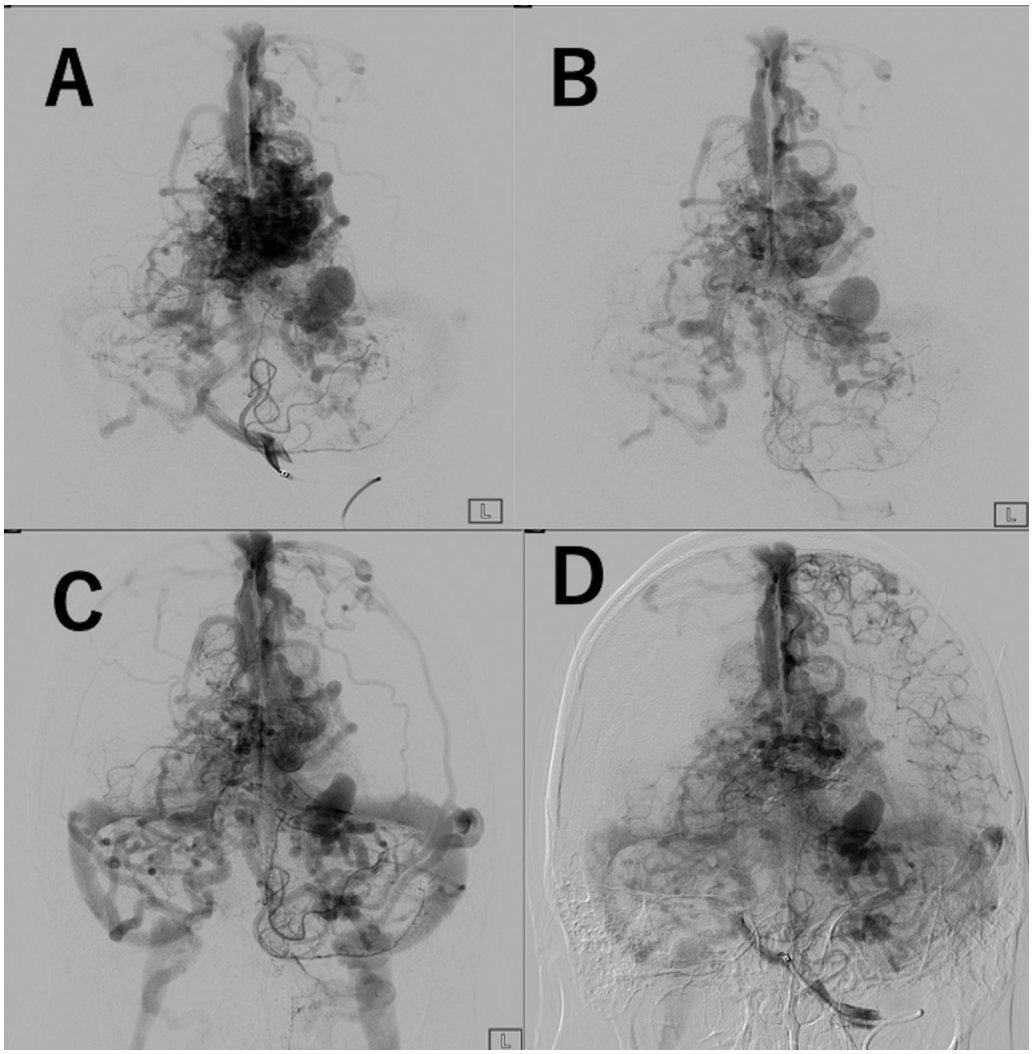

Fig. 3 Changes in the venous varix after each embolization. Changes in the venous varix before and after the (**A**) first, (**B**) second, (**C**) third, and (**D**) fourth embolizations, and radiographs of the embolized material acquired after each embolization. The embolized venous varices shrank after each treatment. The second embolization was performed 1 month after the first embolization, the third embolization was performed 2 months after the second embolization, and the fourth embolization was performed 6 months after the third embolization.

originally performed to ensure complete obliteration of an AVM, it can also be applied to treat venous varices. In TVE, coils are placed in the venous varix to promote thrombosis of the varix; thus, a mass effect may occur due to the coils. Whether to perform TAE versus TVE should be comprehensively based on the location of the venous varix, its relationship with the surrounding brain tissue, and the catheter approach.

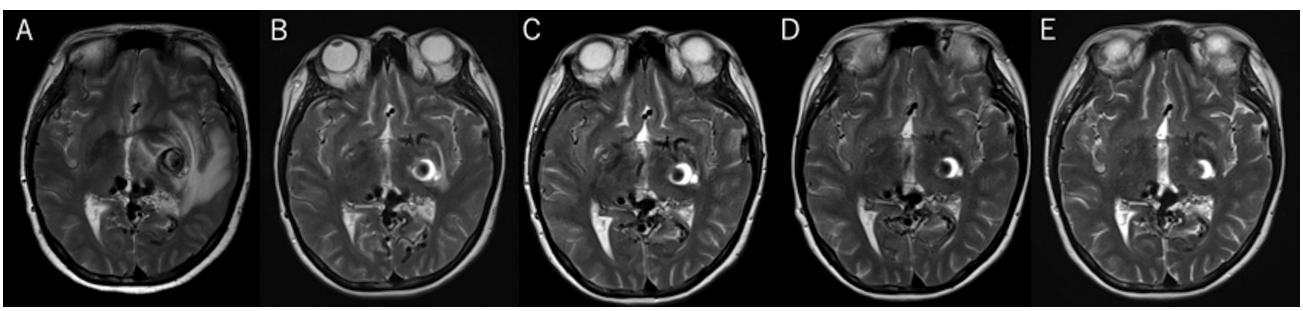

Fig. 4 Magnetic resonance images acquired after the fourth embolization. (**A**) T2-weighted magnetic resonance image acquired after the second embolization shows that the venous varix had decreased in size; however, the surrounding cerebral edema persisted. (**B**) Further decrease in the size of the venous varix was observed after the fourth embolization, and a dramatic reduction in the cerebral edema was also observed. Magnetic resonance images acquired (**C**) 1 year, (**D**) 3 years, and (**E**) 4 years after embolization revealed no enlargement of the venous varix or worsening of cerebral edema.

Table 1 A review of the literature on venous varices in the draining vein

Case	Study	Age (y)	Sex	Location	Spetzler–Martin grade	Symptom	Thrombosis	Edema	Treatment	Complication	Further treatment	Radiologic outcome	Follow-up (mo)
1	Mineura et al[9]	21	M	Cerebellum	IV	Trigeminal neuralgia	–	–	IVR (TAE)	Venous varix	Surgery	Venous varix, resolved	NM
2	Sato et al[10]	52	M	Cerebellum	III	Hemorrhage	–	+	Surgery	No	No	AVM, complete removal	3
3	Kim et al[2]	63	M	Frontal	III	Headache	+	+	Conservative	No	No	Venous varix, resolved	NM
4	Kim et al[2]	60	M	Parietal	III	Convulsion	–	+	IVR	Intracranial hemorrhage	NM	NM	NM
5	Chakraborty et al[11]	63	M	Frontal	I	Hemorrhage	–	–	IVR (TAE)	No	No	AVM and venous varix, complete obliteration	12
6	Rahme et a[12]	69	M	Frontal	III	Hemorrhage	–	–	IVR (TAE) + radiosurgery	Intracranial hemorrhage after radiosurgery	Surgery	AVM and venous varix, complete removal	10
7	Haryu et al[13]	46	M	Temporal	III	Neurologic deficit (aphasia)	+	–	IVR (TAE) + surgery	Postoperative edema	Surgery	AVM, complete removal	5
8	Kushi et al[8]	40	M	Parietal	III	Neurologic deficit (numbness)	+	+	IVR (TVE) + radiosurgery	No	No	Venous varix, resolved	6
9	This study	43	F	Corpus callosum	V	Neurologic deficit (hemiparesis)	+	+	IVR (TAE)	No	No	Venous varix, resolved	48

Abbreviations: AVM, arteriovenous malformation; IVR, interventional radiology; NM, not mentioned; TAE, transarterial embolization; TVE, transvenous embolization.

Conclusion

This report presents a case wherein TAE reduced the size of a venous varix in the draining vein and relieved the neurologic symptoms for at least 4 years. To date, there have been no reports on the long-term efficacy of palliative treatments for AVMs. Patients with AVMs with a venous varix in the draining vein are prone to neurologic deficits. High-grade AVMs are uncommon, and palliative embolization is recommended for symptom resolution.

Ethical Declarations

Informed consent was obtained from the patient. The authors declare that this manuscript does not contain previously published materials and is not under consideration for publication elsewhere.

Funding

None.

Conflict of Interest

None declared.

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