

Imaging

Spontaneous pial single-channel arteriovenous fistulae with angiographically occult small feeding arteries: case report

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Abstract

Background: Pial single-channel arteriovenous (AV) fistulae are usually cured by interrupting only the feeding arteries identified in cerebral angiography.

Case Description: A 27-year-old woman presented after 3 complex partial seizures due to a pial single-channel AV fistula with the varix located in the right temporal lobe. When all feeding arteries identified by preoperative cerebral angiography were temporarily occluded, varix blood flow was confirmed with Doppler ultrasonography. Many small feeding arteries were identified, and the varix was dissected and removed. The postoperative course was uneventful and postoperative cerebral angiography revealed no abnormal vascular lesions.

Conclusion: Pial single-channel AV fistulae are not always cured by interrupting only the feeding arteries identified in cerebral angiography and might require varix removal.

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Keywords:

Arteriovenous fistulae; Doppler ultrasonography; Varix; Occult feeding arteries

1. Introduction

Intracranial pial single-channel arteriovenous (AV) fistulae are rare vascular lesions that are considered distinct from arteriovenous malformations (AVM) [15]. They consist of 1 or more arterial connections to a single venous channel without any intervening nidus of vessels or capillaries. Because of their high-flow nature, they are often associated with a venous varix [1,2,4,8,27]. Various reports have described surgical or endovascular experiences in dealing with these somewhat intimidating lesions [1,2,4,8,10,12,27]. Most authors have recommended that only the feeding arteries identified in cerebral angiography should be interrupted as closely to the fistulae as possible, thereby leaving the varix intact [5,7,10-12,24,28].

We report a patient with spontaneous intracranial pial single-channel AV fistulae treated by surgical resection of the varix. The lesion had many small feeding arteries that were not identified by preoperative cerebral angiography.

Abbreviations: AV, arteriovenous; AVM, arteriovenous malformation; SPECT, single-photo emission computed tomography.

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Fig. 1. Magnetic resonance T₂-weighted image shows abnormal flow voids with perifocal edema in right temporal lobe.

2. Case report

A 27-year-old, otherwise healthy woman was admitted to our hospital after enduring 3 complex partial seizures within 1 year. Magnetic resonance images of the head demonstrated abnormal flow voids with perifocal edema in the right temporal lobe (Fig. 1). Cerebral angiography showed a large aneurysm-like dilation, nourished by tortuous and dilated right middle and posterior cerebral arterial branches that drained into the right sigmoid sinus (Fig. 2). A nidus of AVM was not evident. Brain perfusion SPECT images revealed no regions with decreased perfu-

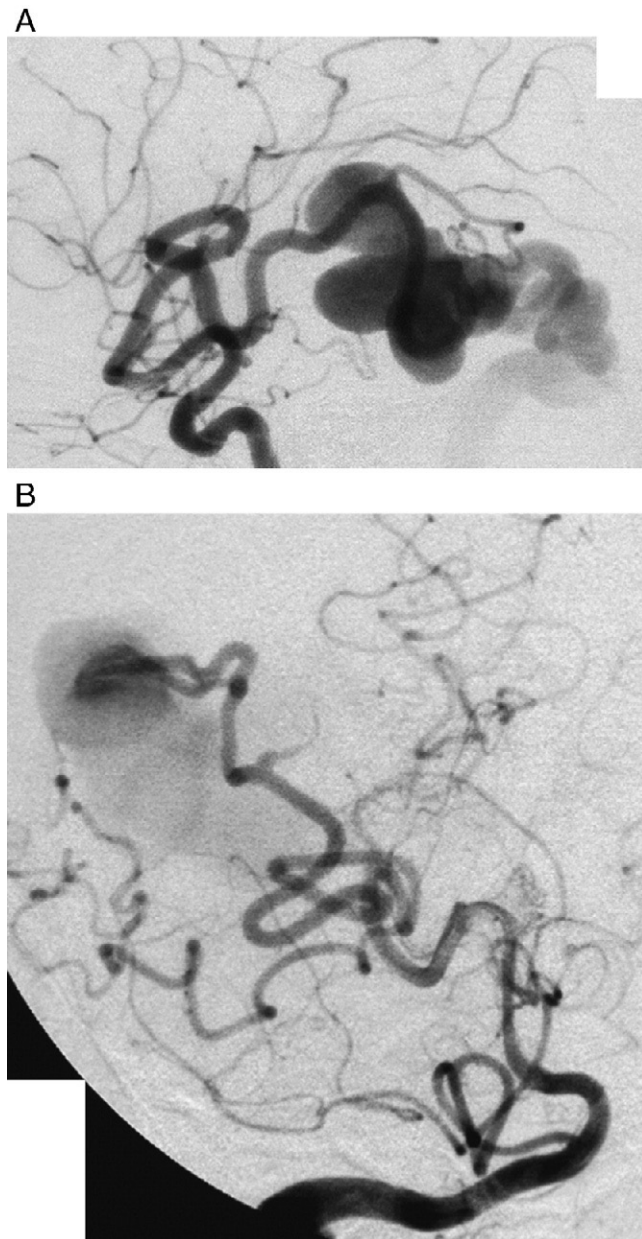


Fig. 2. Lateral view of right carotid angiogram (A) and Towne's view of right vertebral angiogram (B) show single-channel arteriovenous fistula fed by both right middle and posterior cerebral arteries and drained by a variceal vein toward the right sigmoid sinus.

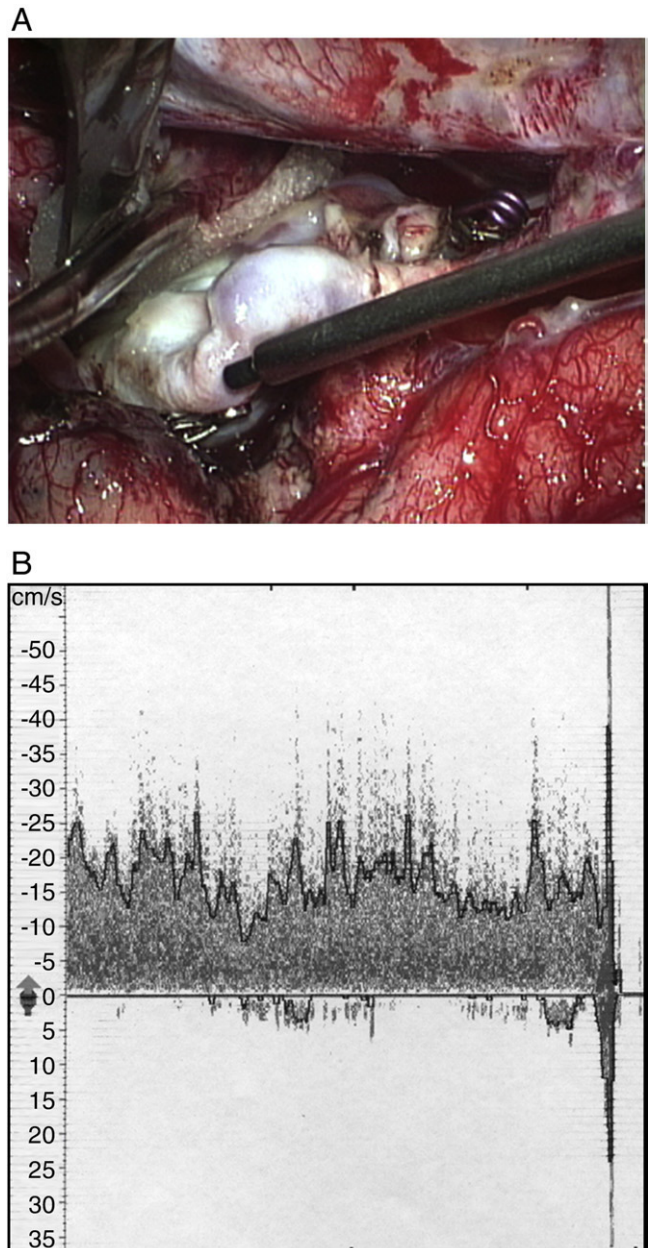


Fig. 3. A: Intraoperative photograph shows Doppler ultrasonographic measurements of blood flow in the varix during temporary occlusion of feeding arteries with aneurysm clips. B: Doppler ultrasonography velocity waveforms show arterial supply of blood into the varix.

sion and vasoreactivity to acetazolamide was limited around the lesion [22].

The patient underwent a right temporoparietooccipital craniotomy. Opening of the dura revealed 3 dilated feeding arteries connected to a huge varix at different points. We temporarily occluded the 3 feeding arteries immediately proximal to their entrance into the varix by using aneurysm clips, and confirmed the presence of varix blood flow by Doppler ultrasonography using a TC2020 (Pioneer, Uberlingen, Germany) and a flexible 20-megahertz probe with a diameter of 1.5 millimeters in the pulse wave mode (Fig. 3). After coagulation and the 3 dilated feeding arteries were cut,

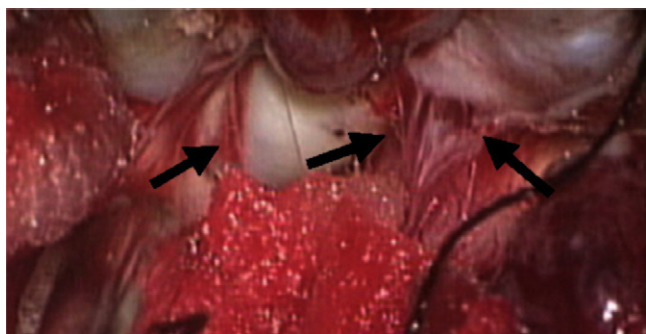


Fig. 4. Intraoperative photograph shows many small feeding arteries into the varix (arrows).

the varix was dissected from surrounding brain tissues with a step-by-step coagulation technique. Thereafter, many small feeding arteries were identified and coagulated (Fig. 4). The varix was dissected and immediately cut before draining into the right sigmoid sinus and removed. A propofol coma with controlled hypotension was initiated immediately after surgery and maintained for the next 24 hours. A computed tomography scan and brain perfusion SPECT imaging performed on the first postoperative day revealed a minor hemorrhage in the right temporal lobe and no hyperperfused regions, respectively. Neurologic deficits were not evident after propofol coma was withdrawn. The subsequent clinical course remained uneventful. Cerebral angiography on the 20th postoperative day revealed that the patient was free from abnormal vascular lesions (Fig. 5).

3. Discussion

The natural history of pial single-channel AV fistulae is unknown. Literature describing AVM indicates that single-vein drainage contributes to the risk of AVM hemorrhage [12,20] as well as small AVM [13,25], both of which are characteristics of pial AV fistulae [12]. Flow and perfusion pressure through a pial AV fistula is certainly high. Impaired venous drainage in AVMs has been implicated in the risk of hemorrhage [20], but whether venous varices accompanied by pial AV fistulae are associated with an increased risk of hemorrhage remains unclear. Magnetic resonance images of our patient demonstrated brain edema surrounding a varix, which might have caused the 3 episodes of complex partial seizures. Thus, we considered that the varix was actively growing and should be surgically treated.

Most authors have recommended that only the feeding arteries identified by cerebral angiography should be interrupted as closely as possible to pial single-channel AV fistulae through craniotomy or by endovascular means, leaving the varix intact [5,7,10–12,24,28]. However, 1 report has described that after microsurgically occluding a feeding artery near a fistula, the varix was punctured and an arterial supply of blood into the varix persisted [1]. In addition, postoperative cerebral angiography revealed a lesion filled

via small arterial branches that were not identified by preoperative cerebral angiography. Ultimately, the patient underwent a second craniotomy, and the varix was dissected from the cortex and removed en bloc. Another report describes that despite endovascular and subsequent microsurgical occlusion of the arteries feeding AV fistulae, postoperative cerebral angiography demonstrates incomplete occlusion of the lesion [27].

We found that when the feeding arteries identified on preoperative cerebral angiography were occluded just

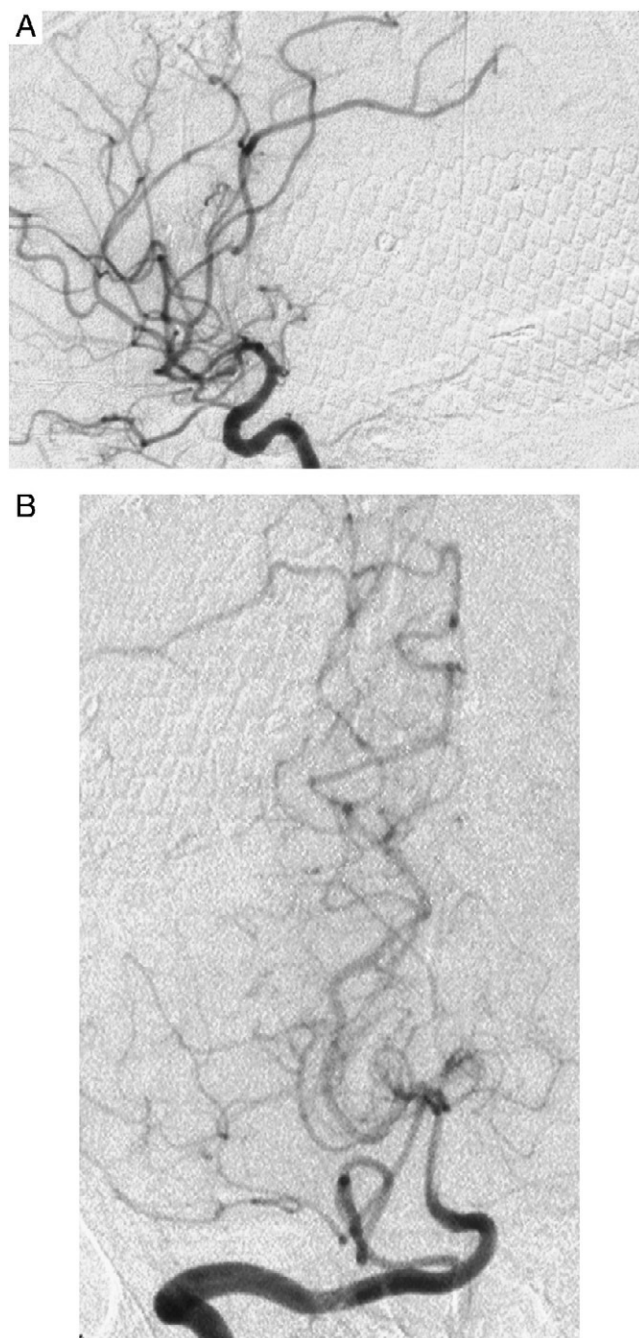


Fig. 5. Lateral view of right carotid angiogram (A) and Towne's view of right vertebral angiogram (B) at 18 days after surgery show absence of lesion and normalization of right middle and posterior cerebral arterial diameters.

proximal to their entrance into the varix, the varix was still supplied with blood. In addition, many small feeding arteries that were invisible on preoperative cerebral angiograms were identified intraoperatively. These findings corresponded with those of the reported cases, suggesting that pial single-channel AV fistulae are not always cured with interruption of only the feeding arteries identified in cerebral angiography and might require removal of the varix.

Many authors have advocated the use of Doppler ultrasonography during AVM surgery [3,9,14,16-19,21,23,29], because the presence or absence of Doppler signals can determine the completion of AVM resection [6,26]. After feeding arteries were temporarily occluded in our patient, Doppler ultrasonography revealed blood flow to the varix. Thus, intraoperative Doppler ultrasonography might be a useful device for pial single-channel AV fistula surgery.

4. Conclusions

Interrupting only the feeding arteries identified by cerebral angiography does not always cure pial single-channel AV fistulae and thus the varix might require removal. Intraoperative Doppler ultrasonography might be a useful device for pial single-channel AV fistula surgery.

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Commentary

The authors have reported a single case of a pial single-channel AV fistula that remained patent after occlusion of all feeding arteries identified by preoperative cerebral angiography. It is well recognized that only complete interruption