Ischemic stroke in a young adult with a known epileptogenic arteriovenous malformation: illustrative case

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BACKGROUND Brain arteriovenous malformations (AVMs) usually manifest as hemorrhages or seizures. They rarely present with ischemic symptoms, especially in young patients. We present a case of an epileptogenic AVM that led to cerebral infarction due to paradoxical embolic occlusion of the middle cerebral artery (MCA) involving the main feeder of the lesion.

OBSERVATIONS A 35-year-old male had been suffering from AVM-associated epilepsy for 10 years and was scheduled for surgery. He suddenly developed right-sided hemiconvulsions followed by hemiparesis and impaired consciousness. Computed tomography revealed no intracerebral hemorrhage, and symptoms were initially thought to indicate epilepsy and Todd’s palsy. Because of his prolonged symptoms, he underwent magnetic resonance imaging, which revealed a large cerebral infarction due to occlusion of the MCA involving the main feeder of the AVM. The patient underwent AVM resection, and the partially thrombosed nidus was completely removed. Histopathological investigation revealed a fresh thrombus in totally occluded nonarteriosclerotic feeders. He had no atrial fibrillation; however, subsequent transesophageal echocardiography revealed a patent foramen ovale, suggesting a paradoxical embolism.

LESSONS This case serves as a reminder that AVMs can present with considerable variability. Acute cerebral infarction should be considered a possible mechanism of seizures, even in patients with epileptogenic AVM.

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KEYWORDS arteriovenous malformation; cerebral infarction; paradoxical embolism

Brain arteriovenous malformation (AVM) is a congenital malformation associated with the risk of hemorrhage and seizures. AVM causes intracerebral hemorrhage in more than half of patients and symptomatic epilepsy in 20% to 25%.1 In contrast, only 0.07% of patients with AVM experience cerebral infarction.23 Vascular steal has been thought to be the leading cause of ischemic events in patients with AVM. We report a case of an AVM with cerebral infarction caused by occlusion of a major artery involving the main feeder, which was suspected to be a paradoxical embolism.

Illustrative Case

A 35-year-old male with no specific medical or family history had been suffering from AVM-associated epilepsy for 10 years. Two years prior, he had been referred to and was evaluated at our institution after experiencing convulsions in the right upper limb. Digital subtraction angiography (DSA) showed the presence of an AVM in the left parietal lobe, mainly fed by the left middle cerebral artery (MCA) and draining into the transverse sinus (Fig. 1). The AVM was scheduled for surgical removal with preoperative embolization. While waiting for surgery, he was reevaluated for recurrent right-sided hemiconvulsions, followed by hemiparesis and impaired consciousness. Neurological examination on admission revealed a disturbance of consciousness, rated on the Glasgow Coma Scale as 8 (E4M1V3), with global aphasia and right hemiplegia. The deep tendon reflex of the right lower limb was hyperactive in the presence of the Babinski sign. Computed tomography (CT) scanning did not

ABBREVIATIONS AVM = arteriovenous malformation; CT = computed tomography; DSA = digital subtraction angiography; LVO = large vessel occlusion; MCA = middle cerebral artery; MRI = magnetic resonance imaging; PFO = patent foramen ovale.

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reveal intracerebral hemorrhage or early CT signs of acute ischemic stroke. Additionally, there was no increase in the density of the nidus suspected of thrombosis (Fig. 2). Electrocardiography revealed no evidence of atrial fibrillation. The patient was initially diagnosed with recurrent symptomatic epilepsy and Todd’s palsy. However, magnetic resonance imaging (MRI) performed 2 days later demonstrated acute cerebral infarction in the left MCA region (Fig. 3A) and occlusion of the proximal part of the MCA (Fig. 3B). DSA revealed partial recanalization of the left MCA, although the main feeder remained occluded (Fig. 3C and D). The patient gradually regained consciousness, and rehabilitation therapy was initiated during the acute stage. We decided to proceed with resection of the AVM to prevent bleeding; surgery was performed on the 23rd day after onset (Fig. 4A and B) without complications. Histologically, the sections showed a dilated feeder and newly formed thrombus in the lumen of the feeder, which was suspected to be an embolus (Fig. 4C). Postoperative DSA revealed complete AVM resection and major MCA recanalization (Fig. 4D and E). Subsequently, the patient underwent transesophageal echocardiography revealing a patent foramen ovale (PFO). The patient received anticoagulants to prevent repeated paradoxical embolisms and was transferred to another hospital to continue rehabilitation on the 42nd day with a modified Rankin Scale score of 4.

Patient Informed Consent
The necessary patient informed consent was obtained in this study.

Discussion
Observations
Only 0.07% of AVM cases are associated with cerebral infarction, and 0.08% of cerebral infarction cases are caused by AVMs.3 There are very few reports concerning infarctions due to occlusion of the AVM feeder.2,3 Cerebral infarction associated with AVM may result from coincidental cardioembolic and atherothrombotic infarctions,2,4 endothelial damage in high-velocity blood flow, or vascular steal from the normal brain into the low-resistance nidus of the AVM.2,5–7 In addition to large vessel occlusion, lacunar-like infarctions associated with vascular steal have also been reported.2 Previous studies have identified the risk factors for vascular disease in those with occlusion of the main feeder. Oral contraceptive use, dissection, and embolization of the heart or aorta have been suggested as causes of main artery occlusion in younger patients.2

In addition, spontaneous thrombosis of the AVM has been reported to occur in fewer than 1% of cases.9 However, this phenomenon is clinically silent in most cases. In a few instances (<4%), resolution was immediately preceded by hemorrhage.10 There are no reports of infarction resulting from the spontaneous thrombosis of AVM.

In the present case, transesophageal echocardiography was performed to detect the source of the embolism. Based on these findings, the patient was suspected to have a PFO, and a paradoxical embolism was thought to be the cause of the main feeder occlusion. Although no studies have reported patients with brain AVM combined with PFO, it is well known that PFO is associated with ischemic stroke in younger patients.11 Little is known regarding the mechanisms of embolus migration toward the feeder, which could be a coincidence. It is possible that an AVM with a large nidus can attract blood flow, although this has not been systematically studied. Based on these hypotheses, occlusion of the main feeder may easily occur in an AVM with a huge nidus, especially in patients with embolic sources and a PFO.

Some controversy surrounds the resection of AVMs after large vessel occlusion (LVO) and ischemic stroke. The spontaneous recanalization rate after LVO has been shown to be 24.1% within 24 hours and 52.7% after >24 hours,12 suggesting a risk of AVM rupture. Alternatively, the nidus may become completely thrombosed during LVO. Further investigation is warranted to determine the rate of AVM rupture after LVO, including when the main feeder is affected. In the present case, we observed partial recanalization of the MCA on follow-up DSA. The AVM was being supplied by the MCA as well as the anterior cerebral arteries. Additionally, the patient was a young adult with gradually improving symptoms; hence,
we decided to remove the AVM to prevent bleeding, after consulting with the patient and his family.

**Lessons**

This case serves as a reminder of the significant variability observed in AVMs. Seizures are a common symptom in patients with AVM. However, acute cerebral infarction should be considered as a possible mechanism of seizures, even in young patients with epileptogenic AVM. In the present case, the symptoms and radiological findings were consistent with symptomatic epilepsy during initial evaluation, leading to delayed diagnosis and missed opportunities for hyperacute thrombolytic therapy for ischemic stroke. It is crucial to consider immediate MRI if neurological recovery is delayed.

![Image of AVM and related medical images](image-url)
References

Disclosures
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions
Conception and design: Maeda, Kuribara, Yanagawa, Kohyama, Kurita.
Acquisition of data: Kuribara, Yanagawa, Kurita. Analysis and interpretation of data: Kuribara, Yanagawa, Kurita. Critically revising the article: Maeda, Kuribara, Kurita. Reviewed submitted version of manuscript: Maeda, Kuribara, Kohyama, Kurita. Approved the final version of the manuscript on behalf of all authors: Maeda. Administrative/technical/material support: Kuribara, Tsukagoshi, Kohyama, Kurita. Study supervision: Yanagawa, Kurita.

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