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A complete thrombotic large aneurysm at the nonbranching segment of the distal anterior cerebral artery mimicking a cavernous malformation: illustrative case

Yu Hirokawa, MD, Yuhei Michiwaki, MD, PhD, Tatsuya Tanaka, MD, PhD, Masatou Kawashima, MD, PhD, Tomihiro Wakamiya, MD, PhD, Ryohei Sashida, MD, Kazuaki Shimoji, MD, PhD, Eiichi Suehiro, MD, PhD, Keisuke Onoda, MD, PhD, Fumitaka Yamane, MD, PhD, and Akira Matsuno, MD, PhD

Department of Neurosurgery, International University of Health and Welfare, School of Medicine, Narita Hospital, Narita City, Chiba, Japan

BACKGROUND Thrombotic aneurysms at the nonbranching segment of the distal anterior cerebral artery (ACA) are extremely rare and difficult to differentiate from cavernous malformations by radiographic features alone.

OBSERVATIONS Computed tomography and magnetic resonance imaging of a 30-year-old female patient with a chronic headache complaint revealed a 22-mm frontal lobe mass. The mass showed heterogeneous mixed intensity and hemosiderin deposits on magnetic resonance images. It was not visualized by conventional angiography, indicating that the mass and ACA/other vessels were not connected. The patient was preoperatively diagnosed with a cavernous malformation. However, during resection, the mass surface was white and smooth, different from a cavernous malformation. Although the mass was adherent to the pericallosal artery branch, no luminal continuity was observed. After detachment, the mass was completely resected. Pathological and immunohistochemical findings indicated a vessel wall and interior thrombus. The patient was rediagnosed with a thrombotic aneurysm at the distal ACA nonbranching segment and discharged 10 days postsurgery without neurological deficits.

LESSONS Because radiographic findings of thrombotic aneurysm and cavernous malformation are similar, mass lesions in contact with major arteries should be differentiated as thrombotic aneurysms, even when the artery lumen appears disconnected from the mass.

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KEYWORDS thrombotic aneurysm; distal anterior cerebral artery; nonbranching; cavernous malformation; surgery

Saccular aneurysms of the nonbranching segment of the distal anterior cerebral artery (ACA) are extremely rare.[1] Moreover, thrombotic aneurysms at this segment are even more rare. Thrombotic aneurysms are occasionally difficult to differentiate from cavernous malformations, intracranial hematomas, or brain tumors by radiographic features alone.[2] In this report, we describe a case involving the resection of a complete thrombotic large aneurysm at the nonbranching segment of the distal ACA that was diagnosed as a cavernous malformation preoperatively.

Illustrative Case

Presentation and Neuroimaging Findings

A 30-year-old female presented to a local clinic with a complaint of chronic headache. Magnetic resonance imaging (MRI) revealed a mass lesion in the right frontal lobe, and the patient was referred to our hospital for further examination and treatment. The patient had a medical history of mental illness and chronic headache 10 years prior but had no history of infections or head trauma. On admission, she presented with no abnormal neurological findings other than a forehead headache. Plain computed tomography (CT) revealed a mass lesion with high density in the periphery and low density in the interior of the right frontal lobe (Fig. 1 left). The mass lesion did not show any apparent enhancement on contrast-enhanced CT images (Fig. 1 right). T1- and T2-weighted images exhibited a mass lesion with a maximum diameter of 22 mm and heterogeneous mixed intensity (Fig. 2A and B). The mass was surrounded by hemosiderin deposits, which showed hypointensity on T2*-weighted images (Fig. 2C). Enhanced MR images in the sagittal view demonstrated that the nonenhanced mass was

ABBREVIATIONS ACA = anterior cerebral artery; CT = computed tomography; MRI = magnetic resonance imaging.

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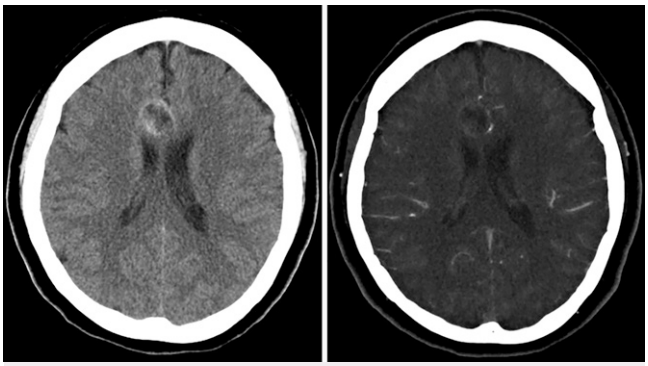

FIG. 1. Left: CT image shows a mass lesion with high density in the periphery and low density in the interior of the right frontal lobe. **Right:** The mass lesion shows no apparent enhancement on a contrast-enhanced CT image.

located above the corpus callosum (Fig. 2D). The lumen of the pericallosal artery and branches of the ACA were not connected to the mass (Fig. 2E). The mass was not visualized by conventional angiography, indicating a lack of connection between the mass and the ACA or other vessels (Fig. 3). Based on these findings, the patient was preoperatively diagnosed with a cavernous malformation. Resection was performed because the patient was young, the mass had evidence of hemorrhage as shown in the T2*-weighted images, and the pathology needed confirmation.

Surgical Findings

Resection of the mass was performed via an interhemispheric approach. Hemosiderin deposition was observed on the mass surface, which was white and smooth, and did not pulsate (Fig. 4A). This appearance was quite different from that of a cavernous malformation, and it appeared to correspond to a vessel wall. Although the mass was adherent to a branch of the pericallosal artery, no luminal continuity was observed (Fig. 4B). After the mass was detached from the artery, discontinuity with the arterial wall occurred, and complete resection was achieved (Fig. 4C). The pericallosal artery and branches were intact.

Postoperative examinations revealed the complete absence of the mass, no adverse events were reported, and the patient was discharged 10 days after surgery with no neurological deficits.

Pathological Findings

Pathological examination of the mass primarily demonstrated collagen fibers, and Elastica van Gieson staining revealed no elastic fibers in the tissue specimen (Fig. 5A and B). Immunohistochemically, α-smooth muscle actin–positive (Fig. 5C) and desmin-positive cells (Fig. 5D) were found on the luminal side, indicating smooth muscle and collagen fibers in the wall. These findings were consistent with the structure of the media and adventitia of a blood vessel wall without the endothelium. There was a thrombus inside the wall, and no findings of cavernous malformation were observed. These findings confirmed that the mass was not a cavernous malformation but rather a thrombotic aneurysm.

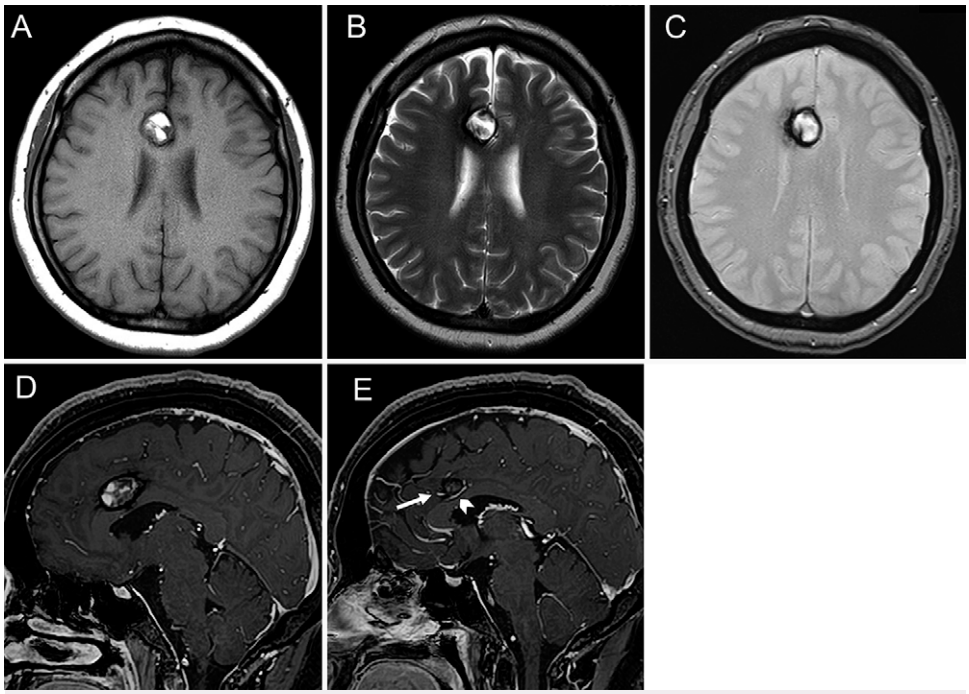

FIG. 2. Axial T1-weighted (**A**) and T2-weighted (**B**) MR images revealing the mass lesion with a maximum diameter of 22 mm with heterogeneous mixed intensity. Axial T2*-weighted image (**C**) shows the mass surrounded by hemosiderin deposits visible as hypo-intensity. Enhanced MR images in the sagittal view (**D and E**) demonstrate a nonenhanced mass located above the corpus callosum (**D**). No connection is observed between the lumen of the pericallosal artery (*arrowhead*) and the branch (*arrow*, **E**) of the anterior cerebral artery and the mass.

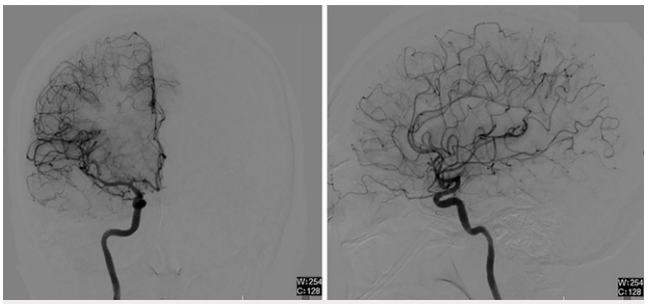

FIG. 3. Anteroposterior (**left**) and lateral (**right**) right internal carotid artery angiographs. The mass is not visualized.

Discussion

Observations

This case was extremely rare for two reasons. First, the aneurysm originated at the nonbranching segment of the distal ACA. Second, the aneurysm developed complete thrombosis. The aneurysm was preoperatively diagnosed as a cavernous malformation because no evidence of its continuation with the lumen of the arteries on preoperative examinations was observed. MRI findings of heterogeneous mixed intensity without enhancement also appeared to support the diagnosis of a cavernous malformation. However, intraoperative findings and pathological examinations revealed that the mass was not a cavernous malformation, as suggested by radiographic findings, but rather a thrombotic aneurysm. A previous study also reported that completely thrombotic aneurysms were difficult to differentiate from cavernous malformations and intracranial hematomas by radiographic features alone.[2]

Here, the primary concern was whether the aneurysm was continuous with the ACA. During surgery, the aneurysm adhered to the ACA, but the continuity of the lumen of the ACA could not be confirmed. This might be explained by the findings of a previous study, which showed that the endothelial cell lining develops on the granulation tissue toward the center of the aneurysmal neck during the repair process after coil or stent treatment for saccular aneurysms.[3] Considering this, we hypothesized that the lack of continuity between the vascular lumen and the aneurysm neck was due to the endothelium covering the aneurysm neck after thrombosis of the aneurysm. However, the exact mechanism for the loss of continuity between the thrombotic aneurysm and the parent artery remains unclear.

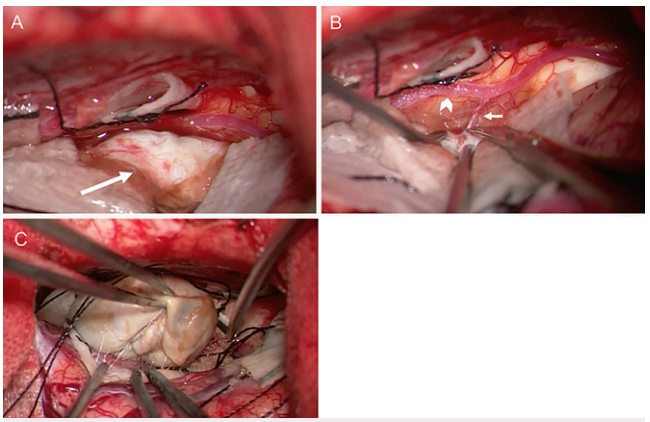

FIG. 4. Intraoperative photographs. **A:** White and smooth surface of the mass (*arrow*); the mass did not pulsate. **B:** The mass adherent to the branch (*arrow*) of the pericallosal artery (*arrowhead*), but no luminal continuity is observed. **C:** Complete resection of the mass after detachment from the artery.

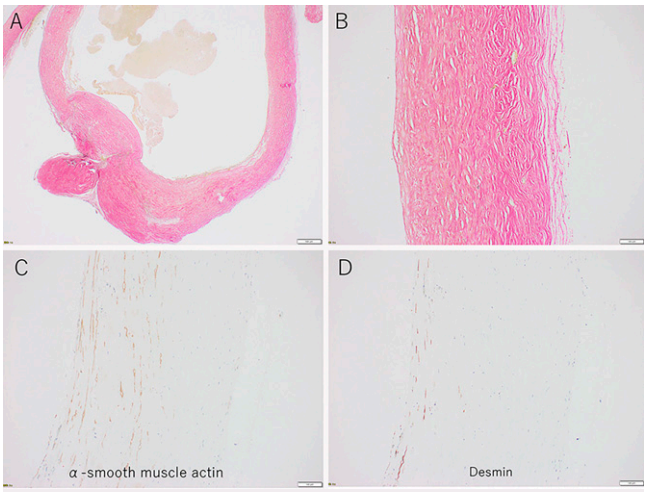

FIG. 5. Pathological findings. Elastica van Gieson staining reveals mainly collagen fibers and no elastic fibers in the tissue specimen (**A and B**). α-smooth muscle actin–positive cells (**C**) and desmin-positive cells (**D**) are observed on the luminal side.

Aneurysms at the distal ACA account for approximately 1%–9% of intracranial aneurysms and most commonly occur at the bifurcation of the pericallosal and calloso-marginal arteries.[4–7] Saccular aneurysms at the nonbranching segment of the distal ACA are extremely rare,[1] and pseudoaneurysms, which are mostly traumatic or infectious, at this site are also relatively rare.[8,9] However, as the patient had no history of infection or trauma, the cause and mechanism of the aneurysmal origin were unknown. According to a systematic review, female gender, anterior circulation location, and larger aneurysms were identified as slightly prevalent factors for nongiant intracranial aneurysms with completely spontaneous thrombosis.[10] This case included these factors.

Hemosiderin deposition was observed on preoperative radiographic images, intraoperatively, and during the pathological examination. This indicated that the aneurysm was possibly previously ruptured and was related to the history of headaches. Previous studies have reported rare cases of completely thrombosed aneurysms that presented with recanalization, reenlargement, and ischemic events.[10–12] Therefore, craniotomy or endovascular treatment should be considered along with a follow-up, and the resection performed in this case is considered reasonable.

Lessons

Thrombotic saccular aneurysms rarely develop in the nonbranching segment of the distal ACA, as was observed in this case. Because the radiographic findings are similar, a complete thrombotic aneurysm could be misdiagnosed as a cavernous malformation. Therefore, mass lesions in contact with major arteries should be differentiated as thrombotic aneurysms, even when the lumen of the arteries appears disconnected from the mass.

Acknowledgments

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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: Hirokawa, Michiwaki. Acquisition of data: Hirokawa, Michiwaki, Tanaka, Sashida. Analysis and interpretation of data: Hirokawa, Kawashima, Wakamiya. Drafting the article: Hirokawa, Onoda. Critically revising the article: Hirokawa, Kawashima, Sashida, Shimoji, Yamane. Reviewed submitted version of manuscript: Michiwaki, Kawashima, Sashida, Suehiro, Yamane, Matsuno. Study supervision: Kawashima.

Correspondence

Yu Hirokawa: International University of Health and Welfare, School of Medicine, Narita Hospital, Chiba, Japan. yuhirokawa13@gmail.com.