

# **Giant Serpentine Aneurysm Arising From the Middle Cerebral Artery Successfully Treated With Trapping and Anastomosis**

## **—Case Report—**

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### **Abstract**

**A 56-year-old man presented with a giant serpentine aneurysm arising from the middle cerebral artery (MCA) manifesting as right hemiparesis and motor aphasia. Magnetic resonance imaging and digital subtraction angiography identified the giant serpentine aneurysm arising from the MCA. The patient was treated surgically. Temporary clipping of the distal channel induced thrombosis in the vascular channel, and the thrombosis was aspirated with an ultrasonic suction device after superficial temporal artery-MCA anastomosis. This case shows that initial occlusion of the distal channel is effective to treat giant serpentine aneurysm.**

Key words: giant serpentine aneurysm, thrombosis, trapping, superficial temporal artery-middle cerebral artery anastomosis, middle cerebral artery

### **Introduction**

Giant serpentine aneurysm (GSA) was first reported in 1965,<sup>16)</sup> and is defined as a giant, partially thrombosed aneurysm (greater than 25 mm diameter) with tortuous vascular channels forming a serpentine inflow and outflow pathway.<sup>6-10,14,15,17,18)</sup> The entry and exit points of the residual aneurysm lumen are clearly identifiable, and are separated by a variable distance. GSAs have been described as large globoid or pear-shaped masses with a 1.0- to 4.0-mm thick fibrous wall that may contain numerous small vessels similar to vasa vasorum.<sup>2,6,13)</sup> The vascular channels do not seem to be residual lumens of the parent artery but are typically intrathrombotic canals that are not endothelialized and do not contain normal elastic lamina or media. GSAs may arise from fusiform aneurysms.<sup>4,18,19)</sup>

Treatment of GSAs should aim to arrest growth, to eliminate the mass effect, and to obliterate the abnormal vascular channel. Flow through the vascular channels is typically slow, but the outflow channel

gives rise to normal distal cerebral arterial branches that supply areas of the brain that would otherwise be supplied by the same parent vessel.<sup>5,6,17)</sup> Therefore, surgical strategies often require both elimination of blood flow into the aneurysm, and revascularization of vital distal cerebral territories.

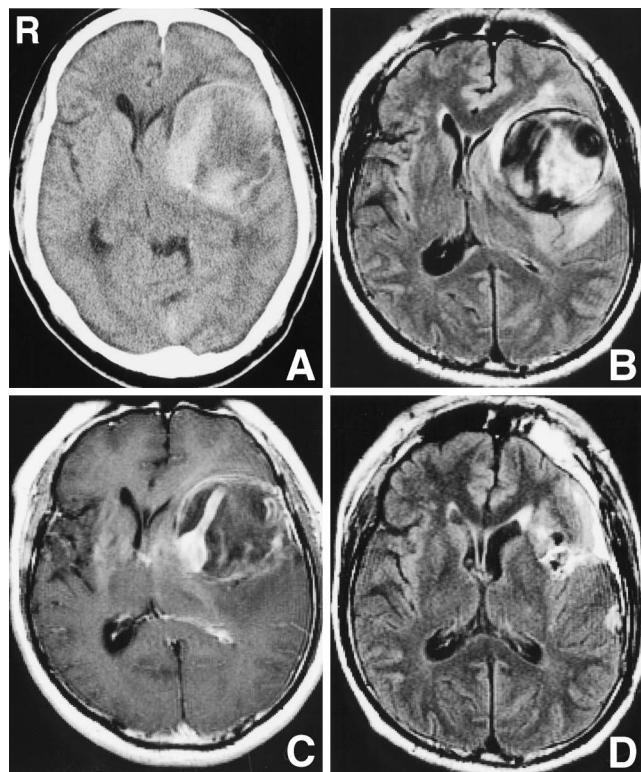
Here we discuss the successful surgical treatment of a GSA arising from the middle cerebral artery (MCA).

### **Case Report**

A 56-year-old man presented with a 2-month history of gradually worsening motor aphasia and disorientation. Neurological examination identified mild right hemiparesis. Computed tomography (CT) revealed a mixed density globoid mass and partial peripheral calcification in the left frontotemporal region (Fig. 1A). Magnetic resonance (MR) imaging revealed a 77-mm diameter heterogeneous mass with a flow void on both T<sub>1</sub>- and T<sub>2</sub>-weighted images, with significant perifocal edema on the fluid-at-

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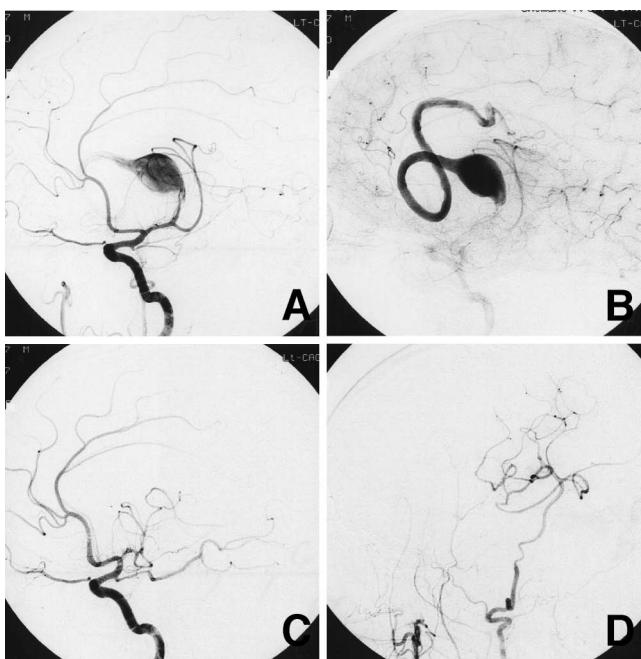
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**Fig. 1** A: Computed tomography scan revealing a heterogeneous mass with slight calcification in the peripheral portion. B: Fluid-attenuated inversion recovery (FLAIR) magnetic resonance (MR) image showing a heterogeneous mass in the left frontotemporal region with adjacent edema. C: T<sub>1</sub>-weighted MR image with gadolinium revealing enhancement of the periphery and the eccentric channel within the aneurysm. D: Postoperative FLAIR MR image showing disappearance of the mass and edema.

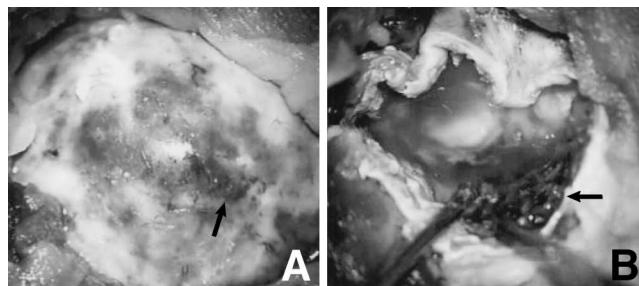
tenuated inversion recovery image (Fig. 1B), and heterogeneous enhancement and residual flow through the serpiginous lumen within the mass after gadolinium administration (Fig. 1C). Cerebral angiography demonstrated a partially thrombosed giant aneurysm arising from the superior branch of the left MCA. The serpiginous lumen was filled slowly and supplied blood to the distal area (Fig. 2A, B). These findings were consistent with a partially thrombosed giant aneurysm arising from the left MCA with a serpentine channel. Therefore, the patient was readmitted for surgical treatment.

A simple superficial temporal artery (STA)-MCA bypass was planned to vascularize the distal superior trunk of the MCA and trap the aneurysm. After the STA was dissected from the scalp, a left frontotemporal craniotomy was carried out. Ventricular



**Fig. 2** A-C: Selective left internal carotid angiograms, lateral view, revealing a serpentine channel arising from the superior trunk of the middle cerebral artery in the early phase (A), and the channel filled slowly and supplied blood to the distal area during the late phase before surgery (B), and disappearance of the channel after surgery (C). D: Selective left external carotid angiogram, lateral view, showing good filling of the superficial temporal artery and distal superior trunk.

drainage was performed for swelling and tightness of the brain. After the dura was opened, a giant aneurysm was seen within the sylvian fissure, with a thinned temporal lobe. Careful microsurgical dissection around the lateral aspect of the aneurysm exposed the distal portion of the M<sub>2</sub> superior trunk at the distal side of the aneurysm. Doppler imaging revealed blood flow through the aneurysm, which prevented dissection of the proximal aspect of the aneurysm because mass reduction was impossible and the proximal entrance of the aneurysm could not be exposed. After placement of a temporary clip on the distal portion of the M<sub>2</sub>, the STA was anastomosed in end-to-side fashion. Excellent flow was recognized through the STA and the distal superior trunk on the distal side of the temporary clip, so the temporary clip was exchanged for a permanent clip. During this procedure, Doppler imaging revealed partial disappearance of the blood flow in the aneurysm. Dissection around the aneurysm with coagulation of vasa vasorum of the aneurysm wall



**Fig. 3** Intraoperative photographs. A: The giant aneurysm and vasa vasorum of the aneurysm wall (arrow). B: Thrombus within the aneurysm (arrow).

was continued (Fig. 3A). Within 1 hour of distal occlusion of the M<sub>2</sub> trunk, blood flow through the aneurysm had disappeared, so the aneurysm wall was partially incised. The serpiginous lumen within the aneurysm contained old blood and new thrombus that was aspirated with an ultrasonic suction device, and the aneurysm wall was resected (Fig. 3B). The lenticulostriate arteries were strongly adhered to the aneurysm, and had to be dissected from the aneurysm very carefully. The proximal entrance was exposed from inside and outside of the aneurysm, and permanent clips were placed on the proximal channel of the aneurysm.

The patient awoke without new neurological deficits. Postoperative angiography showed good filling of the STA and superior trunk, and the serpiginous lumen disappeared (Fig. 2C, D). Follow-up MR imaging after the operation showed disappearance of the mass and perifocal edema (Fig. 1D). The patient's symptoms displayed significant improvement.

## Discussion

GSA usually manifests as intracranial mass rather than intracranial hemorrhage. The symptoms are dependent on the aneurysm location, but the predominant signs and symptoms are headache, hemiplegia and hemiparesis, visual disturbance, cranial nerve paresis, dysphasia and aphasia, nausea, vomiting, and seizure and vertigo.<sup>1,3)</sup>

The neuroimaging findings of GSAs are distinctive. CT demonstrates an oval or globoid mass of mixed density. Heterogeneous regions of increased attenuation represent thrombus, and tubular regions of decreased attenuation represent the patent vascular channel. After contrast administration, enhancement of the serpentine vascular channel is apparent. MR imaging shows a mass lesion as a heterogeneous

mass that represents various stages of hemoglobin degradation and flow void lesions. MR angiography shows the aneurysm is clearly separated from the normal parenchyma, and the vascular channel is visible. Angiography is the most accurate method for evaluating the location and state of flow in a GSA. Our diagnosis of GSA was based on these features.

Some GSAs have been treated successfully by endovascular procedures intended to induce thrombosis and shrinkage of the mass.<sup>1,3,12)</sup> However, these GSAs arose from the anterior cerebral artery, posterior cerebral artery, vertebral artery, and internal carotid artery. Two cases of GSA arising from the left MCA were complicated with aphasia after parent artery occlusion with endovascular treatment.<sup>11)</sup> GSAs arising from the MCA might be treated surgically with revascularization of the distal area by STA-MCA anastomosis. However, the proximal entrance of a GSA can be difficult to isolate, because the spherical solid mass may obstruct the operative field of vision.

In our case, we were able to isolate the proximal entrance by initial placement of a temporary clip on the distal channel. This clip induced thrombosis in the vascular channel, so we could aspirate the thrombus with an ultrasonic suction device after STA-MCA anastomosis. We think that the slow flow through the vascular channel was probably congested by the distal occlusion and thrombosis. This approach seems counterintuitive, as occlusion of the outflow would make the aneurysm more prone to rupture because of increased intraluminal pressure. However, such distal channel occlusion is a reasonably safe undertaking based on the Bernoulli equation.<sup>9)</sup>

In the present case, the aneurysm thrombosed after distal channel occlusion and wall coagulation. Oozing at the thrombus persisted during thrombus aspiration. Coagulation of the vasa vasorum of the aneurysm wall was effective in controlling the oozing, which suggests transmural vascular connections between the vasa vasorum and the thrombus.

Preservation of the lenticulostriate artery is extremely important during surgical removal of a GSA of the MCA. In our case, the lenticulostriate arteries were strongly adhered to the aneurysm because of its giant size, and had to be dissected from the aneurysm very carefully.

Surgical treatment of a GSA is challenging. The present case of GSA arising from the MCA was treated by placement of a temporary clip on the distal channel resulting in coagulation of the vasa vasorum after STA-MCA anastomosis.

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