

Case Report

Giant Serpentine Aneurysm of the Middle Cerebral Artery

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Giant serpentine aneurysms are rare and have distinct angiographic findings. The rarity, large size, complex anatomy and hemodynamic characteristics of giant serpentine aneurysms make treatment difficult. We report a case of a giant serpentine aneurysm of the right middle cerebral artery (MCA) that presented as headache. Treatment involved a superficial temporal artery (STA)-MCA bypass followed by aneurysm resection. The patient was discharged without neurological deficits, and early and late follow-up angiography disclosed successful removal of the aneurysm and a patent bypass graft. We conclude that STA-MCA bypass and aneurysm excision is a successful treatment method for a giant serpentine aneurysm.

KEY WORDS : Angiographic feature · Bypass surgery · Giant serpentine aneurysm.

INTRODUCTION

Giant serpentine aneurysms were first documented by Segal and McLaurin¹⁴. The distinctive neuroimaging features of these aneurysms are a large size (> 25 mm), and a twisted vascular course within the aneurysm with multiple entrance and exit points. Giant serpentine aneurysms contain a thrombus, show peripheral calcification, have a mass effect including a midline shift, and have adjacent edema mimicking a brain tumor in computed tomography (CT)^{1,3,5}. Patients with giant serpentine aneurysm usually show symptoms associated with the mass effect, including headache, hemiparesis and seizure^{4,15}. Aggressive treatment is generally recommended for giant serpentine aneurysms since conservative measures often result in continual aneurysm growth, neurological deterioration and poor outcomes^{11,12,15}. Prior to the use of revascularization surgery in the neurosurgical field, surgical treatment of giant serpentine aneurysms was associated with unacceptable morbidity and mortality rates as high as 33%^{1,7}.

However, the application of bypass surgery has resulted in satisfactory outcomes¹⁶. Treatment of giant serpentine aneurysms should be individualized due to the complex and varied hemodynamics and morphological features⁶.

The present report describes a case of a giant serpentine aneurysm of the middle cerebral artery (MCA). Treatment involved a superficial temporal artery (STA)-MCA bypass and resection of the aneurysm. Neurological and angiographic studies two years after postoperatively indicated the treatment was successful.

CASE REPORT

A 43-year-old woman presented at the emergency department complaining of severe headache. She had a two year history of intermittent headache which had gradually worsened over the past several months. She also experienced transient left hand weakness that persisted for ten minutes and left facial numbness. Her past medical history was unremarkable, and she did not take any regular medication. Neurological examinations at admission revealed no focal deficits. CT showed a 7 × 8 × 5 cm mass with an approximately 10 mm midline shift, and adjacent edema. There was peripheral calcification and mixed density within the mass (Fig. 1A). Contrast enhancement showed that the mass had a heterogeneous enhancing pattern, and a large and tortuous blood

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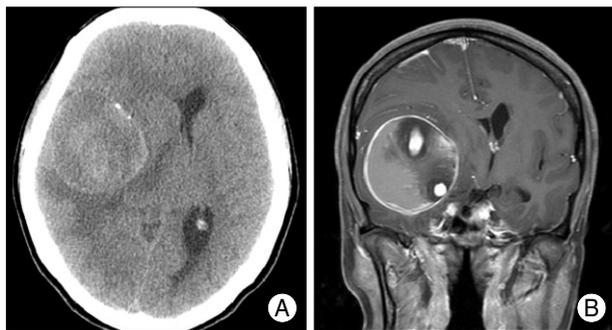


Fig. 1. Preoperative images of the giant serpentine aneurysm. A : A preoperative axial CT image showing a round, iso-dense mass with peripheral calcification, midline shifting and peripheral edema. B : A preoperative coronal MR image demonstrating the mass effect with midline shifting, and a thrombus within the aneurysm.

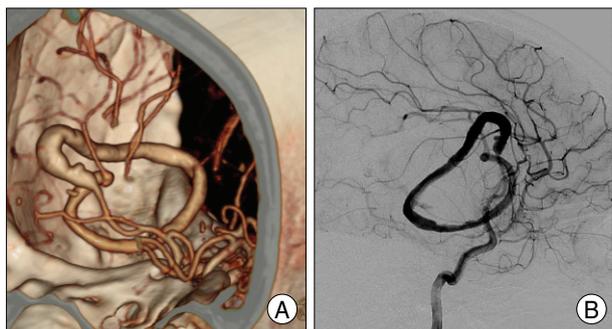


Fig. 2. Preoperative angiographic features of the giant serpentine aneurysm. A : Left anterolateral projection CT angiography image showing tortuous and thick "serpiginous" blood vessels. B : A preoperative angiographic lateral view showing superior displacement of the right middle cerebral artery and distal blood supply from the serpiginous artery.

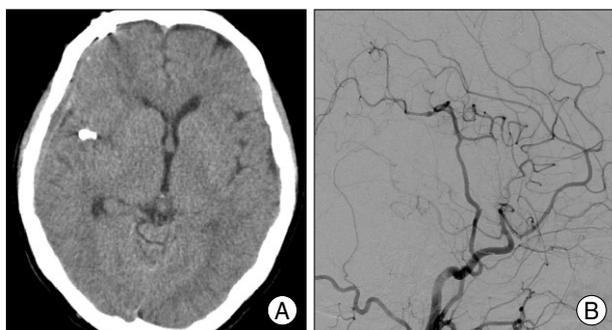


Fig. 3. Postoperative images showing with proximal and distal clipping of the giant serpentine aneurysm and bypass surgery. A : A postoperative axial CT image showing improved midline shifting, lack of mass effect and peripheral edema. B : A lateral view of common carotid arteriography showing lack of the aneurysm and dye filling to the distal MCA territory through the bypass.

vessel passed through the mass. Magnetic resonance image (MRI) scans showed different stages of organizing hematoma within the mass, and continuity with the right MCA (Fig. 1B). A diagnosis of a giant aneurysm was made. Angiography showed the right MCA was displaced upward, and demonstrated an eccentric vascular course through the aneurysm with multiple entrance and exit points that differentiated the mass from a regular saccular giant aneurysm. The

morphology of the vascular course within the aneurysm was snake-like and serpiginous. Flow stagnation resulted from a long transit time that was attributed to the formation of a thrombus within the aneurysm (Fig. 2).

The patient was treated surgically. The parietal branch of the STA was prepared for anastomosis. A wide sylvian dissection was performed and the whole length of the aneurysm and proximal and distal MCA were identified. End-to-side bypass surgery was performed between the STA and the efferent M4 branch. After bypass, permanent clips were placed at the proximal and distal artery of the aneurysm and the aneurysm excised. Several perforators from the aneurysm were coagulated and cut. The postoperative course was uneventful and the patient was discharged without any neurological problems. A postoperative CT scan revealed complete removal of the mass and normalized midline shifting (Fig. 3A). A one week postoperative angiography demonstrated good patency of the bypass graft with sufficient distal perfusion (Fig. 3B), and a two year follow-up CT angiography revealed the same findings.

DISCUSSION

The origin and pathogenesis of giant serpentine aneurysms remain uncertain. They may represent the expansion of a fusiform aneurysm with partial thrombosis and repeated intramural hemorrhage through a weak point in the arterial wall¹⁰. Christiano et al.⁴ reported that giant serpentine aneurysms most commonly occur in the MCA circulation (50%) followed by the PCA circulation (18%) and vertebrobasilar artery (15%). The MCA circulation has no barriers such as dural or bony structures to prevent aneurysm growth, and this anatomical characteristic is thought to favor development of serpentine aneurysms.

Fanning et al.⁷ suggested that the lower force of jet blood flow of the MCA brought about the flow stagnation within the aneurysm which was attributed to the formation of thrombus. Additionally, giant serpentine aneurysms have the different eccentric vascular channels, resulting the Coanda effect, describing jet blood flow forces are stronger in the deflected surface of the wall rather than central portion of the aneurysm which makes thrombus within central portion of the aneurysm.

In computed tomography, the giant serpentine aneurysm looks like brain tumor in several aspects, well circumscribed mass lesion with edema and midline shifting. Therefore, it may be often misdiagnosed as a brain tumor, initially. In contrast enhancing CT scan, the giant serpentine aneurysm demonstrates as herogenous enhancing mass with peripheral calcification and thick, wavy vessel within the mass.

These findings imply that the thrombus is located within the aneurysm and the formation of the giant serpentine aneurysm has been taken a long time. There are no consistent findings associated with MRI resulting from coexistence of different stages of thrombus¹⁰.

The natural history of giant serpentine aneurysms has not been established due to their rarity and severity. The few reports on giant serpentine aneurysms contain only a small number of cases. While some authors reported spontaneous occlusion of the aneurysm¹², the majority reported rapid neurological deterioration leading to death after ischemic stroke, or recanalization of the spontaneously occluded aneurysm^{9,11,14}. Suzuki et al.¹⁵ followed 39 giant serpentine aneurysm cases and reported that angiographic enlargement occurred in 12 of those patients over the follow-up period. They suggested that this enlargement was the result of repetitive intramural hemorrhage and accumulation of the thrombus. Sari et al.¹² reported a case of spontaneous, complete thrombosis of a giant serpentine aneurysm. Mahadevan et al.¹¹ reported on a giant serpentine aneurysm that caused rapid neurological deterioration and death due to ischemic stroke. The neurological deterioration in that case resulted from stretching or compression of the parent cortical vessels or acute occlusion of the vascular channel due to thrombosis accumulation. Lee et al.⁹ reported a case where a giant serpentine aneurysm recanalized after spontaneous occlusion. Therefore, monitoring and further treatment should always be considered even after spontaneous occlusion. Although subarachnoid hemorrhage (SAH) is not considered a common presentation of giant serpentine aneurysms, Suzuki et al.¹⁵ reported 28% of giant serpentine aneurysm patients initially presented with SAH.

Surgery appears to be the optimal treatment for most giant serpentine aneurysm cases. Suzuki et al.¹⁵ documented outcomes following conservative or aggressive management of 18 cases of MCA giant serpentine aneurysm. Of the six conservatively treated patients, five suffered neurological deterioration or death, while one showed no neurological change. Of the 12 surgically treated patients, all showed neurological improvement and preserved distal cortical blood flow. These findings indicate that aggressive treatments should be considered.

Important issues to consider in aneurysm surgery include accessibility, aneurysm morphology and perforators. In the past, carotid artery ligation was used to treat giant serpentine aneurysms, but was associated with poor outcomes and ischemic stroke rates as high as 33%¹³. Surgical wrapping or coating was attempted, but resulted in the deaths of two of 18 patients¹⁵. Horowitz et al.⁸ reported that occlusion of the distal outflow only was an effective surgical management, but

this approach has yet to be established. Prior to the introduction of bypass surgery, giant serpentine aneurysm surgery resulted in overall morbidity and mortality rates of 30-35%¹¹. Outflow from the giant serpentine aneurysm feeds distal cortical vessels, and occlusion or narrowing of those vessels results in stroke⁷. Amin-Hanjani et al.² reported a case of a giant serpentine aneurysm treated using an STA-MCA bypass and proximal MCA clipping. The patient was neurologically intact, and follow-up angiography revealed no aneurysm and a patent bypass 13 years later². Recently, endovascular approaches have been used to treat giant serpentine aneurysms, especially those in surgically inaccessible areas, and successful results have been reported^{6,10}. However, endovascular approaches do not immediately relieve the mass effect and cannot be performed in cases of insufficient collateral distal blood flow. The present patient suffered headache and ipsilateral compressive trigeminal neuropathy due to the severe local mass effect of the aneurysm, making a key goal of treatment the immediate elimination of the mass effect.

Bypass surgery is indispensable for avoiding ischemic complications after aneurysm resection. When performing an STA-MCA bypass to provide distal blood flow, it is easier to create the anastomosis at the surface of the brain rather than deep in the sylvian fissure. However, it is sometimes difficult to identify the correct recipient vessel on the brain surface. Cerebral angiographs do not provide sufficient information regarding the correct distal vessel for bypass, especially near the sylvian point where MCA branching is maximal and the course of branches is complex and crowded. Therefore, we used a microvascular Doppler on the cortical artery to detect loss of arterial sound when we applied a temporary clip to the MCA just distal to the aneurysm, and the return of sound after removal of the temporary clip. After a successful bypass, the aneurysm can be excised and the perforators from the aneurysm to the insular cortex can be sacrificed as they appear to be non-functioning.

CONCLUSION

We found that STA-MCA bypass and aneurysm excision was a successful treatment method for a giant serpentine aneurysm. This approach immediately relieved symptoms related to the mass effect, and provided distal blood flow.

References

1. Aletich VA, Debrun GM, Monsein LH, Nauta HJ, Spetzler RF : Giant serpentine aneurysm : a review and presentation of five cases. *AJNR Am J Neuroradiol* 16 : 1061-1072, 1995
2. Amin-Hanjani S, Chen PR, Chang SW, Spetzler RF : Long-term follow-up of giant serpentine MCA aneurysm treated with EC-IC bypass and proximal occlusion. *Acta Neurochir (Wien)* 148 : 227-228,

- 2006
3. Belec L, Cesaro P, Brugieres P, Gray F : Tumor-simulating giant serpentine aneurysm of the posterior cerebral artery. *Surg Neurol* 29 : 210-215, 1988
 4. Christiano LD, Gupta G, Prestigiacomo CJ, Gandhi CD : Giant serpentine aneurysms. *Neurosurg Focus* 26 : E5, 2009
 5. Coley SC, Hodgson TJ, Jakubowski J : Coil embolization of giant serpentine aneurysms : report of two cases arising from the posterior cerebral artery. *Br J Neurosurg* 16 : 43-47, 2002
 6. Drake CG, Peerless SJ : Giant fusiform intracranial aneurysm : review of 120 patients treated surgically from 1965 to 1992. *J Neurosurg* 87 : 141-162, 1997
 7. Fanning NF, Kelleher MO, Ryder DQ : The pretzel sign : angiographic pattern of tortuous intra-aneurysmal blood flow in a giant serpentine aneurysm. *Br J Neurosurg* 17 : 67-71, 2003
 8. Horowitz MB, Yonas H, Jungreis C, Hung TK : Management of a giant middle cerebral artery fusiform serpentine aneurysm with distal clip application and retrograde thrombosis : case report and review of the literature. *Surg Neurol* 41 : 221-225, 1994
 9. Lee KC, Joo JY, Lee KS, Shin YS : Recanalization of completely thrombosed giant aneurysm : case report. *Surg Neurol* 51 : 94-98, 1999
 10. Lukin RR, Chambers AA, McLaurin R, Tew J Jr : Thrombosed giant middle cerebral aneurysms. *Neuroradiology* 10 : 125-129, 1975
 11. Mahadevan A, Tagore R, Siddappa NB, Santosh V, Yasha TC, Ranga U, et al. : Giant serpentine aneurysm of vertebrobasilar artery mimicking dolichoectasia--an unusual complication of pediatric AIDS. Report of a case with review of the literature. *Clin Neuropathol* 27 : 37-52, 2008
 12. Sari A, Kandemir S, Kuzeyli K, Dinc H : Giant serpentine aneurysm with acute spontaneous complete thrombosis. *AJNR Am J Neuroradiol* 27 : 766-768, 2006
 13. Scott M, Skwarok E : The treatment of cerebral aneurysms by ligation of the common carotid artery. *Surg Gynecol Obstet* 113 : 54-61, 1961
 14. Segal HD, McLaurin RL : Giant serpentine aneurysm. Report of two cases. *J Neurosurg* 46 : 115-120, 1977
 15. Suzuki S, Takahashi T, Ohkuma H, Shimizu T, Fujita S : Management of giant serpentine aneurysms of the middle cerebral artery--review of literature and report of a case successfully treated by STA-MCA anastomosis only. *Acta Neurochir (Wien)* 117 : 23-29, 1992
 16. van Rooij WJ, Sluzewski M, Beute GN : Endovascular treatment of giant serpentine aneurysms. *AJNR Am J Neuroradiol* 29 : 1418-1419, 2008