



# Clinical characteristics and endovascular treatment in patients with intracranial giant serpentine aneurysms

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**Background:** Giant serpentine aneurysms (GSAs) are a rare subtype of intracranial aneurysm. Recently, GSAs have been successfully treated with endovascular parent artery occlusion with or without distal bypass. The present study retrospectively analyzed the clinical outcomes of endovascular parent artery occlusion for intracranial GSAs.

**Methods:** Medical records and cerebral angiograms from our endovascular center were retrospectively analyzed. Twenty-two patients with serpentine aneurysms were treated with endovascular occlusion of the parent artery at the site of the aneurysm. These patients had selective treatment. Clinical and angiographic outcomes of the patients were assessed between the 3-month to 3-year stage.

**Results:** The clinical manifestations of GSA included headache, hemiparesis, SAH, epilepsy, memory loss, right oculomotor palsy, and intracerebral hemorrhage. No cerebral infarction occurred. The average Glasgow Outcome Scale (GOS) score was 5. There were no clinical complications and none of the patients had recurrence after selective embolization. All of the patients recovered well, and no recurrence or rebleeding was noted at the follow-up.

**Conclusions:** Endovascular parent artery occlusion may be a safe and effective way to treat intracranial GSAs.

**Keywords:** Endovascular treatment; serpentine aneurysm; parent artery occlusion

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## Introduction

Giant serpentine aneurysms (GSAs) are a rare subtype of intracranial aneurysm with a distinct appearance. They are large ( $\geq 25$  mm in diameter), fusiform, partially thrombosed, tortuous aneurysms with a partial filling of the thrombus in the sac (1-4). However, they do not originate from the folding of arteries in the residual vessels (1-3). Clinically, GSAs typically manifest as space-occupying lesions in the brain parenchyma around the aneurysm and cerebral edema in the adjacent tissue. Moreover, a ruptured aneurysm can also lead to extensive subarachnoid hemorrhage (SAH),

causing headache, epilepsy, and other clinical symptoms (5-7). At present, the diagnosis of this disease is mainly based on the imaging characteristics of GSA and the clinical features consistent with it (8).

Previous studies have reported that the treatment of GSA involves occlusion of the parent artery (5,9). However, perhaps due to the relative scarcity of serpentine aneurysms, these studies do not specifically address the treatment of these large aneurysms, and most of them are case reports. In this paper, we used retrospective methods to systematically analyze the clinical characteristics and endovascular treatment of intracranial GSA patients for the first time.

## Methods

We retrospectively analyzed the medical records and cerebral angiograms in our hospital from October 2002 to October 2019. Computed tomography angiography (CTA), magnetic resonance imaging/angiography (MRI/A), and/or digital subtraction angiography (DSA) were analyzed to identify patients with GSAs. We further evaluated the medical records and cerebral angiography results (including the definition of the morphological characteristics of the aneurysms) of these patients to determine those treated with endovascular occlusion of the parent artery. For these patients, the coil was removed by electrolysis under anesthesia and heparinization.

Prior to treatment, GSA patients underwent temporary occlusion of the parent artery to observe whether there were any neurological deficits. If no positive symptoms or signs of neurological deficit were observed, the aneurysm was filled with coils. When the coil protruded from the parent artery, the aneurysm was filled until the parent artery near the GSA was occluded. Following occlusion, the distal part of the occluded artery was observed by contrast cerebral angiography. Finally, collateral blood vessels were perfused.

If the patient could not tolerate the temporary occlusion of the parent artery, the occlusion was relieved, and the aneurysm was permanently occluded following surgical distal bypass. After the surgery, patients received anticoagulant medication (Warfarin sodium) to maintain the international normalized ratio (INR) value between 2 and 3, and ensure that blood pressure was controlled between 120–130/70–80 mmHg.

The study was approved by the ethics committee of Renmin Hospital, Wuhan University (NO.: 2018-X-016) and informed consent was taken from all the patients or substitute decision-maker.

## Results

A total of 22 GSA patients were retrospectively analyzed, including 15 males and 7 females (aged between 16 and 67 years). The clinical manifestations of GSA included headache, hemiparesis, SAH, epilepsy, memory loss, right oculomotor palsy, and intracerebral hemorrhage (*Table 1*). Of the 22 patients, 8 experienced headache and SAH (*Figure 1*), 5 had intracerebral hemorrhage (*Figure 2*), 3 had right hemiparesis (*Figure 3*), 11 had epilepsy, 5 had memory loss, and 2 had right oculomotor palsy. The average Glasgow Outcome Scale (GOS) score was 5. There were no clinical complications and none of the patients had recurrence after

selective embolization. In the early treatment stage, five patients suffered memory loss and had oculomotor nerve palsy; however, symptoms disappeared at the 3-months postoperative stage. None of the patients received surgical distal bypass (*Table 1*).

## Discussion

At present, the diagnosis of GSA is fundamentally based on imaging characteristics and clinical features (3-5). Generally, GSA refers to a large ( $\geq 2.5$  cm) fusiform aneurysm with local thrombosis, and its incidence is rare (3,5). Most GSAs are located in the middle cerebral artery (MCA) and its branches; however in our study cohort, GSAs were primarily located in the posterior cerebral artery (PCA). The patients included in our study were younger, with ages ranging between 16 and 67 years old. Their occurrence in the PCA segment may indicate an embryological similarity between the MCA and the PCA. The MCA is a branch of the anterior cerebral artery (ACA) while the PCA is a branch of the internal carotid artery. GSAs of the superior cerebellar artery (SCA) have also been reported (9).

The clinical manifestations of GSA are a result of the space-occupying lesion, adjacent tissue compression, and SAH, and can lead to seizures or other neurological dysfunctions (8-10). In our study, one patient with ruptured P2 segment GSA presented with memory loss, hemianesthesia, oculomotor nerve palsy, and visual disturbance. The other eight cases of P2 segment GSA exhibited hemianesthesia due to middle brain compression. Although massive aneurysmal hemorrhage due to thrombosis is uncommon, SAH occurred in two cases. Five cases (5/22) were anterior circulation aneurysms. The imaging features of GSA can be found in CTA, MRI, and DSA (*Figure 2*). Thus, evaluating the anatomy and dynamics of cerebral circulation prior to surgery is necessary in order to avoid local cerebral ischemia.

Etiologically, endothelial damage plays an important role in the formation of GSAs (7). Injury is typically induced by blood turbulence and subsequent intramural hemorrhage. Additionally, platelet aggregation to the endothelium and cell matrix can accelerate thrombosis, as well as the formation of thrombus aneurysms (which is another explanation for the local ischemia).

Treatment of GSAs should eliminate the pathway of the parent vessels, prevent the growth of aneurysms, and reduce the cerebral space-occupying effect on adjacent tissue (5). In the past, surgical resection was typically employed to achieve these goals. Recently, however, GSAs

**Table 1** Surgery, treatment, and results of 22 cases of typical GSA

No.	Age/gender	Clinical manifestation	Vascular	Angiographic reexamination results	Clinical angiographic follow-up
1	16/M	Headache, SAH	P2, L	Complete occlusion	GOS=5, asymptomatic (8M)
2	22/F	Epilepsy	P2, L	Complete occlusion	GOS=5, asymptomatic (8M)
3	33/M	Epilepsy, hemiparesis	P2, L	Complete occlusion	GOS=5, asymptomatic (6M)
4	35/M	SAH, ICH	P2, L	Complete occlusion	GOS=5, asymptomatic (4M)
5	42/F	Memory loss	P2, R	Partly occlusion	GOS=5, asymptomatic (8M)
6	43/M	SAH, epilepsy	P2, L	Complete occlusion	GOS=5, asymptomatic (3M)
7	45/M	Headache epilepsy	P2, R	Complete occlusion	GOS=5, asymptomatic (8M)
8	50/F	SAH, memory loss	P2, L	Complete occlusion	GOS=5, asymptomatic (3Y)
9	53/M	Epilepsy, memory loss	P2, L	Complete occlusion	GOS=5, asymptomatic (1Y)
10	56/M	SAH, hemiparesis	P2, R	Complete occlusion	GOS=5, asymptomatic (10M)
11	63/F	Epilepsy, memory loss	P1, L	Complete occlusion	GOS=5, asymptomatic (8M)
12	37/M	Epilepsy, memory loss	P1, R	Partly occlusion	GOS=5, asymptomatic (8M)
13	42/M	Headache, epilepsy	MCA, R	Complete occlusion	GOS=5, asymptomatic (8M)
14	19/F	ICH, epilepsy	MCA, L	Complete occlusion	GOS=5, asymptomatic (3Y)
15	21/M	Epilepsy, memory loss	MCA, L	Partly occlusion	GOS=5, asymptomatic (3M)
16	27/F	ICH, epilepsy	MCA, R	Complete occlusion	GOS=5, asymptomatic (1Y)
17	39/M	ICH, hemiparesis	MCA, L	Complete occlusion	GOS=5, asymptomatic (7M)
18	46/F	Oculomotor palsy	MCA, L	Complete occlusion	GOS=5, asymptomatic (12M)
19	57/M	ICH, hemiparesis	MCA, L	Complete occlusion	GOS=5, asymptomatic (3M)
20	59/M	Headache	ACA, L	Complete occlusion	GOS=5, asymptomatic (7M)
21	61/M	Oculomotor palsy	ACA, L	Complete occlusion	GOS=5, asymptomatic (6M)
22	63/M	Headache, blurred vision	ACA, L	Complete occlusion	GOS=5, asymptomatic (6M)

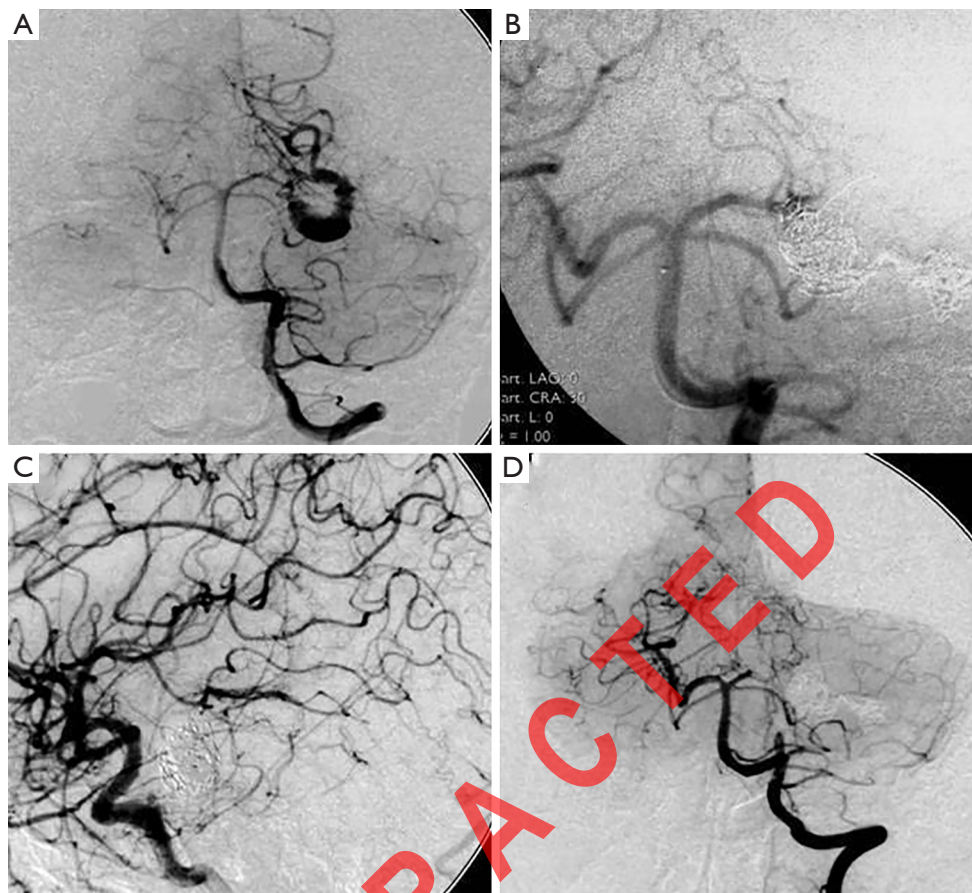
SAH, subarachnoid hemorrhage; MCA, middle cerebral artery; GOS, Glasgow Outcome Score; ICH, intracerebral hemorrhage; ACA, anterior cerebral artery.

have been successfully treated with endovascular occlusion with or without distal artery bypass (2,11,12). Although preserving the parent artery is an important objective of the endovascular treatment of aneurysms, it is impossible to treat serpentine aneurysms in this way at present because there is no definable neck.

We believe that surgical revascularization and flow diverters do not guarantee the reconstruction of serpentine aneurysms, although they can be combined with parent artery occlusion. In fact, four cases of GSAs located in the MCA even worsened following carotid ligation and superficial temporal middle artery anastomosis (8). Moreover, two cases of GSAs of the MCA reportedly underwent complete spontaneous thrombosis after angiography (13),

although recanalization was also possible (14,15). The rate of lumen stenosis and the degree of collateral circulation will ultimately determine clinical results. In the near future, multi-shell diffusion MRI acquisition maybe used to evaluate the brain injury in more accuracy strategy (16). The establishment of good collateral circulation and the absence of ischemic symptoms indicate a slower rate of aneurysm stenosis. Occlusion of the SCA, PCA, and the parent artery of MCA are common and well tolerated (2,11,12).

Among the 22 patients included in our study, the treatment of GSA of the PCA required permanent artery occlusion. However, we observed no visual field defects, which is a lower incidence than that previously reported 14.2% (10). Yet, the transient seizure in the patients might



**Figure 1** A 35-year-old male with intraventricular and subarachnoid hemorrhage. (A) Left vertebral artery angiography revealed a large serpentine aneurysm in the left P2A segment; (B) the left vertebral artery angiographic aneurysm was completely embolized following coil embolization; (C) the left internal carotid artery angiography (lateral) showed that the distal part of the left posterior cerebral artery was supplied by anastomosis of the cortical artery after embolization; (D) seven months later; the left vertebral artery angiography showed no recurrence of the aneurysm, and some retrograde filling of the distal portion of the aneurysm was also discovered.

have been caused by the delayed space-occupying effect. There are no permanent neurological sequelae in the treatment of serpentine aneurysms. The low incidence of aneurysm-bearing artery occlusion with visual field defect and other neurological dysfunctions is due to the abundant vascular anastomosis among the PCA, ACA, SCA, and MCA.

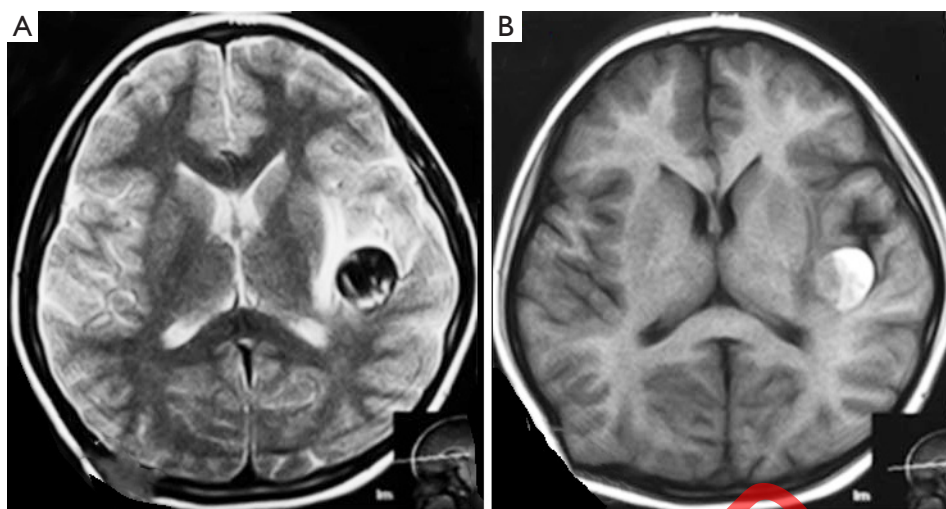
For the success and durability of the initial treatment, the method of sacrificing the parent artery is suitable and feasible, and seems to be an excellent intravascular treatment. Analysis of angiogram results shows that the collateral vessels at the distal end of the occluded point were not occluded by the embolic material, forming a siphon effect and allowing the tumor-carrying artery to supply blood through the meninges for reconstruction. In our research, 22 cases of GSA were treated with parent

artery occlusion. There were no postoperative clinical complications, indicating that parent artery occlusion is an effective and exact therapeutic method for GSA.

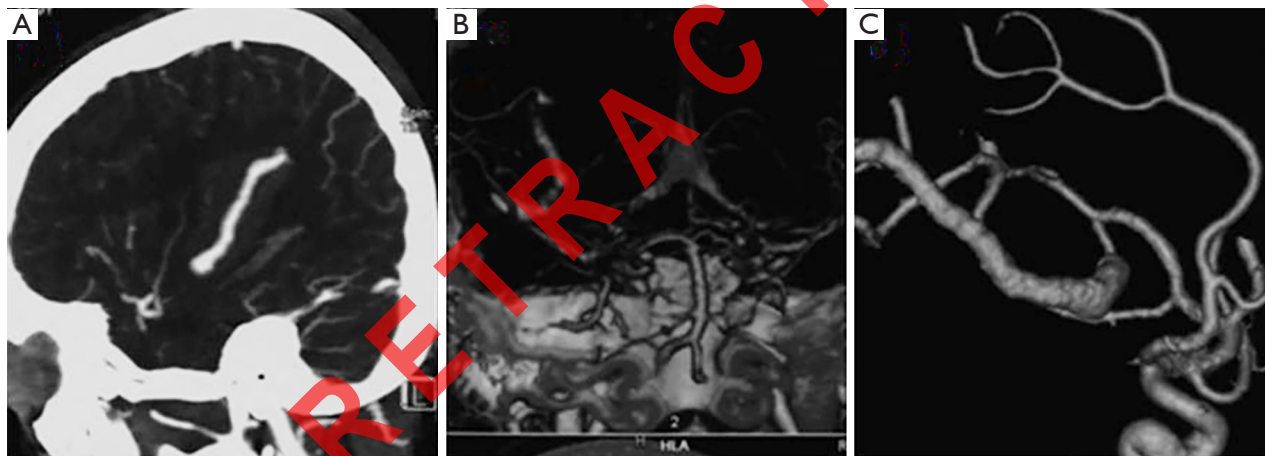
There are some limitations in this study that should be noted. Firstly, this was not a large-scale clinical control study, and it had no relevant control study. Also, since the incidence rate of this disease is relatively low, the sample size was small and could not be calculated using G-Power. Lastly, the data collected in this study spanned a long period of time, and thus, the level of surgical treatment among the included patients might have differed.

## Conclusions

The long-term prognosis of GSA patients who have



**Figure 2** A 19-year-old female with intraventricular and subarachnoid hemorrhage. MRI T2 (A) and T1 (B) showed giant aneurysms of the left middle cerebral artery with thrombosis around the internal lumen and cerebral edema around the aneurysms, suggesting local cerebral ischemia.



**Figure 3** A 57-year-old male with hemiparesis. (A and B) CTA showed tortuous prolongation of the aneurysms. (C) 3D Angiography showed serpentine aneurysms of the left middle cerebral artery. There were normal vessels in the distal part of the aneurysms, and the blood flow in the aneurysms was slow.

undergone intravascular treatment is positive with no clinical complications, indicating that artery occlusion therapy is a safe and effective treatment option for GSA.

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