



# Giant Serpentine Aneurysms: A Review and Presentation of Five Cases

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**Summary:** We present five cases of giant serpentine aneurysms (large, partially thrombosed aneurysms containing tortuous vascular channels with a separate entrance and outflow pathway) and review 28 cases reported in the literature. Giant serpentine aneurysms should be considered as a subgroup of giant aneurysms, distinct from saccular and fusiform varieties, given their unique clinical presentation and radiographic features.

**Index terms:** Aneurysm, giant; Interventional neuroradiology

In 1977 Segal and McLaurin introduced the term *giant serpentine aneurysm* as a subcategory of giant aneurysms, distinct from the saccular variety, based on observations of two cases and literature review. Subsequently, several case reports and reviews have further defined the clinical presentation and pathophysiology of these aneurysms. This article presents five cases and reviews the literature in an attempt to clarify further the presentation, diagnosis, and treatment of giant serpentine aneurysms.

## Case Reports

### Case 1

A 44-year-old, right-handed woman with history of seizures, well-controlled hypertension, and obesity was admitted to an outside institution in June 1989 after a grand mal seizure. Neurologic examination revealed intact cranial nerves and motor systems. Sensory modalities were preserved, and laboratory examinations were unremarkable. A computed tomographic (CT) scan showed a large right frontotemporal mass approximately  $4.5 \times 4.0 \times 3.7$  cm of increased heterogeneous attenuation with peripheral ring and central calcification. After contrast administration, eccentric and central regions of enhancement were visible. There was no evidence of hemorrhage. Magnetic resonance (MR) imaging showed a partially thrombosed aneurysm arising from the region of the right middle cere-

bral artery with slow flow along its medial and superior aspect and lamellated thrombus throughout the remaining portion. An angiogram performed the next day demonstrated a giant serpentine aneurysm at the right middle cerebral artery trifurcation. The aneurysm had a well-defined neck associated with a surrounding large avascular component displacing the right middle cerebral artery. The main branches of the middle cerebral artery, including the angular branch, originated proximal to the aneurysm neck. A small middle cerebral artery branch, however, arose from a separate outflow pathway. The patient requested a second opinion before therapy and was discharged pending further consultation.

The patient was admitted to Johns Hopkins Hospital in November 1989 for reevaluation. She had been neurologically intact and seizure free during the time interval. A CT scan again demonstrated the large central and peripherally calcified mass in the right frontotemporal region. After contrast administration, however, no enhancement could be identified within the center of the lesion. Peripheral enhancement not previously noted was identified. On MR the lesion had a heterogeneous signal compatible with thrombus of variable age (Fig 1A). No central flow could be identified on gradient-echo images (Fig 1B). A repeat cerebral angiogram and a subsequent angiogram 3 months later demonstrated vascular displacement compatible with the known mass in the frontotemporal region and complete thrombosis of the aneurysm. MR performed 1 year later demonstrated a persistent thrombosed aneurysm with chronic changes. The patient remains clinically stable without focal neurologic abnormalities.

### Case 2

A 34-year-old woman presented with left-sided weakness, finger agnosia, tinnitus, and right homonymous hemianopsia in July 1990. CT evaluation showed a lobulated mass  $6.0 \times 4.5 \times 4.0$  cm of heterogeneous increased density, partially calcified in its periphery, that involved the right frontotemporal region with moderate mass effect (Fig 2A). After contrast administration, an eccentric, serpigi-

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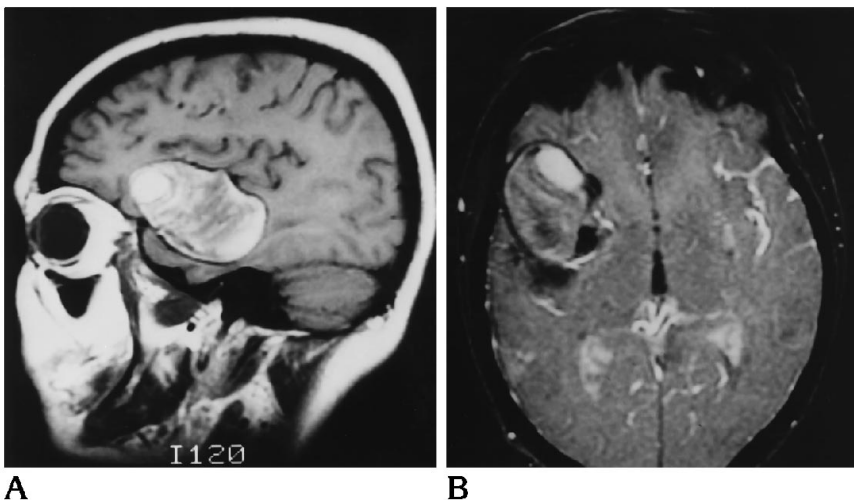
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Fig 1. A, T1-weighted sagittal image. Large right temporal lobe mass with regions of heterogeneous increased and decreased signal, representing various degradation products of hemoglobin.

B, Axial gradient-echo image. No central flow can be identified. Regions of increased signal that consist of extracellular methemoglobin correlate with regions of increased signal on T1 (A).



nous enhancing channel as well as peripheral enhancement equivalent to a “target sign” were visible (Fig 2B). MR confirmed the CT findings and demonstrated a lobulated mass of complex signal representing variable degrees of hemoglobin degradation products. Standard spin-echo imaging showed serpentine regions of flow void (Fig 2C). The signal intensity of these regions increased on gradient-echo images and represented flow. An angiogram showed a tortuous ectatic vascular channel arising from a sylvian branch of the right middle cerebral artery distally and ultimately supplying a distal posterior parietal cortical artery (Fig 2D–G). Flow through the ectatic channel was extremely slow, and displacement of normal middle cerebral artery branches was compatible with the large thrombotic mass identified on CT and MR.

A Tracker catheter (Target Therapeutics, Fremont, Calif) was placed coaxially through a 7F guiding catheter into the middle cerebral artery branch supplying the aneurysm. Amobarbital (40 mg) injected into the ectatic vascular channel did not change the patient’s neurologic status. Initially, occlusion of the feeding vessel immediately proximal to the aneurysm neck was attempted with detachment of two helical coils, 3 mm and 5 mm (Target Therapeutics). The coils, however, migrated into the proximal portion of the serpentine vascular channel of the aneurysm, and occlusion was not obtained. Coils were initially used because of the potential risks of bucrylate reflux in the more proximal lenticulostriate branches.

The Tracker catheter was then advanced into the entrance of the aneurysm, which was occluded with direct injection of bucrylate into the proximal aspect of the aneurysm. The previously placed coils, which were freely moving in the proximal aspect of the serpentine vascular channel, were immobilized and incorporated within the bucrylate. Control angiograms immediately after embolization and 3 months later demonstrated complete thrombosis of the aneurysm. A follow-up CT scan, MR, and an MR angiogram 1 year later confirmed complete thrombosis of the aneurysm. She progressively improved with only

elements of a right homonymous hemianopsia as residua as of January 1992.

### Case 3

For 15 years a 44-year-old, right-handed man experienced dysequilibrium, which progressively worsened over a 1-year period. Neurologic examination revealed a cluster of neurologic signs localized to the right brain stem and cerebellum, including right-sided appendicular ataxia, absent right corneal reflex, right-sided peripheral facial weakness, and right-sided hearing loss. Extraocular movement testing demonstrated breakdown of smooth pursuits and a lack of saccadic eye movements. Nystagmus was present with full excursions in all directions. A CT scan demonstrated a  $5.0 \times 4.0$ -cm mass of increased heterogeneous attenuation and partial peripheral calcification in the mid-posterior fossa with its epicenter at the level of the pons. After contrast administration, an enhancing, eccentric, serpentine tubular structure and peripheral ring were visible. On MR, this mass had a complex heterogeneous signal that consisted of lamellated thrombus and an eccentric, slow-flowing, ectatic vascular channel that was consistent with a partially thrombosed giant aneurysm displacing the pons and midbrain anteriorly and the vermis and cerebellum posteriorly (Fig 3A). Surgical clipping was unsuccessful, as no identifiable neck could be found and subsequent angiography failed to define a definite neck. The vertebral artery just distal to the origin of the posterior inferior cerebellar artery made an abrupt transition to an ectatic, irregular vascular channel that was tortuous and filled the basilar artery (Fig 3B and C).

The right vertebral artery was occluded with a balloon just distal to the origin of the posterior inferior cerebellar artery for 15 minutes. During occlusion, the patient was neurologically stable with no signs of clinical deterioration. Selective injections of the right internal carotid artery demonstrated a patent posterior communicating artery filling the posterior cerebral arteries and retrograde filling of

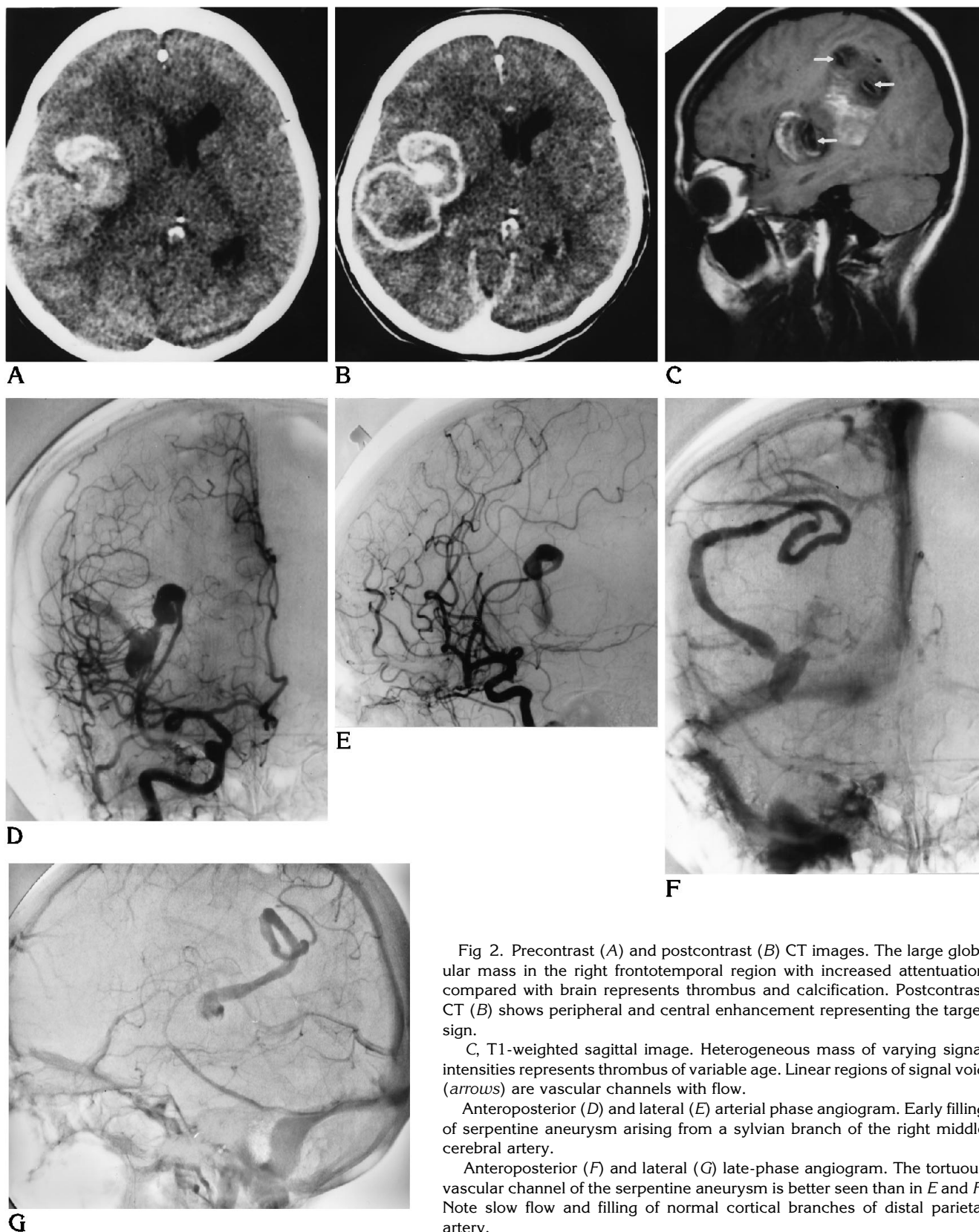


Fig 2. Precontrast (A) and postcontrast (B) CT images. The large globular mass in the right frontotemporal region with increased attenuation compared with brain represents thrombus and calcification. Postcontrast CT (B) shows peripheral and central enhancement representing the target sign.

C, T1-weighted sagittal image. Heterogeneous mass of varying signal intensities represents thrombus of variable age. Linear regions of signal void (arrows) are vascular channels with flow.

Anteroposterior (D) and lateral (E) arterial phase angiogram. Early filling of serpentine aneurysm arising from a sylvian branch of the right middle cerebral artery.

Anteroposterior (F) and lateral (G) late-phase angiogram. The tortuous vascular channel of the serpentine aneurysm is better seen than in E and F. Note slow flow and filling of normal cortical branches of distal parietal artery.

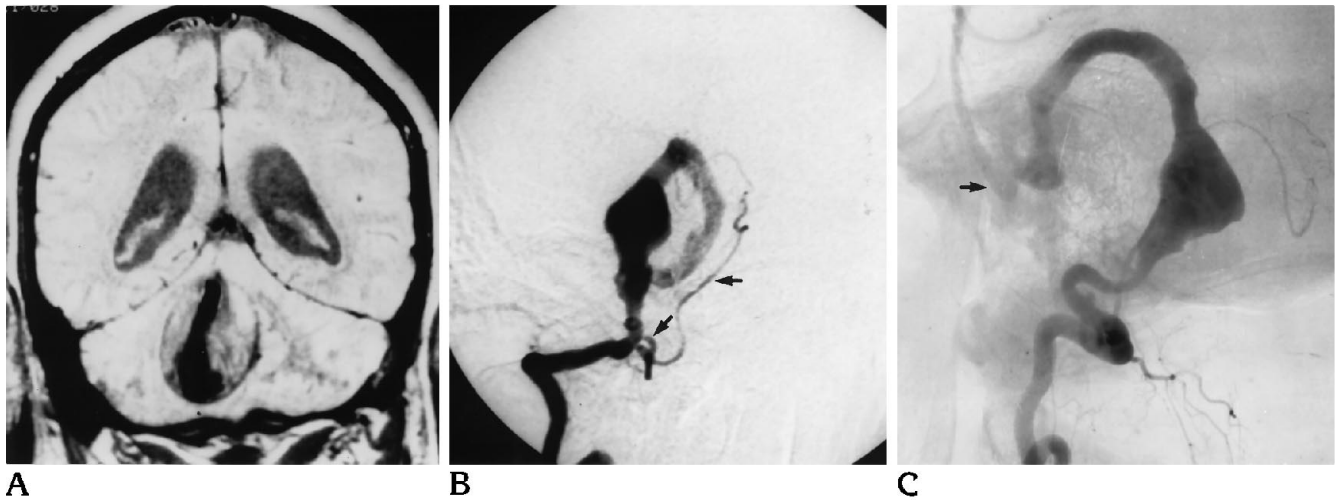


Fig 3. A, T1-weighted coronal image. The large midline posterior fossa mass of heterogeneous signal represents thrombus of various age, and the linear region of flow void represents flow in a vascular channel.

Anteroposterior (B) and lateral (C) arterial phase angiogram. Filling of ectatic vascular channel involving the distal vertebral artery distal to the posterior inferior cerebellar artery (arrows in B demonstrate the posterior inferior cerebellar artery), and ending at the base of the basilar artery (arrow in C) corresponds to the vascular channel identified by MR.

the distal basilar artery. A left occipital artery injection filled the left vertebral artery, which subsequently filled the basilar artery. The patient underwent ligation of the right vertebral artery distal to the posterior inferior cerebellar artery origin after the angiogram without complications. His symptoms slowly abated and have not recurred.

#### Case 4

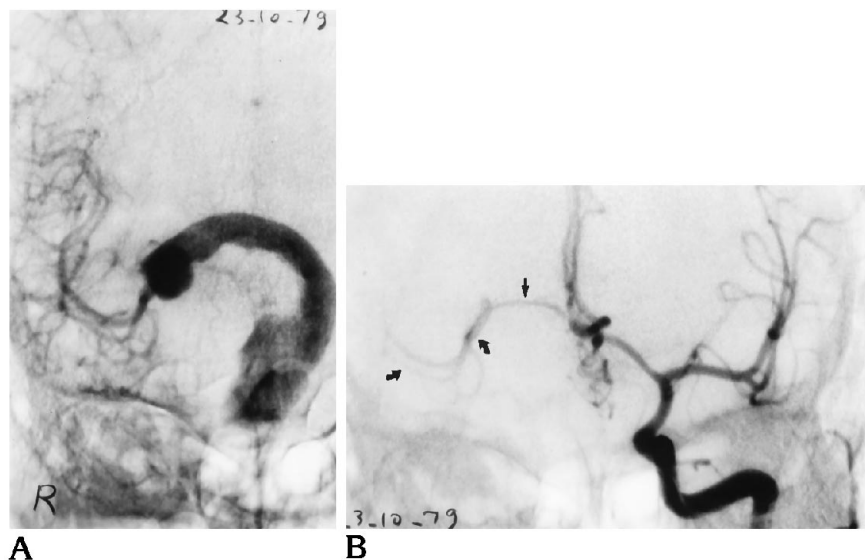
A 20-year-old man was referred to the University Hospital, University of Western Ontario (London, Canada) for further evaluation of a progressive left homonymous hemianopsia. A CT scan and angiogram from the referring

institution showed a giant,  $5.5 \times 4.5$ -cm, predominantly thrombosed aneurysm of the right supraclinoid carotid artery extending to the origin of the middle cerebral artery (Fig 4A and B). The anterior communicating artery was patent, and there was contralateral filling of the right cerebral circulation with cross-compression testing.

The patient underwent a right superficial temporal artery-middle cerebral artery bypass in October 1979 and balloon occlusion of the right internal carotid artery below the ophthalmic artery 5 days later. A control angiogram in April 1980 confirmed complete thrombosis of the aneurysm and substantial reduction of the mass effect. The patient was neurologically intact except for a residual left homonymous hemianopsia.

Fig 4. A, Anteroposterior arterial phase of right internal carotid angiogram. Large ectatic vascular channel involving the supraclinoid segment of the internal carotid artery. Note filling of normal M1 and M2 branches of the middle cerebral artery.

B, Anteroposterior arterial phase of left internal carotid angiogram. Same patient as in A. Contralateral filling of normal right A1 segment (straight arrow) and M1 and M2 branches of the right middle cerebral artery (curved arrows). Mass effect of the thrombosed portion of the aneurysm demonstrated by superior displacement right A1 and M1 segments.



### Case 5

In August 1991 a fever developed in a 14-year-old, right-handed boy. Evaluation at a local emergency room included a plain radiograph sinus series that showed a calcified mass within the cranial vault. Neurologic examination was normal with intact cranial nerves and motor systems. The sensory system was preserved, and deep tendon reflexes were symmetric. A CT scan showed a 10.0 × 9.0 × 8.0-cm heterogeneous mass of increased density with central and peripheral calcifications in the middle cerebral artery distribution extending frontally (Fig 5A). Contrast examination showed a large eccentric, ectatic vascular channel and peripheral rim enhancement (Fig 5B). An angiogram demonstrated a partially thrombosed giant aneurysm involving the left distal M1, M2, and M3 segments with associated avascular mass that displaced the surrounding vasculature (Fig 5C and D). The aneurysm lumen extended throughout a thrombosed portion of the aneurysm and filled a distal posterior parietal branch on the late phases of the arteriogram (Fig 5E and F). Several lenticulostriate vessels originated from the left M1 segment just proximal to the aneurysm origin. The patient was then referred to the Barrow Neurological Institute in Phoenix, Ariz (1).

In October 1991 an angiogram again showed the giant left middle cerebral artery serpentine aneurysm, and the patient underwent a left double superficial temporal artery-middle cerebral artery bypass. The distal outflow pathway of the middle cerebral artery aneurysm was clipped, and a ventriculostomy tube was placed. A control angiogram the next day demonstrated patency of the anastomoses and filling of the middle cerebral artery branches distal to the aneurysm via the superficial temporal artery. The previously identified serpiginous lumen, which coursed through the aneurysm from a proximal, more spherical portion of the aneurysm, had thrombosed. The proximal spherical portion, however, remained patent.

On the ninth postoperative day, a second craniotomy was performed, and the proximal aneurysm neck was clipped. A partial thrombectomy was also performed without complications. After surgery, the patient remained neurologically intact. A control angiogram 2 days after the second surgery did not reveal any residual aneurysm lumen (Fig 5G) and showed filling of the distal middle cerebral artery branches via the superficial temporal artery-middle cerebral artery anastomoses (Fig 5H). The patient was discharged in good neurologic condition (1).

### Discussion

Giant serpentine aneurysms have been defined as giant, partially thrombosed aneurysms (greater than 25 mm) with tortuous vascular channels that have a separate entrance and outflow pathway (2-7). Previous pathologic reports (2, 4-13) describe the aneurysms 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 (2, 8, 13). Extensive laminated thrombus, which may or may not be calcified, is present within the aneurysm and contains vascular channels. These 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 (2, 6-9, 11-13). The canals may be central or eccentric within the aneurysm with small branching channels that end blindly. Flow through the vascular channels is typically slow (3-5, 10, 13) and supplies distal branches of the cerebral vasculature to vital or nonvital areas of the brain (2-9, 12, 13). In the literature we found 33 cases of giant aneurysms, including the 5 presented here, that fulfill the criteria of giant serpentine aneurysms (Table 1).

Based on 29 of 33 cases in which clinical information is available, the initial presentation of patients with giant serpentine aneurysm is of an intracranial mass rather than intracranial hemorrhage and is dependent on aneurysm location. The predominant signs and symptoms are summarized in Figure 6 and are headache, 72% (21 of 29); hemiplegia and hemiparesis, 55% (16 of 29); visual disturbance, 48% (14 of 29); cranial nerve palsy, 31% (9 of 29); dysphasia and aphasia, 27% (8 of 29); nausea and vomiting, 24% (7 of 29); seizure, 21% (6 of 29); papilledema, 14% (4 of 29); mental deterioration and depression, 14% (4 of 29); dysesthesia, 7% (2 of 29); and vertigo, 3% (1 of 29). Based on the 33 cases in the Table, giant serpentine aneurysms have been identified on the middle cerebral artery or its branch vessels in 51% (17 of 33) of the patients, on the vertebral artery in 18% (6 of 33), the posterior cerebral artery in 12% (4 of 33), the internal carotid artery in 12% (4 of 33), the anterior cerebral artery in 3% (1 of 33), and the posterior communicating artery in 3% (1 of 33). Most patients are male, 65.6% (21 of 32), with a mean age of 41 years (range, 14 to 69 years).

The radiographic findings of giant serpentine aneurysms are distinctive. Plain-film radiographs may show pineal displacement caused by mass effect, curvilinear calcifications, present in case 5, or erosive changes involving the skull base (2, 5, 8, 9, 11, 14, 15). CT demonstrates an oval or globoid mass of mixed density (Figs 2A and 5A). Heterogeneous regions of increased attenuation represent thrombus,

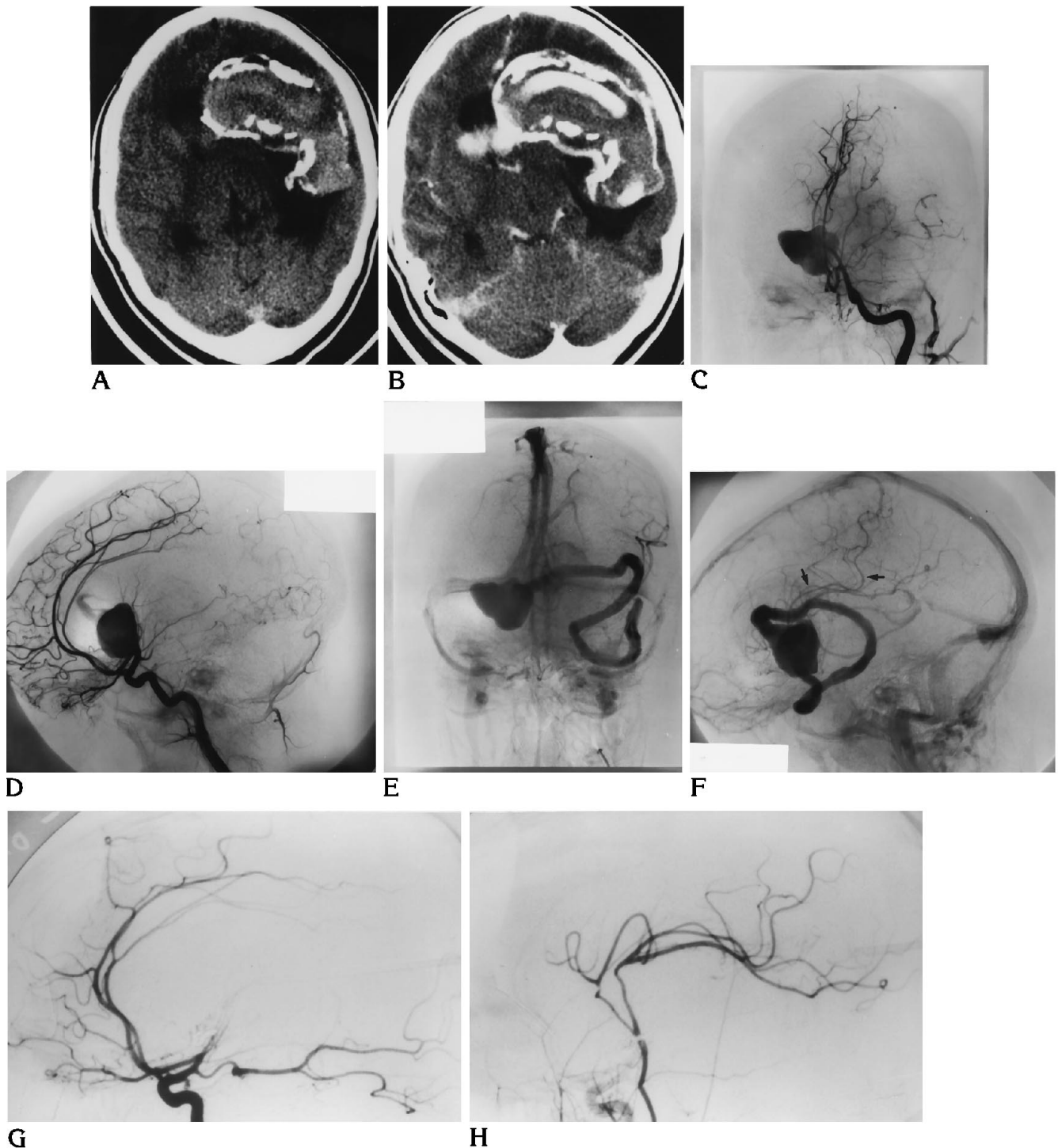


Fig 5. Precontrast (A) and postcontrast (B) CT images. The large globular mass in the left frontotemporal region with increased central attenuation compared with brain represents thrombus and peripheral calcification. The linear region of decreased attenuation enhances after contrast administration (B) and represents the serpentine vascular channel. Additionally there is peripheral enhancement target sign.

Anteroposterior (C) and lateral (D) arterial-phase angiogram. Early filling of serpentine aneurysm arising from left middle cerebral artery. Note marked mass effect with midline shift and displacement of the anterior cerebral arteries.

Anteroposterior (E) and lateral (F) late-phase angiogram better demonstrates the tortuous vascular channel of the serpentine aneurysm. Note slow flow and filling of normal cortical branches of the middle cerebral artery (arrows).

Final postoperative (G) lateral angiogram demonstrates the complete obliteration of the aneurysm lumen. Patency of the double extracranial-intracranial bypass is demonstrated (H).

## Literature review in patients with giant serpentine aneurysms

Author	Age, y/Sex	Symptoms	Signs	Bleed?	Aneurysm Site	Aneurysm Size, cm	Surgery	Outcome
Castelman and Kibbee (22) 1963	56/M	Headache, seizure, intermittent collapse	Symmetric hyporeactive reflexes	Yes	R ACA	7.0 × 5.0 × 5.0	None	Death secondary to small-cell lung carcinoma
Sadik et al (12) 1965	47/M	Headache, progressive visual loss, N/V, L-sided weakness	Decreased visual acuity, papilledema, L facial palsy, L hemiplegia	No	R MCA	8.5 × 5.5 × 5	Attempted resection, biopsy	Progressive neurologic deterioration with death 7th postoperative d
Cantu and LeMay (9) 1966	58/M	Headache	Dysnomia, dyscalculia, dyspraxia, finger agnosia	No	R MCA	8.0 × 5.5 × 5.5	Clipping, resection	Dysphasia
Terao and Muraoka (13) 1972	38/M	Headache, N/V, dysphasia, weakness L arm and leg	L hemiparesis, papilledema	No	R MCA	8 × 5.5 × 6.0	Aspiration curettage	Death secondary to R hemispheric stroke 6th postoperative d
Lukin et al (21) 1975	67/M	Headache, depression	Expressive dysphasia, R hemiparesis	...	L MCA	6	Coating	Expressive dysphasia, R hemiparesis
Case 1								
Case 2	30/M	Headache, dysarthria	Mild R central 7th CN palsy, partial R hemiparesis	...	L MCA	...	Resection	Mild diplopia
Segal and McLaurin (6) 1977								
Case 1	39/M	Headache, vertigo, blurred vision	Papilledema	No	R MCA	3.8 × 3.6 × 2.0	Resection	Partial L homonymous hemianopsia
Case 2	30/M	Headache, aphasia, depression	R central 7th CN palsy, R hemiparesis, receptive aphasia	No	L MCA	6.5 × 6.0 × 5.0	Resection	Persistent diplopia
Ammerman and Smith (16) 1977	20/M	Seizures, R-sided weakness	R hemiparesis	...	L MCA	...	STA-MCA anastomosis, aneurysm trapping	Seizures
Fodstad et al (2) 1978	27/F	Diplopia, L hemifacial paresthesias, L ocular pain	Partial L 3rd and 6th CN palsy, L hemifacial hypesthesias and hypalgesia	...	L ICA, developed after clipping of saccular aneurysm	7.5	L ICA ligation, partial resection	Partial L ophthalmoplegia 5th CN palsy
Pinto et al (17) 1979								
Case 12	...	Diplopia	...	...	R ICA	...	...	...
Tomasello et al (7) 1979	40/M	Headache, N/V	L central 7th CN palsy, L hemiparesis	Yes	R MCA temporal branch	6.5 × 5.0 × 4.5	Aneurysm trapping, resection	Neurologically intact
Patel et al (5) 1981	33/F	Headache, L weakness and hypoesthesia, vomiting	Mild L hemiparesis, homonymous hemianopsia	No	R MCA	...	Attempted resection, common carotid occlusion	Mild L hemiparesis, L hemianopsia (Table continues)

Table continued

Author	Age, y/Sex	Symptoms	Signs	Bleed?	Aneurysm Site	Aneurysm Size, cm	Surgery	Outcome
Kuwabara et al (26) 1981	28/M	...	...	...	Bilateral vertebral	1.5 × 0.9 × 0.8	Ventriculoperitoneal shunt	...
Fukamachi et al (10) 1982	48/F	Headache, R-sided weakness	R hemiparesis, R homonymous hemianopsia	No	L PCA	6.0 × 5.0 × 4.0	Aneurysm trapping, partial resection	Progressive clinical deterioration, death postoperative after pulmonary emboli
Whittle et al (19) 1982	59/M	Headache, seizures	Facio-brachial paresis, dysphasia	No	L MCA	5.0 × 3.0	Aneurysm wrapping	Progressive clinical deterioration, death postoperative after pulmonary emboli
Vlahovitch et al (18) 1982	32/F	...	...	...	L carotid	...	Anastomosis, occlusion ICA	...
Case 3	48/M	...	...	...	PCom	6.0 × 1.7	Occlusion PCom	...
Case 4	20/M	Headache, mental deterioration	L hemiparesis, L homonymous hemianopsia	No	R PCA	...	STA/PCA anastomosis, aneurysm trapping, resection	Complete recovery
Chang et al (24) 1986	50/M	Headache, double vision, nausea	Neurologically intact, chronic renal failure	No	L vertebral	1.7 × 2.5 × 1.5	None	Death from intracranial hemorrhage presumed to be secondary to aneurysm rupture
Terada et al (20) 1988	27/F	Headache, visual blurring, dysarthria, vomiting	L central 7th CN palsy, L hemiparesis, papilledema	No	R MCA	8.0 × 7.0 × 6.0	Resection	Neurologically intact
Li et al (11) 1988	69/F	Dysphasia	R homonymous hemianopsia, R hemiparesis, sensory motor aphasia	No	L PCA	6.0 × 4.0 × 4.5	None	Progressive deterioration leading to death in 5 m
Belec et al (18) 1988	53/F	Seizures	Hyperactive R biceps reflexes	No	L MCA	3.5 × 2.0 × 5.0	Aneurysm trapping, resection	Minimal R hand fine motor dysfunction, R arm hyperreflexia
Haddad and Haddad (3) 1988	51/F	Headache, Lower CN dysfunction, ataxia, vomiting	L hemiparesis, truncal ataxia, nystagmus	No	R vertebral	4.5	Vertebral occlusion test followed by trapping and resection	Improvement of neurologic findings
Sugital et al (25) 1988	Case 1							(Table continues)



Table continued

Author	Age, y/Sex	Symptoms	Signs	Bleed?	Aneurysm Site	Aneurysm Size, cm	Surgery	Outcome
Case 2	61/M	Headache, dysarthria	L hypesthesias, and hemiparesis	No	L vertebral	3.5	Two-stage procedure: 1, proximal ligation; 2, distal ligation resection	Stabilization of defects after some improvement
Kumabe et al (4) 1990 Case 1	39/M	R ocular pain	Neurologically intact	No	L MCA	5	None	Progressive aneurysm growth with neurologic deterioration leading to death
Case 2	53/F	Headache, diplopia, nausea	R medial longitudinal fasciculus syndrome	No	Basilar-L PCA	2.5	None	Progressive aneurysm growth with neurologic deterioration leading to death
Present study Case 1	44/F	Seizures	Neurologically intact	No	R MCA	4.5 × 4 × 3.7	None	Spontaneous thrombosis of aneurysm without developing neurologic defects
Case 2	34/F	Headache, seizures	Mild L hemiparesis, L homonymous hemianopsia	No	R MCA	6 × 4.5 × 4.0	Endovascular occlusion of feeder with helical coils and barytate	Neurologically intact
Case 3	44/M	Loss of fine motor control R hand	Peripheral 7th CN palsy, R appendicular ataxia, R decreased motor tone	No	R vertebral	5.0 × 4.0	Ligation R vertebral artery	Clinical recovery
Case 4	20/M	Progressive visual symptoms	R optic nerve neuropathy	No	R supraclinoid ICA	5.5 × 4.5	STA-MCA anastomosis, balloon occlusion ICA and ophthalmic artery	Complete thrombosis of aneurysm, neurologically intact
Case 5	14/M	None, calcified intracranial mass found incidentally on sinus series	Neurologically intact	No	MCA	10.0 × 9.0 × 8.0	Two-stage procedure: 1, double L STA-MCA bypass, distal aneurysm clipping; 2, proximal clipping and thrombectomy	Neurologically intact

Note.—ACA indicates anterior cerebral artery; CN, cranial nerve; ICA, internal carotid artery; MCA, middle cerebral artery; N/V, nausea and vomiting; PCA, posterior cerebral artery; PCOM, posterior communicating artery; and STA, superficial temporal artery.

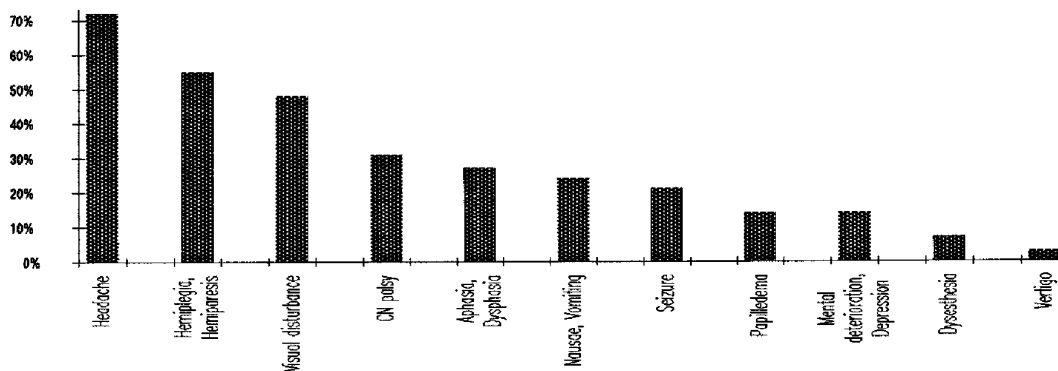


Fig 6. Percentage of patients with signs or symptoms, based on 29 patients in the Table.

and linear regions of slightly decreased attenuation represent a patent vascular channel on nonenhanced scans (Fig 5A). After contrast administration, a target sign may be observed (Figs 2B and 5B) and represents enhancement of the vascular capsule and serpentine vascular channel, (3–5, 8, 10, 16–19) as seen in four of our five cases.

The MR findings of giant serpentine aneurysms have not been extensively reported but consist of a mass lesion with a complex signal that represents various stages of hemoglobin degradation (Figs 1A, 2C, and 3A), slow flow, and flow void regions (8, 20) (Figs 2C and 3A). Belec et al (8) describe a case in which MR demonstrated a hyperintense signal in the center of the aneurysm surrounded by a hypointense ring. The aneurysm was clearly separated from normal parenchyma, and the vascular channel was visible. Gradient-echo imaging, with its sensitivity to slow flow, used in conjunction with standard spin-echo images, may better define the slowly flowing vascular channels. A primarily T1-weighted pulse sequence, however, is sensitive to the T1-shortening effects of methemoglobin and may limit its usefulness (Fig 1B). Phase-contrast MR angiography, which is insensitive to the T1-shortening effects of methemoglobin, may be used to evaluate the serpentine vascular channel, although its efficacy needs further clinical evaluation. Angiography is pathognomonic of a giant serpentine aneurysm (2–7, 10, 12). The typical features of giant serpentine aneurysms previously discussed are well demonstrated in all our cases (Figs 2D–G, 3B and C, 4A, and 5C–F).

The cause of giant serpentine aneurysms is unclear. They may develop from saccular aneurysms by continued expansion (12, 14, 15, 21) or arise from fusiform aneurysms (7, 16,

20). In 1979 Tomasello et al (7) reported a small fusiform aneurysm of the posterior temporal branch of the middle cerebral artery that progressed to a giant serpentine aneurysm over 5 years. Although the pathogenic mechanisms were unclear, this report suggested that giant serpentine aneurysms may arise from fusiform aneurysms, which are most likely a degenerative response to arteriosclerosis, infection, or unknown causes. Many authors consider giant serpentine aneurysms as separate entities distinct from saccular and fusiform aneurysms (2, 3, 6) and possibly even congenital (5, 9, 13, 22). In 1978 Fodstad et al (2) reported a left intracavernous aneurysm that developed a serpentine channel 6 months after internal carotid ligation. The development of the serpentine channel in the original globular aneurysm was attributed to the Coanda effect or boundary wall effect, in which the direction of a jet stream, produced by a relative stenosis in a vessel, is deflected to one wall of the vessel and stabilized by changes of the counter current flow in the relative low-pressure zones along the walls of the vessel immediately distal to the stenosis. Modified from mechanical fluid hemodynamics, the Coanda effect was first considered a mechanism resulting in some postaneurysm clipping complications by Robinson and Roberts in 1972 (23). This mechanism of giant serpentine aneurysm formation has also been proposed by other authors (4, 19). We concur with the conclusion that giant serpentine aneurysms are not derived from saccular aneurysms because they do not arise at vessel bifurcations or origins of vestigial vessels, lack an anatomic neck, and have separate entrance and exit sites of the vascular channel (3, 6).

Haddad et al (3) further conclude that giant serpentine aneurysms do not represent a sub-

group of fusiform aneurysms in which mural thrombosis has reduced the lumen to a serpentine channel. In their cases, the feeding and draining arteries did not have the typical funnel-shaped appearance of fusiform aneurysms. Unlike giant serpentine aneurysms, fusiform aneurysms are most commonly encountered in the vertebrobasilar and carotid arteries.

The therapeutic options for giant serpentine aneurysms depend on the presentation of the aneurysm, location, and anatomic features of the feeding and draining vessels (4, 7, 10, 16, 24, 25). Untreated aneurysms have an unpredictable course. Some aneurysms grow and cause progressive neurologic deterioration (4, 8, 20); others may remain stable. Spontaneous thrombosis may occur as seen in case 1. Intracranial hemorrhage is unlikely secondary to the thickened aneurysmal wall and extensive thrombosis; however, in 1988 Terada et al (20) reported a patient who died after intracranial hemorrhage presumed to be secondary to aneurysm rupture. Before advanced endovascular techniques developed, and extracranial and intracranial bypass was an option, perioperative morbidity and mortality for giant serpentine aneurysms was 30% to 35%. The mortality rate related to intraoperative hemorrhage and thrombosis of the slowly flowing vascular channel that supplied vital distal branches of the parent artery, which were untested before surgery, approached 17%.

The need to provide a functional bypass before definitive therapy of the aneurysm can be determined by an endovascular balloon occlusion test (cases 3 and 4). The entrance channel to the aneurysm is temporarily occluded by a balloon, and the patient's neurologic status is closely monitored. A change in neurologic findings indicates that the vascular channel supplies vital distal branches of the parent artery without sufficient collateral filling of the distal vasculature and that a functional bypass is needed (25). Standard balloon occlusion testing at this institution is performed in a fully heparinized patient for 15 minutes. During the occlusion period, continuous neurologic evaluation and electroencephalographic monitoring is performed and compared with baseline. Additionally, patency of the circle of Willis is determined by digital angiography. Subselective injections of amobarbital into the aneurysm provides complimentary information to the balloon occlusion test with a functional evaluation

of the brain parenchyma supplied by the distal outflow vessels as performed in case 2. If the balloon occlusion and amobarbital tests are negative (outflow pathway of the serpentine channel does not supply vital distal cerebral vessels), permanent occlusion can be obtained with detachable balloons or liquid polymerizing agents (case 2). Standard coils currently available are not used initially for occlusion of large arterial vessels, because they do not consistently provide immediate complete occlusion, thereby increasing the risk of thrombotic emboli. However, in cases in which the risk of possible bucrylate reflux into more proximal vessels supplying eloquent areas of brain parenchyma is high, coils may be considered as a therapeutic option. If a functional bypass is indicated, the aneurysm can be occluded permanently after the bypass procedure, obviating the need for a second operation.

Many surgical procedures have been used to treat giant serpentine aneurysms: simple coating or wrapping (19, 21), thrombectomy or resection (3, 6, 7, 9-13, 21, 25), thrombectomy or resection after a functional bypass procedure (1, 16, 24) as performed in case 5 (Fig 5H), carotid ligation with or without partial resection (2, 5, 18), carotid occlusion after a functional bypass procedure as performed in case 4, and ventriculoperitoneal shunting to relieve hydrocephalus in inoperable cases (26).

Therapeutic options must be selected carefully and depend on initial presentation, location, and anatomic features of the aneurysm. Before therapy, be it endovascular, surgical, or a combined approach, adjunct diagnostic procedures including temporary balloon occlusion testing and amobarbital evaluation are necessary to determine whether the serpentine vascular channel supplies vital distal cerebral arteries. Failure to provide a functional bypass before resection or embolization may result in a progressive downhill postoperative course (7, 12, 13, 16, 24, 25). If the vascular channel does not supply vital distal branch arteries or a functional bypass is provided, endovascular occlusion of the proximal serpentine channel is attractive and may obviate the need for surgical intervention. However, after endovascular occlusion of large aneurysms, persistent symptoms may require thrombectomy or aneurysm resection because of mass effect. Additionally, in determining whether endovascular occlusion alone or a surgical approach with or without

concomitant endovascular occlusion is to be performed as the initial therapy of serpentine aneurysms, the available resources and experience levels of the responsible physicians need to be considered. Complete or near-complete recovery can be expected (1, 16, 24), after the appropriate diagnostic evaluations and a multi-disciplinary discussion of available therapeutic options as demonstrated by cases 4 and 5.

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