Intraoperative Functional and Perfusion Monitoring During Surgery for Giant Serpentine Middle Cerebral Artery Aneurysms

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Key words
- Cerebral revascularization
- Giant aneurysm
- Indocyanine-green
- Microvascular bypass
- Motor-evoked potentials monitoring
- Serpentine aneurysm
- Ultrasound flowmetry

Abbreviations and Acronyms
CT: Computed tomography
DSA: Digital subtraction angiography
GOS: Glasgow Outcome Scale
ICGA: Indocyanine-green angiography
MCA: Middle cerebral artery
MEP: Motor evoked potentials
MRI: Magnetic resonance imaging

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BACKGROUND: **Giant serpentine aneurysms are a rare entity, which can be managed using either endovascular or surgical techniques. Although the perioperative morbidity and mortality have decreased since the development of bypass revascularization procedures, their surgical treatment is still challenging. Intraoperative functional and perfusion monitoring techniques can be precious to make better decisions and improve outcomes.**

CASE DESCRIPTION: **We report on the case of a giant, unruptured, partially thrombosed, serpentine middle cerebral artery aneurysm that was treated with partial endovascular coiling of intra-aneurysmal vascular channels, surgical resection of the aneurysm, and end-to-end M1-temporal M2 anastomosis.**

CONCLUSIONS: **Intraoperative continuous motor evoked potentials monitoring, flowmetry, and indocyanine-green angiography provide precise and reproducible information about cerebral function and perfusion, respectively, allowing for more rational decision making during surgery for these challenging malformations.**

INTRODUCTION

The treatment of giant middle cerebral artery (MCA) aneurysms by either endovascular means or direct clipping is difficult to achieve and carries a high risk of mortality and morbidity (7, 15, 53). Giant aneurysms of the MCA represent 13%—15% of all giant intracranial aneurysms (7, 13). Approximately 4% of the aneurysms located on the MCA are giant and most frequently occur at the bifurcation. The natural history of these aneurysms carries a poor prognosis (14, 35). The treatment of large and giant aneurysms is therefore generally indicated (17, 27, 29, 53). In rare cases a giant aneurysm may be partially thrombosed, cause mass effect, and have a residual serpiginous vascular channel that ends in a distal branch of the feeding artery. These aneurysms are located on the MCA in about 50% of cases and often present with rapid neurological deterioration secondary to an ischemic evolution (11). At admission these malformations may be misdiagnosed as brain tumor or hemorrhagic stroke (16) because they usually present with mass-related symptoms and neuroimaging shows well-circumscribed edges, perilesional edema, and obvious brain shift (1, 6, 11, 12, 39). The available therapeutic options for serpentine aneurysms include surgery, endovascular therapy, or their combination. Direct microsurgical clipping of giant serpentine aneurysms is associated with morbidity and mortality rates as high as 30%—35% (17, 31). The cerebral revascularization obtained by means of bypass surgery, alone or combined with endovascular approaches, has resulted in a reduced risk of cerebral ischemia and more satisfactory outcomes (10, 14, 24, 35, 37, 41-44, 49, 51). Intraoperative monitoring of motor evoked potentials (MEPs) combined with flowmetric measurements and indocyanine-green angiography (ICGA) enhances the capacity of rational decision making during such a complex surgical procedure. However, a thorough review of the pertinent literature showed that only rarely have ultrasound flowmetry and MEPs monitoring been used.

We describe for the first time the simultaneous use of intraoperative MEPs, ultrasound flowmetry, and ICGA during bypass surgery to treat a giant serpentine aneurysm of the bifurcation of the MCA. Neurological and angiographic studies showed the complete disappearance of the malformation with an effective restoration of the cerebral blood flow and a good neurological outcome.

CASE REPORT

A 66-year-old obese and hypertensive woman presented at the emergency department complaining of a gradually worsening left hemiparesis, intermittent headache, and psychomotor impairment. Neuroradiological workup with angio-CT, MRI, and digital subtraction angiography (DSA) showed a 5.5-cm unruptured giant partially thrombosed serpentine MCA aneurysm, surrounded by adjacent edema with midline shift (**Figure 1**).

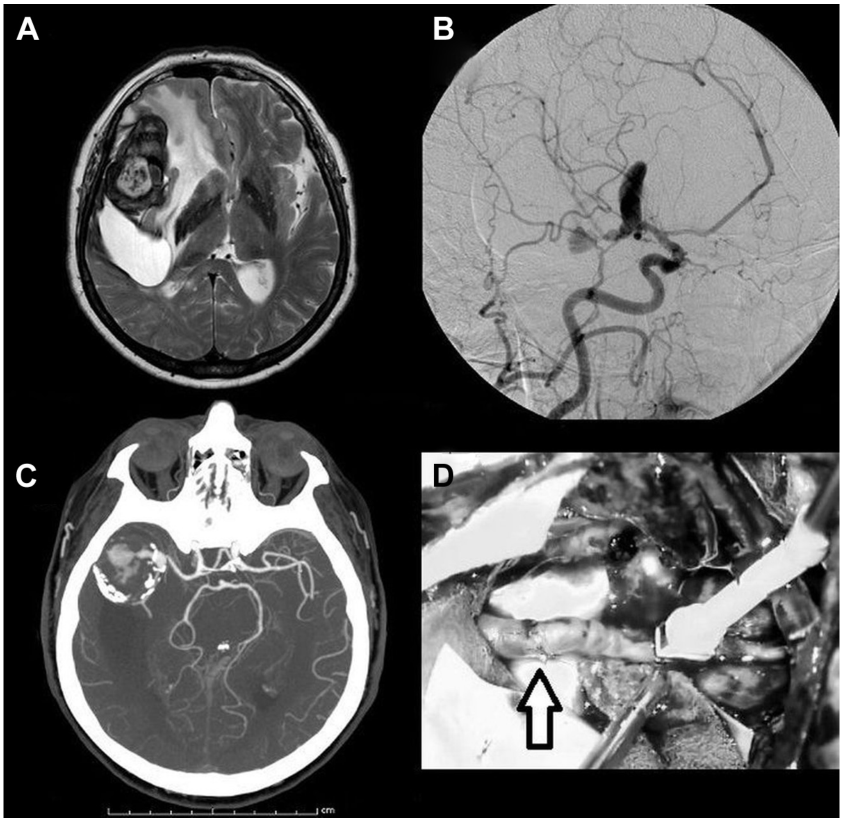

Figure 1. (**A**) T2-weighted axial magnetic resonance imaging (MRI) showing a giant thrombosed aneurysm dome surrounded by cerebral edema and a posterior cyst. (**B**) On digital substraction angiography (DSA) the aneurysm is only partially filled, with a serpentine pattern. (**C**) Angio—computed tomography showing calcifications of the thrombosed part. (**D**) Intraoperative view of the microanastomosis between M1 and temporal M2, while checking the flow on distal M2.

A revascularization procedure with aneurysm thrombectomy and clip reconstruction was scheduled. Two days before surgery, the patient underwent an endovascular treatment with partial coiling of intra-aneurysmal vascular channels in order to reduce intraoperative bleeding, especially during thrombectomy. In order to minimize the risk of new infarction, neuroradiologists embolized only the areas of the aneurysm that were safely distant from the channels potentially feeding the brain parenchyma.

A frontotemporal craniotomy was performed (13). Intraoperative motor evoked potentials (MEPs) were monitored continuously. After exposing and trapping the aneurysm with temporary clipping, we opened its thickened wall and performed a decompressive thrombectomy with the ultrasonic surgical aspirator. Intraoperative clip reconstruction of the MCA bifurcation proved to be impossible due to the stiffness of the wall of the aneurysm neck. For this reason we decided to resect the aneurysm cutting the inflowing M1 just proximal to the aneurysm and both the outflowing M2 vessels right after their exit. The dissected superficial temporal artery was too small and not suitable for a high-flow revascularization procedure. Therefore we performed an end-to-end M1 to temporal M2 anastomosis with a 9-0-monofilament nylon, obtaining a cerebral blood flow of 30 mL/min as measured by flowmeter microprobe. The frontal M2 artery was not revascularized on the basis of a sufficient backflow, as measured by intraoperative flowmetry (−12 mL/min) and confirmed by ICGA. Accordingly, MEP monitoring showed no variations of amplitude and latency during surgery. Total occlusion time of M1 was 56 minutes. The total intraoperative blood loss was 450 mL.

At postoperative day 2 the patient was awakened, and the neurological examination showed a left hemiparesis. A perfusion CT scan revealed a fronto-insular ischemic area. Angiographic control confirmed the patency of the M1-M2 anastomosis and the presence of collateral perfusion in the frontal cortex (**Figure 2**). At 1-year follow-up after rehabilitation the patient has only a mild hyposthenia of the inferior left limb, requiring occasional assistance (GOS 4).

DISCUSSION

The treatment of giant serpentine MCA aneurysms by endovascular means or direct clipping is seldom achievable and carries a high risk of mortality and morbidity (1, 49). Although different endovascular techniques have been introduced in the management of serpentine giant aneurysms, they cannot reduce the aneurysm mass effect nor properly deal with cases when insufficient collateral distal blood flow occurs (7, 21, 49). However, preoperative endovascular coiling of serpentine aneurysmal channels can be helpful in reducing intraoperative bleeding during thrombectomy. Anyway, endovascular treatment in such complex cases must be tailored according to the specific characteristics of anatomy and hemodynamics. Only 31 cases of giant serpentine aneurysms have been surgically treated (**Table 1**). Techniques of cerebral revascularization, alone or combined with clipping or endovascular approaches, offer an effective protection against cerebral ischemia, making aneurysm exclusion a safer procedure (7, 36). Before the availability of the bypass option, giant serpentine aneurysm surgery resulted in overall morbidity and mortality rates of 30%—35% (31). It has also been shown that aneurysm surgical resection associated with bypass surgery may, in selected cases, lead to successful aneurysm obliteration and mass effect symptoms relief, limiting the risks of postoperative ischemic complications

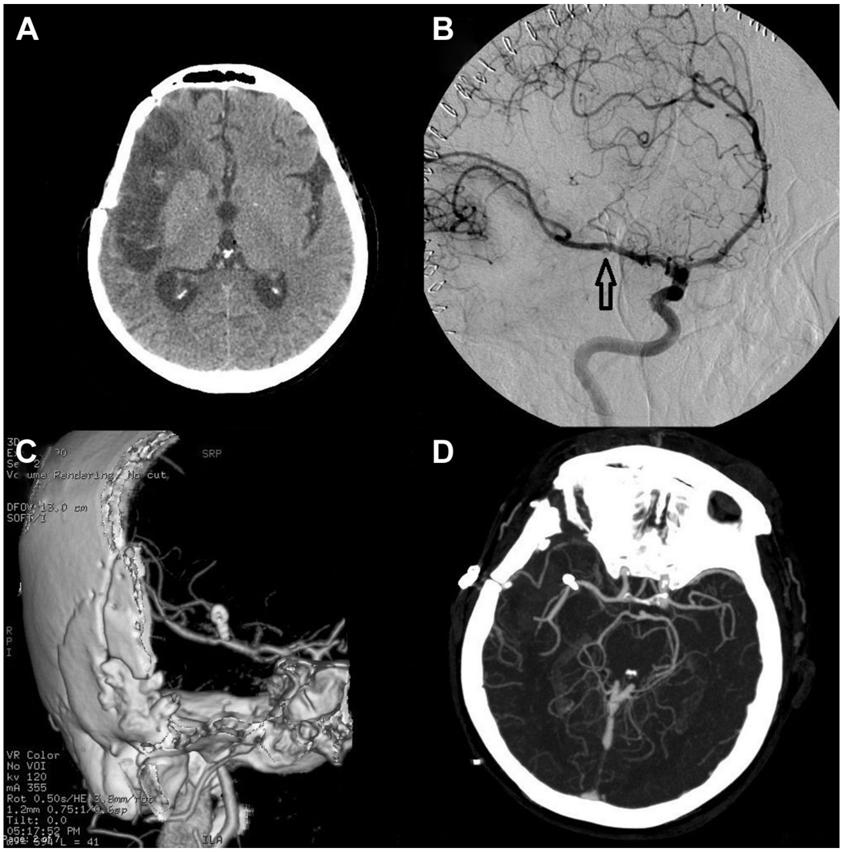

Figure 2. (**A**) Follow-up computed tomography (CT) scan showing a right insular infarction; (**B**) Postoperative DSA showing the bypass (*open arrow* pointing at microsuture site). (**C**) and (**D**) Postoperative angio-CT confirming the patency of middle cerebral artery anatomosis (clip is on the outflowing frontal branch of M2).

(1, 7, 28, 35, 36, 44). An adequate intraoperative monitoring of both neurophysiological functions and local cerebral blood flow measurements is mandatory during such a complex surgical procedure. Ultrasound flowmetry has been used in 3 out of 31 surgical cases reported in the literature and neurophysiologic monitoring only in one (see **Table 1**). In our case the stability of the recorded MEPs during M1 temporary clipping provided a powerful functional control during the micro-suturing procedure of the M1-temporal M2 anastomosis, regardless of the occlusion time, which might be prolonged. Moreover, flowmetric measurements combined with ICGA confirmed the patency of the high-flow bypass. ICGA is particularly useful to assess patency and flow in vessels that are difficult to explore with ultrasound flowmetry probe. In our case, ICGA was precious to identify the small perforators arising from the MCA, in order to carefully spare them during the trapping-bypass procedure, and the collateral circulation toward the frontal lobe. Interestingly, although ICGA does not provide a quantitative flow measure and flowmetry probe does not perfectly fit on very small vessels, they proved to be complementary when we detected a good collateral backflow along the frontal M2 branch (4, 17, 22). The combination of both techniques made us confident enough to avoid further and potentially dangerous procedures on the frontal M2 branch and prompted us to change our surgical strategy. The collateral circulation along with the M1-M2 anastomosis protected the cerebral hemisphere from developing a massive infarction, while the patient suffered only a mild fronto-insular ischemic injury. In the meantime, aneurysm excision achieved the immediate resolution of the mass effect accounting for the preoperative symptoms. The use of all three methods during a single complex surgical operation, which we report for the first time, can be of great value in selected cases. Actually, MEP, flowmetry, and ICG-angiography measure different entities. While MEP provides a continuous neurophysiological picture of the function of motor pathways, flowmetry quantifies the arterial flow and ICG-angiography provides a direct visualization of the vessels' patency. While flowmetry is able to measure the value of blood flow of sufficiently large intracranial vessels, it is not useful to monitor small perforators that don't properly fit on the flowmeter probe. In such cases ICG-angiography can better visualize the perforators' patency. However, a patent vessel is not necessarily a vessel with a proper blood flow, which can be impaired by a distal partial occlusion.

Although a complete revascularization should always be the first option, in our case the balance between risks and benefits led us to perform only a partial revascularization. This exceptional option must be taken into account only when both perfusion and functional monitoring techniques make the surgeon confident enough, and only after a careful balance of all the alternative possibilities. It is always difficult to understand which will be the optimal solution when dealing with such complex vascular malformations. In any case, surgeons should always adopt every available technique when they balance between risks and benefits in order to make the best decision for the patient's outcome.

CONCLUSION

Intraoperative MEPs, flowmetry, and indocyanine-green angiography provide precise and reproducible information about cerebral function and perfusion, respectively, allowing for more rational decision making during surgery. Intraoperative functional and perfusion monitoring is always advisable when dealing with such challenging giant vascular malformations.

Table 1. Giant Serpentine Aneurysms Surgically Treated: Literature Review

Authors, Year	Age (years), Sex	Location	Size (cm)	Presentation	Treatment	Intraoperative Monitoring	Outcome
Sadik et al., 1965 (38)	47, M	rt MCA	8.5 × 5.5 × 5	Headache, decreased visual acuity, lt hemiparesis	Resection	None	Died
Cantu & LeMay, 1966 (8)	58, M	rt MCA	8 × 5.5 × 5.5	Headache, dysnomia, dyscalculia, dys-praxia, finger agnosia	Clipping & resection		Dysphasia
Terao & Muraoka, 1972 (47)	67, M	lt MCA	8 × 5.5 × 6	Headache, dysphasia, lt hemiparesis	Aspiration curettage	None	Died
Lukin et al., 1975 (33)	67, M	lt MCA		Headache, expressive dysphasia, rt hemiparesis	Surgical wrapping		Expressive dysphasia, rt hemiparesis
	30, M	lt MCA		Headache, dysarthria, rt hemiparesis, central CN VII palsy	Resection		Diplopia
Segal & McLaurin, 1977 (39)	39, M	rt MCA	3.8 × 3.6 × 2	Headache, blurred vision, papilledema	Resection	None	lt Homonymous hemianopsia
	30, M	lt MCA	6.5 × 6 × 5	Headache, aphasia, central CN VII palsy, rt hemiparesis, receptive aphasia	Resection	None	Persistent diplopia
Ammerman & Smith, 1977 (3)	20, M	lt MCA		Seizure, rt hemiparesis	STA-MCA anastomosis, aneurysm trapping		Seizure disorder
Fodstad et al., 1978 (18)	27, F	lt ICA	7.5	Diplopia, partial CN III & VI palsy, hemifacial paresthesias	lt ICA ligation, partial resection	None	Partial lt ophthalmoplegia, CN V palsy
Tomasello et al., 1979 (48)	40, M	rt MCA	6.5 × 5 × 4.5	Headache, central CN VII palsy, lt hemi-paresis	Surgical aneurysmal trapping & resection		Neurologically intact
Fukamachi et al., 1982 (19)	48, F	lt PCA	6 × 5 × 4	Headache, rt hemiparesis, rt hemianopsia	Surgical aneurysm trapping, partial resection		rt Hemiparesis, rt hemianopsia
Whittle et al., 1982 (52)	59, M	lt MCA	5 × 3	Headache, seizure, faciobrachial paresis, dysphasia	Aneurysm wrapping	None	Died
Vlahovitch et al., 1985 (50)	32, F	lt ICA			Anastomosis, ICA occlusion		
Chang et al., 1986 (9)	20, M	rt PCA		Headache, lt hemiparesis, lt hemianopsia	STA-PCA anastomosis, aneurysm trapping & resection		Neurologically intact
Li et al., 1988 (32)	27, F	rt MCA	8 × 7 × 6	Headache, dysarthria, central CN VII palsy, lt hemiparesis	Resection		Neurologically intact
Haddad & Haddad, 1988 (23)	53, F	lt MCA	3.5 × 2 × 5	Seizure	Surgical aneurysm trapping & resection		Minimal rt hand fine motor dysfunction
Sugita et al., 1988 (45)	51, F	rt VA	4.5	Headache, lower CN dysfunction, lt hemiparesis, truncal ataxia, nystagmus	VA occlusion test followed by surgical trapping & resection	None	Improvement of neurological findings
	61, M	lt VA	3.5	Headache, dysarthria, lt hemiparesis	Proximal ligation, then distal ligation & resection	Ultrasound flowmeter	Unchanged neurological findings

Suzuki et al., 1992 (46)	39, M	rt MCA	6.5 × 3.7 × 4	Headache, partial CN VI palsy	STA-MCA bypass		Neurologically intact
Horowitz et al., 1994 (25)	19, M	lt MCA	6	Seizure	Sodium amobarbital testing, STA-MCA bypass, clipping of the parent vessel distal to the aneurysm		Neurologically intact with thrombosed aneurysm at 18-month follow-up
Isla et al., 1994 (26)	37, M	lt MCA	10 × 4 × 4	Seizure; recurrence: dysphasia, lethargy, rt hemiparesis	STA-MCA bypass & ICA ligation; recurrence after 2 years: clipping of lt MCA & aneurysm resection	None	Mild aphasia, rt hemiparesis
Aletich et al., 1995 (1)	20, M	rt ICA	5.5 × 4.5	Rt optic nerve neuropathy	STA-MCA anastomosis, balloon occlusion ICA & ophthalmic artery	None	Complete thrombosis of aneurysm, neurologically intact
	14, M	lt MCA	10 × 9 × 8	Neurologically intact	2 stages: 1) double lt STA-MCA bypass, distal aneurysm clipping; 2) proximal clipping & thrombectomy	None	Neurologically intact
Bakac et al., 1997 (5)	17, F	rt MCA	4 × 3 × 2.5	Headache, right ptosis, meningismus, SAH	Proximal & distal clipping of the aneu-rysm & resection		Neurologically intact, aneurysm formation on another MCA branch that was clipped
Lee et al., 1999 (30)	18, F	lt PCA		Headache, thrombosed aneurysm recanalizing over 3-week period	Surgical aneurysm trapping	None	Transient CN III palsy
Amin-Hanjani et al., 2006 (2)	14, M	MCA		Headache	STA-MCA bypass & clipping of parent artery	None	Transient hemiparesis, neurologically intact at 13-year follow-up
Van-Rooij et al., 2008 (49)	51, F	lt ACA		Frontal syndrome	BTO, endovascular aneurysm lumen occlusion w/glue, resection of thrombosed aneurysm		Neurologically intact
Lee et al., 2010 (31)	43,F	rt MCA	7 × 8 × 5	Severe headache, transient lt hand weakness	STA-MCA bypass, resection	None	Neurologically intact
González-Darder et al., 2011 (20)	35, F	rt MCA	5.5 × 3.9 × 3.3	Headache, lt hemiparesis, psychomotor impairment	High-flow bypass from the petrous internal carotid artery	SEP, MEP, microdoppler flowmeter	Neurologically intact
Senbokuya et al., 2012 (40)	38, M	lt Distal ACA	4	Headache	BTO, trapping, and resection	None	Neurologically intact
Moon et al., 2012 (34)	49, F	Distal ACA	4.5 × 1.8	Headache, intermittent loss of consciousness	Trapping, rt end to lt side A3 bypass	Microdoppler flow meter	Neurologically intact

M, male; rt, right; MCA, middle cerebral artery; lt, left; CN, cranial nerve; STA, superficial temporal artery; ICA, internal carotid artery; PCA, posterior cerebral artery; VA, vertebral artery; SAH, subarachnoid hemorrhage; ACA, anterior cerebral artery; BTO, balloon test occlusion; SEP, somatosensory evoked potential; MEP, motor evoked potential.

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