



Case Report

Brain ischemia due to direct vascular compression associate with rapid enlargement of unruptured middle cerebral artery aneurysm: A case report

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ABSTRACT

Background: Acute cerebral infarction is a rare complication resulting from an unruptured cerebral aneurysm (UCA). There is presently no consensus on the optimal strategy for the management of UCAs with cerebral infarctions.

Case Description: A 53-year-old man presented with transient dysarthria and left hemiparesis. Magnetic resonance imaging (MRI) demonstrated the presence of a 7 mm UCA originating from the middle cerebral artery bifurcation, and diffusion-weighted imaging showed no evidence of cerebral infarction. One month later, his transient left hemiparesis recurred, and the patient was admitted to our hospital. Computed tomography angiography showed enlargement of the aneurysm. His left hemiparesis worsened 3 days later. MRI showed cerebral infarction in the area of perforating arteries and further enlargement of the aneurysm with surrounding parenchymal edema. Therefore, the rupture risk was considered to be rarely high and dome clipping was performed immediately. Postoperatively, his neurological status improved without any recurrent brain ischemia.

Conclusion: We report a rare case of a rapidly enlarging aneurysm that presented with cerebral infarction. This is the first report describing aneurysmal sac enlargement that can lead to perforating artery obstruction and brain ischemia. The case illustrates the importance of performing close follow-up examinations to confirm findings that suggest a high rupture risk.

Keywords: Brain ischemia, Direct compression, Rapid enlargement, Suction and decompression technique, Unruptured cerebral aneurysm

INTRODUCTION

Unruptured cerebral aneurysms (UCAs), especially large or giant cerebral aneurysms, are often present with neurological symptoms such as visual disturbances and oculomotor palsy without subarachnoid hemorrhage (SAH).^[7] These symptoms are generally considered to be the result of direct compression to the surrounding cranial nerves by UCAs. Brain ischemia is also known to be an uncommon complication resulting from UCAs, the frequency of which has been reported to be in the range of 0.6–11%, although a more recent study reported a range of 1.4–3.3%.^[5,14] These ischemic lesions commonly develop in the distal part of the aneurysm due to thrombotic materials formed within the aneurysmal sac,^[8] whereas ischemic lesions in the proximal part of

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the aneurysm are rare.^[2,4,9,11] Herein, we report a patient who presented with acute ischemic stroke in the area supplied by perforating arteries in the adjacent proximal part of an unruptured middle cerebral artery (MCA) aneurysm with rapid enlargement, and discuss the management of ischemic stroke associated with UCAs.

CASE DESCRIPTION

A 53-year-old man with transient dysarthria and left hemiparesis was admitted to our hospital. The patient had recovered from neurological deficits before being transferred to our hospital, and serological examination showed no presence of vascular risk factors. Magnetic resonance imaging (MRI) showed an UCA originating from the MCA bifurcation (7 mm in diameter) without evidence of acute cerebral infarction [Figure 1a]. The patient was diagnosed with transient ischemic attack (TIA); subsequently, careful observation was performed because of the UCA. He had a medical history of hypertension and a familial history of SAH. Cerebral angiography was performed 1 week after the initial TIA, wherein a saccular aneurysm including the superior trunk of the M2 branch was confirmed [Figure 1b]. Surgical treatment for the aneurysm was scheduled a few months later because of the acute state of brain ischemia. A month later, the patient was transferred again and admitted to our hospital on account of TIA recurrence. MRI showed no acute brain ischemia, but revealed that the aneurysm had expanded from 7 mm to 14 mm in diameter [Figure 2a]. Three days after admission, the left hemiparesis and dysarthria reappeared, and MRI showed acute cerebral infarction in the area supplied by the MCA perforating artery [Figure 2b and c]. In addition, parenchymal edema in the insular cortex surrounding the aneurysm was observed on a fluid-attenuated inversion recovery image [Figure 2d]. Intimal thrombosis within the aneurysmal sac was not clearly detected on MRI. Computed tomography (CT) angiography clearly depicts rapid enlargement of the aneurysmal sac [Figure 3]. We considered that the direct compression to the adjacent proximal perforating artery was caused by the rapidly enlarged UCA, which led to cerebral infarction. Although the cerebral infarction was in an acute state, we considered that the rapid enlargement of the aneurysm was due to an imminent rupture. Thus, the patient was assigned to undergo surgery. Surgical clipping was performed 2 days after admission. The MCA aneurysm was exposed through the pterional approach. Because the UCA was enlarged, the intimal pressure was suspected to be high, and the perforating arteries from M1 located behind the aneurysmal sac were difficult to observe under the microscope. Initially, the proximal M1 and distal M2 trunks from the aneurysm were occluded by temporal clipping to decompress the aneurysm. However, the aneurysmal intimal pressure remained high after blocking blood flow by temporal clipping. Therefore, we

decompressed and shrunk the enlarged aneurysmal sac using the suction and decompression technique.^[16] After adequate decompression, angioplastic clipping using multiple clips was successfully completed without obstruction of the parent artery and perforating arteries. Post application of surgical

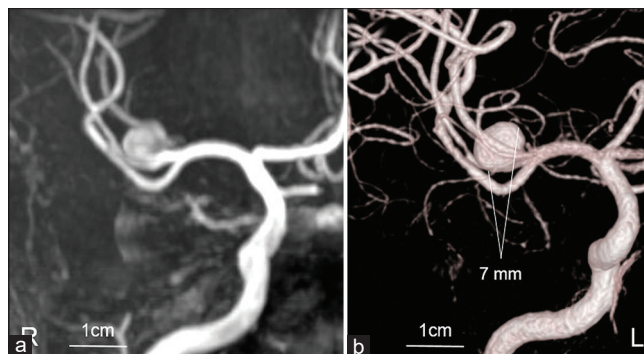


Figure 1: Initial radiological findings. Magnetic resonance angiography showed unruptured cerebral aneurysm in the right middle cerebral artery bifurcation (7 mm in diameter) (a). Cerebral angiography performed 1 week after the initial transient ischemic attack did not exhibit aneurysm growth (b).

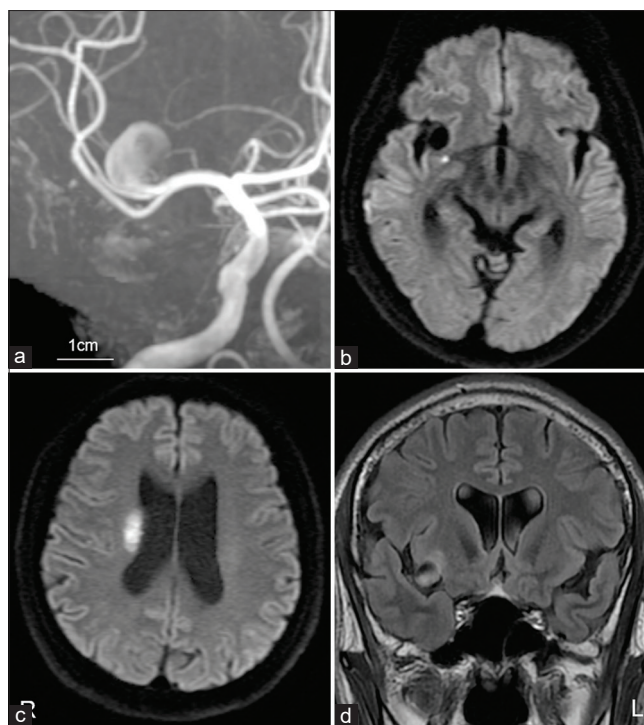


Figure 2: Magnetic resonance image (MRI) findings immediately after the recurrent transient ischemic attack (TIA). MRA performed immediately after the recurrent TIA detected rapid enlargement of the unruptured cerebral aneurysm (a). MRI showing acute cerebral infarction in the area supplied by a proximal perforating artery (b and c). Fluid-attenuated inversion recovery image showing peripheral edema in insular cortex attached to an enlarged aneurysmal sac (d).



Figure 3: Computed tomography angiography (CTA) findings. Preoperative CTA showing an enlarged right middle cerebral artery aneurysm (arrowhead).

clips, intraoperative endoscopy clearly showed an association between the aneurysmal sac and the perforating artery [Figure 4a-f]. Postoperatively, CT angiography showed no evidence of the M2 trunk obstruction and the aneurysmal neck remnant [Figure 5]. The patient's mild hemiparesis improved, and recurrence of infarctions could not be verified clinically for 2 years after surgery.

DISCUSSION

Clinical and radiological features of brain ischemia associated with UCA

Brain ischemia associated with UCA is an uncommon complication.^[8] Earlier, such an incidence was reported in the range of 3.0–6.3%, except for dissecting aneurysm cases.^[3] Brain ischemia associated with UCA most commonly develops in middle-aged patients, symptomatic UCA being commonly located in the internal carotid artery and MCA bifurcation.^[1,3,5,14] A previous review described that 9.7% of patients had experienced recurrent TIAs, and 2.4% of the patients experienced worsening of symptoms in long-term follow-up, whereas no recurrence of TIA was observed in most cases.^[14] Thrombosis is the most common finding in UCA, whereas partial and complete thrombosis was observed in only half of the cases in symptomatic UCA.^[1,5,8] In addition, the symptomatic aneurysm size was relatively small, the mean maximal diameter as reported was in the range of 7.5–12.5 mm, although the partial thrombosis factor that can obscure the size of UCA was also considered.^[14] In the present case, multiple recurrence of TIAs were experienced, and clinical symptoms progressed till the patients finally developed ischemic stroke. Thrombosis was not detected; however, UCA showed rapid enlargement.

Mechanism of brain ischemia associated with UCA

The two main mechanisms of brain ischemia associated with UCA have been considered.^[2,4,5,8,14]

First, distal embolization from intra aneurysmal thrombosis and/or extension of the thrombosis to the parent artery have been considered. Aneurysmal thrombosis is more common in larger UCAs. Factors associated with aneurysmal thrombosis include size and chamber volume to the orifice area, blood stagnation, slow flow, and increased blood flow viscosity. Turbulent flow within the aneurysmal sac leads to endothelial injury. Subsequently, the subendothelial matrix is exposed, which favors platelet deposition and thrombus formation.^[5] Intracavitary thrombosis may cause distal embolization or parent artery occlusion due to the local extension of thrombosis.

The second considered mechanism are direct compression from the aneurysmal sac to parent artery and these branches, or displacement of the parent artery or adjacent structures. A small number of cases of brain ischemia caused by direct compression of the aneurysmal sac to perforating arteries have also been reported [Table 1].^[2,4,9,11] According to the previous reports, a large or giant MCA aneurysm tends to cause brain ischemia by direct compression by the aneurysm. However, the area of cerebral infarction was not clearly described, and the parent artery obstruction due to compression by the aneurysm may have caused distal side infarction in one case.^[2] In most cases, direct compression with the UCA is diagnosed by cerebral angiography or MRI. As previously mentioned, thrombosis in the aneurysmal sac was observed in half of the cases, whereas thrombosis was ruled out by intraoperative findings in the present case. In addition, intraoperative endoscopic examination revealed the MCA perforating artery, such as the lateral distal striate arteries, compressed by enlargement of the aneurysmal sac. We considered that the rapid enlargement of the aneurysmal sac caused direct compression to the perforating artery, which was proximally close to the aneurysmal sac and caused acute cerebral infarction in the corona radiata. To the best of our knowledge, this is the first and only reported case of cerebral infarction in the area supplied by a perforating artery located on the proximal side of the UCA that clearly confirmed direct compression to the perforating artery with an aneurysmal sac.

Management of brain ischemia associated with UCA

Because of the low frequency of this complication, the management of patients with brain ischemia associated with UCA remains unclear. Based on the previous reports, the risk of ischemic recurrence is low regardless of treatment option, and medical and surgical treatments are not superior to conservative treatment in reducing the risk of ischemic event recurrence.^[1,3,14] Antiplatelet therapy (APT) is undertaken

Table 1: Summary of brain ischemia associated with UCA.

References	Location of UCA	Size	Area of infarction	Clinical symptom	Peripheral edema	Thrombosis	Timing from treatment	Prognosis
Mehdorn, 1980	MCA	20×25	No listed	TIA	No listed	–	1 month	Good
Fukuoka, 1981	MCA	20×11	No listed	TIA	No listed	+	No listed	Good
Oka, 1986	ACA (A1)	4×3	No listed	CI	No listed	–	19 days	Good
Choen, 2007	Acorn	No listed	ACA, Acom	CI	No listed	+	No listed	No listed
Present case	MCA	14×9	MCA perforating artery	CI	+	–	2 days	Good

UCA: Unruptured cerebral aneurysm, MCA: Middle cerebral artery, ACA: Anterior cerebral artery, Acom: Anterior communicating artery, TIA: Transient ischemic attack, CL: Cerebral infarction

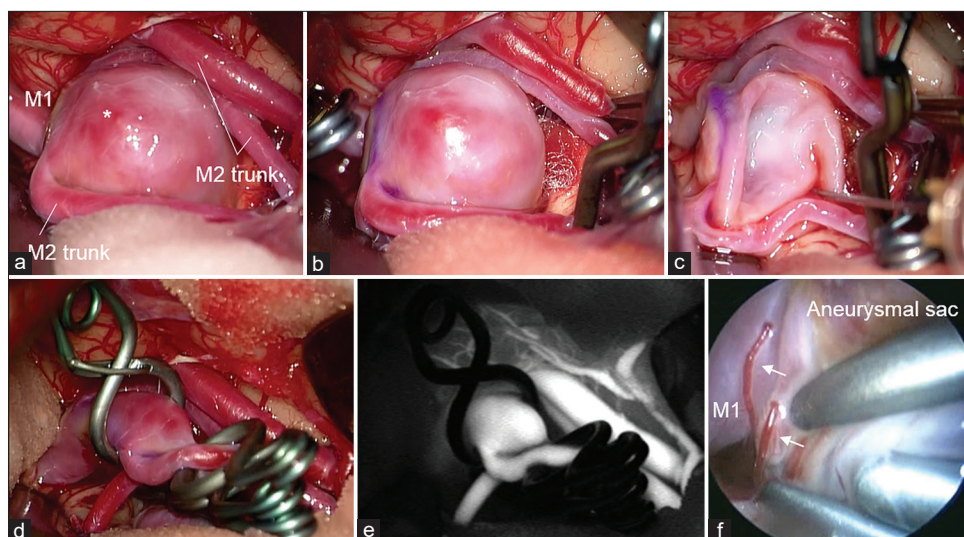


Figure 4: Surgical findings. Aneurysmal sac with bleb (*) and MCA trunks exposed through the pterional approach (a). Temporal clipping of MCA trunks performed, but intimal pressure remained high (b). Aneurysmal sac shrunk using the suction and decompression technique (c). Angioplastic clipping performed to avoid obstruction of the parent artery by multiple clips (d). Indocyanine green imaging (e). Endoscopy showed directly compressed perforating arteries (arrow) with an enlarged aneurysmal sac from behind the shrunk aneurysmal sac (f).

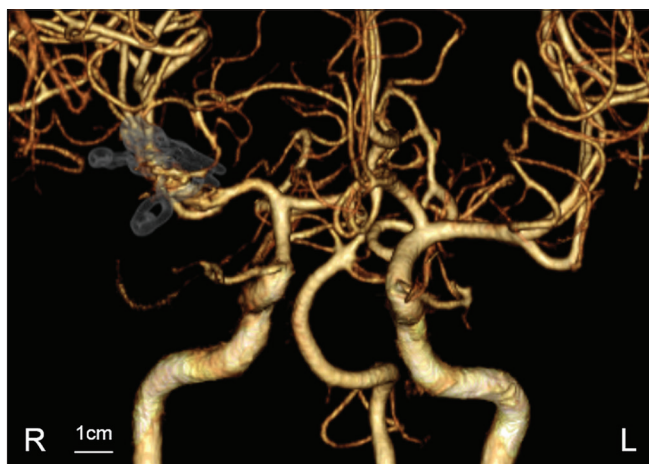


Figure 5: Postoperative computed tomography angiography (CTA) finding. Postoperative CTA showed that the multiple clipping for the middle cerebral artery aneurysm were successfully performed without the M2 trunk obstruction and aneurysmal neck remnant.

in several cases and is considered as a reasonable option for patients who are not in good condition for surgical treatment or are still waiting for beginning of their treatment.^[5] However, patients treated with APT occasionally experience aneurysm rupture, massive SAH can occur in both early and late states from the ischemic event.^[1,5] Based on those previous reports, in most cases, a conservative approach has been often recommended for brain ischemia associated with UCA.

Surgical treatment for imminent rupture of UCA presenting with acute brain ischemia

However, to prevent aneurysm rupture, surgical treatment should be considered in the case when imminent rupture is suggested because high frequency of early SAH in patients with UCA after brain ischemia is reported.^[5] As is well known, enlargement of aneurysm size indicates increasing

rupture risk, and perianeurysmal brain edema on MRI also suggests an association between bleb formation and enlargement of the aneurysm.^[6,12] In the present case, recurrence and progression of ischemic events developed, MRI revealed enlargement of aneurysm with perianeurysmal edema in the surrounding cortex of the UCA. Although the risk of surgical treatment for cerebral aneurysms in the presence of acute cerebral infarctions should be considered, surgical treatment for imminent rupture of the UCA should be prioritized over treatment for acute cerebral infarction. The previous reports have described good prognosis for surgical clipping for UCAs presenting with brain ischemia due to direct compression to arteries, and no further recurrent brain ischemia was experienced after the operation.^[4,9,11]

Surgical treatment for large and giant UCA is among the challenging lesions. A previous report described that surgery significantly reduced the mortality (from 31% to 4%) but increased the morbidity (from 8% to 19%) as compared with conservative treatment in patients with giant UCA.^[10] In this report, the authors described that direct surgical clipping may be indicated for giant aneurysms with perforating arteries that can be preserved. As is well known, complex MCA aneurysms often involve the M2 trunk with an enlarged aneurysmal sac, and those perforating arteries from the M1 located behind the enlargement aneurysmal sac, which is difficult to observe before a surgical clip is applied. Extracranial-intracranial bypass with parent artery obstruction was a considerable procedure for an enlarged MCA aneurysm involving the parent artery or branches. A previous report also described the use of multiple clipping techniques to preserve and reconstruct the parent artery, whereas sufficient decompression of aneurysm is required.^[15] Therefore, we decompressed the aneurysmal intimal pressure using the suction and decompression technique; these techniques have been previously described as one of the most effective techniques for surgical angioplastic clipping to relax aneurysmal intimal pressure.^[16] In fact, we prepared an extracranial-intracranial bypass in the case of a parent artery obstruction, but it was not required. Parent artery and M2 branch reconstruction were achieved using a multiple clipping technique, after adequate decompression of the aneurysm using suction and decompression techniques. In addition, endoscope-assisted microsurgery for surgical clipping is also an effective technique to prevent perforating artery injury.^[13] As previously mentioned, surgical clipping for complex large MCA aneurysm is challenging, several considerations by surgeons are required before and during surgery to avoid complications.

CONCLUSION

Brain ischemia associated with UCA is a rare complication. Most cases present with distal embolization are associated

with aneurysmal thrombosis, whereas rapid enlargement of the UCA can lead to brain ischemia due to direct compression to a nearby parent or perforating artery. Clinicians should be aware of the risk of brain ischemia due to enlargement of the UCA and should consider urgent treatment of the UCA even immediately after brain ischemia. For surgical clipping, several techniques should be considered to ensure safety and success.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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