Repeated rupture of a middle meningeal artery aneurysm in moyamoya disease

Case report

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A case of moyamoya disease associated with subarachnoid hemorrhage and intracerebral hematoma resulting from repeated rupture of a middle meningeal artery aneurysm is reported. The aneurysm was progressively enlarged over a period of 1 month and was treated with middle meningeal artery embolization. The treatment method is discussed. (DOI: 10.3171/2009.11.JNS09895)

KEY WORDS • cerebral aneurysm • middle meningeal artery • moyamoya disease • subarachnoid hemorrhage

Between 5 and 15% of patients with MMD have intracranial aneurysms.5 Moyamoya disease–related aneurysms are classified into major artery aneurysms from the circle of Willis and peripheral aneurysms from collateral circulation.6,13 Approximately 40% of the aneurysms present in MMD are peripheral and are mainly located at the distal portions of perforating arteries and at anterior and posterior choroidal arteries. These aneurysms are caused by an increased stress in the vessel wall related to the high flow imposed by occlusion of the anterior circulation. Peripheral aneurysms have been reported by various authors who used diverse methods of treatment to obtain variable results; however, the MMA is a very rare location for peripheral artery aneurysms in MMD. We experienced an MMD case with an MMA aneurysm presenting with SAH and ICH. A review of the literature revealed that there are only 3 cases of MMA aneurysms associated with MMD.2,5,15 In this paper we report on the patient’s clinical course and the process of management, and also discuss a treatment strategy.

Case Report

History and Presentation. This previously healthy 43-year-old woman experienced a sudden onset of headache followed by loss of consciousness. On admission, she was unconscious and exhibited right hemiparesis and left mydriasis. The left extremities showed a pain withdrawal response. A CT scan revealed the presence of an SAH with ICH in the left temporal lobe (Fig. 1). Computed tomography angiography showed bilateral occlusion of the distal internal carotid arteries and prominent posterior cerebral arteries, which was compatible with MMD. Catheter angiography demonstrated that the right MCA territories were supplied from the right posterior communicating artery. The left MCA territories were supplied from the left MMA, dural branches of the ophthalmic artery, posterior circulation, and dural branches of the internal maxillary artery (Fig. 2). An irregularly shaped aneurysm was observed at the entrance of the left MMA into the cranium (Fig. 2). Multiple vascular channels that were scattered around the aneurysm made it difficult to delineate the aneurysmal neck. The patient’s consciousness gradually improved to the point at which she was able to communicate nonverbally. A follow-up brain CT scan performed 7 days later showed a resolving state of SAH and an increased amount of ICH (Fig. 3), which implied minor rebleeding. Angiography demonstrated that the size of the aneurysm was slightly increased (Fig. 4). This aneurysm was believed to be a pseudoaneurysm that bleeds easily and should be urgently obliterated from the blood flow.

Operation and Postoperative Course. In the management of this aneurysm the authors took a few options into consideration. As first-line treatment, we believed endovascular embolization would be effective. However, obliterating the aneurysm itself was found to be impossible because the aneurysmal neck consisted of multiple channels. Although the next option was a proximal occlusion of the MMA, pos-
sibly near the neck, after an extracranial-intracranial bypass, we considered bypass surgery not feasible because the STA was too small in diameter to be suitable for a bypass (Fig. 5), and according to our limited experience, the wall of the recipient cerebral artery in an advanced stage of MMD was expected to be too friable and not adequate for suturing. Moreover, we believed a distal extracranial-intracranial bypass would not be enough to cover the proximal portion of the MCA territory.

A left craniotomy was performed for aneurysmal neck clipping. A dural incision was performed while paying particular attention to achieving minimal injury of dural vessels. Numerous vascular channels were present along the temporal base. The channels had thin fragile walls and were engorged toward the brain. Easy rupture of these vessels could cause brisk arterial bleeding, which would not allow a further approach. Although the patient woke up without additional neurological deficits, a postoperative brain CT scan showed a low density in the temporal lobe (Fig. 6). Subsequent angiography revealed that the aneurysm was further enlarged. Because of the failure to perform aneurysmal neck clipping, we attempted to endovascularly occlude the MMA. A balloon test that lasted 30 minutes was well tolerated and the MMA was completely embolized just proximal to the aneurysm (Fig. 7). One month after the first hemorrhage, the patient returned to her independent daily life as a housewife, although she complained of mild subjective weakness of the right arm.

Discussion
Peripheral Artery Aneurysms in MMD
Aneurysms associated with MMD are classified into major artery aneurysms arising from the major cerebral arterial segments, and peripheral artery aneurysms arising from the distal segments of the peripheral cerebral arteries. Major artery aneurysms are found more frequently in the vertebrobasilar system and peripheral artery aneurysms are located in the basal ganglia, near or in the moyamoya vessels.

Peripheral artery aneurysms in MMD have been reported sporadically. Their natural course, pathological characteristics, and treatment are not well known. Rupture of peripheral aneurysms results in intraventricular hemorrhage, ICH, or SAH, according to the location of the aneurysm. Repeated bleeding and subsequent aneurysm enlargement have been reported as early as 5 days after the initial rupture. Recurrent bleeding predicts serious neurological deficits or death, similarly to

Fig. 1. Axial CT scans revealed the presence of a diffuse SAH (left) with intracerebral hematoma (right) in the left temporal lobe.

Fig. 2. Results from the initial angiography showing that the left hemisphere was supplied from dural branches of the ophthalmic artery (A), posterior circulation (B), and branches of the internal maxillary artery including the MMA (C and D). Arrows indicate MMA aneurysm.

Fig. 3. Follow-up axial CT scans obtained 7 days after admission showed a resolving state of an SAH (left) and an increased amount of intracerebral hematoma (right), which implied minor rebleeding.
Repeated rupture of an MMA aneurysm in moyamoya disease

saccular aneurysm rupture.\textsuperscript{9} The pathological feature of a peripheral artery aneurysm was depicted as a false aneurysm,\textsuperscript{3,10,12,17} in which the wall is composed of collagen fibers and fibrin without elastic fibers.\textsuperscript{3} These pseudoaneurysms have a tendency to undergo an early increase in size.\textsuperscript{3,12,17} True aneurysms associated with MMD have been reported,\textsuperscript{8,11} which were located at the AChA. The anterior and posterior choroidal arteries are commonly involved in MMD-related peripheral aneurysms. The location of the aneurysm or imaging features does not discriminate true from false aneurysms.

There are several options available for the management of these aneurysms, such as surgical obliteration, extraintracranial revascularization surgery, endovascular surgery, and observation (that is, expectation of a spontaneous regression). Endovascular treatment has shown good results for major artery aneurysms associated with MMD.\textsuperscript{1} Performing surgery on aneurysms associated with MMD located around the circle of Willis is difficult because of interruption of anastomotic channels, poor tolerance to retraction and ischemia, and poor reserve capacity of hemodynamics.\textsuperscript{4}

For peripheral artery aneurysms associated with MMD, Kuroda et al.\textsuperscript{7} reported the efficacy of STA-MCA anastomosis with encephaloduromyoarteriosynangiosis in 3 cases with a thalamoperforating artery aneurysm, a posterior choroidal artery aneurysm, and an AChA aneurysm, respectively. The authors confirmed the disappearance of these aneurysms 1 and 2 months after revascularization surgery.\textsuperscript{7} Sugiura and Matsuzawa\textsuperscript{14} reported on endovascular coil embolization of an AChA aneurysm. Parent artery trapping and/or excision of aneurysms was attempted by other authors.\textsuperscript{3,8,11,12} Yuasa et al.\textsuperscript{17} described the extraction of an aneurysm located on the temporal lobe but did not find the parent artery. Hamada et al.\textsuperscript{3} did not find the aneurysm fed by the distal AChA; therefore, these authors performed STA-MCA anastomosis and clipped the parent artery. Nakai et al.\textsuperscript{11} and Lee et al.\textsuperscript{8} trapped the parent artery and excised the aneurysm. Spontaneous regression was also noted in an 11-year-old girl within 2 months of the initial attack\textsuperscript{10} and in an 18-year-old woman within 9 months of the attack.\textsuperscript{16}

The treatment modality should be chosen cautiously. We believe that several factors should be considered when deciding on the treatment method for the management of peripheral aneurysms in MMD, which include: 1) the possibility of identification of the parent artery and aneurysmal neck; 2) the presence or absence of multiple vascular channels around the aneurysm; 3) the intracranially supplying territory of the parent artery; 4) the possibility of introducing a catheter near the aneurysm; and 5) the location of the aneurysm.

Middle Meningeal Artery Aneurysms in MMD

Takahashi\textsuperscript{15} first reported an MMA aneurysm associated with MMD in a 10-year-old girl who presented with gait and speech disturbances. The aneurysm, which did not bleed, was located at the junction of the MMA with a branch of the anterior cerebral artery. Borota et al.\textsuperscript{2} reported a saccular aneurysm in the posterior branch of the MMA that localized partly outside and partly inside the dura. This patient was a 41-year-old man who presented with SAH and subdural hematoma. He underwent surgery but died 4 days later. Koebbe and Horowitz\textsuperscript{2} reported on a 31-year-old woman with Down syndrome and MMD. The aneurysm was noted in the collateral branches between
the MMA and the occipital artery and was treated with parent branch occlusion via an endovascular approach.

For aneurysms located just inside the middle cranial fossa, it is difficult for surgeons to identify the parent vessel intraoperatively. For other neurosurgeons who may encounter MMA aneurysms, we believe our experience may be helpful in choosing treatment options, even though our management process produced complications. The natural history of peripheral aneurysms in MMD is unknown, but frequent and short-term rebleeding and rapid expansion of aneurysms require urgent and reasonable treatments. Surgical excision or endovascular embolization would sacrifice intracranial collaterals. Treatment modalities should be carefully chosen according to the location of the aneurysm, the nature of the parent artery, and catheter accessibility.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author contributions to the study and manuscript preparation include the following: Conception and design: JS Suk. Acquisition of data: YS Park. Analysis and interpretation of data: YS Park. Administrative/technical/material support: JT Kwon. Study supervision: JS Suk.

References