Combination of neuroendoscopic hematoma evacuation and endovascular coil embolization for a ruptured anterior choroidal artery aneurysm in patients with moyamoya disease: illustrative cases

Kohei Uemasu, MD,1 Hiroyuki Koizumi, MD, PhD,1,2 Daisuke Yamamoto, MD, PhD,1,2 Sumito Sato, MD, PhD,1 Hideto Komai, MD,1 Madoka Inukai, MD, PhD,1 Takuichiro Hide, MD, PhD,1 Yasushi Asari, MD, PhD,2 and Toshihiro Kumabe, MD, PhD1

Departments of 1Neurosurgery, and 2Emergency and Critical Care Medicine, Kitasato University School of Medicine, Sagamihara, Kanagawa, Japan

BACKGROUND The treatment strategy for hemorrhagic moyamoya disease (MMD) due to a ruptured aneurysm at the distal portion of the anterior choroidal artery remains controversial. The authors successfully treated the ruptured aneurysm with neuroendoscopic hematoma evacuation, followed by endovascular coil embolization.

OBSERVATIONS The authors encountered two patients with massive hemorrhagic MMD whose MMD had already been diagnosed and who had a periventricular anastomosis due to a ruptured aneurysm of the distal portion of the anterior choroidal artery involving the periventricular anastomosis. In both cases, neuroendoscopic hematoma evacuation was performed for hemorrhagic MMD in the acute phase, followed by endovascular coil embolization of the ruptured aneurysm in the chronic phase. In both endovascular treatments, the patient's condition was stabilized by hematoma evacuation, allowing a detailed preoperative evaluation of the anatomical findings of the vessel and functional findings of intraoperative neurophysiological monitoring using continuous monitoring of motor evoked potentials to preserve motor function.

LESSONS Combination therapy can be useful for hemorrhagic MMD in patients with diagnosed MMD with a periventricular anastomosis. Additionally, a preoperative understanding of the vascular construction and intraoperative neurophysiological monitoring will aid in the successful coil embolization of aneurysms at the distal portion of the anterior choroidal artery with hemorrhagic MMD.

https://thejns.org/doi/abs/10.3171/CASE23677

KEYWORDS anterior choroidal artery aneurysm; endoscopic surgery; endovascular treatment; hemorrhagic moyamoya disease; hybrid surgery

The incidence of cerebral aneurysms associated with moyamoya disease (MMD) is 4% to 15%.1,2 MMD is a disease of unknown etiology. The two most prominent symptoms are ischemia and hemorrhage. The randomized controlled Japan Adult Moyamoya Trial demonstrated the effectiveness of direct bypass surgery for hemorrhagic MMD.3,4 Subgroup analysis of the trial results also demonstrated that choroidal anastomosis is one of the abnormal periventricular collaterals in MMD, which is a factor in hemorrhagic MMD.5 A periventricular anastomosis is defined as anastomosis of the choroidal and medullary arteries in the periventricular region, which consists of fragile vessels in MMD and is associated with hemorrhagic MMD.5,6 Recent studies have reported that neuroendoscopic hematoma evacuation is effective in the acute treatment of patients with MMD with no tolerance to ischemia due to increased cerebral pressure.6,7 However, the number of reported neuroendoscopic hematoma evacuations for patients with hemorrhagic MMD is still limited. The treatment options for hemorrhagic MMD due to a ruptured aneurysm include conservative treatment, craniotomy, and endovascular treatment. However, the choice of treatment strategy is controversial.8,9 We had experience with two cases of combination surgery: neuroendoscopic hematoma evacuation for hemorrhagic MMD in the acute phase and endovascular embolization for a ruptured aneurysm of the distal anterior choroidal artery.

ABBREVIATIONS CT = computed tomography; DSA = digital subtraction angiography; GCS = Glasgow Coma Scale; ICG = indocyanine green; ICH = intracerebral hemorrhage; ICP = intracranial pressure; IVH = intraventricular hemorrhage; MEP = motor evoked potential; MMD = moyamoya disease.

INCLUDE WHEN CITING Published March 11, 2024; DOI: 10.3171/CASE23677.

SUBMITTED November 18, 2023. ACCEPTED February 2, 2024.

© 2024 The authors, CC BY-NC-ND 4.0 (http://creativecommons.org/licenses/by-nc-nd/4.0/)
Illustrative Cases

Case 1

A 61-year-old male, previously diagnosed with MMD and refusing bypass surgery for ischemia, experienced sudden headache, vomiting, and loss of consciousness with a Glasgow Coma Scale (GCS) score of 3. Initial brain computed tomography (CT) revealed a right intracerebral hemorrhage (ICH) near the ventricle in the caudal nucleus, intraventricular hemorrhage (IVH), and acute hydrocephalus (Fig. 1). Initial brain CT angiography revealed no obvious aneurysms.

Neuroendoscopic surgery was immediately performed to decrease the intracranial pressure (ICP) due to the hemorrhage. Intraoperative endoscope-integrated indocyanine green (ICG) video angiography showed abnormally meandering vessels in the ventricular wall; however, no aneurysms were present within the visual range (Fig. 2). Postoperatively, his consciousness improved, but left paralysis persisted.

Digital subtraction angiography (DSA) was performed a month after neuroendoscopic surgery. Because the patient had a deterioration in his general condition due to complications such as aspiration pneumonia, it took a long time until angiographic examination. The patient had abnormal periventricular collaterals and a right distal anterior choroidal artery aneurysm in the plexal segment (Fig. 3A and B). The right anterior choroidal artery was dilated. Therefore, we concluded that embolization was possible. As coil embolization can cause additional motor impairment, intraoperative transcranial motor evoked potential (MEP) monitoring was performed with the patient under general anesthesia. The MEPs remained unchanged until the end of the procedure. Postoperatively, complete obliteration of the aneurysm was achieved without occlusion of the cisternal portion of the anterior choroidal artery (Fig. 3C and D). He had no complications caused by this endovascular treatment, his consciousness improved to a GCS score of 15, and he had no complications from the endovascular treatment (Fig. 3C and D). After continued postoperative rehabilitation, the patient's left paralysis improved, and he was able to walk with light assistance. The patient was discharged to a rehabilitation hospital with a modified Rankin Scale score of 4.

Case 2

A 47-year-old female with MMD had a history of subcortical hemorrhage. Magnetic resonance angiography revealed multiple
collateral vessels branching from the basal arteries and connecting them to the medullary arteries in the periventricular area (white arrows). B: Coronal CT scan of the head revealing subcortical hemorrhage in the right parietal lobe with IVH.

Initially, we performed hematoma removal via neuroendoscopic surgery. Postoperatively, her consciousness improved, but the left paralysis persisted. DSA confirmed the diagnosis of bilateral MMD and pseudoaneurysm. However, the aneurysm was located at the distal portion of right distal anterior choroidal artery, and the distal aneurysm had leptomeningeal anastomosis from the right middle cerebral artery (Fig. 5A–C).

To prevent rerupture of the aneurysm, the patient underwent coil embolization of the right distal anterior choroidal artery aneurysm using transcranial MEP monitoring with the patient under general anesthesia. Guiding the catheter into the aneurysm interrupted the blood flow, but leptomeningeal anastomosis preserved the distal blood flow (Fig. 5D). Therefore, embolization was possible (Fig. 5E and F). We performed coil embolization of the aneurysm with proximal occlusion of the distal portion of the anterior choroidal artery immediately before the aneurysm without any changes in the MEPs (Fig. 5E and F). Postoperatively, her left paralysis persisted; however, her consciousness improved to a GCS score of 15. The patient was discharged to a rehabilitation hospital with a modified Rankin Scale score of 2. In addition, because she had a good outcome after rehabilitation, we treated her with additional indirect surgical revascularization to prevent bleeding 17 months after coil embolization.

**FIG. 4.** Case 2. A: Coronal, thin-slab maximum intensity projection magnetic resonance angiogram showing multiple collateral vessels branching from the basal arteries and connecting to the medullary arteries in the periventricular area (white arrows). B: Coronal CT scan of the head revealing subcortical hemorrhage in the right parietal lobe with IVH.

**FIG. 5.** Case 2. A: Lateral DSA showing moyamoya vessels and an aneurysm (white arrow) at the distal segment of the right anterior choroidal artery, which was postulated to be the cause of bleeding. B: Frontal DSA showing moyamoya vessels and an aneurysm (white arrow). C: Frontal superselective angiography showing an aneurysm (black arrow) and moyamoya vessels distal to aneurysm (arrowheads). D: Frontal DSA showing that guiding the catheter into the aneurysm interrupted blood flow, but the leptomeningeal anastomosis preserved distal blood flow (arrowheads). E: Frontal DSA showing embolization of the right distal anterior choroidal artery aneurysm with N-butyl cyanoacrylate. F: Lateral DSA after embolization with N-butyl cyanoacrylate showing disappearance of the aneurysm (white arrow) at the distal segment of the right anterior choroidal artery.
Patient Informed Consent
The necessary patient informed consent was obtained in this study.

Discussion
Observations
We identified the following two important clinical issues: 1) the combination of neuroendoscopic surgery and endovascular treatment is useful for the treatment of hemorrhagic MMD, and 2) detailed anatomical exploration of complex vessel structures and functional findings of intraoperative neurophysiological monitoring will aid in the endovascular treatment of hemorrhagic MMD.

First, neuroendoscopic hematoma evacuation during the acute phase is useful in treating hemorrhagic MMD. Therefore, patients with hemorrhagic MMD require urgent evacuation of the hematoma as soon as possible to save their lives. In addition, for MMD patients with no tolerance to ischemia due to increased cerebral pressure, evacuation of the hematoma as soon as possible is required to preserve neurological function. However, hematoma evacuation via craniotomy in patients with hemorrhagic MMD is difficult because of the presence of moyamoya vessels and collateral flow. Therefore, we opted for less invasive neuroendoscopic surgery during the acute phase. Mino et al. reported the following three advantages of neuroendoscopic surgery for hemorrhagic MMD patients: 1) can maximally preserve preexisting collateral anastomosis; 2) can preserve the potential donor vessels used for future revascularization surgery, and 3) can normalize the high ICP immediately by surgical intervention in a minimally invasive manner. In addition, the ICG endoscope system was used for our surgical procedure. ICG angiography is very useful for real-time imaging, and its high spatial resolution facilitates the detection of fragile periventricular collaterals in patients with MMD, especially invisible moyamoya vessels hiding in the periventricular subependymal. Therefore, we are convinced that neuroendoscopic surgery becomes an effective tool in the removal of a hematoma in patients with MMD with no tolerance of ischemia due to increased cerebral pressure and difficult to approach by craniotomy in the acute stage safer and more secure.

Endovascular treatment as a secondary procedure after neuroendoscopic hematoma evacuation is useful for treating hemorrhagic MMD caused by a ruptured aneurysm. The therapeutic options for aneurysms associated with MMD include conservative treatment, craniotomy, and endovascular treatment. During the disease course, the most feared outcome is rebleeding. The rebleeding rate with conservative therapy is approximately 40%. Unless an aneurysm is treated, the risk of rebleeding remains. Because of the high rebleeding rate, we selected surgical treatment instead of conservative treatment. In endovascular treatment and craniotomy for peripheral aneurysms associated with MMD, the percentage of patients who maintained neurological symptoms before treatment was 95.2% for endovascular treatment versus 89.6% for craniotomy. Endovascular treatment is reported to be preferable in cases in which a catheter guide to the aneurysm is possible. Moyamoya vessels are often fragile, thin, and run in various ways. Approaching the catheter close to the aneurysm may not be possible, especially when the aneurysm is located at the distal portion of the anterior choroidal artery, as in our two cases. With sufficient effort and time, DSA can provide a detailed assessment of the angioarchitecture to identify the best approach route. The aneurysm was in the distal portion of the anterior choroidal artery but was prominently present due to the large load. Catheter selection is critical for treating aneurysms located in small tortuous vessels. To select the right direction in thin and tortuous vessels, we used a flow-related microcatheter and a hydrophilic-coated microguidewire with high vascular selectivity. Support was also enhanced using a distal access catheter. By selecting an appropriate catheter, we were able to perform a less invasive endovascular procedure.

Second, both the anatomical and functional findings of the vessels and intraoperative neurophysiological monitoring will aid in the endovascular treatment of hemorrhagic MMD. Aneurysms associated with hemorrhagic MMD are often considered pseudoaneurysms. Therefore, mother vessel occlusion is the treatment of choice. The question remains whether occlusion of the mother vessel causes new neurological symptoms. We used two solutions based on anatomical and functional information to prevent complications. The first was to determine whether the lesion could be embolized anatomically. In case 1, the aneurysm was in the right distal anterior choroidal artery on the plexal segment. The anterior choroidal artery is classified into cisternal and plexal segments. The latter is anastomosed with the lateral posterior choroidal artery, a branch of the posterior cerebral artery, but can be safely embolized clinically. In case 2, a guiding catheter was inserted into the mother vessel, and contrast injection was performed while wedging. Contrast imaging revealed retrograde collateral vessels, and embolization was performed. The second was MEP monitoring during endovascular treatment; in both cases, MEP monitoring was performed with the patient under general anesthesia to prevent the appearance of intraoperative complications and further motor disturbance. MEP monitoring is often used in arteriovenous malformation embolization and has recently been reported to be effective in the endovascular treatment of choroidal artery aneurysms with hemorrhagic MMD. The MEP was maintained without any changes in both cases. Thus, we determined that the distal portion of the anterior choroidal artery with an aneurysm could be embolized without additional functional deterioration, based on both anatomy and function.

Lessons
We performed combination surgery for hemorrhagic MMD. In cases of hemorrhagic MMD, which is difficult to treat by craniotomy, minimally invasive neuroendoscopic surgery in the acute phase and endovascular treatment as a secondary procedure result in a good prognosis. Combination surgery can be useful to protect against ischemic tolerance and collateral vessels in hemorrhagic MMD, especially in patients who have already been diagnosed with MMD and have abnormal periventricular collaterals. In addition, anatomical findings of vessels based on DSA and functional findings of intraoperative neurophysiological monitoring will aid in the endovascular treatment of hemorrhagic MMD.

References


Disclosures
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
Conception and design: Koizumi, Uemasu. Acquisition of data: Koizumi, Uemasu, Yamamoto, Sato, Komai. Analysis and interpretation of data: Uemasu, Yamamoto, Komai, Inukai. Drafting the article: Koizumi, Uemasu, Hide. Critically revising the article: Koizumi, Uemasu, Yamamoto, Kumabe. Reviewed submitted version of manuscript: Koizumi, Uemasu, Yamamoto, Komai, Kumabe. Approved the final version of the manuscript on behalf of all authors: Koizumi. Statistical analysis: Uemasu. Administrative/technical/material support: Uemasu, Yamamoto. Study supervision: Koizumi, Uemasu, Asari, Kumabe.

Correspondence
Hiroyuki Koizumi: Kitasato University School of Medicine, Sagamihara, Kanagawa, Japan. koizumi-h@cl.cilas.net.