Cerebrovascular Treatment with Superselective Balloon Test Occlusion for Multiple Cerebral Aneurysms Associated with Middle Cerebral Artery Anomaly: A Case Report

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Abstract

A 38-year-old woman presented with impaired consciousness and anisocoria due to a subarachnoid hemorrhage and an intracerebral hemorrhage of the left temporal lobe. Examination revealed severe tortuosity in the left middle cerebral artery and three sequential bead-like aneurysms. She underwent a craniotomy in the acute phase to stop rebleeding at the rupture site and remove the intracerebral hematoma. During the chronic phase, endovascular treatment with superselective balloon test occlusion (ssBTO) was performed for the remaining aneurysms. Preoperative ssBTO was useful in evaluating collateral circulation and assessing the curability of the treatment.

Keywords: cerebral aneurysm, middle cerebral artery, superselective BTO, collateral network, vascular anomaly

Introduction

There are few descriptions of anomalies of the bifurcation of the internal carotid artery (ICA) or middle cerebral artery (MCA). Moreover, there are several MCA anomalies, such as accessory MCA, duplicated MCA, fenestration of MCA, and duplicated origin of MCA. Although unfused or twiglike MCA, indicating MCA trunk occlusion with the collateral plexiform arterial network, has been reported recently, the present case has a very rare fenestration-like structure. It does not fall into any of these categories. We report our experience with a case of multiple cerebral aneurysms accompanied by an M1 vascular anomaly, which we treated with parent artery occlusion (PAO) alone after preoperative superselective balloon test occlusion (ssBTO).

Case Report

The patient was informed and agreed to the study. A 38-year-old woman was brought to the hospital by an ambulance owing to sudden onset of headache, vomiting, impaired consciousness (Glasgow Coma Scale E1VTM3), and anisocoria. A computed tomography (CT) scan of the head revealed a subarachnoid hemorrhage (SAH) with an intracerebral hematoma (Fig. 1A). Digital subtraction angiography of the left ICA revealed three sequential aneurysms of sizes 4.0 × 3.5 × 3.0 mm, 7.2 × 3.9 × 3.9 mm, and 8.0 × 4.8 × 4.3 mm, respectively, in the left MCA (M1 segment) with severe tortuosity. The most distal aneurysm had a bleb, which seemed to be the point of rupture. The branch from left A2 traveled around M1 with the aneurysms; it branched into multiple lateral striate arteries (LSA) before ultimately joining the left M1 distally. Thin anastomotic vessels were also observed at M1 and M2 (Fig. 1B-D). The most distal aneurysm could not be approached with the help of a microcatheter because of severe bending and tortuosity of the vessels and the very small diameter of M1. Therefore, radical endovascular treatment would be difficult in the acute phase. We decided to perform a craniotomy to clip the rupture site and remove the intracerebral hematoma. First, the hematoma was removed via a transsylvian approach, and the most distal aneurysm with the bleb was clipped via a sub frontal approach. The vessels that anastomosed M1 and M2 were observed visually during the procedure (Fig. 2).
Fig. 1  Initial computed tomography shows subarachnoid hemorrhage (SAH) and hematoma in the left temporal lobe (A). Three-dimensional (3D) rotation angiography of left internal carotid artery (ICA) (B), conventional left ICA angiography (C), and vascular structure schema (D) shows three consecutive aneurysms at the left M1 segment, A1-M1 anastomosis (arrowheads), and M1-M2 anastomosis (arrows). The most distal aneurysm is a ruptured cerebral aneurysm with a bleb (asterisk).

Fig. 2  Clipping of a ruptured aneurysm was performed by securing the internal carotid artery (star) via a sub frontal approach. The anastomosis between the left M1 and M2 was confirmed during the operation (arrows).

(RNF213), associated with Moyamoya disease, was sequenced in the hospital, but no mutations were found. The postoperative course was uneventful, and approximately 6 months later, the patient had only mild visual field impairment. Radical endovascular treatment was planned for the remaining aneurysms. Because of severe bending and the thin diameter of the M1 segment proximal to the aneurysms, a microcatheter could not pass through. Therefore, ssBTO was first performed at this site. In case of a negative finding, proximal occlusion of the artery or so-called PAO was planned.

A 6-Fr. Fubuki (ASAHI INTECC J-sales, Inc. Tokyo, Japan) catheter was introduced via the left ICA under local anesthesia. A 3.4-Fr. Guidepost (Tokai Medical Products Inc. Aichi, Japan) was guided to the proximal portion of the siphon. From there, the left M1 was accessed with an Excelsior SL-10 microcatheter (Target Therapeutics/Stryker, Fremont, CA) and a CHIKAI 14 micro guidewire (ASAHI INTECC J-sales, Inc. Tokyo, Japan). When the microcatheter was engaged in the M1 for ssBTO, the blood flow in the MCA was completely discontinued (Fig. 3A). An angiography during ssBTO showed no retrograde perfusion of the aneurysm, and the area distal to M2 was visualized from the blood vessels anastomosing the anterior cerebral artery (ACA) and MCA (Fig. 3B). No neurological symptoms appeared after approximately 20 min of occlusion; thus, PAO was performed at this site (Fig. 3C, D). Postoperative head MRI showed no cerebral infarction and no new neurological findings. We also confirmed by visualization that the aneurysm had completely disappeared. On postoperative day 10, the patient was discharged from the hospital with a modified Rankin scale score of 1.

Discussion

Accessory MCA, duplicated MCA, and fenestrated MCA are well-known MCA anomalies. Our case involved hypoplasia and severe tortuosity of the left M1, but distal to the left M2, the vessel had a normal diameter, and its pathway was maintained. Furthermore, while a branch from the left A2 traveled around M1, it branched into mul-
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Fig. 3  Superselective angiography performed by inserting a microcatheter into M1 shows three consecutive aneurysms, and the most distal aneurysm has lost its bleb due to clipping (asterisk). (A) Conventional left internal carotid artery (ICA) angiography (B) and vascular structure schema (C) visualized superselective balloon test occlusion performed by occluding the same site (shaded area), which reveals a slight stump (arrow) but no aneurysms. The distal left M2 was perfused by the A1-M1 anastomosis. Three-dimensional-rotation angiogram of the left ICA after endovascular treatment (D) shows that the blood flow to the aneurysms had completely disappeared. Furthermore, LSA branching from A1-M1 anastomosis (arrows) is preserved.

Accessory MCA is understood to provide blood flow to a fixed portion of the cortex, mainly in the MCA region. The collateral circulation in the present case did not correspond to accessory MCA but rather had large fenestration-like structures. Embryologically, accessory MCA is considered a dilation of the recurrent artery of Heubner or a remnant of the anastomosis connecting the ACA and MCA. The present case could have resulted from accessory MCA formation that halted before completion. The known MCA anomalies, including accessory MCA, cannot explain the abnormal pathway in the present case.

Seo et al. reported that aplastic or twiglike MCA (Ap/T-MCA) as a remnant of the primitive vascular system associated with abnormal development. While the cause of Ap/T-MCA is not completely understood, it is believed to be caused by developmental abnormalities in the proximal MCA during the fetal period. Recently, a subtype called "twiglike networks of an anomalous collateral artery" (T-NACA) has been proposed, in which twiglike networks remain inside the collateral circulation from A1 or A2 to the periphery and bypass the M1 portion. Although the collateral circulation in the present case did not correspond to T-NACA because it lacked a twiglike network, a characteristic vascular structure branch from the left A2 that joined the left M1 distally also found in T-NACA. Therefore, a vascular anomaly in the present case can be considered a rare anomaly with the incomplete arrest of MCA development.

Liu et al. found that the normal adult MCA relieves the pressure transmitted from the ICA. In contrast, in Ap/T-MCA, flow-related aneurysms tend to form because of hemodynamic stress on narrow and fragile collateral routes, which rupture easily. Although the present case does not consist of an Ap/T-MCA, it also had sequential aneurysms in the proximal portion of the incomplete and small diameter of M1 immediately after bifurcation from the ICA strongly suggests the involvement of hemodynamic stress.

In the present case, the aneurysms passed beyond the planned MCA embolization site, making it necessary to predict the therapeutic effect in advance. We previously reported the utility of ssBTO for predicting ischemic complications in PAO. In the present case, ssBTO showed no retrograde perfusion of the aneurysm and was useful in preventing ischemic complications and evaluating the curability of the treatment. We did not find previous reports of patients with sequential, bead-like aneurysms similar to the present case or those who were completely cured with ssBTO and PAO. Previous studies have demonstrated that in BTO of the ICA, false negatives occur in 13%-15% when only neurological assessments are performed and in 3%-10% when cerebral blood flow examinations are added to assessments. The false-negative rate of
ssBTO remains unknown, and more cases need to be examined in the future. We considered radical treatment by M1 trapping and hematoma removal with motor evoked potential ( MEP) in this case. But the patient was intubated due to severe SAH, and the lesion was on the dominant hemisphere, so it was difficult to evaluate the preservation of language function. Furthermore, the patient also had anisocoria that did not allow time for detailed evaluation of the collateral flow. Therefore, despite the risk of ssBTO, we decided to treat the patient in two phases: hematoma removal and clipping to prevent rerupture.

In conclusion, we report a case of a vascular anomaly with two large fenestration-like structures, suggesting that the formation of the accessory MCA may have halted before completion. Treatment with PAO alone may be possible in these cases with multiple collateral routes. Preoperative ssBTO can be useful in evaluating the collateral circulation and assessing the curability of the treatment.

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Abbreviations

ACA: anterior cerebral artery, Ap/T-MCA: aplastic or twiglike MCA, CT: computed tomography, ICA: internal carotid artery, LSA: lateral striate arteries, MCA: middle cerebral artery, MEP: motor evoked potential, PAO: parent artery occlusion, RNF213: ring finger protein 213, SAH: subarachnoid hemorrhage, ssBTO: superselective balloon test occlusion, T-NACA: twiglike networks of an anomalous collateral artery

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Conflicts of Interest Disclosure

All authors declare that there is no conflict of interest.

References

1) Uchiyama N: Anomalies of the middle cerebral artery. Neurol Med Chir 57: 261-266, 2017
2) Komiyama M: Essential anatomical knowledge for neurointervention—functional neurovascular anatomy. Jpn J Neurosurg 13: 116-125, 2004
3) Seo BS, Lee YS, Lee HG, Lee JH, Ryu KY, Kang DG: Clinical and radiological features of patients with aplastic or twig-like middle cerebral arteries. Neurosurgery 70: 1472-1480, 2012
4) Shin HS, Lee SH, Ryu CW, Koh JS: Flow-related intracranial aneurysms associated with unfused arterial twigs relevant to different vascular anomalies: embryologic and hemodynamic considerations. Acta Neurochir 156: 1637-1646, 2014
5) Liu HM, Lai DM, Tu YK, Wang YH: Aneurysms in twiglike middle cerebral artery. Cerebrovasc Dis 20: 1-5, 2005
6) Isozaki M, Arai H, Neishi H, Kitai B, Kikuta KI: Super-selective balloon test occlusion of the posterior communicating artery in the treatment of a posterior cerebral artery fusiform aneurysm: a case report. NMC Case Rep J 3: 129-131, 2016
7) Dare AO, Gibbons KJ, Gillihan MD, Guterman LR, Loree TR, Hicks WL: Hypotensive endovascular test occlusion of the carotid artery in head and neck cancer. Neurosurg Focus 14: e5, 2003
8) Linskey ME, Jungreis CA, Yonas H, et al.: Stroke risk after abrupt internal carotid artery sacrifice: accuracy of preoperative assessment with balloon test occlusion and stable xenon-enhanced CT. AJNR Am J Neuroradiol 15: 829-843, 1994

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