Online July 12, 2022
Journal of Neuroendovascular Therapy 2022; 16: 510–514 DOI: 10.5797/jnet.cr.2022-0025

Case Report

Lessons Learned from Mechanical Thrombectomy of an Acute Occlusion of a Duplicated Middle Cerebral Artery: A Case Report

Tomoaki Akiyama,[1,2] Tomohiro Okuda,[2] and Satoshi Inoha[1]

Objective: Detection of acute arterial occlusion in an anomalous middle cerebral artery (MCA) is challenging in an emergency setting because of its rarity.

Case Presentation: We report an 81-year-old woman who presented with acute occlusion of a duplicated middle cerebral artery (DMCA). Although the absence of the superior trunk of the left MCA was identified on preoperative imaging, initial angiography showed no typical sign of the occluded vessel. Repeated angiography eventually revealed retrograde arterial flow parallel to the other visible MCA trunk, which raised the possibility of a DMCA. The occlusion occurred at the origin of the DMCA originating from the internal carotid artery terminus, which obscured its presence. Mechanical thrombectomy was performed and achieved complete recanalization. The DMCA had two trunks of approximately equal size. The patient completely recovered within 90 days.

Conclusion: Comprehensive knowledge of cerebrovascular anomalies is essential to identify the occluded branch faster and accurately and to avoid thrombectomy-related complications in endovascular recanalization therapy. Relevant DMCA anatomy and tips for identifying an occluded DMCA are discussed.

Keywords ▶ anomaly, duplication, middle cerebral artery, stroke, thrombectomy

Introduction

Acute ischemic stroke due to occlusion of the horizontal (M1) or proximal insular (M2) segments of the middle cerebral artery (MCA) is a common indication for mechanical thrombectomy.[1)] Although M1 normally arises from the terminus of the internal carotid artery (ICA) as a single trunk, failure of the lateral striate arteries to fuse during embryogenesis can cause duplicated middle cerebral artery (DMCA) or formation of other M1 trunk variants.[2)] Reported incidence rates of DMCA range between 0.2% and 2.9%.[3)] Occlusion of this anomaly is rarely encountered in clinical practice and accurate diagnosis is challenging.[4)] We experienced a patient with an acute embolic occlusion of the DMCA origin who underwent successful thrombectomy. This case report discusses the relevant anatomy of DMCA, focusing on the identification of occlusion.

Case Presentation

An 81-year-old woman presented with global aphasia, left conjugate eye deviation, and severe right hemiparesis. The patient had no cerebrovascular risk factors or history of atrial fibrillation. Time from symptom onset to hospital arrival was 33 minutes. The National Institutes of Health Stroke Scale (NIHSS) score was 21. Diffusion-weighted imaging (DWI) of the brain revealed hyperacute infarction in the subcortical right parietal lobe. No ischemic lesions were visualized in the left hemisphere. Left hemisphere DWI-Alberta Stroke Program Early Computed Tomography score was 11 (**Fig. 1A** and **1B**). FLAIR imaging demonstrated arterial hyperintensity in the left

[1]*Department of Neurosurgery, Hachisuga Hospital, Munakata, Fukuoka, Japan*
[2]*Department of Neurosurgery, Graduate School of Medical Sciences, Kyushu University, Fukuoka, Fukuoka, Japan*

Received: April 22, 2022; Accepted: May 31, 2022
Corresponding author: Tomoaki Akiyama. Department of Neurosurgery, Hachisuga Hospital, 2650, Nosaka, Munakata, Fukuoka 811-3423, Japan
Email: akiyama.tomoaki@kyudai.jp

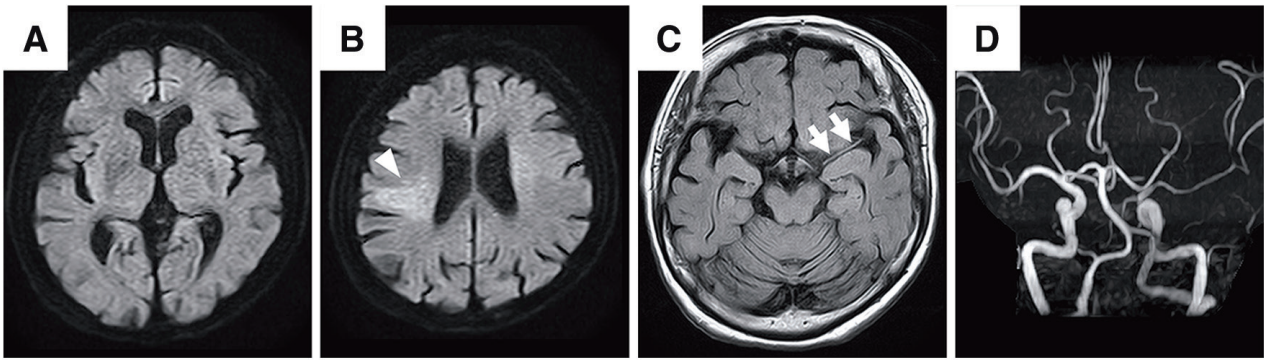

Fig. 1 MRI on admission. (**A** and **B**) DWI shows no acute infarct in the left MCA territory; hyperacute infarction is visualized in the subcortical right parietal lobe (white arrowhead). (**C**) FLAIR imaging reveals a vascular hyperintensity in the left proximal Sylvian fissure that courses parallel to the visible MCA trunk (white arrows). (**D**) MRA demonstrates absence of the superior trunk in the insular segment of the left MCA; the diameter of the horizontal segment of the left MCA is slightly smaller compared to that of the right MCA. DWI: diffusion-weighted imaging; MCA: middle cerebral artery

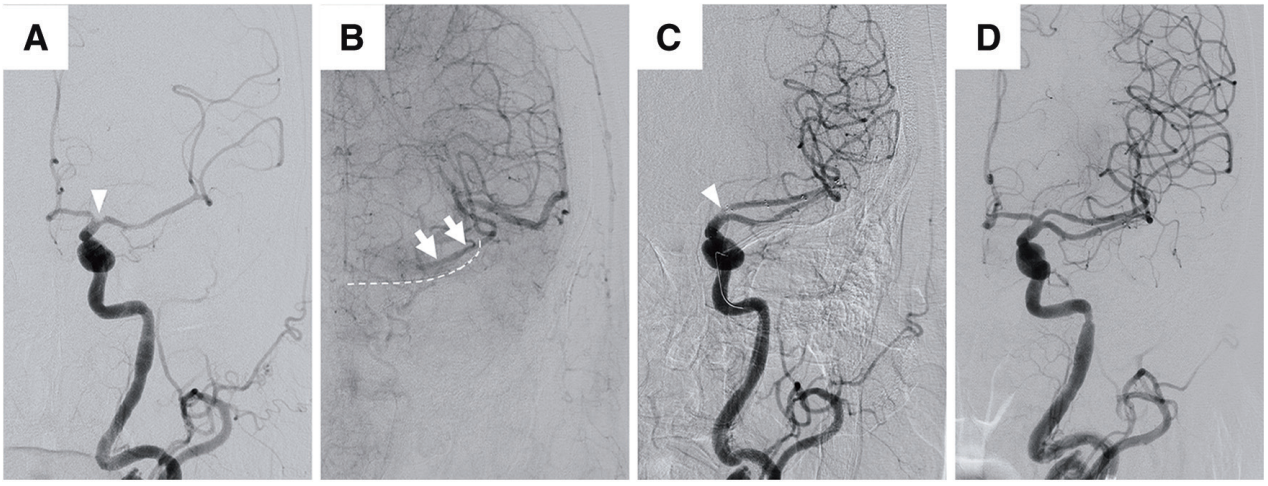

Fig. 2 Anteroposterior angiographic views. (**A**) Initial angiography shows no cerebral flow to the frontal lobe. No obvious sign of the occluded vessel was observed. Later, a small filling defect at the ICA terminus was identified (white arrowhead). (**B**) Delayed arterial phase angiography shows retrograde collateral blood flow via collaterals parallel to the other MCA trunk (white arrows). The microguidewire (white dotted line) is located in the non-occluded MCA trunk. (**C**) A stent retriever was deployed. Flow was restored immediately and a thrombus located at the origin of the superior trunk of the DMCA (white arrowhead) was identified. (**D**) Complete recanalization was achieved after one pass. This DMCA had two trunks of approximately equal size. DMCA: duplicated middle cerebral artery; ICA: internal carotid artery; MCA: middle cerebral artery

Sylvian fissure (**Fig. 1C**). On MRA, the left M1 trunk was slightly smaller than the right and the left M2 superior trunk was absent; no right MCA occlusion was demonstrated (**Fig. 1D**). The patient was diagnosed with acute left embolic M2 occlusion associated with clinical-DWI mismatch. The asymptomatic acute right hemisphere infarction indicated immediate spontaneous revascularization of a temporarily occluded peripheral right MCA branch. We elected to treat the patient with intravenous recombinant tissue plasminogen activator followed by mechanical left M2 thrombectomy.

Mechanical thrombectomy was performed under local anesthesia. A 9-Fr Optimo balloon guiding catheter (Tokai Medical, Aichi, Japan) was advanced via the femoral artery into the left cervical ICA. This was time consuming because of severe aortic and carotid tortuosity. Initial left ICA angiography revealed no cerebral flow to the frontal lobe, which was primarily supplied by the superior M2 trunk (**Fig. 2A**). In the delayed phase, retrograde flow to the point of occlusion via collaterals was observed near the presumed MCA bifurcation. We first attempted to detect the hidden origin of the occluded vessel using a microguidewire in the vicinity of the distal M1; however, we were unsuccessful. Repeated carotid angiography revealed delayed-phase retrograde collateral filling of the superior branch of the occluded MCA, which passed parallel to the other M1 trunk (**Fig. 2B**). 3D rotational angiography revealed anomalous retrograde flow that coursed parallel to the visible MCA trunk and

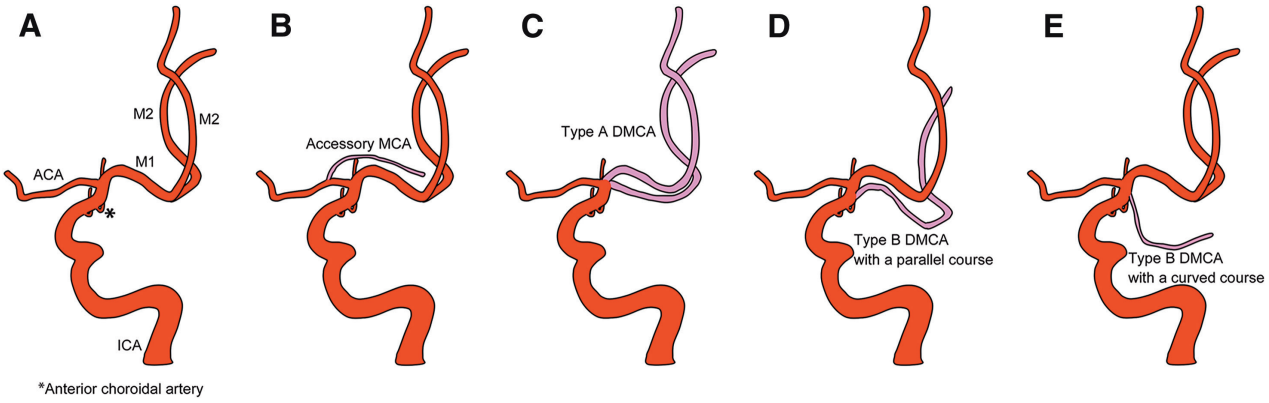

Fig. 3 Illustration of a normal MCA and double MCA trunks. (**A**) Normal MCA. (**B**) Accessory MCA originating from the ACA. (**C**) Type A DMCA separates at the ICA terminus into two trunks of approximately equal size. (**D**) Type B DMCA with parallel course branches off between the ICA terminus and the anterior choroidal artery and courses parallel to the main MCA trunk. (**E**) Type B DMCA with curved course passes toward the temporal lobe. ACA: anterior cerebral artery; DMCA: duplicated middle cerebral artery; ICA: internal carotid artery; M1: horizontal segment of the MCA; M2: insular segment of the MCA; MCA: middle cerebral artery

confirmed the possibility of an occluded DMCA or accessory MCA. A detailed search identified a small filling defect at the ICA terminus (**Fig. 2A**). Lesion crossing with a small J-shaped tip microguidewire (Traxcess; MicroVention Terumo, Aliso Viejo, CA, USA) was carefully performed and Rebar microcatheter (Medtronic, Minneapolis, MN, USA) was advanced into the DMCA. Evaluation of vessel diameter of occluded DMCA was performed using superselective angiography through a microcatheter. Then, a 4 mm × 40 mm Solitaire X revascularization device (Medtronic) was deployed, which immediately restored flow and revealed thrombosis at the origin of the superior DMCA trunk (**Fig. 2C**). The first pass resulted in complete reperfusion of the occluded DMCA and achieved grade 3 modified thrombolysis in cerebral infarction flow (**Fig. 2D**). Door-to-puncture, puncture-to-recanalization, and onset-to-recanalization times were 60, 98, and 191 minutes, respectively. Immediately after the procedure, the patient's NIHSS score improved to 10. She was diagnosed with cardioembolic stroke after the procedure as paroxysmal non-valvular atrial fibrillation was identified and other causes were excluded. Edoxaban was administered to prevent recurrent stroke. Twenty-five days after thrombectomy, the patient was discharged home with an NIHSS score of 0. At the 90-day follow-up visit, her modified Rankin Scale score was 1 and she had resumed her usual activities of daily living.

Discussion

The case presented here emphasizes the difficulty of diagnosing acute DMCA occlusion in an emergency clinical stroke setting. Prompt identification of the responsible vessel and understanding relevant variations in cerebrovascular anatomy are essential to shorten the time between symptom onset and reperfusion and to avoid thrombectomy-related complications.

In human embryogenesis, the MCA arises from the primitive ICA proximal to the anterior cerebral artery (ACA) as plexiform arterial twigs. These twigs undergo repeated fusion and regression before forming a prominent large single branch (**Fig. 3A**). Any interruption of this maturation may lead to various vascular anomalies including DMCA and accessory MCA.[2] In 1973, Teal et al. demonstrated that the accessory MCA originates from the ACA (**Fig. 3B**), while the DMCA branches off the ICA (**Fig. 3C**–**3E**).[5] However, no apparent embryologic basis for this has been explained. Both variant arteries feed a portion of the cortex that is usually supplied by an MCA branch. Congenital MCA variants are rare compared with those of other intracranial arteries.[3] Kai et al. classified DMCA into two types according to the site of separation from the ICA.[6] In type A, the separation occurs at the ICA terminus (**Fig. 3C**), whereas the separation occurs between the ICA terminus and the origin of the anterior choroidal artery in type B (**Fig. 3D** and **3E**). Type B is twice as frequent as type A.[7] Although the larger of the two trunks in a DMCA is usually defined as the main MCA,[2] the main trunk is difficult to define when the diameter of both trunks is the same. Our patient's DMCA had two trunks of approximately equal size that originated separately from the ICA terminus. An M1 terminal bifurcation or trifurcation was absent. The superior branch of the DMCA supplied the frontal and parietal lobes, while the inferior branch perfused the temporal and parietal lobes, which are typically fed by the superior

and inferior trunks of a normal M2, respectively. Yamamoto et al. described the type A DMCA as a direct bifurcation, which can be considered a normal MCA bifurcation variant.[8] This description applies in our case. The type A DMCA commonly has two similar-sized MCA vessels running in parallel in the Sylvian fissure (**Fig. 3C**).[7] In contrast, the type B DMCA vessels tend to be smaller than those of the main MCA and may be classified according to course (parallel course or curved course toward the temporal lobe; **Fig. 3D** and **3E**).[6,7] Several authors have proposed that type A DMCA and type B DMCA with parallel duplicated arteries correspond to direct bifurcation of the MCA, while type B DMCA with a curved course toward the temporal lobe should be considered as an early temporal branch of the MCA.[3,6–8]

Although the use of mechanical thrombectomy for M2 occlusion is increasing and has demonstrated good clinical outcomes,[1] the branching location of the occluded vessel is difficult to notice in cases where the thrombosis is located at the origin of the branch.[9] Moreover, its use for an occluded DMCA is extremely rare, and to our knowledge, only one previous case treated with mechanical reperfusion therapy has been reported.[4] Based on our experience, we would like to point out several points regarding identification of an occluded DMCA. First, retrograde collateral flow parallel to an MCA trunk in delayed phase angiography is an important clue that indicates a DMCA is present. We identified absence of the superior branch of the left M2 on preoperative MRA in our patient; however, initial angiography showed no typical sign of the occluded vessel.[10] Although retrograde collateral flow was observed in the delayed phase, this flow remained in the distal portion of M2. Therefore, we did not immediately identify a vessel occlusion or the presence of a DMCA. Repeated angiography eventually revealed retrograde arterial flow parallel to the other M1 trunk, which raised the possibility of an occluded accessory or DMCA. Second, neurointerventionalists should carefully check the MR imaging studies. FLAIR vascular hyperintensity (FVH) can be indicative of slow flow within an affected vessel, which may be retrograde collateral flow distal to the occlusion.[11] When FVH follows a path parallel to the other MCA trunk even in the presence of a full M1 trunk, an occluded DMCA is suggested. In our case, we did not notice these findings before thrombectomy was performed. In addition, susceptibility vessel sign on T2*-weighted imaging is a useful marker of clot location in cardioembolic stroke.[12] Although the patient did not undergo this sequence due to the reduction of MR imaging time, M1 susceptibility sign adjacent to the non-occluded MCA stem might help identify the occluded DMCA. Third, although we used an MR imaging-based imaging protocol for this patient, CT-based neurovascular imaging including CTA is becoming the gold standard in the diagnosis of acute ischemic stroke.[9] Especially in patients with medium-vessel occlusion, detection of the occlusion site on conventional CTA or MRA has been challenging, but advanced imaging techniques including multiphase CTA, 3D CTA, 4D CTA, and CT perfusion have enabled fast and reliable detection of occluded vessels and assessment of collateral status.[9] Even when CTA appears patency of the intracranial vessels, a large area of penumbra on CT perfusion suggests occlusion of DMCA or other large artery anomalies.[4] These imaging techniques can also render identifying the location of the clot and anatomic variants in the distal region possible, resulting in safe lesion passing during thrombectomy. Finally, comprehensive knowledge of cerebrovascular anomalies is essential to identify the occluded vessel faster and more accurately in endovascular recanalization therapy.

In thrombectomy for acute embolism associated with the DMCA or accessory MCA, based on superselective angiography after lesion passing, careful selection of stent retriever size is needed. Koge et al. reported vessel wall injury by stent retriever thrombectomy in a case of ICA occlusion with DMCA.[13] They did not recognize the MCA trunk that had a slightly smaller caliber than that of the normal M1 segment due to the presence of DMCA, and the use of a larger stent retriever that was deployed from the distal ICA into the MCA trunk caused iatrogenic vascular injury. A direct aspiration first pass technique that does not require lesion crossing may also be effective and safe in cases with atypical occlusion site or abnormal vascular structure.[13,14]

Conclusion

We described a rare case of acute DMCA occlusion treated with mechanical thrombectomy. Safe and effective performance of mechanical thrombectomy in stroke patients requires detailed understanding of cerebrovascular anatomy and anatomic variants.

Ethical Approval

In accordance with the hospital's protocol, this report did not require the Institutional Review Board's approval.

Written informed consent was obtained from the patient for publication of this report and the accompanying images.

Disclosure Statement

The authors declare that they have no conflicts of interest.

References

1) Menon BK, Hill MD, Davalos A, et al. Efficacy of endovascular thrombectomy in patients with M2 segment middle cerebral artery occlusions: meta-analysis of data from the HERMES Collaboration. *J Neurointerv Surg* 2019; 11: 1065–1069.
2) Uchiyama N. Anomalies of the middle cerebral artery. *Neurol Med Chir (Tokyo)* 2017; 57: 261–266.
3) Komiyama M, Nakajima H, Nishikawa M, et al. Middle cerebral artery variations: duplicated and accessory arteries. *AJNR Am J Neuroradiol* 1998; 19: 45–49.
4) Pressman E, Amin S, Renati S, et al. Middle cerebral artery duplication: a near miss for stroke thrombectomy. *Cureus* 2021; 13: e15220.
5) Teal JS, Rumbaugh CL, Bergeron RT, et al. Anomalies of the middle cerebral artery: accessory artery, duplication, and early bifurcation. *Am J Roentgenol Radium Ther Nucl Med* 1973; 118: 567–575.
6) Kai Y, Hamada J, Morioka M, et al. Treatment of unruptured duplicated middle cerebral artery aneurysm: case report. *Surg Neurol* 2006; 65: 190–193; discussion, 193.
7) Chang HY, Kim MS. Middle cerebral artery duplication: classification and clinical implications. *J Korean Neurosurg Soc* 2011; 49: 102–106.
8) Yamamoto H, Marubayashi T, Soejima T, et al. Accessory middle cerebral artery and duplication of middle cerebral artery--terminology, incidence, vascular etiology, and developmental significance. *Neurol Med Chir (Tokyo)* 1992; 32: 262–267.
9) Ospel JM, Goyal M. A review of endovascular treatment for medium vessel occlusion stroke. *J Neurointerv Surg* 2021; 13: 623–630.
10) Liang W, Wang Y, Du Z, et al. Intraprocedural angiographic signs observed during endovascular thrombectomy in patients with acute ischemic stroke: a systematic review. *Neurology* 2021; 96: 1080–1090.
11) Sanossian N, Saver JL, Alger JR, et al. Angiography reveals that fluid-attenuated inversion recovery vascular hyperintensities are due to slow flow, not thrombus. *AJNR Am J Neuroradiol* 2009; 30: 564–568.
12) Cho KH, Kim JS, Kwon SU, et al. Significance of susceptibility vessel sign on T2*-weighted gradient echo imaging for identification of stroke subtypes. *Stroke* 2005; 36: 2379–2383.
13) Koge J, Kato S, Hashimoto T, et al. Vessel wall injury after stent retriever thrombectomy for internal carotid artery occlusion with duplicated middle cerebral artery. *World Neurosurg* 2019; 123: 54–58.
14) Arakawa T, Hiramatsu H, Kida S, et al. A case of acute embolism of the accessory middle cerebral artery treated using ADAPT thrombectomy without lesion passing. *NMC Case Rep J* 2021; 8: 805–810.