www.surgicalneurologyint.com



Surgical Neurology International

Editor-in-Chief: Nancy E. Epstein, MD, Clinical Professor of Neurological Surgery, School of Medicine, State U. of NY at Stony Brook.

SNI: Neurovascular

Editor Kazuhiro Hongo, MD



Shinshu University, Matsumoto, Japan

Anterior cerebral artery dissection for a patient with ipsilateral aplastic or twig-like middle cerebral artery: An illustrative case report

Toshitsugu Terakado¹, Yuji Matsumaru², Eiichi Ishikawa³

¹Department of Neurosurgery, Koyama Memorial Hospital, Kashima, Departments of ²Stroke Prevention and Treatment, and ³Neurosurgery, University of Tsukuba, Tsukuba, Japan.

E-mail: *Toshitsugu Terakado - toshi_0211jp@yahoo.co.jp; Yuji Matsumaru - yujimatsumaru@md.tsukuba.ac.jp; Eiichi Ishikawa - e-ishikawa@md.tsukuba.ac.jp



Case Report

*Corresponding author: Toshitsugu Terakado, Department of Neurosurgery, Koyama Memorial Hospital, Kashima, Japan.

toshi_0211jp@yahoo.co.jp

Received : 17 February 2023 Accepted : 12 April 2023 Published: 28 April 2023

DOI 10.25259/SNI_170_2023

Quick Response Code:



ABSTRACT

Background: An aplastic or twig-like middle cerebral artery (Ap/T-MCA) is a rare anomaly, which sometimes causes ischemic infarction. Collateral flow from the ipsilateral anterior cerebral artery (ACA) is important for patients with Ap/T-MCA. If ipsilateral ACA stenosis or occlusion occurs, a large infarction with a wider field than the ACA territory could happen. First, mechanical thrombectomy was performed for the right ACA near occlusion caused by arterial dissection with ipsilateral Ap/T-MCA in this case. Second, Wingspan stenting was performed for the right ACA restenosis.

Case Description: A 77-year-old female presented to the hospital with the left hemiparesis. We diagnosed a right ACA infarction caused by right ACA occlusion. Digital subtraction angiography showed right Ap/T-MCA and ipsilateral ACA near occlusion. Thrombectomy was performed, and recanalization was achieved with mild ACA stenosis. The lesion was the dissection due to angiographical finding. Two months after treatment, transient left hemiparesis occurred and right ACA stenosis progressed. Computed tomography perfusion showed hypoperfusion of the right hemisphere. Wingspan stenting was performed from the left internal carotid artery through the anterior communicating artery with an intermediate catheter. The patient was discharged without any neurological deficit.

Conclusion: We reported the first case of a patient who underwent Wingspan stenting for the right ACA dissection with Ap/T-MCA. Short-term follow-up and aggressive intervention should be considered for collateral pathway dissection with Ap/T-MCA because the symptoms can become serious. The patients with Ap/T-MCA should be cautious about the collateral pathway arterial changes in particular ipsilateral ACA due to the increasing hemodynamic stress.

Key words: Anterior cerebral artery dissection, Aplastic or twig-like middle cerebral artery, Wingspan

INTRODUCTION

An aplastic or twig-like middle cerebral artery (Ap/T-MCA) is a rare occlusive lesion with reticular vessels in the unilateral middle cerebral artery (MCA); its prevalence is very low, at approximately 0.088-1.17%.[1,16,20,26] Only a few studies have reported that Ap/T-MCA causes cerebral infarction (CI).^[1,5,6,17,19,20,22,23,25,28] In patients with Ap/T-MCA, collateral circulation develops, particularly from the ipsilateral anterior cerebral artery (ACA). We report a restenosis

This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-Share Alike 4.0 License, which allows others to remix, transform, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms. ©2023 Published by Scientific Scholar on behalf of Surgical Neurology International

case of the right A2 acute occlusion due to arterial dissection with ipsilateral Ap/T-MCA treated by thrombectomy first, and then by Wingspan stenting. Arterial dissection could occur due to the hemodynamic stress for the collateral artery of Ap/T-MCA, and we offer several suggestions for the mechanism of ACA dissection.

CASE DESCRIPTION

A 77-year-old woman with untreated hypertension and hyperlipidemia presented to our hospital with sudden left hemiparesis. National Institutes of Health Stroke Scale for the patient was 6. Electrocardiogram at admission revealed sinus rhythm and no atrial fibrillation. Brain magnetic resonance imaging (MRI) showed right medial side of the anterior lobe infarction. Brain magnetic resonance angiography (MRA) showed right A2 occlusion and poor visualized right MCA [Figure 1]. Emergent digital subtraction angiography showed right A2 near occlusion and reticulated vessels developed from the proximal right MCA [Figures 2a and b]. These results indicated that reduced flow of the right MCA due to occlusion of the right A2 caused the left hemiparesis. We immediately performed thrombectomy of the right A2. After deploying stent retriever to the right A2 segment, the lesion was identified as a stenotic lesion. Oral administration of loading-dose dual-antiplatelet therapy (200-mg aspirin and 300-mg clopidogrel) was initiated. After the stent was slowly retrieved, recanalization of mild stenosis was accomplished. The lesion was considered dissection due to the presence of dilatation and string sign in retrospect [Figure 2c]. The symptoms of the patient improved without any recoiling of MRI 1 week after the treatment, and the MRI showed no enlargement of the CI, and MRA showed adequate dilatation of the right A2 segment [Figures 3a-c]. Basi-pararel anatomical scanning showed no right M1 form [Figure 3d]. We continued dual-antiplatelet therapy (100-mg aspirin and 75-mg clopidogrel). However, transient left hemiparesis frequently occurred 2 months after treatment. MRI showed worsening of stenosis in the right A2 segment and an increase in the right ACA area infarction [Figures 3e and f]. Hence, we switched Clopidogrel with prasugrel (3.75 mg).

Since perfusion study of the CT scan showed a remarkable decrease in cerebral blood flow (CBF) in the right



Figure 1: (a) Diffusion-weighted imaging on magnetic resonance imaging showed cerebral infarction in the right anterior cerebral artery area. (b) Brain magnetic resonance angiography showed right A2 occlusion (white arrow) and poor right middle cerebral artery blood flow (white arrowhead).



Figure 2: (a) Preoperative DSA of the right common carotid artery showing reticulated vessels developed from the proximal right MCA. Antegrade blood flow from the distal MCA was slow. The right A2 segment was not visualized. (b) Preoperative DSA of the left internal carotid artery showed right A2 near occlusion (white arrowhead). (c) Lateral view of preoperative DSA showed proximal right A2 near occlusion and distal right A2dissection (arrowhead). DSA: Digital subtraction angiography, MCA: Middle cerebral artery.



Figure 3: (a and b) DWI showed no newly infarction. (c) Brain MRA showed recanalization of the right A2 segment. (d) Basi-parallel anatomical scanning showed no right M1 form. (e) DWI 2 months later showed an enlarged infarction in the right ACA area. (f) Brain MRA showed right A2 restensis. (g) Perfusion study of computed tomography scan showed a decrease in the cerebral blood flow at the right ACA and anterior middle cerebral artery. DWI: Diffusion-weighted imaging, MRA: Magnetic resonance angiography, ACA: Anterior cerebral artery.

hemisphere was achieved [Figure 3g], we performed an additional endovascular treatment under general anesthesia 7 days after second admission. For increasing the stability, AXS Catalyst5 110 cm (Stryker) was placed in the left A1 segment with AXS Offset (Stryker). The catheter was wedged, and the antegrade flow from the left A1 segment was stopped. Right internal carotid angiography showed that the bilateral A2 segment was described, and we judged that continuing treatment with a short-term wedge of the left A2 segment was possible. Wingspan 2.5×15 mm (Stryker) was deployed at the dissecting right A2 segment. The right A2 segment was adequately dilated, and the flow of the right distal ACA was improved [Figure 4]. The wedge time of the left A1 segment was 15 min. The patient was discharged without any neurological deficit or other obvious complications. Restenosis was not observed 6 months after stenting [Figure 5].

For this report, consent was obtained from the patient.

DISCUSSION

Ap/T-MCA is a rare vascular anomaly that sometimes causes ischemic infarction.^[1,5,6,17,19,20,22,23,25,28] In such cases, cerebral ischemia occurs due to low CBF of the Ap/T-MCA area. For preventing stroke recurrences, superficial temporal arteryto-MCA bypass (+indirect bypass) was performed in some previous cases.^[5,19,20,22,23,28] As ipsilateral ACA is the main collateral pathway in the Ap/T-MCA patients, ipsilateral ACA stenosis can cause severe CBF decrease not only in the ACA territory but also in MCA territory, as seen in the present case. Three cases of Ap/T-MCA with contralateral MCA stenosis/occlusion or Ipsilateral internal carotid artery (ICA) stenosis have been reported previously [Table 1]; however, no cases of an ipsilateral A2 lesion have been reported. We noted that patients with Ap/T-MCA at a high risk of atherosclerosis should be monitored through strict and repeated imaging, particularly regarding the progression of collateral artery stenosis.

In our case, the underlying cause of ACA stenosis could be arterial dissection. We hypothesize that ipsilateral ACA that is the main collateral pathway for the Ap/T-MCA hemisphere is under continuous hemodynamic stress and causes arterial dissection. Other arterial changes of the ipsilateral ACA, including aneurysmal formation, probably due to hemodynamic stress, were seen with Ap/T-MCA [Table 2].^[3,4,8,9,13,21,26] In these cases, aneurysm formation occurred due to hemodynamic stress caused by the increased flow of collateral arteries. Pathological findings of the



Figure 4: (a) Right A2 restenosis before the second treatment. (b) Catalyst5 was inserted into the left A1 segment, and antegrade blood flow from the left A1 segment disappeared. (c) The bilateral A2 segment was described from the right A1 segment. (d and e) Catalyst5 was placed in the left A1 segment, and Wingspan was placed in the right A2 segment after stenting. (f) Wingspan was dilated on cone-beam computed tomography.



Figure 5: (a) Diffusion-weighted imaging showed no new infarction. (b) Computed tomography angiography showed recanalization of the right A2 segment.

aneurysm associated with the Ap/T-MCA have been reported previously.^[27] The report described that the mother vessel on the proximal side of the aneurysm was the stratification of the intimal elastic plate, and the intimal elastic plate was disrupted with intimal thickening at the aneurysm neck. These changes occurred due to hemodynamic stress. Hemorrhagic stroke occurred if the outer arterial wall ruptured due to these changes. As only intimal elastic plate was teared, arterial dissection with stenosis occurred in the present case.

For treating ACA stenosis or occlusion with ischemic infarction, superficial temporal artery-radial artery graft-A3 bypass or A3–A3 bypass surgery has been reported to be effective.^[10,14,24] However, these surgical methods need

high skill for bypass surgery. Other issues regarding these methods are the interruption of the affected blood flow during anastomosis and the risk of bilateral ACA occlusion. In addition, these surgeries cannot prevent thromboembolic stroke from the dissection.

Owing to these reasons, stent implantation is considered reasonable for the intracranial artery dissection in ischemic stroke,^[12] as it can improve the antegrade flow and avoid further extension of dissection and occurrence of new thromboembolic event. In a previous study, only one case of using enterprise stent for ACA stenosis has been reported.^[7] An intracranial stent, Wingspan, is covered by insurance in Japan for treating the recurrences of intracranial

Table 1: The list of aplastic or twig-like MCA with the other stenosis lesions.												
Case No.	Authors and year	Age	Sex	Side	Clinical presentation	Vascular risk	Coexiting athreosclerotic vascular change	Treatment				
1	Takahashi <i>et al.</i> 1997 ^[22]	54	F	R	TIA, CI	None	Unilateral ICA stenosis	STA-MCA anastomosis				
2	Edgell et al. 2010 ^[6]	42	F	R	TIA	Tobacco, obesity	Contralateral. MCA stenosis	ND				
3	Takeda <i>et al</i> . 2022 ^[23]	62	М	R	TIA, CI	HT, DM	Contralateral. MCA stenosis	STA-MCA anastomosis				
4	Present case	77	F	R	CI	HT	Unilateral ACA stenosis	ACA stenting				
101 1			1					26261				

ACA: Anterior cerebral artery, CI: Cerebral infarction, DM: Diabetes mellitus, F: Female, HT: Hypertension, ICA: Internal carotid artery, M: Male, MCA: Middle cerebral artery, ND: Not described, R: Right, STA: Superficial temporal artery, TIA: Transit ischemic attack

 Table 2: The list of aplastic or twig-like MCA with anterior cerebral artery aneurysms.

Case No.	Authors and year	Age	Sex	Side	Clinical presentation	Aneurysm location	Treatment				
1	Han <i>et al.</i> 1994 ^[9]	34	F	L	SAH	A1-accesory MCA	Clipping				
2	Amagasaki <i>et al</i> . 1998 ^[3]	64	Μ	L	Incidental	A1-heubner	Clipping				
3	Cekige <i>et al</i> . 2005 ^[4]	32	Μ	L	SAH	A1-accesory MCA	Coil				
4	Kim <i>et al.</i> 2005 ^[13]	64	F	L	SAH	A1, Acom	Clipping				
5	Shin <i>et al</i> . 2014 ^[21]	46	F	L	SAH	A1-accesory MCA	Clipping				
6	Fukuda <i>et al</i> . 2018 ^[8]	60	F	R	SAH	A1-accesory MCA	Coil				
7	Viso <i>et al</i> . 2021 ^[26]	59	F	L	SAH	Acom, Pcom	Coil				
8	Viso <i>et al.</i> 2021 ^[26]	48	F	R	SAH	Acom	Coil				
Acom: Anterior communicating artery, F: Female, MCA: Middle cerebral artery, L: Left, M: Male, R: Right, SAH: Subarachnoid hemorrhage,											

Pcom: Posterior communicating artery

stenotic lesions. However, other intracranial stents for coil embolization and coronary stents are practically used for such intracranial stenotic lesions, because they can easily be deployed.^[11] Wingspan is an open-cell, self-expanding stent that is guided over-the-wire stent housed in a specialized outer sheath. The guidance of Wingspan may be difficult or the stent may not be deployed in the distal access. To solve the problem of deploying Wingspan in the ACA, an intermediate catheter is essential. In Japan, in April 2022, intermediate catheters were started to be used for highly tortuous lesions. As a result, if the intermediate catheter can be guided to the A1 segment, the supporting power will increase and Wingspan deployment may become possible. Note that if the intermediate catheter is guided to the A1 segment, the antegrade distal flow will disappear due to the catheter wedge. If the blood flow from the contralateral A1 segment is not secured, ischemic complications may occur. Moreover, there is a risk of ipsilateral A1 perforator infarction. The duration of intermediate catheter placement in the A1 segment should be minimized. Wingspan placement should be considered only in cases where these conditions are applicable. In applicable cases of Wingspan placement, it has the great advantage of maintaining the antegrade blood flow compared with bypass surgery.

Our statement from this case report has some limitations. First, long-term outcomes are unknown because the

patient was only followed up 3 months after treatment. The restenosis rate after Wingspan stenting was reported to be 11.1–17.6%.^[2,18] More cases of restenosis associated with anterior circulation than posterior circulation have been reported.^[15] In addition, calcification and residual stenosis immediately after treatment are considered restenosis factors.^[29] Because Wingspan stenting for ACA stenosis or occlusion is rare, restenosis requires caution.

CONCLUSION

We reported the first case of a patient with Ap/T-MCA complicated by ipsilateral A2 dissection who underwent Wingspan stenting. Short-term follow-up and aggressive intervention should be considered for treating collateral pathway dissection with Ap/T-MCA because the symptoms can become serious with time. The patients with Ap/T-MCA need to be cautious about the collateral pathway arterial changes, especially ipsilateral ACA, due to the increasing hemodynamic stress.

Declaration of patient consent

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

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Akkan K, Ucar M, Kilic L, Celtikci E, Llgit E, Onal B. Infused or twig-like middle cerebral artery. Eur J Radiol 2015;84:2013-8.
- 2. Alexander MJ, Zauner A, Gupta R, Alshekhlee A, Fraser JF, Toth G, *et al.* The WOVEN trial: Wingspan one-year vascular events and neurologic outcomes. J Neurointerven Surg 2021;13:307-10.
- 3. Amagasaki K, Yagishita T, Kawataki T, Kase M, Nukui H. Middle cerebral artery aplasia associated with an aneurysm of the proximal anterior cerebral artery. Acta Neurochir (Wein) 1998;140:1313-4.
- 4. Cekige HS, Peynircioglu B, Saatci I. Endovascular treatment of an "anterior cerebral artery" aneurysm in a patient with "embryonic unfused middle cerebral artery" anomaly: A case report. Neuroradiology 2005;47:690-4.
- 5. Cho KC, Kim JJ, Jang CK, Hong CK, Joo Y, Kim YB. Rete middle cerebral artery anomalies: A unifying name, case series, and literature review. J Neurosurg 2018;131:453-61.
- 6. Edgell RC, Boulos AS, Haghighi AB, Bernardini GL, Yavagal DR. Middle cerebral artery stenosis associated with moyamoya pattern collateralization. Front Neurol 2010;1:119.
- Feng Z, Duan G, Zhang P, Chen L, Xu Y, Hong B, *et al.* Enterprise stent for the treatment of symptomatic intracranial atherosclerotic stenosis: An initial experience of 44 patients. BMC Neurol 2015;15:187.
- 8. Fukuda Y, Matsunaga Y, Hirayama K, Yoshimura S, Senkawa T, Sato K, *et al.* A case of aplastic or twig-like middle cerebral artery associated with a ruptured A1 aneurysm at the origin of the anomalous collateral artery. Jpn J Stroke 2018;40:75-80.
- 9. Han DH, Gwak HS, Chung CK. Aneurysm at the origin of accessory middle cerebral artery associated with middle cerebral artery aplasia: Case report. Surg Neurol 1994;42:388-91.
- Horiuchi T, Ichinose S, Agata M, Ito K, Hongo K. STA-ACA bypass using the ipsilateral free STA graft as an interposition graft and A3-A3 anastomosis for the treatment of bilateral ACA steno-occlusive ischemia. Acta Neurochir (Wien) 2018;160:779-82.
- 11. Izumi T, Nishihori M, Imamura H, Iihara K, Sakai N, JR-NET Investigators. Endovascular therapy for intracranial artery stenosis: Results from the Japanese Registry of Neuroendovascular Therapy (JR-NET)3. Neurol Med Chir (Tokyo) 2020;60:256-63.
- 12. Kim DJ, Kim BM, Suh SH, Kim DI. Self-expanding stent placement for anterior circulation intracranial artery dissection presenting with ischemic symptoms. Neurosurgery

2015;76:158-64.

- 13. Kim MS, Oh CW, Hur JW, Lee JW, Lee HK. Aneurysms located at the proximal anterior cerebral artery and anterior communicating artery associated with middle cerebral artery aplasia: Case report. Surg Neurol 2005;64:534-7.
- 14. Kiyofuji S, Inoue T, Hasegawa H, Tamura A, Saito I. A3-A3 anastomosis and superficial temporal artery-radial artery graft-A3 bypass to treat bilateral ACA steno-occlusive hemodynamic ischemia with cognitive and executive dysfunction: A technical note. Acta Neurochir (Wien) 2014;156:2085-93.
- 15. Levy EI, Turk AS, Albuquerque FC, Niemann DB, Aagaard-Kienitz B, Pride L, *et al.* Wingspan in-stent restenosis and thrombosis: Incidence, clinical presentation, and management. Neurosurgery 2007;61:644-50.
- Liu HM, Lai DM, Tu YK, Wang YH. Aneurysms in twig-like middle cerebral artery. Cerebravasc Dis 2005;20:1-5.
- 17. Lutz T, Monnings P, Ayzenberg I, Lukas C. Twig-like middle cerebral artery: A seldom vessel anomaly of important relevance. Clin Neuroradiol 2018;28:441-3.
- Ma N, Zhang Y, Shuai J, Jiang C, Zhu Q, Chen K, *et al.* Stenting for symptomatic intracranial arterial stenosis in China: 1-year outcome of a multicentre registry study. Stroke Vasc Neurol 2018;3:176-84.
- Matsunaga Y, Izumo T, Morofuji Y, Horie N, Hayashi K, Matsuo T. Revascularization for aplastic or twiglike middle cerebral artery: A case report. J Stroke Cerebrovasc Dis 2018;27:e78-9.
- 20. Seo BS, Lee YS, Lee HG, Lee JH, Ryu KY, Kang DG. Clinical and radiological features of patients with aplastic or twiglike middle cerebral arteries. Neurosurgery 2012;70:1472-80.
- 21. 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 (Wein) 2014; 156:1637-46.
- 22. Takahashi M, Fujimoto T, Suzuki R, Asai J, Miyo T, Hokaku H. A case of spontaneous middle cerebral artery occlusion associated with a cerebral aneurysm angiographically disappearing after STA-MCA anastomosis. No Shinkei Geka 1997;25:727-32.
- 23. Takeda H, Yanaka K, Onuma K, Nakamura K, Ishii K, Ishikawa E. Aplastic or twiglike middle cerebral artery with contralateral middle cerebral artery stenosis showing transient ischemic attack: Illustrative case. J Neurosurg Case Lessons 2022;3:CASE22121.
- 24. Tsunoda S, Inoue T, Segawa M, Okubo S, Akabane A. Revascularization to the ACA: Effectiveness and variation of the STA-RAG-A3 bonnet bypass. Acta Neurochir (Wien) 2021;163:3483-93.
- 25. Uchiyama T, Okamoto H, Koguchi M, Tajima Y, Suzuyam K. A case of aplastic or twig-like middle cerebral artery presenting with an intracranial hemorrhage two years after a transient ischemic attack. No Shinkei Geka 2016;44:143-8.
- 26. Viso R, Lylyk I, Albiña P, Lundquist J, Scrivano E, Lylyk P. Hemorrhagic events associated with unfused or twig-like configuration of the middle cerebral artery: A rare vascular anomaly with clinical relenvance. Interv Neuroradiol

2021;27:285-90.

- 27. Watanabe N, Marushima A, Hino T, Minamimoto S, Sato M, Ito Y, *et al.* A ruptured aneurysm in aplastic or twig-like middle cerebral artery: A case report with histological investigation. NMC Case Rep J 2022;9:7-12.
- Yamada D, Ishibashi R, Kinosada M, Kurosaki Y, Handa A, Chin M, *et al.* Aplastic or twig-like middle cerebral artery with short-term ischemia and bleeding. Jpn J Stroke 2020;42:190-5.
- 29. Zhang K, Li TX, Wang ZL, Gao BL, Gu JJ, Gao HL, et al.

Factors affecting in-stent restenosis after angioplasty with Enterprise stent for intracranial atherosclerotic diseases. Sci Rep 2021;11:10479.

How to cite this article: Terakado T, Matsumaru Y, Ishikawa E. Anterior cerebral artery dissection for a patient with ipsilateral aplastic or twiglike middle cerebral artery: An illustrative case report. Surg Neurol Int 2023;14:154.

Disclaimer

The views and opinions expressed in this article are those of the authors and do not necessarily reflect the official policy or position of the Journal or its management. The information contained in this article should not be considered to be medical advice; patients should consult their own physicians for advice as to their specific medical needs.