

Acute Thrombectomy for Contralateral Internal Carotid Artery Occlusion after Revascularization Surgery for Quasi-moyamoya Disease: A Case Report

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Abstract

Ischemic complications can occur after revascularization surgery for moyamoya disease, but acute contralateral internal carotid artery (ICA) occlusion is an extremely rare complication. The patient was a 51-year-old woman with no medical history. Left frontal lobe infarction and bilateral ICA terminal stenosis were identified by repeated transient right paresis and aphasia. We diagnosed her with quasi-moyamoya disease associated with hyperthyroidism and performed revascularization surgery for the symptomatic left side. Although neurological symptoms did not worsen immediately after the surgery, disturbance of consciousness, right conjugate deviation, and left paresis appeared 4 hr after the surgery. New infarction appeared in the right frontal lobe, and the blood signal beyond the right middle cerebral artery (MCA) disappeared on MRI and MRA. Mechanical thrombectomy (MT) using a suction catheter improved antegrade blood flow in the MCA. The left paresis remained at discharge (modified Rankin Scale score = 4), but she was able to walk independently 3 months after the operation and was independent at home. Acute contralateral ICA occlusion after revascularization for moyamoya disease is an extremely rare complication, but the symptoms can be severe and treatment should be considered. To the best of our knowledge, there have been no reports of MT for postoperative acute contralateral ICA occlusion. Since the results of endovascular treatment such as percutaneous transluminal angioplasty and stent placement for patients with moyamoya disease are poor, MT using an aspiration catheter could be a good treatment option.

Keywords: quasi-moyamoya disease, contralateral ischemic complication, mechanical thrombectomy, revascularization

Introduction

Ischemic complications after revascularization surgery for moyamoya disease occur in 4.7–22.2% of cases.^{1–3)} Most ischemic complications occur on the surgical side, and occlusion of the contralateral internal carotid artery (ICA) in the acute phase after revascularization surgery is an extremely rare complication. The frequency is approximately 0.6%, and when it occurs, it often follows a serious course.⁴⁾ We encountered a case of quasi-moyamoya

disease associated with hyperthyroidism, in which right ICA occlusion occurred immediately after left revascularization surgery and mechanical thrombectomy (MT) was performed.

Case Report

The patient was a 51-year-old woman with no medical history. She was diagnosed with left frontal lobe infarction and bilateral ICA terminal stenosis due to repeated transient right paresis and aphasia (Fig. 1A and 1B). She had no family history of moyamoya disease. Blood examination showed no other findings suggestive of abnormal coagulation or vasculitis, except a slightly high von Willebrand factor (vWF) activity of 196%. Free T3 22.71 pg/ml, free T4 >7.77 ng/dl, thyroid stimulating hormone <0.005 μ IU/ml, and

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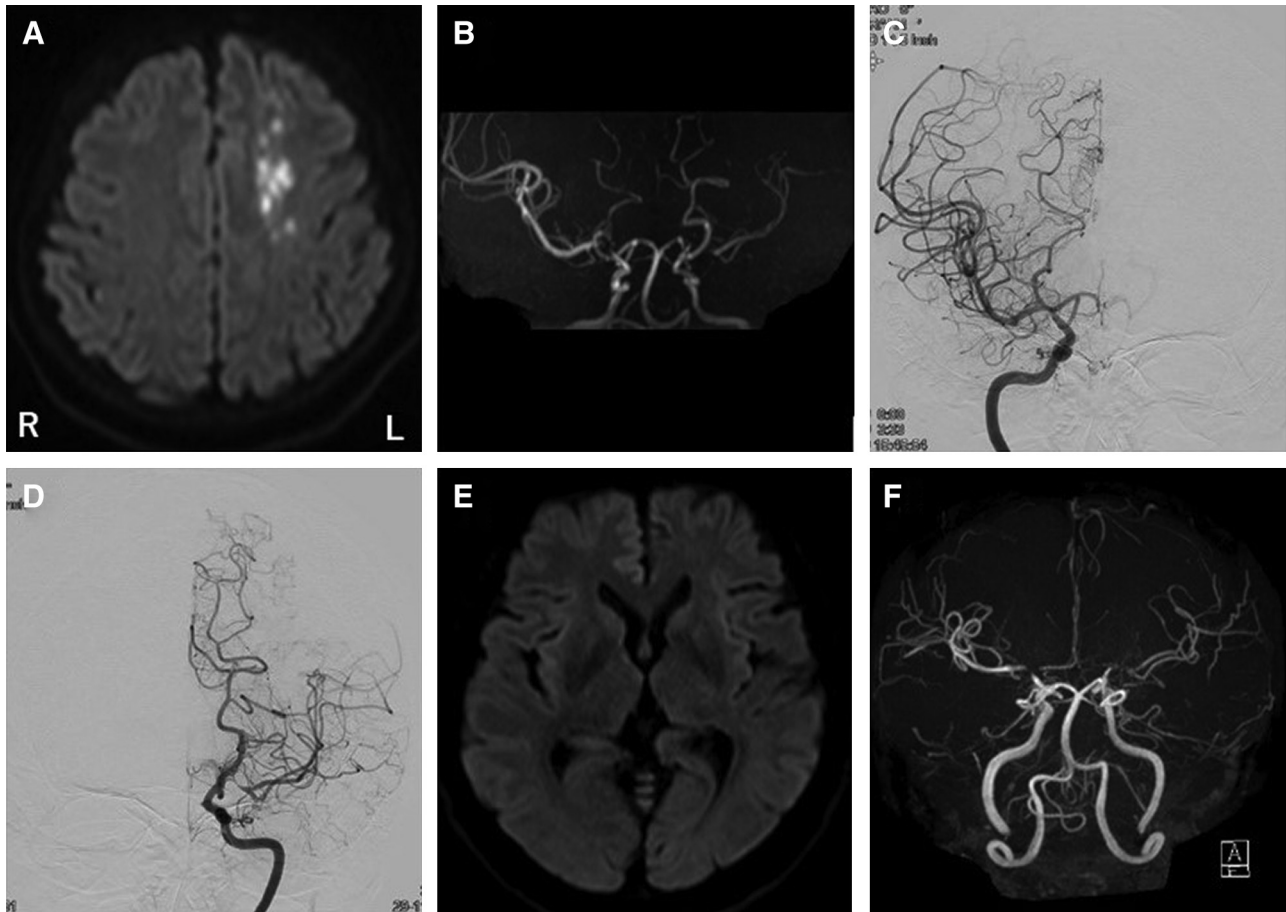


Fig. 1 Preoperative MRI and digital subtraction angiography. (A) High intensity was observed in the left frontal watershed area on initial MRI DWI. (B) MRA showed stenosis of the bilateral ICA terminus. Depiction of the left MCA was reduced compared to the right MCA. (C and D) The right and left ICAG (A–P view) before surgery. ICA stenosis was more severe on the left side than on the right side. (E and F) MRI obtained the day before surgery shows no new cerebral infarction and slightly improved blood flow signal within the ACA; bilateral ICA terminal stenoses remain. A–P: anterior–posterior, ACA: anterior cerebral artery, DWI: diffusion-weighted imaging, ICA: internal carotid artery, ICAG: internal carotid angiography, MCA: middle cerebral artery.

thyroid stimulating hormone receptor antibody 7.2 IU/L indicated hyperthyroidism, so we diagnosed her with quasi-moyamoya disease associated with hyperthyroidism. She started taking clopidogrel 75 mg and potassium iodide 50 mg, and we confirmed normalization of thyroid function. Digital subtraction angiography (DSA) revealed severe stenosis at the terminal of the left ICA. Her left middle cerebral artery (MCA) had antegrade blood flow, but the flow was slow, and her left anterior cerebral artery (ACA) was shed. Although stenosis was also found at the terminal of the right ICA, we decided to treat the symptomatic left ICA stenosis (Fig. 1C and 1D).

Four months after the onset of symptoms (3 months after the DSA), we performed a left superficial temporal artery–MCA bypass and encephalo-duro-myopercranial synangiosis. A preoperative MRI

showed no new cerebral infarction and slightly improved blood flow signal within ACA; however, bilateral ICA terminal stenoses remained (Fig. 1E and 1F). The operation time was 4 hr and 58 min, the amount of bleeding was 180 ml, and the intraoperative in–out balance was 800 ml. Postoperative CT showed no bleeding or infarction, and she was awake and had no deterioration of neurological findings immediately after the operation. Intraoperative blood pressure and postoperative blood pressure were maintained at approximately 100–120 mmHg.

Four hours after the surgery, consciousness disorder (Glasgow Coma Scale E2V1M6), right conjugate deviation, and left hemiparalysis appeared. Multiple new area of infarction within the right frontal lobe, insular cortex, caudate nucleus, and internal capsule were noted on MRI. Alberta Stroke Program Early

Computed Tomography-DWI (ASPECTS + W) was 3.⁵⁾ Bilateral MCA blood flow signal disappeared in MRA (Fig. 2A and 2B). There were no signal changes noted on the FLAIR image. Blood flow beyond the right ICA terminus was extremely delayed in DSA, and MT was performed using a suction catheter (Fig. 2C–2F). Under local anesthesia, 9 Fr Optimo 90 cm (Tokai Medical Products, Aichi, Japan) was applied to the right ICA. Rebar 18 153 cm (Medtronic, Minneapolis, MI, USA) and Chikai 14 guide wires (Asahi Intecc, Aichi, Japan) were introduced to the ICA terminus, and only the micro guide wire was lesion-crossed to the MCA. A Penumbra 4MAX

catheter (Penumbra, Alameda, CA, USA) was pushed toward the proximal part of the obstruction and removed while sucking. Two passes of thrombus aspirations were performed, and modified thrombolysis in cerebral infarction was 2c.⁶⁾ The procedure was completed after confirming that complete occlusion did not occur after 15 min of follow-up (onset puncture: 39 min, puncture recanalization: 21 min, onset recanalization: 60 min). Immediately after the MT, the disturbance of consciousness improved, and the patient was able to communicate. MRI showed new infarcts in the frontal cortex and basal ganglia instead of extensive infarcts in the

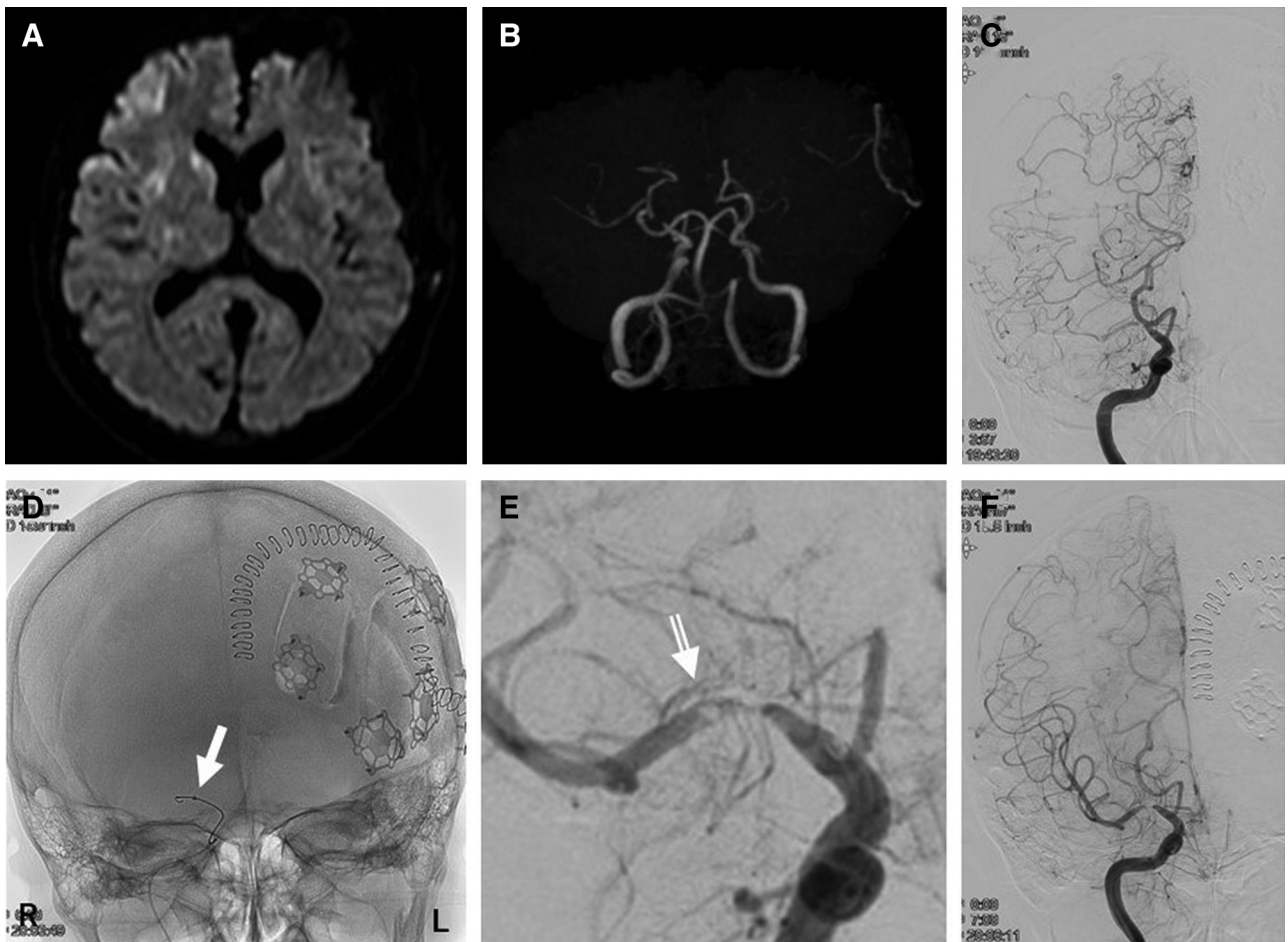


Fig. 2 MRI, MRA, and digital subtraction angiography just after the appearance of postoperative symptoms. (A) A high-intensity area appeared in the right frontal lobe on MRI DWI. (B) The MCA blood flow signal disappeared in MRA. (C) The right ICAG (A–P view). Blood flow beyond the ICA terminus was delayed. (D) Rebar 18 153 cm (Medtronic, Minneapolis, MI, USA) and Chikai 14 guide wires (Asahi Intech, Aichi, Japan) were guided to the ICA terminus, and only the Micro guide wire was lesion-crossed to MCA. A Penumbra 4MAX catheter (Penumbra, Alameda, CA, USA) was pressed against the obstruction (white arrow). (E) The right ICAG after one pass. The MCA recanalized slightly, and thrombus was revealed just distal of stenosis (white double arrow). (F) Post ICAG showed antegrade blood flow of the MCA. A–P: anterior–posterior, DSA: digital subtraction angiography, DWI: diffusion-weighted imaging, ICA: internal carotid artery, ICAG: internal carotid angiography, MCA: middle cerebral artery.

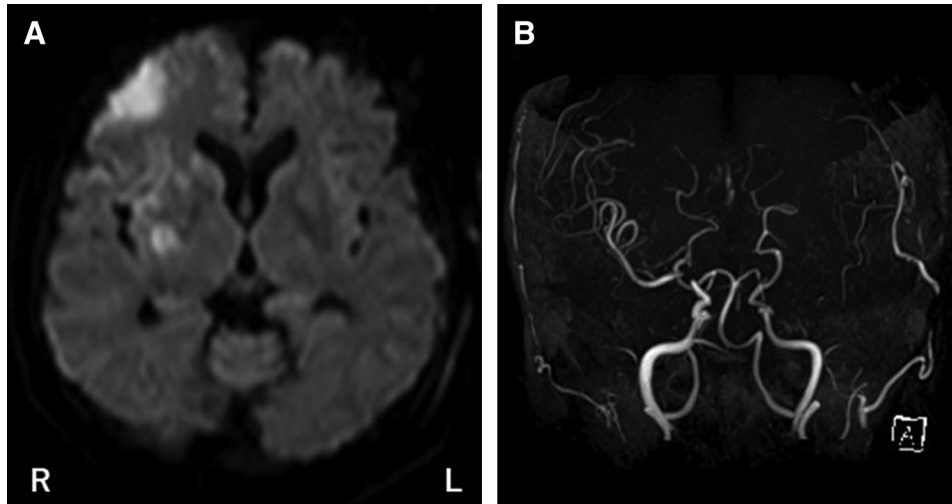


Fig. 3 MRI and MRA after MT. (A) A high-intensity area was observed in part of the frontal lobe and basal ganglia on the follow-up MRI DWI. **(B)** The right MCA blood flow signal improved in MRA at day 12 after the procedure. The signal of the left MCA remained absent, but no infarction appeared. DWI: diffusion-weighted imaging, MCA: middle cerebral artery, MT: mechanical thrombectomy.

right cerebral hemisphere (Fig. 3A). No local hyperperfusion was found in the cerebral blood flow test performed the day after the operation. Echocardiography, carotid echo, Holter electrocardiogram, and blood sampling were performed again to determine the cause of cerebral infarction, but no evidence of embolism was found. Oral administration of clopidogrel was continued, and MRA on the 12th day after the surgery showed that the blood flow signal of the right MCA was improved (Fig. 3B). Left paresis (manual muscle test 3) remained at discharge (modified Rankin Scale score = 4), but she was able to walk independently 3 months after the operation and was independent at home. There was no relapse of symptoms after 18 months of follow-up. The patient provided consent for publication of this case report.

Discussion

We experienced a case of acute contralateral ICA occlusion that occurred after revascularization surgery for quasi-moyamoya disease. We performed MT using an aspiration catheter and obtained relatively good results. To the best of our knowledge, there have been no reports of MT cases performed after revascularization surgery for moyamoya disease.

There have been few reports of cerebral infarction on the contralateral side during the perioperative period of revascularization for moyamoya disease (Table 1). The frequency of contralateral infarction is 0.62%–5.1% and often presents with single or multiple small infarctions in the cortical region.

The onset is frequent after POD1 and is associated with postoperative excessive hypotension in the presence of contralateral lesions and advanced stage according to the Suzuki classification. Perioperative hemodynamic blood flow changes are thought to be one of the causes of infarction.^{1,3,4,7–10)} Sussman et al. reported that among 1446 cases of revascularization for moyamoya disease, 8 cases (0.6%) had cerebral infarction due to contralateral ICA occlusion. It appeared earlier after surgery than a small infarction caused by hemodynamic changes, and the symptoms became more serious. It was considered that the decrease in blood flow due to the decrease in demand for antegrade blood flow in the skull may have caused thrombus formation and contributed to obstruction.⁴⁾ The present case was consistent with a previous report in which the onset was as early as 4 hr after surgery, the infarct area included the basal ganglia, and the symptoms were severe.⁴⁾ In addition, in this case, a thrombus was found at the terminal of the ICA on digital subtraction angiography immediately after the appearance of symptoms. Since there were no findings suggestive of artery-to-artery embolism or paradoxical cerebral embolism associated with intraoperative bed rest, it is possible that thrombosis was promoted by decreased blood flow in the contralateral ICA associated with postoperative hypotension.

It has been reported that patients with hyperthyroidism also have embolism, including sinus thrombosis.¹¹⁾ Patients with hyperthyroidism often have high fibrinogen and low protein C levels, and T3

Table 1 Literature review about contralateral ischemic complication after surgery for moyamoya disease

No.	Authors (year)	No. of operations	Postoperative contralateral infarction (%) / ICO (%)	Adult/ Child (mean age)	Onset of symptoms (no.)	Site of infarction (no.)	Etiology	Treatment
1	Khan et al. (2003) ⁷⁾	23	1 (4.3)	Adult: 4 (34) and child: 19 (8)	POD1	MCA territory	n.d.	Medication
2	Kim et al. (2005) ⁸⁾	170	5 (2.9)	Child (6.8)	After POD1 (5)	n.d.	n.d.	n.d.
3	Jung et al. (2011) ⁹⁾	79	4 (5.1)	Adult (37.9)	POD1 (1), POD2 (3)	Parietal cortex (1), frontal/parietal/occipital cortex (1), frontal cortex (1), temporal/parietal/occipital cortex (1)	Hemodynamic change	Medication
4	Hatano et al. (2013) ³⁾	84	1 (1.2)	Adult and child (n.d.)	POD3	Frontal cortex	Hemodynamic change	Medication
5	Kazumata et al. (2014) ¹⁾	358	3 (0.84)	Adult (≥18): 177 and child (<18): 181	n.d.	n.d.	n.d.	n.d.
6	Tu et al. (2017) ¹⁰⁾	162	1 (0.62)	Adult (n.d.)	POD2	Frontal cortex	Hemodynamic change	Medication
7	Sussman et al. (2018) ⁴⁾	1446	34 (2.4)/8 (0.6)	Adult and child (n.d.)	Within 12 hr after surgery (8) in ICO cases	Frontal cortex and corona radiata (1), entire cerebral hemisphere (1) in ICO cases	Thrombus formation suspected in ICO cases	Medication and contralateral surgery (5), decompressive craniectomy (1) in ICO cases
8	Present case	37	1 (2.7)	Adult (≥18)	4 hr after surgery	Frontal/insular cortex, caudate/lenticular nucleus, radial crown	Thrombus formation	Mechanical thrombectomy

ICO: Internal carotid artery occlusion, MCA: middle cerebral artery, n.d.: not described.

in the blood has been reported to increase the production of vWF, leading to thrombotic tendencies.^{12,13)} In the present case, we found a slight increase in vWF activity at the first visit, but there were no other findings suggestive of increased coagulation ability. Surgery was performed after confirming normalization of thyroid function, but it is possible that abnormal coagulation associated with

hyperthyroidism affected the course of this case. Atrial fibrillation associated with hyperthyroidism can also cause infarction. Her pulse was stable throughout the surgery and Holter electrocardiography showed no atrial fibrillation, but it cannot be denied that she had paroxysmal atrial fibrillation.

Severe stenosis was revealed at the right ICA terminal after the MT despite there being no signs

on preoperative DSA. While the right ICA terminal stenosis might have progressed prior to the bypass surgery, MRI was negative for progression of right ICA terminal stenosis. We, therefore, believe that the stenotic lesion at the right ICA terminus was not progressing prior to the surgery.

Medication is considered an effective treatment for quasi-moyamoya disease associated with hyperthyroidism.¹⁴⁾ Utku et al. reported that medication alone improved a patient's condition with intracranial vessel stenosis.¹⁵⁾ Our patient was medicated prior to surgery and demonstrated normal thyroid function. The symptoms did not recur after starting the medication; however, the stenosis noted on MRI remained. Ischemia is more likely to occur in the thyrotoxic state and improve as thyroid levels normalize. Despite this, some patients will experience recurrence even after undergoing effective antithyroid therapy. Endo et al. reported the return of neurological symptoms associated with recurrent hyperthyroidism and discussed the efficacy of revascularization surgery.¹⁶⁾ Revascularization surgery appears acceptable for use when areas of vascular stenosis remain, despite adequate medication.

Acute occlusion of the contralateral ICA after surgery is a rare complication, but the prognosis when it develops is poor; therefore, revascularization in the acute phase should be considered.⁴⁾ There have been many reports that endovascular treatment for moyamoya disease is not effective. Revascularization using a stent or percutaneous transluminal angioplasty is said to cause early restenosis and intrastent occlusion, and serious perioperative complications such as dissection have been reported.¹⁷⁻¹⁹⁾ MT using a stent retriever is considered dangerous because the media of blood vessels is broken or thinned in moyamoya disease²⁰⁾ and the wall of blood vessels is inflamed in quasi-moyamoya disease.²¹⁾ There has only been one case report of recombinant tissue plasminogen activator administration and MT using a stent retriever for embolic infarction complicated with moyamoya disease; MT can be performed only in mild cases (Suzuki stages 1 and 2) in which obvious thrombus formation is suggested preoperatively.²²⁾ In the present case, we performed MT using an aspiration catheter. The advantages of an aspiration catheter compared to a stent catheter are that it does not require the passage of the stenosis with the microcatheter and vascular wall injury due to the stent can be avoided. Large-diameter aspiration catheters have been developed to efficiently capture the thrombus; however, in this case, we used a small-diameter aspiration catheter to reduce the risk of vascular injury. Though ASPECTS + W: 3

is generally considered a low score to perform MT, we opted for MT considering the severity of our patient's symptoms and our observation of no signal change on the MRI FLAIR image. Given that recombinant tissue plasminogen activator administration after surgery is not indicated and additional introduction of antiplatelet drugs is generally not recommended, MT using an aspiration catheter is considered a better treatment option.

Conclusion

We performed MT using an aspiration catheter for acute contralateral ICA occlusion that occurred after revascularization surgery for quasi-moyamoya disease. MT using an aspiration catheter could be a treatment option for acute recanalization therapy in patients with moyamoya disease.

Conflicts of Interest Disclosure

No authors have any conflicts of interest.

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