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Case Report

Carotid Cavernous Fistula during Thrombectomy for Acute Ischemic Stroke: A Case Report

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Objective: We report a rare complication, carotid cavernous fistula (CCF), due to vessel perforation during thrombectomy for acute ischemic stroke (AIS).

Case Presentation: An 88-year-old woman underwent thrombectomy for left C4 occlusion of the internal carotid artery. There was strong resistance at the medial C4 while the microguidewire was guided distally, and a CCF was found after deploying and retrieving the stent. It was thought to have been caused by perforation due to intracranial atherosclerotic stenosis of the internal carotid artery.

Conclusion: During thrombectomy for intracranial large vessel occlusion underlying intracranial atherosclerotic stenosis, the risk of vascular injury should be kept in mind.

Keywords ▶ acute cerebral infarction, thrombectomy, vessel perforation, carotid cavernous fistula

Introduction

The incidence of vascular perforation related to thrombectomy for acute ischemic stroke (AIS) is reported 0.6–4.9%[1–5] and the neurological prognosis is poor.[6] In this study, we report a patient in whom perforation occurred at the same site during thrombectomy for AIS related to occlusion at the cavernous sinus of the left internal carotid artery, inducing a carotid cavernous fistula (CCF).

Case Presentation

Patient: An 88-year-old woman.

Medical history: Hypertension, hyperlipidemia, and fracture of the femur.

Present illness: She developed aortic dissection (Stanford type A) and was brought to our hospital by ambulance. Ascending aortic replacement was performed at the Department of Cardiovascular Surgery and she was admitted. On the 11th postoperative day, paroxysmal atrial fibrillation was confirmed and anticoagulant therapy with heparin was started. On the 12th postoperative day, motor aphasia and right hemiparesis developed at 8:00, and there was no improvement in her condition. Brain computed tomography (CT) did not reveal intracranial hemorrhage or early ischemic findings. On the same day, she was referred to our department at 17:00. The National Institutes of Health Stroke Scale (NIHSS) score was 16 and brain magnetic resonance imaging (MRI) was performed at 18:00 according to the in-hospital stroke protocol. Diffusion-weighted imaging (DWI) demonstrated multiple high-intensity areas in the bilateral cerebral hemispheres and cerebellum. Magnetic resonance angiography (MRA) revealed occlusion of left internal carotid artery (**Fig. 1**), suggesting cardiogenic embolism. There was a DWI-clinical mismatch and we considered recanalization therapy to be indicated.

Neuroendovascular treatment: Cerebral angiography was started through left femoral artery puncture at 19:45. Occlusion at the cavernous sinus (C3) of left internal carotid artery, and a collateral flow to distal internal carotid artery via left ophthalmic artery, and a collateral flow via anterior communicating artery from right internal carotid artery were confirmed, and thrombectomy was started (**Fig. 2A** and **2B**).

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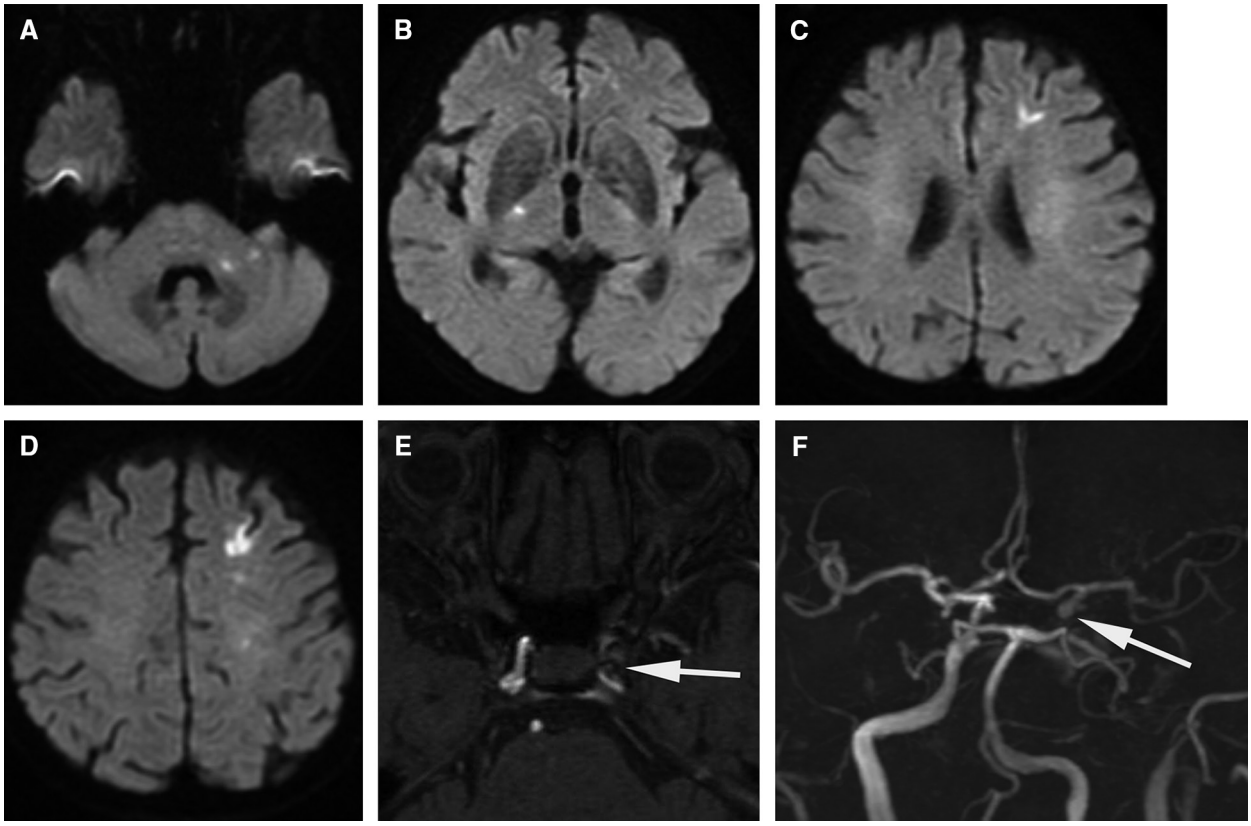

Fig. 1 DWI at onset showed a high-intensity area in left superior cerebellar peduncle (**A**), right corona radiata (**B**), and left frontal lobe (**C**, **D**). MRA-TOF showed a significantly low-intensity area around left internal carotid artery (**E**, arrow). MRA revealed occlusion of left internal carotid artery (**F**, arrow). DWI: diffusion-weighted imaging; MRA: magnetic resonance angiography; TOF: time-of-flight

A 25-cm 9Fr sheath (Terumo, Tokyo, Japan) was inserted and we attempted to insert an Optimo 9 Fr 90 cm (Tokai Medical Products, Aichi, Japan) into left internal carotid artery coaxially with a 6Fr JB2 125-cm CX catheter (Katecs, Kanagawa, Japan) using a 0.035-inch 150-cm SURF guidewire (PIOLAX, Inc., Kanagawa, Japan), but arteriosclerosis made guiding difficult; therefore, it was inserted into left common carotid artery. We attempted to guide a Penumbra 5MAX ACE68 (Medicos Hirata, Osaka, Japan) into distal internal carotid artery using a Marksman 150 cm (Medtronic, Minneapolis, USA) and CHIKAI black14 200 cm (Asahi INTECC, Aichi, Japan). Angiography through the Optimo 9 Fr confirmed C4-C3 occlusion (**Fig. 2C** and **2D**). The CHIKAI black14 was resistant on the medial side of the C4 segment and it was difficult to guide it to a distal site. It was impossible to guide the Marksman beyond the C4 segment of internal carotid artery and the ACE68 to the distal site to the C4/C5 junction. Angiography through the Marksman was performed and the procedure was continued because the Marksman existed in the thrombus of internal carotid artery (**Fig. 2E**). Continuous aspiration from the ACE68 was tried using a direct aspiration first pass technique (ADAPT). The thrombus was confirmed in ACE68, but recanalization of internal carotid artery was not achieved. Using a similar system, the Marksman was guided to the C4 segment of internal carotid artery and the microguidewire was switched to a Traxcess 14 200 cm (Terumo). However, it was resistant on the medial side of the C4 segment, same as 1st pass; it was difficult to guide the Traxcess 14 to a distal site. The Traxcess 14 was considered to have been successfully guided to the M1 segment of left middle cerebral artery (**Fig. 2F** and **2G**), but the Marksman was unable to be guided to a distal site to the C3 segment. Angiography through the microcatheter was not performed and Solitaire 6 mm/40 mm (Medtronic) was deployed through the same site. Expansion of stent was insufficient distal to the 3rd marker of the stent and a stent was deployed to the same size of internal carotid proximal to the 3rd marker (**Fig. 3A** and **3B**). The Marksman was removed, ACE68 was pulled up to the C3 segment using the Solitaire as an anchor while performing continuous aspiration with the ACE68, and the stent was retrieved. Although

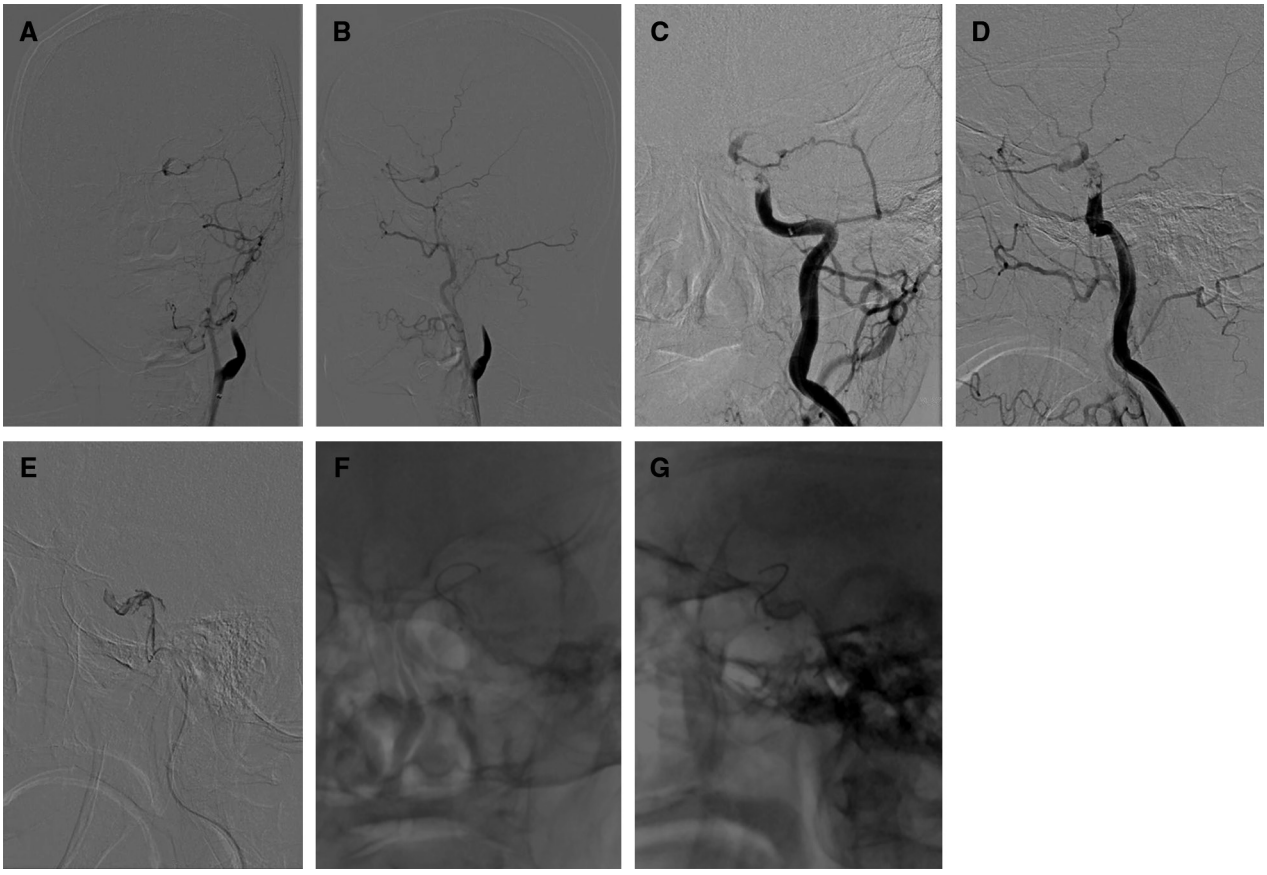

Fig. 2 Angiography before mechanical thrombectomy demonstrated occlusion of the left C4 of internal carotid artery (**A–D**). Angiography from the microcatheter revealed a cavernous sinus around internal carotid artery (**E**). The microguidewire was thought to have been guided from internal carotid artery to middle cerebral artery, but it was actually guided to the cavernous sinus (**F, G**).

the thrombus was retrieved, angiography demonstrated that recanalization was not achieved, confirming a CCF (**Fig. 3C** and **3D**). Common carotid artery was obliterated by inflating the balloon of 9Fr Optimo for 10 minutes without reversing heparin. Then, reocclusion at the C4 segment of internal carotid artery and disappearance of the CCF were confirmed (**Fig. 3E**). A collateral flow via anterior communicating artery was sufficient (**Fig. 3F**), and we decided to complete the procedure with internal carotid artery remaining occluded. Hemostasis using an Angioseal Plus 8 Fr (Terumo) was performed and endovascular procedure was completed.

Postoperative course: Brain MRI the day after surgery revealed the fresh infarction in the territory of the anterior choroidal artery and deep white matter. There was no recurrent of CCF and the occlusion of left internal carotid artery extended to the C1 segment (**Fig. 4**). Aphasia and right hemiparesis were slightly worsened (NIHSS score: 17). The patient was referred to a rehabilitation hospital with a modified Rankin Scale score of 4.

Discussion

The incidence of vascular perforation during thrombectomy for AIS is reported 0.6–4.9%.[1–5] It may frequently occur in the presence of peripheral embolism or hard thrombi. When perforation occurs, the neurological prognosis is poor in many cases.[6]

In the present case, preoperative brain MRI demonstrated calcification consistent with the site of occlusion. Cardiogenic embolism may have occurred at the site of arteriosclerotic stenosis. When attempting lesion cross at the site of occlusion with a microguidewire, vascular perforation seemed to occur at the medial side of the C4 segment of internal carotid artery and microguidewire seemed to be guided into the cavernous sinus. Indeed, a microcatheter that was considered to have been successfully guided to the C3 segment of internal carotid artery may also have been guided into the cavernous sinus. On the 2nd pass, the microguidewire was thought to have been guided to the M1 segment of left middle cerebral artery. However, at this

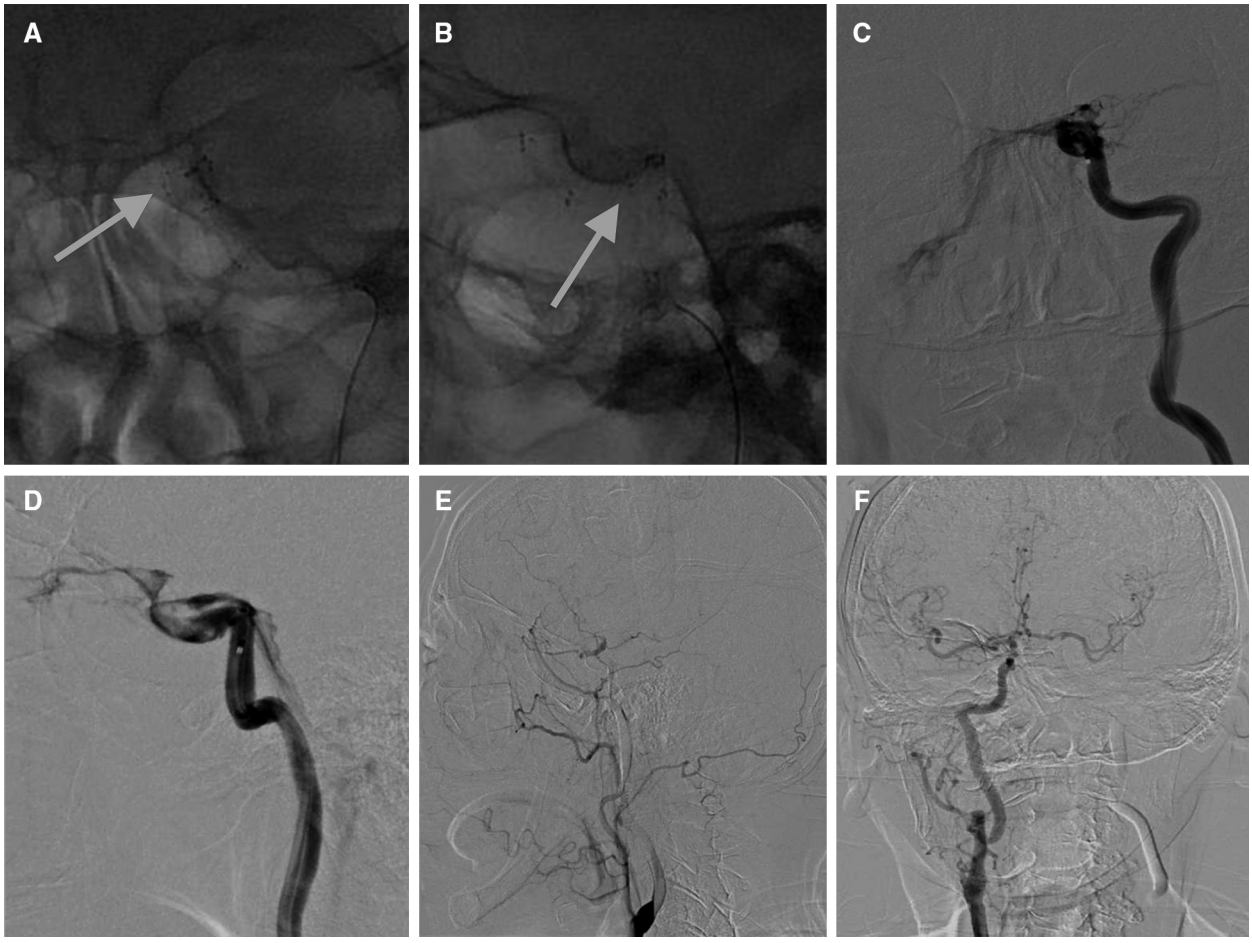

Fig. 3 The stent was not fully deployed from the 3rd marker to the distal end (**A**; frontal view, **B**; lateral view). Angiography after retrieving the stent showed a CCF (**C**; frontal view, **D**; lateral view). Right common carotid angiography showed collateral blood flow via anterior communicating artery (**E**). Left common carotid angiography after temporary occlusion of left common carotid artery confirmed the disappearance of the CCF (**F**). CCF: carotid cavernous fistula

point, the microguidewire may also have been guided into the cavernous sinus through the site of perforation. A CCF may have been formed in the early phase of surgery, but it was not visualized due to occlusion of internal carotid artery. Stent guiding and deployment may have enlarged the site of perforation, making the CCF clear. Four patients with CCF development during thrombectomy, as demonstrated in the present case, have been reported.[7,8] Matsumoto et al. suggested that internal carotid artery, which was essentially tortuous, was extended while pulling out the stent during stent deployment for occlusion of proximal middle cerebral artery (M1), inducing a CCF due to injury of meningohypophyseal trunk.[7] In the present case, arteriosclerotic stenosis may have been present at the site of occlusion; it may have been difficult to guide a microguidewire from the site of stenosis to the true lumen of internal carotid artery on the distal side, causing perforation. We should have noticed microguidewire-related perforation when the cavernous sinus was visualized on angiography through a microcatheter.

In patients with occlusion in the presence of arteriosclerotic stenosis, such as our patient, the recanalization rate after thrombectomy decreases[9] and rescue treatments, such as stenting and balloon angioplasty, may be required.[10] On the other hand, stent deployment at the cavernous sinus of internal carotid artery, balloon angioplasty, or frequent trials of catheter passage may injure the vascular wall, being a risk factor for CCF.[8] In patients with acute occlusion at the cavernous sinus of internal carotid artery in the presence of arteriosclerotic stenosis, such as our patient, thrombectomy should be carefully performed.

Recanalization therapy for AIS may improve the outcome markedly if treatment is rapidly started.[11] At our hospital, to start treatment rapidly, brain MRI is prioritized for

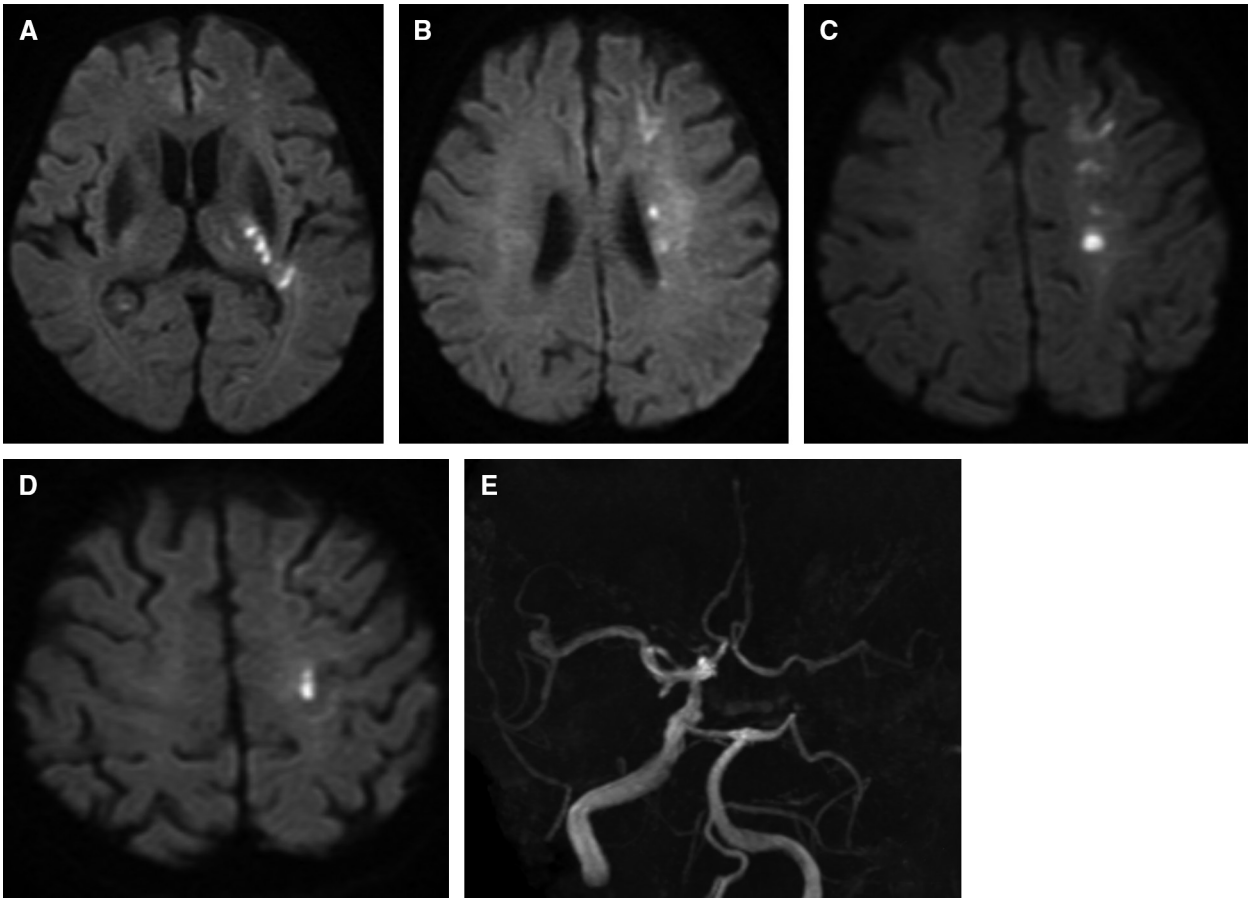

Fig. 4 DWI the day after treatment showed a high-intensity lesion at the territory of anterior choroidal artery (**A**), left corona radiate (**B**), and the watershed area of anterior cerebral artery middle cerebral artery (**C, D**). MRA showed no recurrence of the CCF or occlusion of left internal carotid artery (**E**). CCF: carotid cavernous fistula; DWI: diffusion-weighted imaging; MRA: magnetic resonance angiography

diagnostic imaging in stroke patients. As periarterial calcification exhibits low signal intensity on MRI,[12] it is necessary to confirm the presence of a low-intensity lesion suggestive of arteriosclerotic change around an occluded blood vessel using time-of-flight (TOF) MRA in advance. Marked perivascular calcification on plain CT suggests arteriosclerotic changes and stenosis.[13] Even when performing CT and CT angiography for diagnostic imaging in patients with AIS, it is necessary to confirm the presence of marked perivascular calcification.

In the present case, the microguidewire was formed in a J shape. In a previous study, it was reported the usefulness of pigtail shaped of microguidewire as a method to safely guide a microguidewire into a perforator or occluded vessel that is not visible on angiography.[14] This method enables the assessment of thrombus rigidity based on intraoperative changes in the microguidewire shape and may be reflected by thrombectomy procedures.[15]

If a CCF develops during endovascular treatment in patients with AIS, as demonstrated in the present case, combination therapy with tissue plasminogen activator (t-PA) and intraoperative heparinization may make hemostasis difficult. In the present case, hemostasis was achieved by inflating the balloon of a guiding catheter. However, if a hemorrhagic complication develops, as observed in the present case, hypotensive therapy and heparin reversal with protamine must be initially considered.[16] Hemostasis using a balloon catheter,[17] coil embolization of a parent artery,[18] and transvenous coil embolization of fistula[7] have been reported. Sufficient hemostatic treatment should be considered according to factors related to hemorrhage.

Conclusion

We reported a patient in whom vascular perforation during thrombectomy for AIS induced a CCF. In patients with intracranial large vessel occlusion in the presence of arteriosclerotic stenosis, microguidewire-related vascular perforation at the site of stenosis must be considered.

Disclosure Statement

The authors declare no conflict of interest.

References

1) Goyal M, Demchuk AM, Menon BK, et al: Randomized assessment of rapid endovascular treatment of ischemic stroke. *N Engl J Med* 2015; 372: 1019–1030.
2) Berkhemer OA, Fransen PS, Beumer D, et al: A randomized trial of intraarterial treatment for acute ischemic stroke. *N Engl J Med* 2015; 372: 11–20.
3) Campbell BC, Mitchell PJ, Kleinig TJ, et al: Endovascular therapy for ischemic stroke with perfusion-imaging selection. *N Engl J Med* 2015; 372: 1009–1018.
4) Jovin TG, Chamorro A, Cobo E, et al: Thrombectomy within 8 hours after symptom onset in ischemic stroke. *N Engl J Med* 2015; 372: 2296–2306.
5) Saver JL, Goyal M, Bonafe A, et al: Stent-retriever thrombectomy after intravenous t-PA vs. t-PA alone in stroke. *N Engl J Med* 2015; 372: 2285–2295.
6) Mokin M, Fargen KM, Primiani CT, et al: Vessel perforation during stent retriever thrombectomy for acute ischemic stroke: technical details and clinical outcomes. *J Neurointerv Surg* 2017; 9: 922–928.
7) Matsumoto H, Nishiyama H, Takemoto H, et al: Carotid-cavernous fistula caused by vessel injury while withdrawing a stent retriever during mechanical thrombectomy for acute ischemic stroke: a case report. *JNET J Neuroendovasc Ther* 2018; 12: 235–240.
8) Alan N, Nwachuku E, Jovin TJ, et al: Management of iatrogenic direct carotid cavernous fistula occurring during endovascular treatment of stroke. *World Neurosurg* 2017; 100: 710.e15–710.e20.
9) Leischner H, Flottmann F, Hanning U, et al: Reasons for failed endovascular recanalization attempts in stroke patients. *J Neurointerv Surg* 2019; 11: 439–442.
10) Jia B, Feng L, Liebeskind DS, et al: Mechanical thrombectomy and rescue therapy for intracranial large artery occlusion with underlying atherosclerosis. *J Neurointerv Surg* 2018; 10: 746–750.
11) Saver JL, Goyal M, van der Lugt A, et al: Time to treatment with endovascular thrombectomy and outcomes from ischemic stroke: a meta-analysis. *JAMA* 2016; 316: 1279–1288.
12) Wildy KS, Yuan C, Tsuruda JS, et al: Atherosclerosis of the carotid artery: evaluation by magnetic resonance angiography. *J Magn Reson Imaging* 1996; 6: 726–732.
13) Taoka T, Iwasaki S, Nakagawa H, et al: Evaluation of arteriosclerotic changes in the intracranial carotid artery using the calcium score obtained on plain cranial computed tomography scan: Correlation with angiographic changes and clinical outcome. *J Comput Assist Tomogr* 2006; 30: 624–628.
14) Sato M, Ohshima T, Ishikawa K, et al: A novel technique of safe and versatile microguidewire shaping with neuroendovascular therapy: modified pigtail method. *JNET J Neuroendovasc Ther* 2017; 11: 266–271.
15) Ohshima T, Imai T, Sato M, et al: A novel technique for higher success rates of recanalization with stent clot retriever: corkscrew penetrating method. *JNET J Neuroendovasc Ther* 2017; 11: 94–98.
16) Leishangthem L, Satti SR: Vessel perforation during withdrawal of Trevo ProVue stent retriever during mechanical thrombectomy for acute ischemic stroke. *J Neurosurg* 2014; 121: 995–998.
17) Layton KF, Cloft HJ, Kallmes DF: Cerebral aneurysm perforations during treatment with detachable coils. Use of remodelling balloon inflation to achieve hemostasis. *Interv Neuroradiol* 2006; 12: 31–35.
18) Nakamura A, Kawashima A, Hayashi M, et al: Internal carotid artery injury due to aspiration technique from balloon guide catheter on acute thrombectomy: a case report. *JNET J Neuroendovasc Ther* 2019; 13: 354–357.