Hypoglossal Canal Dural Arteriovenous Fistula Induced after Mechanical Thrombectomy for Acute Ischemic Stroke

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Objective: In this study, we report a patient in whom a dural arteriovenous fistula (dAVF) developed after mechanical thrombectomy for acute ischemic stroke, and embolization was performed.

Case Presentation: The patient was a 44-year-old male. He was brought to the emergency room of our hospital by ambulance with cervical pain. Head MRI revealed ischemic stroke related to right internal carotid artery occlusion. Mechanical thrombectomy was performed and thrombolysis in cerebral infarction (TICI) 2b recanalization was achieved. Middle cerebral artery (M2) occlusion and internal carotid artery dissection were observed. Follow-up was conducted. The postoperative course was favorable, and he was referred to another hospital for rehabilitation. However, cerebral angiography 4 months after surgery showed a dAVF. Transarterial embolization was performed. The postoperative course was uneventful and she was discharged.

Conclusion: In the present case, dissection-related dilation/elevation of the internal carotid artery may have resulted in venous compression, leading to the development of a dAVF. We could confirm these serial changes in images before and after its development.

Keywords: tandem occlusion, arteriovenous fistula, internal carotid artery dissection, anterior condylar confluence, fibromuscular dysplasia

Introduction

Etiological factors for a dural arteriovenous fistula (dAVF) include sinus thrombosis, venous return disorder, venous hypertension, and trauma. In the present case, vascular dilation/elevation related to dissection of the cervical internal carotid artery (ICA) may have led to compression of the internal jugular vein (IJV), inducing stenosis. Subsequently, massive venous return mediated by the IJV may have flown into the vertebral venous plexus (VVP) as a collateral pathway, and induced venous return disorder through phlebostasis (VVP → suboccipital cavernous sinus [SCS] → posterior condylar vein [PCV] → anterior condylar confluence [ACC] → anterior condylar vein [ACV]), leading to the development of an arteriovenous (AV) shunt between the ACV and ascending pharyngeal artery (AphA). We examined imaging findings before and after dAVF development and reviewed the shape of the cervical ICA characteristic of the present case.

Case Presentation

Case: A 44-year-old male.

Complaint: neck pain.

Medical history: Hypertension, congenital facial hemiatrophy.

Drug therapy/allergy: Absent.

Social history: Physical education teacher, left-handedness.
Present illness: Neck pain started in the morning. He suddenly became unresponsive during a meeting and was brought to the emergency room (ER) of our hospital by ambulance.

Physical findings on admission: The body temperature, heart rate, and blood pressure were 36.7°C, 62 beats/min, and 151/94 mmHg, respectively.

Neurological findings: His Japan Coma Scale (JCS) score was II-10 and his Glasgow Coma Scale (GCS) score was E3V4M5. Dysarthria, aphasia, left hemiparesis, and left hemispatial neglect were noted.

Cerebral MRI-diffusion-weighted imaging (DWI) showed a light high-intensity area (HIA) in the middle cerebral artery (MCA) area involving the right temporal lobe (Fig. 1a). MRA revealed occlusion of the right ICA (Fig. 1b). The DWI-Alberta Stroke Programme Early CT Score (ASPECTS) was 7 points. As cervical pain was present, ICA dissection was suspected, and 3D CTA was performed. There was no aortic dissection, but the right cervical ICA was occluded at the C2-C3 level. Marked kinking of the left cervical ICA was observed (Fig. 1c). Considering the time of onset, tissue plasminogen activator (t-PA) was an option and we subsequently scheduled him for mechanical thrombectomy using A Direct Aspiration First Pass Technique (ADAPT) while receiving t-PA.

Intraoperative findings: Under local anesthesia, a 9Fr Optimo (Tokai Medical Products, Aichi, Japan) was inserted into the right ICA through the right femoral artery. An ASAHI CHIKAI BLACK microguidewire (ASAHI INTECC CO., LTD., Aichi, Japan) was guided to the site of occlusion together with a Penumbra 5MAX ACE reperfusion catheter (5MAX ACE; Penumbra, Alameda, CA, USA) and Penumbra 3MAX ACE reperfusion catheter (Penumbra). These catheters were advanced to the site of the occlusion. Angiography was performed through the 5MAX ACE proximal and distal to the site of occlusion to confirm the size of the thrombus (Fig. 1d). Aspiration was applied with proximal balloon protection while pulling back the 5MAX ACE. A red thrombus was observed, and recanalization of the right ICA was achieved. Dissection of the right cervical ICA with kinking was noted, but stenting was considered to be difficult due to its shape. At this point, stenosis of the IJV, which was consistent with the site of cervical ICA dissection, was confirmed. Cerebral veins had returned via the pterygoid plexus or deep cervical vein (DCV) (Fig. 1e). Occlusion of the M2 inferior trunk was observed, but recanalization of the superior trunk toward the motor cortex was confirmed. After confirming the absence of symptom exacerbation, the procedure was ended. Thrombolysis in cerebral infarction (TICI) 2b was achieved and onset-to-recanalization time was 4 hours and 15 minutes, puncture-to-recanalization time was 1 hour and 15 minutes.

Postoperative course: An MRI-DWI on the 1st postoperative day showed an HIA involving the right temporal lobe only. The HIA involving the insular gyri and frontal lobe had disappeared (Fig. 1f). MRA confirmed the visualization of the right ICA and M2 superior trunk (Fig. 1g). After surgery, 75 mg of clopidogrel was orally administered, and there was no recurrent ischemic stroke or progression of ICA dissection. The patient was referred to another hospital for rehabilitation on the 26th postoperative day, with a modified Rankin Scale (mRS) score of 2. He was discharged from the hospital 3 months after surgery. However, tinnitus was noted at 4 months after surgery. Cerebral angiography showed a dAVF at the ACC, and he was admitted for treatment.

Intraoperative findings: Diagnostic external carotid artery (ECA) angiography showed that the hypoglossal branch of the right AphA functioned as a feeder, and that the ACV functioned as a drainer. The difference between the feeder and drainer was marked, reflecting the difference in the diameter. In the hypoglossal canal, a shunted pouch was observed. There was no cortical venous reflux (Fig. 2a). The drainage routes were as follows: ACV → ACC → PCV → SCS → VVP and marginal sinus (MS) → SCS → VVP. Neither the petrous branch of the contralateral AphA or middle meningeal artery (MMA) nor the mastoid branch of the occipital artery (OA) functioned as a feeder, and there was no reflux to the inferior petrous sinus (IPS), cavernous sinus (CS), or superior ophthalmic vein (SOV) (Fig. 2b).

Only the right AphA functioned as a feeder, suggesting a direct AphA-ACV AV shunt. As stenosis of the IJV was present, transcatheter arterial embolization (TAE) with a coil was selected. Under local anesthesia, a 5Fr Envoy (Codman & Shurtleff, Raynham, MA, USA) was inserted into the right ECA through the right femoral artery. An ASAHI CHIKAI BLACK microguidewire was guided to the AphA together with a Headway 45 microcatheter (MicroVention, Aliso Viejo, CA, USA). After confirming the shunt point, four Axium PRIME detachable coils (Medtronic, Minneapolis, MN, USA) measuring 1.5 mm × 4 cm were inserted into the shunted pouch of the AphA, leading to the complete disappearance of the AV shunt (Fig. 2c). After confirming the absence of hypoglossal nerve paralysis and hemorrhage on DynaCT, the procedure was completed (Fig. 2d).
Fig. 1 Images before and after thrombectomy. (a) Diffusion-weighted-MRI before mechanical thrombectomy. (b) MRI (MRA) before mechanical thrombectomy. (c) 3D-CTA of the cervix (lateral view). Solid arrow: the right cervical internal carotid artery was occluded at the C2-C3 level. Dotted arrow: marked kinking of the left cervical internal carotid artery was noted. (d) Internal carotid angiography (frontal view). (e) Internal carotid angiography immediately after mechanical thrombectomy. The superior trunk toward the M2 motor cortex was patent (TICI 2b) (arterial phase, frontal view). Arrow: Stenosis of the IJV was confirmed, being consistent with the site of dissection. (f) Diffusion-weighted-MRI on the 1st postoperative day. (g) MRI (MRA) on the 1st postoperative day, IJV: internal jugular vein; TICI: thrombolysis in cerebral infarction.
administration of an antiplatelet drug was deemed necessary and the patient was followed-up without stenting. The patient had undergone corrective facial surgery for congenital facial hemiatrophy in a university hospital 23 years before the onset of ischemic stroke. In relatively young men with no medical history, marked kinking of the cervical ICA, as demonstrated in the present case, is rare. Its presence suggests a congenital anomaly, especially one of the 3rd branchial arch. In the embryonic stage, when great vessels, such as the aortic arch, descend into the mediastinal space, a loop at the 3rd branchial arch artery-dorsal aorta anastomotic site may come off, becoming linear and forming the ICA (Fig. 3a). Our patient may have suffered an irregularity during this process (kinking-type in the classification prepared by Weibel et al.), leading to an S-shaped ICA, an anomaly with a particularly severe angle among other kinking-type anomalies (Fig. 3b). Several studies have indicated the association of S-shaped ICA administration of an antiplatelet drug was deemed necessary and the patient was followed-up without stenting. The patient had undergone corrective facial surgery for congenital facial hemiatrophy in a university hospital 23 years before the onset of ischemic stroke. In relatively young men with no medical history, marked kinking of the cervical ICA, as demonstrated in the present case, is rare. Its presence suggests a congenital anomaly, especially one of the 3rd branchial arch. In the embryonic stage, when great vessels, such as the aortic arch, descend into the mediastinal space, a loop at the 3rd branchial arch artery-dorsal aorta anastomotic site may come off, becoming linear and forming the ICA (Fig. 3a). Our patient may have suffered an irregularity during this process (kinking-type in the classification prepared by Weibel et al.), leading to an S-shaped ICA, an anomaly with a particularly severe angle among other kinking-type anomalies (Fig. 3b). Several studies have indicated the association of S-shaped ICA
confirmed that the site of cervical ICA dissection was consistent with that of IJV compression-related stenosis (Figs. 1e and 4b). Concerning the pathogenesis of dAVF in the present case, the tortuous site of the ICA may have been already comprised by a history of hypertension, leading to dissection and venous return disorder through dissection-related dilation/elevation of the ICA and compression/stenosis of the IJV. Although venous return is maintained by collateral pathways, such as the VVP and DCV, at the time of IJV occlusion, it was maintained by the following drainage pattern in the present case: ACV → ACC → PCV → SCS → VVP. Furthermore, the right transverse sinus (TS) and sigmoid sinus (SS) had developed (Fig. 1e), and massive venous return, which should have flown into the right IJV, may have flown through the ACV to VVP drainage pattern. DAVFs are characterized by abnormal arteriovenous shunts on the dura mater, but those of the hypoglossal canal (HCDAVFs), are classified as ventral-type fistulas according to Lasjaunias’ classification with fibromuscular dysplasia (FMD). FMD is a vascular lesion typically characterized by the “string of beads sign,” which reflects stenosis and dilation of the ICA, or long tubular stenosis. One study reported that the incidence of S-shaped ICA in patients with FMD was higher than in healthy adults. Furthermore, another study indicated that dissection of the ICA was frequently observed when bilateral S-shaped ICAs were detected on cerebral angiography in patients with acute ischemic strokes. FMD is common in women aged 40–60 years. Our patient was male, but the presence of FMD as a background factor was suggested. His right ICA was more markedly dilated than his left ICA due to dissection at the site of flexion and upward shift (Fig. 4a). Angiography during mechanical thrombectomy also showed dissection-related dilation/elevation of the ICA. Compression and stenosis of the IJV related to this site were observed, suggesting a venous return disorder (Fig. 1e). Furthermore, angiography immediately after ADAPT and cerebral angiography just before dAVF treatment also
since the shunt point is present in a position that is not related to the dura mater. Therefore, strictly speaking, HCDAVFs are not dAVFs, but rather intracranial epidural AVFs. According to this classification, HCDAVFs may be osteodural AVFs, which refer to the intracranial development of a shunt in the epidural space, similar to spinal epidural AVFs. In the course of dAVF pathogenesis, venous occlusion- or venous hypertension-related tissue hypoperfusion or hypoxia may activate angiogenic factors, such as vascular endothelial growth factor (VEGF), basic fibroblast growth factor (bFGF), serum soluble angiopoietin receptor (sTIE-2), and matrix metalloproteinase-9, leading to morbid canalization of physiological shunts between primary anastomotic arteries (PAA) measuring 100–300 μm in inner diameter and existing on the dura mater and veins.5–8) The HCDAVF’s shunt point is not present on the dura mater, and therefore, may be developed by different mechanisms. However, HCDAVFs account for 3.6%–4.2% of intracranial dAVFs,9,10 and they have been confused with IPS, jugular valve, MS, skull base, or foramen magnum dAVFs. No study has specifically examined the pathogenesis of HCDAVFs. In the present case, we speculated that FMD, as described above, or an embryological anomaly might be the etiological factor for shunt development in the epidural space. Several studies reported the development of an AV shunt between the extracranial vertebral artery (VA) and
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observed the day after revascularization, and another involving the ACV 7 days after surgery, suggesting vasodilation and increased blood flow. An AphA-ACV shunt was detected 1 month after surgery. (b) Contrast-enhanced CT at the time of dAVF development (three directions). Laterality was apparent in the contrast enhancement of the suboccipital cavernous sinus or VVP, suggesting venous dilation and increased blood flow. ACV: anterior condylar vein; AphA: ascending pharyngeal artery; dAVF: dural arteriovenous fistula; HIA: high-intensity area; SCS: suboccipital cavernous sinus; VA: vertebral artery; VVP: vertebral venous plexus.

VVP or renal artery and vein in patients with FMD, suggesting that hyperplasia and defect of the media of the arterial wall in the presence of FMD are involved in AV shunt development. Some studies reported that dynamic 4D contrast-enhanced MRA and time-of-flight MRA in addition to conventional MRA were useful for noninvasively confirming blood flow into the ACC/ACV, as shunt points in HCDAVF patients, and the position of a venous pouch. In the present case, it was possible to evaluate blood flow using MRA. When serially evaluating the periphery of the AphA before and after dAVF development using MRA, an HIA involving the right MS was observed the day after revascularization, and another involving the ACV 7 days after surgery, suggesting vasodilation and increased blood flow. An AV shunt may have gradually developed between the AphA and ACV (Fig. 5a). Contrast-enhanced CT showed SCS/VVP vasodilation 3 months after revascularization (before dAVF treatment), and there was a marked laterality (Fig. 5b). There were no changes in the venous drainage pattern at the time of ischemic stroke or dAVF treatment. A previous study reported a dAVF at the ACC with IJV stenosis with the following drainage pattern: left ACV → IPS → CS → contralateral CS → SOV and Sylvian vein. However, it was unclear...
whether the dAVF developed after IJV stenosis, as seen in the present case. Concerning otorhinolaryngological-field surgery, there was no dAVF development following IJV ligation in any patient. However, this was possibly related to differences between ligation-related acute occlusion and compression-related chronic stenosis or the level of stenosis/occlusion sites. In the presence of ligation-related acute occlusion, the duration of venous hypertension is extremely short, and tissue blood flow disorders or abnormal inflammation-related activation of angiogenic factors may be absent. Furthermore, one basic technique employed by otorhinolaryngologists for radical neck dissection involves an upper margin with a line connecting the inferior mandibular margin with the mastoid process or lower. In the present case, the ICA dissection and compressed IJV were present at the C2-C3 level, and the level of IJV ligation may have been higher than that during standard surgical operations (Fig. 1c). Selective ECA angiography was not performed at the time of thrombectomy. Common carotid artery angiography at the completion of treatment did not show an AV shunt and the possibility of guidewire-related APhA perforation was ruled out. The contralateral APhA, petrous branch of the MMA, and mastoid branch of the OA did not act as feeders. There was no reflux into the IPS, CS, or SOV (Fig. 2a and 2b), suggesting a 1 feeder-1 drainer direct AV shunt between the APhA and ACV. In the present case, ICA dissection, ischemic stroke, and a dAVF developed in a relatively short period, which is rare. Considering the presence of FMD as a background factor, follow-up by regular imaging and hypotensive treatment were considered to be necessary.

## Conclusion

We encountered a patient in whom angiography findings before and after dAVF development suggested the involvement of ICA-dissection-related vasodilation/elevation and venous compression in the pathogenesis. Furthermore, we confirmed that vasodilation related to gradually increased blood flow into the ACV had led to shunt formation by comparing MRA findings before and after dAVF development. In acute ischemic stroke patients with tortuosity of the cervical ICA, dissection of the cervical ICA should be considered. Strict blood pressure control and follow-up by imaging may be necessary, considering the possibility that FMD may be present as a background factor even in patients with asymptomatic dAVFs incidentally detected.

## Disclosure Statement

The authors declare that they have no conflict of interest.

## References