Two Patients with Cerebral Infarction Who Underwent Endovascular Treatment for Internal Carotid Artery Dissection Related to an Elongated Styloid Process

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Objective: We report two patients with cerebral infarction who underwent endovascular treatment for internal carotid artery dissection related to an elongated styloid process.

Case Presentations: Case 1: A 48-year-old male. Paralysis of the left upper/lower limbs occurred. On arrival, the National Institute of Health Stroke Scale (NIHSS) score was 14. MRI revealed acute-stage infarction, narrowing of the high-level internal carotid artery adjacent to the right styloid process, and occlusion of the right middle cerebral artery (MCA). After internal carotid artery stenting (CAS), mechanical thrombectomy for distal embolism was performed, leading to recanalization. Case 2: A 45-year-old male. Aphasia and paralysis of the right upper/lower limbs occurred. On arrival, the NIHSS score was 8. MRI showed acute-stage infarction and narrowing of the high-level internal carotid artery adjacent to the left styloid process. Conservative treatment was administered. As there was a dissecting aneurysmal change at the stenotic site, carotid-stent-assisted coil embolization was performed. In the two patients, endovascular treatment led to a favorable prognosis.

Conclusion: For the treatment of arteriogenic cerebral infarction related to atypical stenosis of the high-level cervical internal carotid artery, it is important to review therapeutic strategies, considering the possibility of an elongated styloid process.

Keywords ► cerebral infarction, elongated styloid process, internal carotid artery dissection, endovascular treatment, carotid artery stenting

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Introduction

Previous studies have shown the efficacy of intracranial thrombectomy using a catheter as a treatment option for acute-stage cerebral infarction. In the future, this procedure may be indicated for an increasing number of patients in Japan. Thrombectomy followed by carotid artery stenting (CAS) for arteriogenic cerebral embolism derived from the cervical internal carotid artery has also been performed, but the possibility of arterial dissection-related embolism other than arteriosclerotic lesions must be considered. In particular, an elongated styloid process, as a type of internal carotid artery injury associated with a styloid-process-related external force, may appear in patients with stenosis of the high-level internal carotid artery adjacent to the styloid process. Several studies reported neuroendovascular treatment for this carotid artery dissection.^{1–3)} In this study, we report two patients with cerebral infarction related to

chiropractic or chronic cervical rotary movement on road bike riding in whom endovascular treatment for an elongated styloid process led to a favorable prognosis, and review the literature.

Case Presentations

Case 1

Patient: A 48-year-old male.

Complaints: Incomplete hemiplegia of the left upper/lower limbs, consciousness disorder.

Family history: His father had diabetes mellitus.

Lifestyle: Occasional alcohol drinking. No history of smoking.

Findings on arrival: Sinus rhythm was noted on electrocardiography. Neither lipid metabolism disorder nor diabetes mellitus was observed.

Present illness: On walking, weakness of the left upper/lower limbs and sudden consciousness disorder occurred, and the patient was brought to our hospital by ambulance. One month earlier, he underwent chiropractic massage involving cervical rotation due to stiff shoulders. Cold symptoms, such as cough, had persisted for 2 weeks before onset. He was brought to our hospital by ambulance 42 minutes after onset. On arrival, the National Institute of Health Stroke Scale (NIHSS) score was 14, and CT revealed a hyperdense middle cerebral artery (MCA) sign in the right MCA. MRI showed a light high-signal-intensity area involving a corona radiata at its center on diffusion-weighted images (DWIs). The Alberta Stroke Program Early CT score (ASPECT-DWI) was 9 points. MRA revealed stenosis of the high-level right cervical internal carotid artery with a false cavity and occlusion at the M1 region of the right MCA. Furthermore, bilateral elongated styloid processes (right: 33 mm, left: 37 mm) were observed. Fusion images of three-dimensional rotational angiography (3D-RA) and cone beam CT confirmed that the right styloid process end was adjacent to the site of internal carotid artery dissection (5 mm). In a portion of the right styloid process, there was no continuity, suggesting the possibility of external-force-related fracture/displacement (**Fig. 1**).

Therapeutic strategies: A diagnosis of intracranial arteriogenic embolism associated with internal carotid artery dissection related to elongated styloid processes was made. To perform treatment for cervical internal carotid artery dissection simultaneously, intravenous thrombolysis with tissue plasminogen activator (t-PA) was not conducted, and revascularization using endovascular treatment procedures

Fig. 1 (A) Diffusion-weighted MRI shows high-intensity lesions in the right corona radiata. (B) MRA of bilateral carotid to cerebral arteries shows a stenosis of the right internal carotid artery with irregularity of the vessel (arrow) and occlusion of the right middle cerebral artery (arrowhead). (C) Volume rendered image shows a close relation between the tip of the elongated styloid process and the internal carotid artery. (D) Axial image from CTA shows the close distance on axial sections between the styloid process (arrow) and an intraluminal intimal flap of the internal carotid artery (arrowhead): 5.08 mm.

was selected. The door to puncture time was 2 hours and 26 minutes.

Endovascular treatment (CAS and thrombectomy): Under local anesthesia, these procedures were performed. After heparin at 5000 units was intravenously administered, a 9 Fr Optimo balloon guiding catheter (Tokai Medical Products, Aichi, Japan) was inserted to an area adjacent to the site of right internal carotid artery dissection. After the Optimo balloon was dilated, a 0.014 Transend EX Platinum (Stryker, Kalamazoo, MI, USA) was guided to the distal site of dissection through the true lumen, and a stent (Protégé 10 × 40 mm; Covidien, Irvine, CA, U.S.A.) was deployed. After 30 mL of blood was aspirated from the Optimo, the balloon was deflated. Internal carotid angiography showed occlusion of the MCA (M2 region, superior trunk). The Optimo balloon was again dilated, and manual aspiration with a Penumbra 4MAX Reperfusion Catheter (Penumbra Inc., Alameda, CA, USA), as well as aspiration using a Penumbra 3MAX Reperfusion Catheter (Penumbra Inc.) and Penumbra Aspiration Pump (Penumbra Inc.), were conducted, leading to thrombolysis cerebral infarction (TICI) 2b recanalization. The puncture to recanalization time was 2 hours and 52 minutes (**Fig. 2**).

Subsequent course: Immediately after surgery, enema with aspirin suppository (300 mg) was performed, and argatroban at 60 mg/day was intravenously injected for 2 days. Furthermore, the oral administration of clopidogrel at 75 mg/day and aspirin at 100 mg/day was started in order, and continued. After 10 days, additional cerebral angiography was conducted to confirm the stent patency and absence of stent contact or mechanical compression by cervical rotation in the permissible range using 3D images. After 3 weeks, the patient was referred to a recovery-phase rehabilitation hospital, with a modified Rankin Scale (mRS) score of 2. He was discharged 3 months after onset. Subsequently, stent falling/displacement into the dissected space was noted, and additional treatment by stent-in-stenting was performed 6 months after onset. Neither cerebral angiography after 9 months nor carotid artery echography after 1 year and 9 months showed stent displacement, restenosis, or falling into the dissected space.

Case 2

Patient: A 45-year-old male.

Complaints: Aphasia, incomplete paralysis of the right upper limb.

Medical history: Second right branchial cleft cyst.

Lifestyle: Occasional alcohol drinking. No smoking history.

Findings on arrival: Sinus rhythm on electrocardiography.

No lipid metabolism disorder or diabetes mellitus.

Present illness: Aphasia and weakness of the right upper/lower limbs suddenly occurred, and he was brought to our hospital by ambulance. At the time of onset, there was no special incentive, but his hobby was riding a road bike. The interval from onset until arrival was 2 hours. On arrival, the NIHSS score was 8 points. DWIs showed a high-signal-intensity area involving the frontal lobe, temporal lobe, and insular gyri. The ASPECT-DWI score was 7 points. MRA revealed occlusion of the left MCA and stenosis of the high-level left cervical internal carotid artery. It was impossible to clarify the presence of a false cavity at the stenotic site of the internal carotid artery using 3D-CTA, but bilateral elongated styloid processes (right: 30 mm, left: 31 mm) were observed. The left styloid process end was adjacent to the site of internal carotid artery stenosis. Therapeutic strategies: A diagnosis of intracranial arteriogenic embolism associated with internal carotid artery dissection related to elongated styloid processes was made. Conservative treatment was conducted without performing acute-phase intravenous thrombolysis with t-PA or acute-phase revascularization for the following

Fig. 2 (A) Right common carotid angiogram (lateral view) shows the dilatation of the right internal carotid artery, causing dissection. (B) Right common carotid angiogram after stenting (lateral view). We expanded the Protégé stent covering the true lumen. (C) Right common carotid angiogram (A-P view) shows the occlusion of the right middle cerebral artery. (D) Right common carotid angiogram (A-P view) shows recanalization of the right middle cerebral artery after aspiration of the thrombus.

reasons: intracranial occlusion was present at the M2 periphery, and a rapid improvement in the symptoms (NIHSS score: 4) was noted during the course in the emergency room.

There was no recurrent cerebral infarction. However, MRI after 5 weeks revealed a false cavity at the stenotic site. Cerebral angiography 38 days after onset showed an aneurysm-like morphological change at the site of dissection, which was adjacent to the left styloid process end (approximately 1 mm) (**Figs. 3 and 4**). Internal carotid artery stent-assisted coil embolization of this lesion was selected. Endovascular treatment (stent-assisted coil embolization): For dual-antiplatelet therapy (DAPT), aspirin at 100 mg/day was orally administered as an additional remedy from 1 week before treatment. Under local anesthesia, the procedure was performed. After heparin at 6000 units was intravenously administered, a 7 Fr Shuttle-SL (Cook Medical, Bloomington, IN, USA) was inserted into the left common carotid artery through the right femoral artery. Under distal

Fig. 3 (A) Diffusion-weighted MRI shows an infarction extending to the territories of the middle cerebral artery in the left hemisphere. (B) MRA of the head shows an occlusion at the distal M2 portion of the left cerebral artery (arrow). (C) Left common carotid MRA shows internal carotid artery stenosis (arrow). (D) Left common carotid MRA shows a dissecting aneurysm (arrow).

protection with a GuardWire (Medtronic, Minneapolis, MN, USA), predilation was performed using a Jackal RX balloon measuring 3.5×20 mm (Kaneka Medix Corp., Osaka, Japan). Using a Transend EX platinum 0.014/205 cm (Stryker), an Excelsior SL-10 Preshaped 45° (Stryker) was inserted into a false aneurysm. Subsequently, a PRECISE stent measuring 8×30 mm (Cordis, Miami, FL, USA) was inserted, and embolization with Axium PRIME Detachable Coils (Covidien) was performed. Using a Jackal RX balloon measuring 4.0×30 mm (Kaneka Medix), postdilation was conducted. The absence of debris was confirmed by aspirating 60 mL of blood using a suction catheter. The GuardWire was deflated, and the procedure was completed (**Fig. 4**).

Postoperative course: There was no perioperative complication. Carotid artery echography 1 week after surgery confirmed that the stent patency was favorable. The patient was instructed to avoid excessive cervical rotation. He was discharged 2 months after admission, with an mRS score of 2. During the postoperative follow-up, there has been no recurrent ischemic symptom. Angiography 4 months after treatment confirmed the favorable patency of the stent lumen and complete disappearance of the dissected space.

Discussion

An elongated styloid process (Eagle's syndrome) is characterized by an elongated styloid process or a calcified

Fig. 4 (A) Volume rendered image shows a close relation between the tip of the elongated styloid process and the internal carotid artery (arrow). (B) Axial image from CTA shows the close distance on axial sections between the styloid process (arrow) and an intraluminal intimal flap of the internal carotid artery (arrowhead): 1.36 mm. (C) Left common carotid angiogram (A-P view) shows the dissection and aneurysmal change. (D) Left common carotid angiogram after stent-assisted coil embolization (A-P view). Little contrast media was retained in the false lumen.

styloid ligament that compress the cranial nerves and the carotid artery.^{1,4,5)} A styloid process is embryologically derived from the second branchial arch. It is interesting that second right branchial cleft cyst developed on the contralateral side in Case 2. Initial reports indicated neuralgia related to stimulation of the 5th, 7th, 9th, and 10th cranial nerves by taut mucosa, with tension related to cicatrization after tonsillectomy or trauma, gliding at the styloid process end on swallowing or talking, as well as facial/head pain related to a styloid process in impingement with pain-sense nerves abundant in the carotid artery wall.^{4,5)} Later, patients with transient cerebral ischemic attacks (TIAs), carotid artery dissection,^{2,3,6)} or cerebral infarction^{1,2,3,7)} resulting from styloid-process-related internal carotid artery compression were reported.

Rapid cervical circumnutation immediately before onset is typically involved in the pathogenesis of TIAs or cerebral infarction.^{2,7)} However, an episode of rapid circumnutation or trauma is not always present immediately before onset.^{1,2,3,8)} As another mechanism, the accumulation of chronically repeated, slight stress may lead to dissection.⁸⁾

In our series, neither trauma nor sudden, excessive cervical circumnutation at the time of onset was confirmed.

The accumulation of chronically repeated movements, such as chiropractic/subsequent anteflexion movement on coughing in Case 1 and cervical rotation in a fore posture on road-bike driving in Case 2, may have induced dissection, resulting in cerebral infarction. In the literature, styloid processes measuring 25 to ≥ 30 mm are defined as elongated ones.³⁾ On the other hand, Raser et al.⁹⁾ examined the association between carotid artery dissection and anatomic characteristics of the styloid process of the temporal bone with respect to the angle/length of the styloid process and distance between the carotid artery and styloid process end, and reported that the most important risk factors were the length and distance between the carotid artery and styloid process end. Muthusami et al. indicated that, when the distance was ≤ 5 mm, the odds ratio for the onset of arterial dissection was 7.58.¹⁰⁾ In Cases 1 and 2, initial MRI showed that stenosis of the internal carotid artery was located at a higher position in comparison with arteriosclerotic stenosis; dissection of the internal carotid artery was suspected. In addition, the distance between the lesion and styloid process end was 2–3 mm, suggesting the involvement of the styloid process in dissection of the internal carotid artery. Patients with dissection of the internal carotid artery in the absence of a history of trauma are often regarded as having idiopathic dissection. However, they may include those in whom elongated styloid processes were etiologically involved.⁹⁾ For differentiation, it is important to carefully evaluate the positional relationship/distance between the site of dissection and styloid process.

To treat elongated styloid processes, conservative treatment, such as restrictions on cervical rotation, topical steroid infusion, topical anesthesia, and therapy with analgesic drugs, is selected in some cases.^{2,5)} However, when elongated-styloid-process-related symptoms appear, surgery (styloid process removal) is indicated.^{4,5)} Surgery-related risks include neurovascular injury (especially facial nerve injury), infection, temporarily compromise speech and swallowing, and a visible neck scar.^{4,5)} Furthermore, inadequate shortening of the process or entrapment within the fibrous tissue of any of the adjoining nerve may lead to recurrence.¹¹⁾ Several studies reported revascularization by endovascular treatment for elongated-styloid-process-related dissection of the internal carotid artery.^{1–3)}

Our patients developed acute cerebral embolism related to arterial dissection. In Case 1, revascularization with a carotid artery stent was performed to secure a peripheral access route. Subsequently, thrombectomy was conducted. In Case 2, stent-assisted coil embolization of a pseudoaneurysm

after carotid artery dissection was performed. Although stenting for carotid artery dissection is controversial, its results are favorable according to a recent systematic review involving carotid artery dissection patients with intracranial arterial occlusion.¹²⁾ As the merits of this procedure, it is minimally invasive, and facilitates intracranial revascularization in a short duration. However, strict postoperative follow-up should be conducted, considering the possibility of additional treatment (stent-in-stenting), as demonstrated in Case 1. In Case 2, there was an aneurysm-like change at the site of carotid artery dissection, and stent-assisted coil embolization was selected. Some studies reported the favorable results of similar treatment,^{13,14)} but the long-term prognosis remains to be clarified. Future investigation is needed.

There are several treatment options for elongated-styloid-process-related dissection of the internal carotid artery: medical treatments, including acute-phase thrombolytic therapy, chronic-phase styloid process removal, acute-phase intracranial revascularization, and acute-/chronic-phase cervical revascularization, which should be selected in accordance with the presence or absence of concomitant intracranial lesions. Recent studies reported CAS-combined endovascular treatment for elongated-styloid-process-related dissection of the carotid artery (**Table 1**).^{1–3,15–18)} To prevent recurrence, styloid process removal should be finally performed.^{1,2,17)} However, intra-stent thrombosis, distal thromboembolism,^{2,17)} or stent fracture³⁾ (however, a diagnosis of an elongated styloid process was not made on initial treatment in the patient with stent fracture) may occur before the procedure. Furthermore, a study indicated the risk of rupture related to surgical operations during styloid process removal in patients with pseudoaneurysms.¹⁸⁾

In Case 1, there was no carotid artery contact at standard rotation/flexion positions due to fracture-related displacement (**Fig. 5A**). As the styloid process course is anatomically present between the internal and external carotid arteries, carotid artery contacts could be reproduced by putting rotation/flexion positions on the unaffected side in Case 2 (**Fig. 5B**). After revascularization by endovascular treatment, the patient was instructed to avoid excessive rotation/flexion movements. We did not plan a resection of the styloid processes on the patient's wishes. Follow-up is being performed at the outpatient clinic. The course has been favorable. However, considering post-stenting complications, as described above, strict follow-up, involving diagnostic imaging, is necessary after revascularization. In some cases, additional treatment, styloid process removal, must be considered.

Table 1 Summary of previously reported cases of Eagle's syndrome treated by endovascular therapy (CAS)

Author Year	Age Sex	Neurological symptoms before initial endovascular therapy	Initial endovascular therapy	Additional endovascular therapy	Surgical resection of SP	Follow-up duration after the last treatment and events
Todo et al. (2012) ⁽¹⁸⁾	57 M	Hemiparesis, aphasia	Endovascular thrombectomy and CAS	None	None	1 year
Sveinsson et al. (2013) ⁽²⁾	38 M	Hemiparesis, dysarthria	Endovascular thrombectomy and CAS	Endovascular thrombectomy, intracranial stenting and carotid artery stent-in- stenting 3 months after initial endovascular treatment	Done	6 months
Ogura et al. (2014) ⁽¹⁷⁾	55 M 55 M	Hemiparesis, aphasia Hemiplegia, sensory disturbance, hemispatial neglect	CAS Endovascular thrombectomy and CAS	None None	Done None	3 months 3 months
Hooker et al. (2016) ⁽³⁾	64 M	Dysarthria, aphasia	CAS CAS	None	None	Stent fracture 1 year after CAS
Miyata et al. (2016) ⁽¹⁾	41 M	Disturbance of consciousness, monoplegia, facial palsy, dysarthria, disturbance of attention	Endovascular thrombectomy and CAS	None	Done	1 year
Mann et al. (2017) ⁽¹⁵⁾	39 W 38 M	Hemiparesis Dysarthria	CAS CAS	None None	None None	5 years unknown
Smoot et al. (2017) ⁽¹⁶⁾	60 M	Hemiparesis, hemiparesis, dysarthria	CAS	None	None	3 weeks
Present case	48 M	Disturbance of consciousness, hemiparesis	Endovascular thrombectomy and CAS	Carotid artery stent-in-stenting 6 months after initial endovascular treatment	None	1 year and 9 months
Present case	45 M	Hemiparesis, aphasia	Carotid artery stent-assisted coil embolization	None	None	4 months

CAS: carotid artery stenting; F: female; M: male; SP: styloid process

Fig. 5 (A) Cone beam CT just after the CAS of Case 1. Head rotation to the right, straight, and to the left. The distances between 33 mm styloid process (yellow arrows) and stent, also between transverse process of atlas and stent dramatically change according to the rotation of head. Note the styloid process has fracture, presumably due to previous chiropractic. (B) Fusion image of the second case at 4-month follow-up. The tip of styloid process is attached to the stent with rotation of the head to the right (left black arrow). CAS: carotid artery stenting

Conclusion

We encountered two patients for whom acute-phase cerebral infarction treatment was performed, considering elongated-styloid-process-related injury of the internal carotid artery, thus leading to favorable results. In patients with arteriogenic cerebral infarction related to high-level cervical internal carotid artery stenosis at a site adjacent to a styloid process, it is important to select diagnostic/therapeutic strategies, considering the possibility of elongated-styloid-process-related injury of the internal carotid artery. Careful follow-up is necessary.

Disclosure Statement

There is no conflict of interest regarding this article.

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