Large Dissecting Aneurysm of the Cervical Internal Carotid Artery Associated with Elongated Styloid Process: A Case Report

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Objective: A rare case of large dissecting aneurysm of the internal carotid artery caused by elongated styloid process is reported.

Case Presentation: A 45-year-old woman, who had noted an uncomfortable feeling of the pharynx from 2 years before, developed swelling of the left neck, pharyngalgia, and dysphagia 3 months before. Cervical CTA revealed an aneurysm 27 mm in diameter in the left extracranial internal carotid artery. The styloid process was elongated and displaced the aneurysm, and dissecting aneurysm due to compression was suspected. A covered stent was placed to induce thrombosis in the aneurysm, resulting in alleviation of symptoms, and imaging examinations confirmed thrombosis and regression of the aneurysm.

Conclusion: Reports of treatment for elongated styloid process complicated by large aneurysm are very rare. A covered stent is a possible therapeutic option if direct surgery is difficult.

Keywords ▶ Eagle syndrome, aneurysm, covered stent

Introduction

Elongated styloid process is a syndrome that exhibits various symptoms caused by displacement of the pharynx and surrounding nerves and arteries by the elongated styloid process. There have been occasional reports of dissection of the neighboring internal carotid artery due to an elongated styloid process, but reports of dissecting aneurysm are scarce, and those of treatment for such aneurysms are very rare. We report our experience in treatment of a case of large dissecting aneurysm of the cervical internal carotid artery associated with elongated styloid process.

Case Presentation

The patient was a 45-year-old woman. Her primary complaints were swelling of the left neck, pharyngalgia, pain on swallowing, and stuffiness of the left ear. She had hypertension but no history of palatotonsillectomy or head and neck trauma. She was a smoker (10 cigarettes/day for 20 years) and a drinker (2 L of beer per day) but had no particular familial history.

She had noted an uncomfortable feeling of the throat and stuffiness of the left ear from 2 years before and occasionally felt neck pain on neck rotation, but their cause was not identified by a local ear nose throat (ENT) clinic. As she noted swelling in the left neck and gradually developed pharyngalgia and dysphagia from 3 months before, she consulted our hospital.

On her visit, there was swelling in the left submandibular region, where she complained of exacerbation of spontaneous pain and pain on neck rotation. Swallowing was possible despite pain, and no other neurologic abnormality was observed.

On ENT endoscopy, the pharyngeal cavity was narrowed on the left side (Fig. 1).
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Head and neck CTA showed a saccular aneurysm 27 mm in maximum diameter in the left extracranial internal carotid artery from the level of the first cervical vertebra to the proximal part of the skull base. The internal carotid artery distal to the aneurysm was delineated less clearly compared with the contralateral side, suggesting a decrease in blood flow. A styloid process elongated to 42 mm and apparently displacing the aneurysm was observed (Fig. 2). While the contralateral styloid process was also elongated to 40 mm, it was not in contact with the internal carotid artery.

Head and neck MRI showed no signal change in the brain parenchyma. The aneurysm displaced the ipsilateral pharyngeal cavity to the other side, the interior of the aneurysm was hypointense in T1- and T2-weighted images, and no clear thrombosation was noted (Fig. 3a).

On angiography, an aneurysm with a maximum diameter of 27.7 mm was noted in the cervical left internal carotid artery, and the segment distal to the aneurysm was visualized with a delay (Fig. 4a). On the balloon occlusion test (BOT) by obstructing the left common carotid and external carotid arteries, no symptoms appeared, but single photon emission computed tomography ($^{99m}$Tc-HMPAO) during the BOT showed about 80% decrease in blood flow of the left middle cerebral artery territory compared with the other side.

Therapeutic strategy
Since an elongated styloid process in contact with an aneurysm was observed in this patient, the condition was diagnosed as dissecting aneurysm due to elongated styloid process. Since the aneurysm caused symptoms by displacing the surrounding tissues and, should it enlarge or rupture, could induce life-threatening conditions, such as airway obstruction, it was judged to be an indication for surgical treatment. As the distal end of the aneurysm was considered difficult to secure by neck incision from the size and location of the lesion, endovascular treatment was selected. We first planned to occlude the parent artery and considered endovascular treatment with concomitant bypass surgery recommendable from the results of the BOT, but the patient did not wish craniotomy. Also, because there was a possibility that symptoms would not be resolved by intraneurysmal coil embolization due to persistence of mass effect, we aimed to induce thrombosation in the aneurysm by stent placement. The aneurysmal orifice was very large, and as bare metal stents were not expected to produce a flow-diverting effect, we decided to use a covered stent. Since the use of a covered stent in the carotid artery is not approved in Japan, approval by the institutional review board and consent by the patient were obtained. Although resection of the styloid process, which caused the aneurysm, was considered an alternative, we placed priority on treatment of the aneurysm, which was considered to be the cause of the symptoms, with additional treatment in mind should resolution of symptoms not be obtained.

Endovascular treatment
The procedure was performed under general anesthesia by initiating oral administration of 100 mg of aspirin and 75 mg of clopidogrel from 1 week before. An 8 Fr Shuttle Guiding Sheath (Cook Medical, Bloomington, IN, USA) was placed in the left internal carotid artery on the proximal side of the aneurysm via the right femoral artery. We attempted to guide an Excelsior SL-10 (Stryker, Kalamazoo, MI, USA) distally to the aneurysm using a 4F Cerulean catheter (Medikit Co., Ltd, Tokyo, Japan) as an intermediate catheter, but direct navigation was difficult, and it could be guided by turning it once in the aneurysm. Since SL-10, turned in the aneurysm, could not be straightened, it was guided distally to the aneurysm by having the Cerulean track it and was replaced with a Carotid GuardWire (GW; Medtronic, Minneapolis, MN, USA) via the Cerulean. After the GW was dilated and anchored in the distal internal carotid artery, it was straightened and replaced with a 400 cm 0.035 inch wire via the Cerulean. The aneurysm was 27.7 mm in maximum diameter and 12.8 mm in neck diameter, and the diameter of the normal part of the vessel was 5.0 mm proximal, and 4.1 mm distal, to the aneurysm. Since endoleak may occur as a problem after expansion of a covered stent, an Niti-S ComVi stent 8 mm × 6 cm (Century

Fig. 1 ENT endoscopy. The left pharyngeal cavity was narrowed compared with the contralateral side. ENT: ear nose throat
noted after the procedure, neck pain was exacerbated, and a burning sensation of the throat appeared, on the first post-procedural day. Head and neck MRI T1- and T2-weighted images showed signal intensification in the aneurysm (Fig. 3b), and symptoms due to spread of inflammation associated with thrombosation to surrounding tissues, particularly the glossopharyngeal nerve, were suspected. The symptoms were gradually alleviated after the initiation of oral administration of prednisolone at 1 mg/kg. Prednisolone was gradually reduced with the concomitant use of pregabalin and non-steroidal anti-inflammatory drugs, and the patient was discharged to home 2 weeks after the procedure. Thereafter, no exacerbation of symptoms was observed, the aneurysm was not delineated on angiography 3 months after

Medical, Inc., Tokyo, Japan), oversized compared with a usual carotid artery stent to improve the tightness of stent fixation and with a length not reaching the curves of the petrous part distal to the lesion, was selected and deployed in the left internal carotid artery. The portion that expanded insufficiently was post-dilated to obtain tight contact with the vascular wall. On final angiography, blood flow into the aneurysm disappeared, and the delay of contrast enhancement of the distal part of the aneurysm was resolved (Fig. 4b).

Postprocedural course
Argatroban was administered continuously from immediately after the procedure. While no neurologic deficits were

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**Fig. 2** Head and neck CTA. An extracranial internal carotid artery aneurysm 27 mm in maximum diameter was observed in contact with the left styloid process, which was elongated to 42 mm (a, b, and d). Delineation was poor distally to the aneurysm, and a decrease in blood flow was suspected (c).
the procedure, and no problems such as in-stent stenosis were noted. The aneurysm size was reduced on MRI 4 months after the procedure (Fig. 3c).

**Discussion**

Elongated styloid process is also called Eagle syndrome since it was first reported by Eagle in 1937. It is a disease that exhibits a wide variety of symptoms including neck pain, pharyngeal symptoms, and ear symptoms due to elongation of the styloid process or ossification of the stylohyoid ligament. The average length of the normal styloid process is 2.5–3.0 cm. Elongated styloid process was diagnosed when its length was ≥3.0 cm in many reports, and a length of ≥4.0 cm was closely correlated with pain. Eagle et al. reported that only 4% of the patients with elongated styloid process were symptomatic, and encounters with the condition are relatively rare in daily practice. Typically, the symptoms include pain and dysphagia due to mechanical stimulation by the elongated styloid process.
Dissecting Aneurysm Caused by Elongated Styloid Process

Since the disorder is likely to be asymptomatic, and since the symptoms are diverse, its diagnosis is often difficult. About 2 years was necessary for the diagnosis of the present case from the onset of symptoms. The disorder is difficult to diagnose by ultrasonography, MRI, or angiography of the neck because the bones cannot be evaluated by these modalities, and CTA, by which the evaluation of the styloid process and surrounding tissues including the carotid artery is possible, is considered useful. Our case could be diagnosed first by CTA. In addition, patients with carotid artery dissection have been reported to have a long styloid process and proximity of the styloid process to the carotid artery compared with average individuals, suggesting an association between elongated styloid process and carotid artery dissection.

Aneurysm of the extracranial internal carotid artery is a very rare disorder reportedly accounting for 1% or less of all intra- and extracranial aneurysms, and many of them are not detected until they cause symptoms. While they rarely rupture, they often present with local compression symptoms such as hoarseness and dyspnea and symptoms of brain ischemia due to thromboembolism. Its etiology varies widely, and trauma, carotid artery surgery, dissection, radiotherapy of the neck, and atheroma have been reported as its possible causes. Ogura et al. reported a case of dissecting aneurysm considered to be related to Eagle syndrome, but reports of large symptomatic aneurysms treated by surgery have been few.

Generally, conservative treatment by the combined use of analgesics, anticonvulsants, antihypertensive agents, and local anesthetics and surgical treatment, that is, resection of the styloid process by oral or cervical incision has been reported to be effective for elongated styloid process. However, the treatment for carotid artery dissection associated with elongated styloid process has not been established. Internal treatments such as antithrombotic therapy are attempted first, but stent placement has been performed for severe stenosis that repeatedly caused ischemic symptoms despite medical treatments. If compression of the artery or stent due to the styloid process persists even after stent placement, the risk of recurrence of dissection or in-stent thrombosis is considered to be present, and it is necessary to consider resection of the styloid process.

Concerning the treatment for extracranial carotid artery aneurysms, aneurysm resection/plication or, in patients difficult to treat by these procedures, combinations of ligation of the internal carotid artery with local cervical or intracranial bypass surgery depending on the state of collateral blood flow were often selected. However, because of the high invasiveness of these treatments including the risk of cranial nerve damage due to operative maneuvers and, in lesions close to the cranial base, the difficulty of the procedure and the necessity of a very wide incision, reports of endovascular treatments by stenting and coil embolization have appeared recently. Particularly, for large aneurysms as in our case, treatments expecting the flow-diverting effect of the stent alone are considered preferable to intra-aneurysmal coil embolization, which itself may cause mass effect. According to the systematic review by Li et al., the success rate of stent placement for extracranial carotid artery aneurysms was high at 92.8%, and covered stents had a more favorable short-term prognosis than bare metal stents in terms of the postprocedural intra-aneurysmal embolization rate, late complication rate, re-treatment rate, etc. As mentioned above, reports of large internal carotid artery aneurysms associated with elongated styloid process have been few, and principles for their treatment have not been established. We, therefore, decided to first attempt less invasive endovascular treatment for the aneurysm, which was considered to be causing the symptoms. Since there is no covered stent approved by the Japanese health insurance system to be used in the head and neck region, the indication must be evaluated carefully. We unavoidably selected a covered stent in our patient, who had a very wide aneurysmal orifice, by judging that the flow-diverting effect would not be expected from a usual bare metal stent. The covered stents available in Japan are for the bile duct or coronary artery, and it was necessary to select one appropriate in length and diameter, effective length of the delivery catheter, and cover material. For our patient, a stent for the bile duct was selected in consideration of the length necessary to sufficiently cover the large aneurysmal orifice and a large diameter of the internal carotid artery. For insertion via the femoral artery, we selected the Niti-S ComVi stent with an effective length of the delivery catheter comparable to, or longer than, a common carotid artery stent and covered with expanded polytetrafluoroethylene (ePTFE). Points of attention in this treatment were that a guiding...
catheter with a relatively large diameter (8-9F or larger) is necessary, that the delivery catheter is hard and difficult to navigate to the carotid artery in patients with tortuous arteries, such as older patients, and that the delivery catheter has an effective length inappropriate for treatment of the carotid artery, which limits the manipulability. In addition, as the high coverage rate suggests the possibility of in-stent thrombosis, it is necessary to perform sufficiently effective antithrombotic therapy before the procedure. Presently, there are no adequate guidelines for antithrombotic therapy for covered stent placement, but combinations of multiple antiplatelets appear to be used frequently as for bare metal stents. Also, the long-term prognosis including the durability of the stent is unclear, and since there is a report of delayed stent fracture due to displacement by the elongated styloid process, continuation of careful follow-up by examinations including angiography is considered necessary.

### Conclusion

In a very rare case of large dissecting aneurysm associated with elongated styloid process, thrombosation of the aneurysm and alleviation of symptoms could be achieved by placement of a covered stent. Although the long-term prognosis is unclear at present, this procedure is expected to be an alternative treatment for large dissecting aneurysm associated with elongated styloid process difficult to resect by direct surgery.

### Disclosure Statement

Neither the first author nor any of the coauthors have any conflicts of interest.

### References