Carotid Stent Fracture due to Eagle Syndrome after Endovascular Stenting for the Treatment of Acute Ischemic Stroke Caused by Internal Carotid Artery Dissection: Case Report

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Objective: We report a case of stent fracture caused by an elongated styloid process, a form of Eagle syndrome.

Case Presentation: A 58-year-old man presented with sudden right hemisensory disturbance and aphasia. MRI revealed multiple acute cerebral infarction in the left parietal and insula cortex. MRA revealed a left-sided internal carotid artery (ICA) occlusion distal to the carotid bifurcation. Digital subtraction angiography (DSA) revealed a left-sided ICA dissection distal to the carotid bifurcation. We performed acute revascularization with aspiration of the thrombus and stenting to treat the carotid dissection. Recanalization with thrombolysis in cerebral infarction (TICI) grade III was achieved. We performed DSA 3 months after an operation, it showed stent fracture and aneurysmal formation in the stent fractured department. From CT findings, the carotid artery dissection and stent fracture appeared to be triggered by the elongated styloid process. We performed stent-assisted coil embolization. After conducting percutaneous transluminal angioplasty (PTA) at high pressure after coil embolization, the elongated styloid process was fractured, and accepted a shift to the outside of the stent contact department. The patient was discharged without medical problems, and no recurrence was observed for 2 months after the surgery.

Conclusion: We experienced a rare case of ICA dissection and carotid stent fracture from Eagle syndrome. Eagle syndrome is an important disease to consider in the differential diagnosis of extracranial carotid artery dissection.

Keywords: Eagle syndrome, elongated styloid process, internal carotid artery dissection, stent fracture, acute revascularization

Introduction

Eagle syndrome (an elongated styloid process) refers to a type of syndrome in which physical compression of a peripheral structure related to an elongated styloid process or ossified stylohyoid ligament causes various symptoms.1) Typical Eagle syndrome is primarily characterized by pharyngeal/facial/cervical pain related to compression of the inferior cranial nerves. However, some studies indicated that cervical internal carotid artery (ICA) dissection related to mechanical compression of this artery occurred,2) inducing transient cerebral ischemic attacks or cerebral infarction.3–17)

In this study, we report a patient with cerebral infarction and acute occlusion of the ICA in whom recanalization was achieved by combining thrombectomy with carotid artery stenting (CAS), but stent fracture and aneurysm formation at the site of fracture were observed after 3 months, and stent-assisted coil embolization was performed as additional treatment, and review the literature.

Case Presentation

Patient: A 58-year-old male.
Complaints: Weakness/numbness of the right half body, aphasia.

Medical history: Not contributory.

Family history: Not contributory.

Present illness: Numbness of the right half body and slight weakness of the right hand and leg were noted at night the day before hospital arrival. Paralysis deterioration and aphasia appeared in the morning on the day of arrival. The patient was brought to our hospital by ambulance. The interval from the onset of the initial symptoms until arrival was 12 hours. Neurologically, right hemiplegia, sensory disturbance of the right half body, and generalized aphasia were observed. The National Institutes of Health Stroke Scale (NIHSS) score was 8. Diffusion-weighted cephalic MRI showed scattered high-signal-intensity areas in the left parietal lobe, temporal lobe, and insular cortex. The diffusion-weighted image (DWI)-Alberta Stroke Programme Early CT Score (ASPECTS) was 8. MRA revealed occlusion of the left cervical ICA and left middle cerebral artery branch (M2) (Fig. 1). DSA showed irregularity and stenosis of the cervical ICA. At the distal site of stenosis, marked thrombus formation was noted. Occlusion involved the origin to the C4 region (Fig. 2). Furthermore, occlusion of the left middle cerebral artery branch (M2) related to distal embolism was observed. This was not considered to be related to acute occlusion in the present case, but a dural arteriovenous fistula involving the left transverse to sigmoid sinuses was noted. As an ICA lesion distal to the frequent site of arteriosclerosis and cerebral infarction occurred at a relatively young age in the absence of risk factors for arteriosclerosis, a diagnosis of idiopathic dissection of the right ICA and arteriogenic embolism was made through exclusive diagnosis. Subsequently, revascularization was started.

Revascularization: Antiplatelet drugs (aspirin at 300 mg, clopidogrel at 300 mg) were orally administered, and 5000 units of heparin were intravenously administered. After thrombectomy with a Penumbra 5MAX (Penumbra, Alameda, CA, USA) for left ICA occlusion (cervical region to C2), carotid artery stenting with a Carotid Wallstent measuring $8 \times 28$ mm (Stryker, Kalamazoo, MI, USA) was performed, leading to recanalization of the left ICA (Fig. 2). To treat occlusion of the left middle cerebral artery branch (M2), thrombectomy was performed using a Trevo XP3 (Stryker). Total recanalization was achieved, and the thrombolysis in cerebral infarction (TICI) grade was evaluated as III.

Fig. 1 Brain MRI and neck MRA on admission. (A) Diffusion-weighted imaging revealed multiple small acute cerebral infarctions in the left parietal and insula cortex. (B) MRA shows a left-sided ICA occlusion distal to the carotid bifurcation. ICA: internal carotid artery
Stent-assisted coil embolization: It was performed under general anesthesia. A 6Fr sheath was inserted into the right femoral artery, and an 8Fr sheath into the left femoral artery. Heparin at 5000 units was intravenously administered. A 6Fr Roadmaster (Goodman, Aichi, Japan) and 8Fr Flowgate (Stryker) were guided into the left common carotid artery. After guiding an Excelsior SL10 (Stryker) into the aneurysm through the 6Fr Roadmaster, a Carotid Wallstent (Stryker) measuring $10 \times 24$ mm was inserted through the 8Fr Flowgate under distal protection with FilterWire EZ (Stryker). After stenting, coils (Target 360 soft measuring 6–4 mm in diameter, Ultra soft measuring 4 to 3 mm in diameter) were inserted into the aneurysm. After intra-aneurysmal embolization, post-dilation was conducted using a Jackal RX balloon (Kaneka Medix, Osaka, Japan) measuring $4.5 \times 30$ mm (14 atm, rated burst pressure). Postoperative angiography did not show any aneurysmal blood flow (Fig. 4).

Course after additional treatment: Combination therapy with aspirin and clopidogrel was continued. Immediately after surgery, right hemiplegia and sensory disturbance reduced. Aphasia ameliorated in comparison with the preoperative state, but mild sensory aphasia remained. Rehabilitation was conducted. The patient was discharged 2 months after surgery. The modified Rankin Scale (mRS) score on discharge was 1. After discharge, there was no new symptom, and there were no changes on MRI 1 or 2 months after surgery. DSA was performed to reassess the dural arteriovenous fistula 3 months after surgery. Stent fracture and aneurysm formation at the site of fracture were observed (Fig. 3). When reviewing postoperative cephalic CT findings, the styloid process measured 80 mm in length, suggesting an elongated styloid process. It had compressed the ICA; stent dilation was unfavorable at the site of compression (Fig. 3). The site of aneurysm formation was consistent with the site of compression of the ICA by the elongated styloid process. In addition, the site of ICA dissection at the time of onset was consistent with the site of stent fracture. Elongated styloid process-related mechanical stimulation of the ICA may have contributed to ICA dissection and stent fracture. Considering the risks of aneurysmal enlargement/rupture and recurrent cerebral infarction, we selected a therapeutic strategy to initially perform stent-assisted coil embolization as additional treatment, followed by extirpation of the styloid process.

Stent-assisted coil embolization: It was performed under general anesthesia. A 6Fr sheath was inserted into the right femoral artery, and an 8Fr sheath into the left femoral artery. Heparin at 5000 units was intravenously administered. A 6Fr Roadmaster (Goodman, Aichi, Japan) and 8Fr Flowgate (Stryker) were guided into the left common carotid artery. After guiding an Excelsior SL10 (Stryker) into the aneurysm through the 6Fr Roadmaster, a Carotid Wallstent (Stryker) measuring $10 \times 24$ mm was inserted through the 8Fr Flowgate under distal protection with FilterWire EZ (Stryker). After stenting, coils (Target 360 soft measuring 6–4 mm in diameter, Ultra soft measuring 4 to 3 mm in diameter) were inserted into the aneurysm. After intra-aneurysmal embolization, post-dilation was conducted using a Jackal RX balloon (Kaneka Medix, Osaka, Japan) measuring $4.5 \times 30$ mm (14 atm, rated burst pressure). Postoperative angiography did not show any aneurysmal blood flow (Fig. 4).

Course after additional treatment: Combination therapy with aspirin and clopidogrel was continued. The patient was rested from immediately after surgery until the day after surgery. During observation, there was no marked cervical extension or circumnutation. There was no surgery-related complication. Conditioned bone CT showed
Internal Carotid Artery Dissection and Carotid Stent Fracture due to Eagle Syndrome

Fig. 3 (A) Left common carotid artery angiogram (lateral view) at 3 months after an operation. (A) The left common carotid artery angiogram shows stent fracture (arrow) and aneurysmal formation at part of the stent fracture. (B) CT from the first visit. The elongated styloid process is seen (arrow). (C) CT performed on the next day of acute revascularization is revealing close proximity of the stent to elongated styloid process (arrow). (D) Cone-beam CT performed with the left-sided carotid artery angiogram shows stent fracture (arrow) and aneurysmal formation at part of the stent fracture. (E) CT after stent-assisted coil embolization. The elongated styloid process accepted a shift to the outside of the stent contact department (arrow).

Fig. 4 Left common artery angiogram at reoperation (lateral view). (A) A left common carotid angiogram before reoperation. (B) Stent-assisted coil embolization was performed. After coil embolization, PTA was performed at high pressure. (C) A left common carotid angiogram after reoperation showing disappearance of the dissecting aneurysm. PTA: percutaneous transluminal angiography
fracture of the styloid process at a site in contact with the stent and reduced compression related to the lateral deviation of a bone fragment the day after surgery (Fig. 5). During surgery, there was no fracture of the styloid process, but fracture may have occurred on post-dilation at a high pressure after coil embolization. After surgery, there was no fracture-related pain. Dynamic cone-beam CT with an angiography was conducted to confirm the absence of compression of the ICA by the fractured styloid process regardless of the head position. Extirpation of the styloid process, which had been scheduled, was not performed, and follow-up was continued. The patient was discharged 7 days after surgery. Angiography was conducted 2 months after additional treatment, but there was no recurrent aneurysm, stenosis, or fracture of the additionally inserted stent. MRI 1 year after stent-assisted coil embolization did not reveal any recurrent cerebral infarction, and plain X-ray did not show any recurrent stent fracture.

**Discussion**

The styloid process is a columnar, bony protrusion adjacent to the anterior stylomastoid foramen on the inferior surface of the temporal bone. Muscles and ligaments responsible for mastication or swallowing adhere to the styloid process. Normally, its length is up to 30 mm. The styloid process measuring >30 mm in length is regarded as an elongated styloid process. Eagle et al. indicated that the length of the styloid process was ≥25 mm in approximately 4% of adults, and that symptoms were observed in approximately 4% of these. No study has reported the correlation between the styloid process length and severity of symptoms. Embryologically, the styloid process is generated from the Reichert cartilage derived from the second branchial arch. The Reichert cartilage consists of four areas: tympanohyal, stylohyoid, ceratohyal, and subhyoid areas. However, the ceratohyal area comprises the funicular connective tissue through retraction in the embryonic phase, forming a styloid hyoid ligament. Ossification of the ceratohyal area of the Reichert cartilage related to some factor may be involved in the pathogenesis of an elongated styloid process. As etiological factors for ossification, disturbance of bone metabolism, anomalies, and trauma are considered, but the details remain to be clarified. Typical symptoms of Eagle syndrome consist of symptoms related to direct physical stimulation by an elongated styloid process and those related to compression of the inferior cranial nerves; various symptoms, such as pharyngeal
Table 1  Summary of previously reported cases of carotid artery dissection caused by an elongated styloid process

<table>
<thead>
<tr>
<th>Case</th>
<th>Author, (year)</th>
<th>Age</th>
<th>Sex</th>
<th>Symptoms</th>
<th>Initial treatment</th>
<th>Additional treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Zuber et al.</td>
<td>43</td>
<td>M</td>
<td>TIA (amaurosis fugax, hemiplegia)</td>
<td>Conservative therapy</td>
<td>None</td>
</tr>
<tr>
<td>2</td>
<td>Soo et al.</td>
<td>41</td>
<td>F</td>
<td>Blindness</td>
<td>Conservative therapy</td>
<td>None</td>
</tr>
<tr>
<td>3</td>
<td>Faivre e t al.</td>
<td>60</td>
<td>M</td>
<td>Confusion, hemiplegia hemianopsia</td>
<td>IV-tPA</td>
<td>Planned surgical resection of styloid process</td>
</tr>
<tr>
<td>4</td>
<td>Ohara et al.</td>
<td>43</td>
<td>M</td>
<td>TIA (aphasia)</td>
<td>Conservative therapy</td>
<td>None</td>
</tr>
<tr>
<td>5</td>
<td>Yamamoto, et al.</td>
<td>51</td>
<td>M</td>
<td>Eye pain, Horner syndrome</td>
<td>Conservative therapy</td>
<td>None</td>
</tr>
<tr>
<td>6</td>
<td>Todo et al.</td>
<td>57</td>
<td>M</td>
<td>TIA (aphasia), neck pain</td>
<td>Endovascular thrombectomy and CAS</td>
<td>None</td>
</tr>
<tr>
<td>7</td>
<td>Razak et al.</td>
<td>41</td>
<td>M</td>
<td>Hemiparesis, hemineglect, visual field deficit</td>
<td>IV-tPA</td>
<td>Planned surgical resection of styloid process</td>
</tr>
<tr>
<td>8</td>
<td>Sveinsson et al.</td>
<td>38</td>
<td>M</td>
<td>Hemiplegia, aphasia headache</td>
<td>IV-tPA, Endovascular thrombectomy and CAS</td>
<td>After recurrent thromboembolism surgical resection of styloid process</td>
</tr>
<tr>
<td>9</td>
<td>Sveinsson et al.</td>
<td>41</td>
<td>F</td>
<td>Sudden headache</td>
<td>Conservative therapy</td>
<td>None</td>
</tr>
<tr>
<td>10</td>
<td>Ogura et al.</td>
<td>55</td>
<td>M</td>
<td>Amaurosis fugax</td>
<td>Conservative therapy</td>
<td>After ICA occlusion surgical resection of styloid process</td>
</tr>
<tr>
<td>11</td>
<td>Ogura et al.</td>
<td>55</td>
<td>M</td>
<td>Hemiplegia</td>
<td>Endovascular thrombectomy and CAS</td>
<td>None</td>
</tr>
<tr>
<td>12</td>
<td>Ogura et al.</td>
<td>80</td>
<td>F</td>
<td>Asymptomatic aneurysm odynophagia</td>
<td>Planned surgical resection of styloid process</td>
<td>None</td>
</tr>
<tr>
<td>13</td>
<td>Hooker et al.</td>
<td>64</td>
<td>M</td>
<td>Dysarthria, aphasia</td>
<td>CAS</td>
<td>None (stent fracture 1 year after)</td>
</tr>
<tr>
<td>14</td>
<td>Miyata et al.</td>
<td>41</td>
<td>M</td>
<td>Disturbance of consciousness monoplegia, facial palsy</td>
<td>Endovascular thrombectomy and CAS</td>
<td>Planned surgical resection of styloid process</td>
</tr>
<tr>
<td>15</td>
<td>Mann et al.</td>
<td>39</td>
<td>F</td>
<td>Hemiparesis</td>
<td>CAS</td>
<td>None</td>
</tr>
<tr>
<td>16</td>
<td>Mann et al.</td>
<td>38</td>
<td>M</td>
<td>Dysarthria</td>
<td>CAS</td>
<td>None</td>
</tr>
<tr>
<td>17</td>
<td>Smoot et al.</td>
<td>60</td>
<td>M</td>
<td>Hemiparesthesia, hemiparesis dysarthria</td>
<td>CAS</td>
<td>None</td>
</tr>
<tr>
<td>18</td>
<td>Shimozato et al.</td>
<td>48</td>
<td>M</td>
<td>Hemiparesis</td>
<td>Endovascular thrombectomy and CAS</td>
<td>Carotid artery stent in stenting</td>
</tr>
<tr>
<td>19</td>
<td>Shimozato et al.</td>
<td>45</td>
<td>M</td>
<td>Hemiparesis, aphasia</td>
<td>Carotid artery stent-assisted coil embolization</td>
<td>None</td>
</tr>
<tr>
<td>20</td>
<td>Irisa et al.</td>
<td>51</td>
<td>F</td>
<td>Hemiparesis, aphasia</td>
<td>Endovascular thrombectomy</td>
<td>Planned surgical resection of styloid process</td>
</tr>
</tbody>
</table>

CAS: carotid artery stenting; IV-tPA: intravenous tissue plasminogen activator; TIA: transient ischemic attack
pain, pharyngolaryngeal discomfort, trismus, and cervical pain on rotation, may occur. Compression of the carotid artery leads to transient cerebral ischemia or cerebral infarction in some cases. However, these disorders are rare. To our knowledge, 20 patients with dissection of the ICA who initially developed cerebral ischemia, such as transient ischemic attacks (TIAs) and cerebral infarction, due to Eagle syndrome have been reported (Table 1). ICA dissection associated with an elongated styloid process is rare, but the number of case reports has recently increased; the involvement of an elongated styloid process in idiopathic cervical ICA dissection may be more frequent than previously recognized.

In the present case, we could not indicate the involvement of an elongated styloid process in the initial phase of treatment. Its involvement was shown to be an etiological factor through a retrospective review of bone conditions for postoperative CT on stent fracture. Based on cerebral angiographic findings, a diagnosis of ICA dissection was made. However, in the initial phase of treatment, we did not differentiate Eagle syndrome, as an etiological factor for idiopathic cervical ICA dissection, from other conditions. For this reason, the assessment of bone conditions using CT before and after surgery was insufficient, and we could not make a diagnosis of an elongated styloid process until the detection of stent fracture. Of the above 20 patients with cerebral ischemia, endovascular thrombectomy was performed as initial treatment in six patients. In three of the six patients, the presence of an elongated styloid process was clear in the initial phase of treatment. In these patients, CTA had been conducted on admission. It is impossible to sufficiently evaluate the styloid process using MRA or DSA because it is not visualized on the former and because bone subtraction is present on the latter. In some cases, plain X-ray (lateral or panorex views) suggests the presence of an elongated styloid process, but diagnosis using CT is more accurate. CTA may be useful for evaluating the presence or absence of an elongated styloid process in the acute phase of ICA dissection. As the ICA is not visualized on acute occlusion, it may be sometimes difficult to evaluate the positional relationship with the styloid process. However, after recanalization treatment by stenting for idiopathic ICA dissection, it may be necessary to evaluate the relationship between the stent and styloid process under bone conditions for CT, considering the presence of an elongated styloid process. Furthermore, it is possible to demonstrate compression of the carotid artery related to changes in the head position using dynamic cone-beam CT with an angiograph.

If patients with cervical ICA dissection in which an elongated styloid process is involved do not respond to medical treatment, extirpation of the styloid process may be effective. In 6 of the 20 patients with ICA dissection associated with an elongated styloid process who initially developed cerebral ischemia, extirpation of the styloid process was performed to prevent recurrent cerebral infarction or the exacerbation of dissection. After extirpation, there was no recurrent cerebral infarction or dissection in the six patients. In Cases 8 and 14, extirpation of the styloid process was conducted due to recurrent cerebral infarction and stent fracture. Furthermore, there was one case report in which stent fracture related to an elongated styloid process was observed after CAS (Case 14). CAS for symptomatic cervical ICA stenosis was performed, but CTA after 1 year showed stent fracture and ICA occlusion, suggesting the involvement of an elongated styloid process in the etiology. In our patient, angiography 3 months after CAS as initial treatment revealed stent fracture and aneurysm formation, and a review of postoperative CT findings showed that compression of an elongated styloid process was an etiological factor. The results suggest that recurrent cerebral infarction or stent fracture may occur if styloid-process-related compression persists after angioplasty by CAS. In the present case, we planned to extirpate the styloid process after stent-assisted coil embolization, but post-dilation on additional treatment led to fracture of the elongated styloid process and an improvement in compression; therefore, extirpation of the styloid process was not performed. Post-dilation was conducted for stent adhesion; fracture of the elongated styloid process was an unexpected event. As a result, in the present case, elongated-styloid-process-related compression of the stent reduced, and extirpation of the styloid process was not performed. However, if compression of a stent by an elongated styloid process persists, extirpation of the styloid process must be considered to prevent recurrent cerebral infarction or dissection and avoid stent fracture. Furthermore, in the present case, post-dilation unexpectedly led to fracture of the elongated styloid process. However, if there had been no fracture, post-dilation of the ICA at a high pressure might have resulted in external damage of this artery; we must reflect on a high-risk procedure. Percutaneous transluminal angioplasty (PTA) for inducing fracture of such an elongated styloid process should be avoided.
**Conclusion**

We reported a rare patient with Eagle-syndrome-related ICA dissection, stent fracture, and aneurysm formation. As an etiological factor for cervical ICA dissection, Eagle syndrome should be differentiated. If an elongated styloid process is continuously adjacent to the ICA after stenting, stent fracture may occur.

**Disclosure Statement**

We declare no conflicts of interest.

**References**