

Case Report

Dynamic assessment of internal carotid artery and elongated styloid process in a case of bilateral carotid artery dissection

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ABSTRACT

Background: Vascular Eagle syndrome is that an elongated styloid process causes ischemic stroke due to internal carotid artery (ICA) dissection. Dynamic assessment using radiological imaging has not been well investigated. We assessed the change in the relative positional relationship between the elongated styloid process and the ICA using a cone-beam computed tomography (CBCT).

Case Description: A 46-year-old female presenting with disturbance of consciousness, right hemiparesis, and aphasia was admitted to our hospital. Initial CT analysis showed a bilateral elongated styloid process. Magnetic resonance angiography (MRA) showed occlusion of the left ICA and a near occlusion of the right ICA. MRA also revealed the intimal flap and intramural hematoma in the bilateral ICA. Digital subtraction angiography showed bilateral ICA occlusion and carotid artery stenting was performed subsequently. After that, we visualized the movement of carotid stent with CBCT fusion methods. The stent moved forward and backward at the attachment point of the styloid process during head rotation, and there was a possibility that mechanical stress was emphasized at this point. Styloidectomy was performed after her rehabilitation. The patient did not experience a recurrence of stroke.

Conclusion: We showed that repeated attachment of the styloid process and ICA may trigger an ICA dissection during head rotation. This finding would be helpful for understanding the causes of vascular Eagle syndrome.

Keywords: Cone-beam computed tomography, Dissection, Eagle syndrome

INTRODUCTION

An elongated styloid process is a major cause of an internal carotid artery (ICA) dissection.^[6,10] Ogura *et al.* proposed the term vascular Eagle syndrome.^[7] Although several trigger movements may cause vascular Eagle syndrome,^[1,3,8,13] dynamic assessment of this condition using radiological imaging has not been well investigated. We observed the case of bilateral ICA dissection due to the elongated styloid process and performed carotid artery stenting (CAS). Subsequently, we assessed the change of the relative positional relationship between the elongated styloid process and ICA with cone-beam computed tomography (CBCT).

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CASE PRESENTATION

A 46-year-old female presenting with disturbance of consciousness, right hemiparesis, and aphasia was admitted to our hospital. Neurological examinations revealed a National Institutes of Health Stroke Scale score of 31. Initial CT analysis showed a bilateral elongated styloid process (right 31.1 cm and left 33.2 cm). Diffusion-weighted magnetic resonance imaging revealed acute cerebral infarction in the left frontal lobe. Magnetic resonance angiography (MRA) showed occlusion of the left ICA and a near occlusion of the

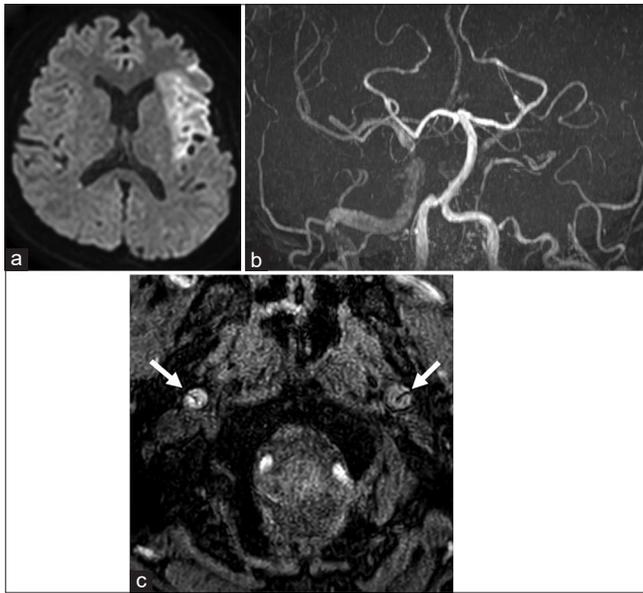


Figure 1: (a) Initial diffusion-weighted imaging showed a hyperintense area in the left middle cerebral artery (MCA) territory. (b) Magnetic resonance angiography (MRA) showed that the right internal carotid artery (ICA) was nearly occluded and the left ICA was fully occluded. The left distal MCA could not be detected using MRA. (c) The intimal flap (white arrow) was detected in the bilateral cervical ICA.

right ICA. MRA also revealed the intimal flap and intramural hematoma in the bilateral ICA [Figure 1]. Digital subtraction angiography (DSA) was performed on the patient followed by an infusion of intravenous tissue plasminogen activator. DSA showed occlusion of the left ICA and near occlusion of the right ICA. After the patient was administered 200 mg aspirin plus 300 mg clopidogrel, CAS was performed on the left ICA. An occlusion in the parietal branch of the left MCA was identified after the left ICA was recanalized (thrombolysis in cerebral infarction scale: TICI 2b). The branch of the left MCA was too distal to perform recanalization therapy. Subsequently, CAS was performed on the right ICA after the flow deteriorated [Figure 2]. After 5 days, there was an asymptomatic occlusion in the right ICA due to an in-stent thrombus. The left ICA was kept patent. CBCT revealed the detailed relationship between the elongated styloid process and the ICA in a neutral position. Specifically, they were positioned very closely together, especially on the left side [Figure 3]. According to CBCT, the distance between them changed on rotation of the head to the right and the left side. We made the fusion image in a neutral position and after the left/right rotations using clinical software (syngo InSpace 3D/3D fusion; Siemens AG, Forchheim, Germany) as previously reported.^[4] The precise registration was performed manually by superimposing the overlapping facial bone and styloid process from two CBCT images. Afterward, we evaluated how the distance of the leading edge of the stent changed. We defined the forward movement as “+” and backward movement as “-.” The left carotid stent moved -15.4 mm when the head was rotated to the right and +3.68 mm when the head was rotated to the left. The right carotid stent moved +3.96 mm when the head was rotated to the left and -3.24 mm when the head was rotated to the right. The fulcrum of movement was located at the attachment point of the styloid process and carotid stent. It was suggested that mechanical stress was emphasized at this point. The movement of the carotid artery was more remarkable in



Figure 2: (a) Digital subtraction angiography showed an occlusion in the cervical portion of the left internal carotid artery (ICA) (white arrow). (b) Carotid artery stenting (CAS) was performed for the left ICA followed by percutaneous transluminal angioplasty (PTA). The proximal and distal edges of the stent are represented with a white arrow. The left styloid process (black arrow) was close to the left ICA. (c) The right ICA was nearly occluded (white arrow). (d) PTA and CAS were also performed on the right ICA. The proximal and distal edges of the stent are represented with white arrows. The right styloid process was also close to the right ICA.

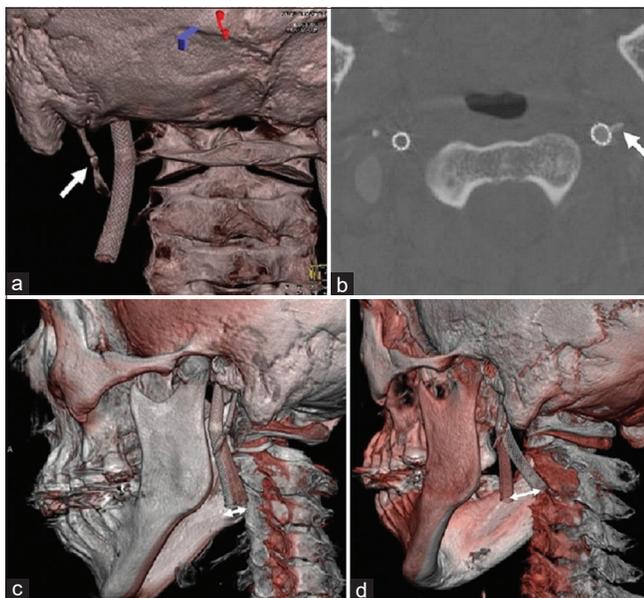


Figure 3: Cone-beam computed tomography after carotid artery stenting showed that the left carotid stent was close to the elongated styloid process (white arrow) according to the 3D (a) and axial images (b). (c) The fusion image between the neutral position (red skull) and the left rotation (white skull) shows forward movement (white double arrows) of the carotid stent. (d) The movement was more remarkable (white double arrows) in the fusion image between neutral position (red skull) and right rotation (white skull).

the left carotid artery. The right hemiparesis and aphasia gradually reduced over time and the patient moved to the rehabilitation hospital 42 days after administration with a Modified Rankin Scale score of 3. After finishing her rehabilitation, the patient underwent a styloidectomy. The patient did not experience a recurrence of stroke.

DISCUSSION

Eagle syndrome is divided into two main types: classical and vascular Eagle syndrome. Classical Eagle syndrome is characterized by pain in the throat, referred otalgia, and a foreign body sensation in the throat caused by attaching the lower cranial nerves with the elongated styloid process.^[2] Vascular Eagle syndrome occurs when an elongated styloid process causes ischemic stroke and carotid artery dissection by physical compression. Ischemic events such as aphasia, weakness, visual loss, or syncope are the most important symptoms in vascular Eagle syndrome.^[7] We reviewed cases of ICA dissection that was caused by vascular Eagle syndrome. The mean age of the vascular Eagle syndrome patients was 50.5 years (range: 38–80 years). Approximately 23.7% of patients were affected on their left side, 47.8% were affected on their right side, and 30.3% of patients experienced bilateral symptoms. The trigger movements were clear in six cases. The trigger movements were as follows: head flexion,^[15]

extension,^[8,10] tilt,^[12] boxing,^[13] and dancing.^[3] In our case, the trigger was unclear because the patient presented with aphasia and she could not explain her symptoms in detail. The proximity of the styloid process to the ICA is also one of the risk factors for ICA dissection. It is reported that the mean diameter between the styloid process and ICA was 8.5 mm in the ICA dissection group.^[9] In our case, the diameter between the styloid process and the ICA was 7.30 mm on the right side and 3.68 mm on the left side. Therefore, we concluded that an elongated styloid process is related to an ICA dissection. The dynamic assessment was avoided because it may exacerbate the dissection.^[10] However, the dynamic assessment also improves the understanding of the anatomical structures between the styloid process and ICA prevents recurrence of stroke. In this case, ICA moved with the closest point of the elongated styloid process as a fulcrum during head rotation, as observed with CBCT. We could evaluate the mechanisms of vascular Eagle syndrome for the 1st time using radiological imaging.

Bilateral ICA dissection due to elongated styloid process is rare and only five cases have been previously reported.^[3,5,7,8,14] To the best of our knowledge, this is the sixth case reported in the literature. Medical treatment is often not effective for vascular Eagle syndrome and may cause recurrence of ischemic stroke within a few days.^[7,11,14] On the other hand, CAS could prevent stroke recurrence in the acute phase. However, stent fracture has been reported due to the repeated attachment of the styloid process.^[6,13] Therefore, CAS therapy is recommended for the prevention of recurrent stroke in the acute phase and during subsequent styloidectomy in the chronic phase is recommended.

CONCLUSION

We used a dynamic assessment with CBCT to analyze the styloid process and the ICA during ICA dissection. Repeated attachment of the ICA and the styloid process may trigger an ICA dissection during head rotation. This finding would be helpful for understanding the causes of vascular Eagle syndrome.

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Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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