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CLINICAL REPORT

Perioperative management of Eagle syndrome complicated by carotid artery dissection

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Abstract Eagle syndrome is characterized by the sensation of a foreign body in the throat, odynophagia, dysphagia, and craniofacial or cervical pain due to an elongated styloid process. Some reports have indicated that an elongated styloid process is one of the causes of carotid artery dissection (CAD). This is the first report describing the anesthetic management of Eagle syndrome complicated by CAD. Careful intratracheal intubation and neurological monitoring are important to prevent intraoperative neurologic deficits. In addition, attention must be paid to postoperative complications such as airway obstruction.

Keywords Eagle syndrome · Elongated styloid process · Carotid artery dissection · Neurological monitoring

Introduction

Eagle syndrome is characterized by the sensation of a foreign body in the throat, odynophagia, dysphagia, and craniofacial or cervical pain due to an elongated styloid process that is part of the styloid chain (styloid process, stylohyoid ligament, and lesser cornu of the hyoid bone). The trigeminal nerve (V), facial nerve (VII), glossopharyngeal nerve (IX), vagal nerve (X), hypoglossal nerve (XII), carotid arteries, and internal jugular vein are all located near the styloid process. The clinical findings of this syndrome were outlined by Eagle in the late 1940s and early 1950s [1, 2]. Compression of the aforementioned cranial nerves and internal carotid artery by an elongated styloid process leads to various symptoms. Some reports have indicated that an elongated styloid process is the cause of carotid artery dissection (CAD) that is not part of Eagle syndrome [3]. In this report, we describe for the first time anesthetic management of a case of Eagle syndrome complicated by CAD.

Case report

A 55-year-old Japanese man (160 cm/55 kg) with no medical history developed sudden dizziness and transient visual loss in his left eye. He had noticed a stiff left-sided shoulder for the past few months. Magnetic resonance imaging showed scattered foci of acute infarction in the left cerebral hemisphere and bilateral internal carotid artery (ICA) dissection at the cervical level. The left ICA was occluded. Three-dimensional (3D) computed tomography (CT) imaging revealed a bilateral elongated styloid process impinging on the ICA (Fig.1), leading to a diagnosis of CAD secondary to Eagle syndrome. Although systemic anticoagulation therapy with heparin and warfarin was provided, he developed transient aphasia and right-sided hemiparesis. An emergent bilateral styloidectomy was performed for the occlusion of the right ICA.

Rapid sequence induction was intravenously performed with 250 mg thiamylal, 60 mg rocuronium, and 0.3 μg/kg/min

The authors received permission from the patient to publish this case report.

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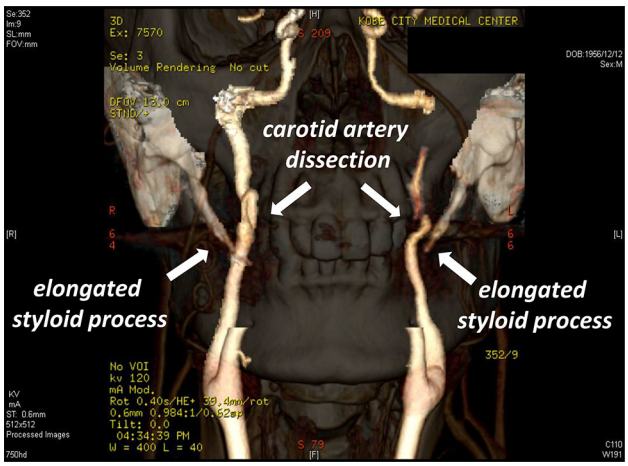

Fig. 1 Three-dimensional-computed tomography (3D-CT) imaging of a patient with Eagle syndrome complicated by carotid artery dissection (CAD). 3D-CT imaging revealed a bilateral elongated styloid process impinging on the internal carotid artery (ICA). The bilateral ICA was dissected at the cervical level and the left ICA was occluded

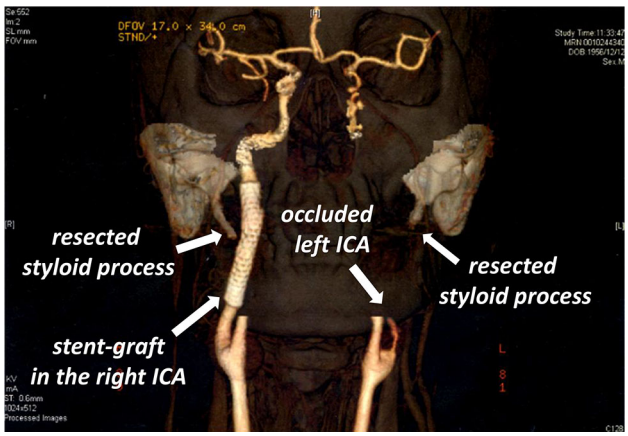

Fig. 2 3D-CT imaging of the neck after the operation. Carotid artery stenting of the left ICA was performed after the resection of the bilateral elongated styloid process. The left ICA was still occluded

remifentanil. Delayed contrast enhancement of the left ICA was observed with anteflexion of the head based on the results of a preoperative cerebral angiography examination. We carefully intubated using a video laryngoscope (Pentax Airway Scope; Ambu, Ballerup, Denmark) to avoid anteflexion of the head. The vocal cords were easily exposed, and intratracheal intubation was accomplished without difficulty. General anesthesia was maintained with 1.2–1.5 % sevoflurane, oxygen–air mixture, and 0.3–0.6 μg/kg/min remifentanil. We also performed intraoperative neurological monitoring (transcranial Doppler sonography, motor-evoked potentials, and somatosensory-evoked potentials) to monitor the exacerbation of CAD, but no significant events were recorded during surgery. When the patient was extubated after surgery, his systolic blood pressure went up to 200 mmHg and the right side of his neck began to swell. Although the patient airway was still patent, he once again developed aphasia. He was reintubated immediately by direct laryngoscopy without difficulty and a hematoma of the right neck was removed. The entire operation time was 4 h and the entire period under anesthesia was 6 h. Total blood loss was 136 ml. The patient was transferred to the intensive care unit under sedation for monitoring of bleeding. He was extubated the next day, and carotid artery stenting of the right ICA was successfully performed under local anesthesia (Fig. 2). Antithrombotic therapy was initiated with aspirin, clopidogrel, and heparin after surgery. Although the patient developed hoarseness for 1 month, his neurological symptoms improved and he was discharged from the hospital on postoperative day 22.

Discussion

The styloid process is a thin osseous projection that lies caudally, medially, and anteriorly from the inferior aspect of the petrous part of the temporal bone. The styloid chain (styloid process, stylohyoid ligament, and lesser cornu of the hyoid bone) develops from endochondral ossification of Reichert's cartilage of the second brachial arch during embryogenesis [4]. Several theories explain the cause of an elongated styloid process, such as congenital elongation of the styloid process and ossification of the stylohyoid ligament [5]. The various lengths and directions of the styloid process lead to variations in the symptoms. There is a consensus that styloid processes with a length exceeding 30 mm can be considered to be elongated [4]. The incidence of elongated styloid process has been found to be around 4 %, with most patients being asymptomatic. It is considered that 4 % of patients presenting an elongated styloid process develop symptoms [6].

Eagle syndrome (elongated styloid syndrome) was first described in 1652 by Pietro Marchetti. The clinical findings of this syndrome were outlined by Watt W. Eagle in the late 1940s and early 1950s [7]. He described two syndromes, classic styloid syndrome (the sensation of a foreign body in the throat, odynophagia, and dysphagia due to intermittent compression of the cranial nerves) and stylocarotid syndrome (pain in the parietal region or superior periorbital region due to compression of the carotid arteries and their perivascular sympathetic fibers).

Some recent reports have indicated that an elongated styloid process is the cause of CAD [8], as in our case. The distinctive feature of this type is that neurological symptoms, such as visual loss, hemiparesis, or aphasia, are aggravated by changing the head position.

The history of the presenting illness and clinical examination are important to correctly diagnose Eagle syndrome. Palpation of the tonsillar fossa reveals an elongated styloid process at this site. A 3D CT image reconstruction reveals the length and direction of an elongated styloid process in detail [9]. Cerebral angiography is also useful to detect abnormalities of the carotid arteries. If complicated by CAD, dynamic studies with head flexion, extension, and rotation are crucial to determine which head position aggravates the symptoms.

Surgical styloidectomy is a radical treatment for Eagle syndrome, although medical treatment is sometimes selected first [10]. If the patient develops CAD, carotid artery stenting should be considered.

Several crucial points should be considered for anesthetic management of patients with Eagle syndrome. This report is the first to describe the anesthetic management of patients with Eagle syndrome complicated by CAD. First, airway management is important. In the case of a patient whose elongated styloid process is attached to the lesser cornu of the hyoid bone, the mobility of the larynx would decrease and difficulties in intubation would be encountered [11, 12]. If the patient develops CAD, head position should be considered during intubation with an intratracheal tube because changes in the head position exacerbates CAD. Preoperative cerebral angiography can be used to identify which head position would delay contrast enhancement of the ICA. Second, intraoperative monitoring is important. Blood pressure and electrocardiogram should be carefully monitored. If sympathetic fibers around the cervical artery or carotid sinus are stimulated, changes in blood pressure and heart rate and even arrhythmia can occur. In cases of CAD, neurological monitoring, such as transcranial Doppler sonography, motor-evoked potentials, and somatosensory-evoked potentials, are essential to monitor the exacerbation of CAD or an embolism from the dissected artery. Third, it is necessary to consider postoperative complications. Unanticipated neuropathy, such as facial droop, hoarseness, and vocal cord paralysis can occur because of surgical manipulation. Progressive airway compromise from a neck hematoma and edema is a life-threatening complication. In cases of CAD, postoperative hematoma would exacerbate the symptoms derived from CAD, as in our case. Therefore, the patient should be carefully monitored in a postanesthetic care unit or intensive care unit after surgery.

In conclusion, CAD is a rare but life-threatening complication of Eagle syndrome. Careful airway management and neurological monitoring are crucial to prevent intraoperative neurologic deficits. In addition, attention must be paid to postoperative complications such as airway obstruction and unanticipated neuropathy.

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