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CLINICAL REPORT

Non-surgical therapy for bilateral glossopharyngeal neuralgia caused by Eagle's syndrome, diagnosed by three-dimensional computed tomography: a case report

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Abstract Eagle's syndrome is an uncommon sequela of elongation of the styloid process. Symptoms include recurrent throat pain and anterolateral neck pain, with referred pain to the ear. We report a 65-year-old man who presented with bilateral glossopharyngeal neuralgia. We performed three-dimensional computed tomography which revealed that the right styloid process was 35.1 mm and the left process was 29.6 mm, leading to diagnosis of an elongated styloid process, i.e. Eagle's syndrome. Because the patient refused surgical treatment, conservative therapy was selected. Oral gabapentin, stellate ganglion block, and 8 % lidocaine spray on the tonsillar branches of the glossopharyngeal nerve resulted in complete resolution of the paroxysms of pain in approximately 3 weeks.

Keywords Eagle's syndrome · Elongated styloid process · Glossopharyngeal neuralgia

Introduction

Glossopharyngeal neuralgia and trigeminal neuralgia are characterized by severe pain in the facial area. The annual incidence of glossopharyngeal neuralgia per 100,000 population is reportedly 0.7. This is one percent of the frequency of trigeminal neuralgia, suggesting that glossopharyngeal neuralgia is a rare disease [1]. Moreover, bilateral glossopharyngeal neuralgia is extremely rare and accounts for 2.5 % of glossopharyngeal neuralgia cases [1]. In addition, most cases of bilateral glossopharyngeal neuralgia are idiopathic and often difficult to treat.

Eagle's syndrome, defined by Eagle in 1949, is a syndrome in which the overgrowth or morphological abnormality of the styloid process causes pains in the distribution of the carotid artery [2]. The epidemiological frequency of this syndrome is 4–8 per 10,000 people [3]. It is characterized by facial pain, pharyngodynia, otalgia, sore throat, odynophagia, globus, or dysphagia, believed to be secondary to elongated styloid processor ossification of the styloid ligament [2, 4]. Patients who are diagnosed with idiopathic glossopharyngeal neuralgia may include those with this syndrome. We report a 65-year-old man with Eagle's syndrome who presented with bilateral glossopharyngeal neuralgia which was relieved by stellate ganglion block (SGB) and self-application of 8 % lidocaine to the tonsillar branches of the glossopharyngeal nerve.

Case report

A 65-year-old man had been suffering from upper respiratory infections approximately 4–5 times annually since childhood, but had never been diagnosed with tonsillitis. His past medical history was otherwise unremarkable. He visited the department of otolaryngology with a 1 year history of bilateral ear fullness and paroxysmal pain at the posterior auricles after infection with acute upper respiratory disease. Bilateral paroxysms of pain occurred simultaneously, approximately 5 times a day, and lasted for approximately 1 min. However, no definitive diagnosis was made, and the symptoms resolved spontaneously in 2 months. Similar symptoms recurred after 4 months. The

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patient visited the pain clinic of another hospital and was prescribed amitriptyline hydrochloride 75 mg/day plus carbamazepine 600 mg/day. However, because of mild vertigo, the drug was changed from carbamazepine to clonazepam 3 mg/day. The incidence of paroxysms of pain decreased after oral administration of the drugs. 4 months later, however, the onset of upper respiratory tract inflammation induced an increase in the number of episodes, and he visited our department.

At his first visit, he complained of "needle-like" pain on both sides of the areas from the posterior auricles to the external ears. Pain of both sides of the external ears was 52 mm on a visual analogue scale (VAS; evaluated as 0–100 by a sliding cursor 100-mm scale; 0 = no pain, 100 = the worst pain imaginable); that for the severest pain was 72 mm. Congestion in the face, ear fullness, and hoarseness were also observed as associated symptoms. Because positive diagnosis of the disease on the basis of the clinical manifestations of the patient was not possible, on the assumption the patient had "atypical facial pain", we first performed SGB on the randomly-selected left side of the face to examine whether this treatment might relieve the congestion of the face. SGB resulted in complete disappearance of the congestion in his face, and the effect was maintained for more than 5 h. However, the left-sided pain did not completely disappear—the score on the VAS decreased from 52 to 26 mm. Then, to examine whether the pain originated from the glossopharyngeal nerve, 8 % lidocaine was sprayed on to the tonsillar branch of the glossopharyngeal nerve on the right side, which completely relieved the pain on this side. 7 days later, 8 % lidocaine was sprayed on the left side (the other side), and the pain disappeared completely.

We performed three-dimensional computed tomography (3D CT) to determine the causes of bilateral glossopharyngeal neuralgia. 3D CT revealed that the right styloid process was 35.1 mm and the left process was 29.6 mm, leading to diagnosis of elongated styloid process (Fig. 1). Although we recommended surgical resection of the styloid processes, the patient preferred conservative therapy. Thus, first we switched from amitriptyline hydrochloride and clonazepam to gabapentin 600 mg/day because this is one of the medications known to be effective for glossopharyngeal neuralgia [5]. The glossopharyngeal neuralgia paroxysms decreased in frequency, but did not disappear completely. Thus, we instructed the patient to spray 8 % lidocaine on the areas of the tonsillar branches of the glossopharyngeal nerve at the onset of a paroxysm and to continue oral administration of the drugs. Nonsteroidal anti-inflammatory drugs had no effect on the attacks, so we did not prescribe them. Subsequently, the paroxysms were completely relieved in approximately 3 weeks. We are following this patient at our outpatient department for approximately 1 year. He experienced glossopharyngeal neuralgia twice during this period, but it was immediately relieved by self-application of 8 % lidocaine each time. The patient has kindly consented to the publication of the data and his images in the journal. We have obtained his written informed consent.

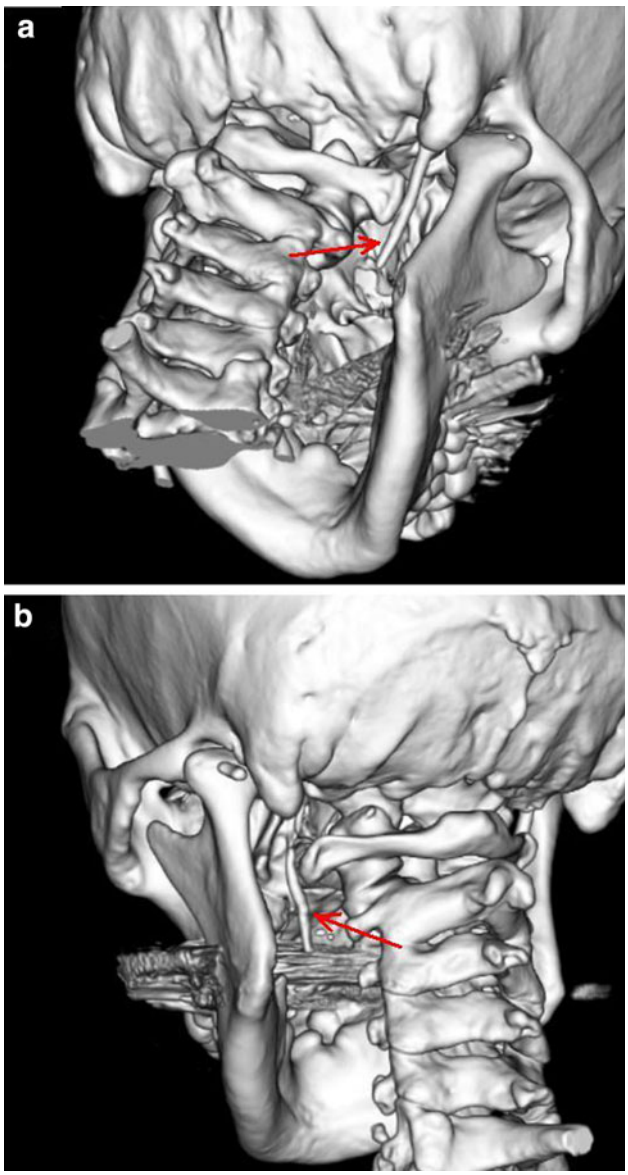

Fig. 1 Three-dimensional computed tomography of the styloid process: the length of the right side is 35.1 mm (**a**) and that of the left side is 29.6 mm (**b**)

Discussion

The styloid process projects downward and forward from the inferior surface of the temporal pyramidal bone, and it is attached to the styloglossus muscle, stylohyoid muscle,

stylohyoid ligament, stylomandibular ligament, stylopharyngeal muscle, etc. The cranial nerves (the glossopharyngeal nerve, facial nerve, trigeminal nerve, and hypoglossal nerve) and the sympathetic nerves are located around the styloid process [6]. The average length of the styloid process is 20–30 mm in adult Europeans and Americans and 15.4–18.8 mm in adult Japanese [6–8]. An elongated styloid process is defined as a process with a length of at least 25 mm [6, 8]. 3D CT is useful for diagnosis of this syndrome [3]. In our case, 3D CT scans revealed bilateral elongated styloid processes, and the patient was diagnosed as having Eagle's syndrome (Fig. 1). The causes of elongation of the styloid process include ossification of the stylohyoid ligament, chronic tonsillitis, past tonsillectomy, and chronic inflammation of the face and neck [9]. Major symptoms include pharyngodynia, odynophagia, otalgia, hearing loss, trismus, and dyskinesia of the tongue. Associated symptoms of this disease include neck stiffness, hot flash in the face, hoarseness, hyperhidrosis, and headache. Our patient complained of otalgia, hot flash in the face, and hoarseness. Although the detailed mechanism of the development of the symptoms has not yet been determined, it is believed they are caused by primary stimulation of nerves adjacent to the styloid process or by inflammation-induced secondary stimulation. The communication between the carotid sinus branches of the glossopharyngeal nerve and the superior cervical ganglia of the sympathetic trunk contributed to the effectiveness of SGB in our case. There is no evidence that SGB is effective for glossopharyngeal neuralgia. However, this case suggest that SGB may be a therapeutic option for facial congestion for patients with Eagle's syndrome.

Treatments for Eagle's syndrome include surgical treatments, for example resection of the styloid process and neurohemal organ decompression, oral administration of anticonvulsants (carbamazepine, baclofen, clonazepam, and phenytoin) and traditional Chinese medicines, intraoral spray or application of local anesthetics, and glossopharyngeal nerve block [4, 6, 10]. However, because Eagle's syndrome is not well known, this condition is often treated as tonsillitis, neuralgia, menopausal symptoms, or a psychogenic disease. Thus, in many cases, the symptoms of Eagle's syndrome could not be relieved. Our patient had developed upper respiratory tract inflammation several times per year; therefore, his symptoms might have been caused by chronic tonsillitis, and tonsillectomy was a possible treatment. However, tonsillectomy itself may cause Eagle's syndrome or bilateral glossopharyngeal neuralgia [11, 12]; therefore, its application requires caution.

Although we informed the patient that surgical intervention is essential treatment for this syndrome, he chose conservative treatment, and oral medication was initiated. However, satisfactory outcomes were not achieved. Consequently, SGB and topical anesthesia with lidocaine spray were added.

Subsequently, with the application of lidocaine 0–3 times per day, his symptoms disappeared completely in approximately 3 weeks from his first visit. It is not clear why the effect of 8 % lidocaine applied to the tonsillar branches of the glossopharyngeal nerve was sustained beyond the duration of action of the drug. The reason may be a synergistic effect with gabapentin. However, if paroxysmal glossopharyngeal neuralgia lasts for a long period, excessive stress on the accompanying sympathetic nerve might also lead to prolonged symptoms. Deafferentation of the glossopharyngeal nerve with lidocaine in the early paroxysmal stage is suggested as being useful.

In conclusion, our patient with the chief complaint of bilateral glossopharyngeal neuralgia was successfully diagnosed as having Eagle's syndrome by 3D CT examination. Many with Eagle's syndrome with an underlying organic cause may be included in the category of patients who present with symptoms of unknown cause, for example pharyngodynia, odynophagia, and pain in the ear, that are difficult to treat or of patients diagnosed with idiopathic glossopharyngeal neuralgia. Thus, 3D CT examination of the styloid process seems useful for differential diagnosis of these patients. Furthermore, noninvasive spraying of local anesthetics, rather than invasive surgery, should be considered as the first line treatment for glossopharyngeal neuralgia caused by Eagle's syndrome. In addition, we regard SGB as an effective treatment for sympathetic symptoms due to glossopharyngeal neuralgia.

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