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Eagle Syndrome: An Unusual Cause Limited Mouth Opening and Surgical Management

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Abstract: Eagle syndrome defined "stylalgia" occurs when an elongated styloid process or calcified stylohyoid ligament causes dysphagia, facial pain recurrent throat pain or foreign body sensation, also associated symptoms such as neck or throat pain with radiation to the ear. The symptoms related to this condition can be confused or miss diagnosis to a wide variety of facial neuralgias. The incidence of Eagle syndrome varies among population. Usually asymptomatic, it occurs in adult patients, can be diagnosed by physical examination and radiologically. A 30-year-old male patient presented to the maxillofacial unit of Sulaimaniyah Teaching Hospital with a complaint of pain in the right side of face interfering with mouth opening and causing deviation to the right side of mouth for 6 months duration. The elongated styloid process of the right side was resected surgically by the intra-oral approach. The patient was asymptomatic and comfortably followed up for 5 months.

Key Words: Eagle syndrome, elongated styloid process, limited mouth opening, pain, stylohyoid ligament, surgical management

It was defined "stylalgia" for the first time by Eagle in 1937 as an autonomous entity related to styloid process elongation or to mineralization of the stylohyoid ligament complex. The styloid process is an elongated conical projection of the temporal bone that lies between the internal and external carotid arteries anteriorly to the mastoid process, and laterally the tonsillar fossa. In this space, the glossopharyngeal, vagus, hypoglossal nerves, internal carotid artery, the internal jugular vein, and facial are located.[1] Eagle syndrome usually asymptomatic, the incidence is 4% of the general population, but the main incidence varies among population, it occurs in adult patients ranging from 30 to 50 years. Males are affected less often than females.[2] The normal length of the styloid process is individually variable, but in most of the patients it is about 20 mm.[3] Still there is debate about Eagle syndrome pathogenesis. Surgical trauma or local chronic irritation could cause periosteitis and osteitis of the stylohyoid complex with consequent reactive ossifying hyperplasia.[4–6]

Eagle syndrome is treated surgically and nonsurgically. A pharmacological approach by transpharyngeal infiltration of anesthetics or steroids in the tonsillar fossa has been used, but the treatment of choice is styloidectomy that can be performed by an extra- or an intraoral approach. The intraoral approach may result in the possibility of an incomplete control over many important vascular and nervous structures due to a restricted operative field and in the risk of deep cervical infections. On the other hand, external surgical approach results in risks of facial nerve injuries, longer hospitalization, and cutaneous scars.[7]

PATIENT PRESENTATION AND SURGICAL MANAGEMENT

A 30-year-old male patient presented to the maxillofacial unit of Sulaimaniyah Teaching Hospital, Kurdistan Region, Iraq, with a complaint of pain in the right side of the face interfering with mouth opening and causing deviation to the right side of mouth for 6 months duration. The pain was referring to the right upper neck region, dull to moderate in intensity and intermittent in nature. The pain intensity was exacerbated by movements such as wide mouth opening, looking up, and turning face to the left side. In addition, the patient during swallowing also had a sensation of foreign body in the throat. The patient had no history of tonsillectomy and trauma. Physical examination intra-orally revealed a tender, small bony hard projection the tenderness on palpation of the right paratonsillar fossa. Radiographically (orthopantomogram and computed tomography) demonstrated elongated bilateral styloid process, measuring 4.6 cm on the right side and 4.4 cm on the left side approximately. Subsequent to the radiographic findings recheck or re-examination of the patient revealed in the left tonsillar fossa a hard bony mass that was palpated but did not produce any symptoms.

The elongated styloid process of the right side was resected surgically by the intra-oral approach through a vertical incision of 2 to 3 cm that was made in the lingual mucosa of the ascending upper jaw. The tip of the elongated styloid process was identified by deep digital palpation and exposed by blunt dissection, resection of 2.2 cm from the styloid process under general anesthesia was done.

Ampiclox injection 500 mg (ampicillin + cloxacillin), Tramadol Ampole was prescribed directly postoperative (with Plasil ampole), and then replaced and continued with Paracetamol vial 500 mg for 3 days postoperatively. Discharge the patient on the first postoperative day. The patient was asymptomatic at follow-up at 5 months and did not have any complaint on the left side also (Fig. 1).

DISCUSSION

Eagle syndrome is defined as the symptomatic elongation or mineralization of the styloid process (calcification or ossification) of the stylohyoid ligament complex, for the first time documented by Watt W Eagle an otorhino laryngologist in the year 1937 over a 20-year period reported over 200 patients by Eagle and explained that the styloid process normally is approximately 25 to 30 mm in length. Also he observed that slight deviation of the styloid process medially could result in severe symptoms of atypical facial pain.[8] However, radiographically the presence of a styloid process elongation or mineralization of the stylohyoid complex with the presence of cervico-pharyngeal pain does not automatically confirm a diagnosis of Eagle syndrome.[9]

The diagnosis of Eagle syndrome can be ascertained by good history, clinical examination, and radiographic assessment with imaging that includes panoramic radiograph, lateral-oblique mandible plain film, lateral head and neck radiograph, Towne radiograph, etc. Lateral views are the best to show the length of the styloid process, despite antero-posterior views being also needed to determine whether there is bilateral involvement and the presence of lateral deviation. CT scans have been used in difficult cases to

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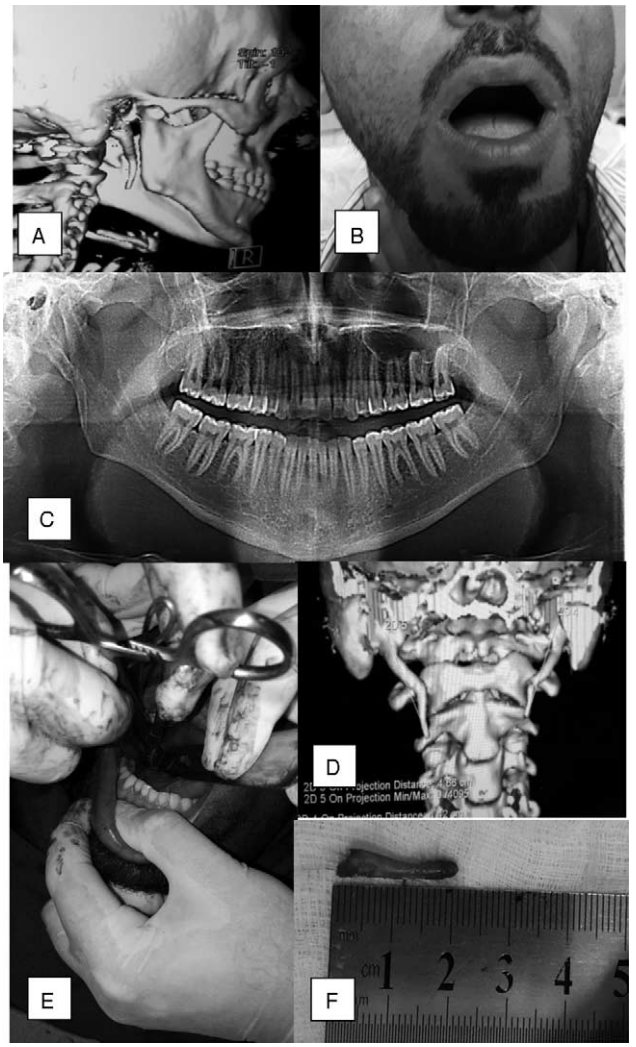

FIGURE 1. (A) Lateral view three-dimensional (3D) of computed tomography (CT) scan showing elongated styloid process. (B) Patient front view showing limited mouth opening. (C) Panoramic radiograph (OPG) showing elongated styloid process. (D) Posterior view 3D of CT scan showing elongated styloid process. (E) Intraoperative view of showing resection of elongated styloid process. (F) Resection of 2.2 cm from styloid process.

support diagnosis. Here, 3D reconstruction made it possible to visualize the exact spatial orientation of the styloid processes. The stylohyoid ligament ossification could definitely be ruled out on the basis of the imaging procedures. The diagnosis of Eagle syndrome is based on multiple clinical and radiographic factors.[10–12]

The physiology of the pain of Eagle syndrome could be attributed to fracture of the ossified stylohyoid ligament, followed by granulation tissue proliferation that causes pressure on surrounding structures and results in pain; or compression of the neural elements, lower branch of the trigeminal nerve, the glossopharyngeal nerve, and/ or the chorda tympani by the elongated styloid process; or even irritation of the pharyngeal mucosa by direct styloid process compression; also styloid process cause impingement on the carotid vessels and producing irritation of the sympathetic nerves in the arterial sheath; or fibrosis and stretching involving the fifth, seventh, ninth, and tenth cranial nerves in the post-tonsillectomy period; and even inflammatory changes and degenerative in the tendonous portion of the stylohyoid insertion, a condition called insertion tendinosis.[10,11,13,14]

The most effective treatment is shortening of the styloid process surgically through either an intraoral or an extraoral approach. Advantage of a transoral resection causes no scars, but we have risk of deep cervical infection and possible neurovascular injury; otherwise, an external approach leads to exposure of the styloid process and the adjacent structures. It also facilitates the resection of a partially ossified stylohyoid ligament. Conservative treatment options have included transpharyngeal injection of steroids and lignocaine, diazepam, the application of heat, nonsteroidal anti-inflammatory drugs, and transpharyngeal manipulation with fracturing of the styloid process manually. Surgeons blind fracturing of the styloid process have risks damage to nearby neurovascular structures and does not usually relieve symptoms.[11,12] The reported patient was successfully treated by using transoral approach. Prognosis of Eagle syndrome is guarded up to 20% of patients by surgical failures. This may be due to subsequent fibrous entrapment syndrome, or inadequate shortening of the process.[13–15]

CONCLUSION

The styloid process elongated syndrome can be diagnosed by a detailed history, physical examination, and radiological investigations. It can be mistaken for many other conditions that must be excluded. In conclusion, Eagle syndrome should always be put in the differential diagnosis of pain and limitation of mouth opening. The best treatment of choice for elongated styloid process is resection.

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