

Lateral Neck Pain - Eagle's Syndrome: A Report of Two Cases

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Abstract

Cervicofacial pain can occur due to elongated styloid process or calcification of stylomandibular ligament or stylohyoid ligament. Other factors also include chronic tonsillitis and tonsillopharyngitis. When the etiology of cervicofacial is elongated styloid process and/or ossification of ligament stylomandibular or stylohyoid it is referred as Eagle syndrome. Here, we are presenting the report of two cases of cervicofacial pain which were due to elongated styloid process and their management. The diagnosis of Eagle syndrome was made and confirm from clinical presentation and radiological investigations. As per present case and the available literature, styloidectomy is the treatment of choice for this condition.

Key words: Cervical pain, Eagle syndrome, Styloid process, Tonsillectomy

INTRODUCTION

Pain in orocervico facial region can occur due to chronic tonsillitis, elongated styloid process (unilateral or bilateral) or ossification of the stylomandibular and stylohyoid ligament. It may exert pressure on various vital structures such as carotid and glossopharyngeal nerve.¹

Stygalgia was first brought to light in 1937 by Eagle. Hence, it is termed as Eagle syndrome. It is also known by the name Ernest syndrome.² The styloid process is a small bony projection of temporal bone, which lies anterior to stylomastoid foramen. The styloid process lies between internal and external carotid arteries, posterior to the tonsillar fossa and lateral to the pharyngeal wall. The styloid process gives attachment to three muscles and two ligaments. The stylohyoid ligament itself extends from the tip of the styloid process to the lesser cornua of the hyoid bone. The stylomandibular ligament extends from the styloid process

to the angle of the mandible. The three muscles include the stylopharyngeus, stylohyoid, and styloglossus. The nerve supply for these muscles comes from glossopharyngeal, facial, and hypoglossal nerves, respectively. The internal jugular vein and the accessory hypoglossal, vagus, and glossopharyngeal nerves are located medial to the styloid process.³

The normal styloid process is 2.5-3 cm long as stated by Eagle.¹ Eagle earlier described two types of clinical presentation of stygalgia.

- Classical stylohyoid syndrome
- Stylocarotid syndrome

Eagle considered that only 4% patients with elongated styloid process experienced the orofacial pain meaning not all the elongated styloid processes cause symptoms.

Classical stylohyoid syndrome occurs mostly the following tonsillectomy with localized pain in tonsillar fossa, dysphagia,odynophagia, and hyper salivation, and it is presumed that tightened tonsillectomy scar moving the tip of the elongated styloid process during functional movement causes the orocervico facial pain.⁴

Stylocarotid syndrome occurs due to irritation sympathetic nerve fibers surrounding the internal or external carotid arteries.

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Ossification of the stylohyoid and the stylomandibular ligaments can result in a similar clinical picture. Few authors consider this condition as a stylohyoid syndrome. However, this condition causing cervicofacial pain is further lesser when compared to the ones caused by the elongation of the styloid process itself.⁵

Diagnosis is made with the help of imaging in form of X-rays, and three dimensional computed tomography with which the length and anatomical relations of the elongated styloid process can be assessed and further management can be planned. (Table 1)⁶

Management of Eagle syndrome has been surgical excision of the styloid process and the ossified ligaments under general anesthesia. Other modalities like steroid injection into the scar tissue and drugs to decrease muscle spasms are also used.⁷

Here, we are discussing two cases of Eagle's syndrome with the cause being elongated styloid process and their management.

CASE REPORTS

Case 1

A 37-year-old male, came to ENT outpatient department (OPD), Maharani Laxmibai Medical College, Jhansi with a chief complaint of pain in right tonsillar fossa for last 6 months. Pain radiated to the posterior part of the tongue, buccal, and cervical region of the neck.

He was asymptomatic before 6 months when he felt pain on swallowing the food which was moderate in intensity, paroxysmal in nature, and persist for 3-5 min/episode in the beginning, but after sometime pain persist throughout the day and radiated to cervical and temporal region.

He also experiences pain on turning the head on either side. The patients gave a history of tonsillectomy 10 years back and no history of trauma in head and neck region.

On intraoral palpation tenderness was present over tonsillar fossa and posterior part of the tongue.

On radiographic examination, orthopantomogram (OPG) and lateral oblique view of the mandible. Styloid process elongated right side > left styloid process (Figures 1 and 2).

On the basis of history and clinical and radiological examination, the patient was diagnosed as a case of stylalgia (Eagle syndrome) and styloidectomy was done under general anesthesia. After styloidectomy patient got excellent improvement in symptoms (Figure 3).



Figure 1: Orthopantomogram of the patient showing bilaterally elongated styloid processes more in right than the left



Figure 2: The lateral oblique view of mandible showing elongated styloid process with the tip extending up to the angle of mandible

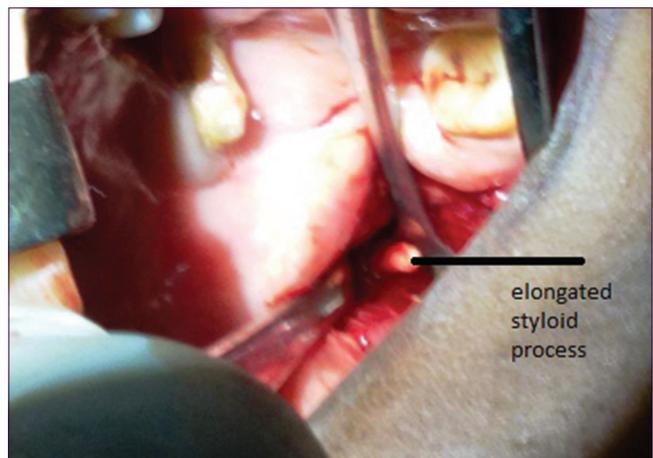


Figure 3: Intraoperative image of elongated styloid process

Case 2

A 60-year-old male presented in ENT OPD, Maharani Laxmibai Medical College, Jhansi with a chief complaint of pain in the orofacial region for 2 months which started initially on deglutination. The pain was moderate in intensity in the beginning but after sometimes pain

persisted throughout the day and referred to the temporal cervical and posterior part of the tongue.

The pain increases on turning head to either side and referred to the temporal region.

On intraoral examination, Grade-II tonsils were present and on palpation of tonsillar fossa pointed end appears.

On radiography orthopantomogram elongated styloid process right > left (Figure 4).

Initially, we did conservative management for 3 weeks with antibiotic and analgesic to treat tonsillitis. After 3 weeks of treatment, symptoms did not alleviated, and we planned for surgical management. We did tonsillectomy with styloidectomy and patients got relief from symptoms and became happy.

DISCUSSION

The stylohyoid chain consists of the styloid process, the lesser cornu of hyoid and stylohyoid ligament. The stylohyoid complexes are developed from second branchial arch or hyoid arch.²

Incidence of osseous metaplasia of stylohyoid ligament varies from 1.4% to 88.7% in asymptomatic individual; but symptomatic ossification of stylohyoid ligament is much rarer <4%.³



Figure 4: Bilateral styloid process

Table 1: Morphologic characteristics of styloid process

| Types | Nomenclature | Radiographic appearances |
|-------|--------------------|---|
| I | Elongated | Uninterrupted integrity of styloid image (>25-28 mm) |
| II | Pseudo-articulated | The styloid process is joined to the mineralized stylomandibular or stylohyoid ligament by a single pseudoarticulation, usually located superior to inferior border of the mandible |
| III | Segmented | Short or long continuous portions of the styloid process or uninterrupted segments of mineralized ligament |

There are various theories to explain its pathogenesis. According to Steinmann, ossified stylohyoid ligament is congenital anomaly.⁴

Fini *et al.*,⁵ reported that past tonsillectomy is related to Eagle syndrome.

Osseous metaplasia of residues of Reichart's cartilage (development, due to surgical trauma or local chronic irritation) resulting in osteitis and periostitis with reactive ossifying metaplasia, a recessive autosomal inheritance, malformation of styloid apparatus associated with malformation of the atlanto-occipital hinge, mucopolysaccharidoses and diffuse idiopathic skeletal hyperostosis, and endocrine disorder in postmenopausal women have all been implicated as pathogenetic factors.⁵

Langlais *et al.*,⁶ classified them into; Type I pattern which is an uninterrupted, elongated styloid process. Type II is characterized by the styloid process apparently being joined to the stylohyoid ligament by a single pseudoarticulation giving the appearance of an articulated elongated styloid process, and Type III consisting of interrupted segments of the mineralized ligament, sometimes creating the appearance of multiple pseudoarticulations.

Palesy *et al.*,⁷ have hypothesized that since the styloid process lies within the lateral pharyngeal space, a chronic soft-tissue injury there may result in edema resulting in an increase in fascial compartment pressure, thereby affecting neurovascular contents of the space, including the cervical sympathetic chain and glossopharyngeal nerve, through direct mechanoreceptor stimulation. The lingual nerve is intimately related to styloglossus muscle (arising from the styloid process) during its course. Ossified ligament may result in stretched styloglossus and resultantly lingual nerve.⁸⁻¹⁰

A patient with Eagle's syndrome may develop non-specific pain, which may change with head movements at the ear or neck. Additionally, a patient with an elongated styloid process may have referred pain to the jaw joint or upper extremities, or dysphagia or foreign body-like irritation throughout the pharynx.^{11,12}

Differential diagnosis includes trigeminal neuralgia, glossopharyngeal neuralgia, sphenopalatine neuralgia, chronic tonsilopharyngitis, temporal arteritis, and temporomandibular joint dysfunction, hyoid bursitis, Sluder's syndrome, etc.

CONCLUSION

Treatment options for Eagle syndrome are both surgical and non-surgical. Based on the above-mentioned cases

and their results and already available literature, we find styloidectomy is the treatment of choice for Eagle syndrome.

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