

Anatomical Perspective of Eagle's Syndrome: Review and a Case Report

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ABSTRACT

Aim: To discuss the anatomical perspective of Eagle's syndrome.

Summary: Eagle's Syndrome is a rare entity which is not commonly suspected in clinical practice, and only a small percentage of the population believed to have an elongated styloid process and a calcified stylohyoid ligament manifest the symptoms. It may develop inflammatory changes or impinge on the adjacent arteries or sensory nerve endings. A large spectrum of signs and symptoms have been mentioned in various reports for Eagle's syndrome. Diagnosis can be made with careful clinical evaluation and confirmed with radiographs showing an elongated styloid process or mineralization of the stylohyoid complex.

Keywords: Eagle's syndrome, Orofacial pain, Stylohyoid ligament, Styloid process

INTRODUCTION

Eagle's syndrome is a rare condition characterized by an elongated temporal styloid process (greater than 30 mm) or calcified stylohyoid ligament irritating the adjacent anatomical structures. The condition was first described by the American otorhinolaryngologist Watt Weems Eagle in 1937.¹



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Although the exact etiology is not known, dystrophic and degenerative changes in the hyoid complex of the styloid process is the cause of Eagle's syndrome. Purulent facial and cervical inflammations, tumors, tonsillectomies and trauma play a major role.^{2,3} The present paper intends to review and discuss the anatomical perspective of a case of Eagle's syndrome.

LITERATURE REVIEW

Eagle's syndrome comprises a constellation of symptoms which may include facial pain, otalgia, dysphagia, voice changes, and a foreign body sensation in the throat that prompts frequent swallowing which occurs secondary to an elongation of the styloid process. This elongation was first described in 1652 by Italian surgeon Pietro Marchetti, who attributed it to an ossifying process. In 1937, Watt W. Eagle^{1,4,7} coined the term stylalgia to describe the pain associated with this abnormality. In studies conducted over a period of twenty years, he reported that the length of the normal styloid process is approximately 25 mm to 30 mm. Other authors also acknowledge the elongation of the styloid process to be an etiologic cause of Eagle's syndrome.⁸⁻¹⁰ Eagle postulated that there are two types of the syndrome: the *classic* type and the *carotid artery* type which was also described in the studies of Breault¹¹ and Lorman¹². The *classic* type is characterized by pain secondary to the stimulation of branches of any of the following cranial nerves V (trigeminal), VII (facial), IX (glossopharyngeal), and X (vagus),¹³ and it is often seen in patients following tonsillectomy. Eagle theorized that these patients develop scarring near the styloid apex that subsequently compresses or stretches nerve structures in the space surrounding the styloid process. The *carotid artery* type occurs when the styloid process becomes involved with the carotid nerve plexus and causes a foreign body sensation in the pharynx and neck pain on rotation of the head.

Study done by Sokler *et al.*¹⁴ have shown that the average length of the styloid process is less than 3 cm, with the normal length ranging from 1.52 to 4.77 cm. Massey¹⁵ reported that only 11 of 2,000 cranial dissections detected a styloid process longer than 4 cm. Harma¹⁶ reported that the incidence of elongated styloid process is 4 to 7%. According to Murtagh *et al.*,¹⁷ only 4 to 10.3% of patients with an elongated styloid

process experience pain. The length of the styloid process has not been found to be correlated to the severity of pain.¹⁸ Continuing with the variability of the length of the styloid process, Kaufman *et al.*¹⁹ reported that 30 mm is the upper limit for normal styloid processes. Moffat *et al.*²⁰ performed a cadaveric study on the styloid process and reported that the normal length is between 1.52 cm and 4.77 cm. Monsour and Young²¹ concluded that the diagnosis of an elongated styloid process could be made whenever the styloid process was longer than 40 mm. In a radiological study by Montalbetti²², the length of the styloid process was reported to be no longer than 25 mm. Others like Wang²³, Basekim²⁴, Savranlar²⁵ and Jung²⁶ have also studied the length from radiographs and three dimensional CT. According to Montalbetti²² and Prasad *et al.*²⁷ prevalence of Eagle's syndrome in the population is reported to be 4% and is more frequent among women. However, other authors have reported the epidemiological incidence to be between 1.4-30%.^{4,28} In their studies they found that most patients with Eagle's syndrome were more than 50 years old.^{22,27} Rizzati²⁹ found a greater tendency in patients between 60-79 years of age. Conversely it was reported in an 11 years old patient by Quereshy *et al.*³⁰

Ilguy *et al.*³¹ reviewed 850 panoramic radiographs (PRs) and reported the incidence of elongated styloid process as 3.7%, and a 1:3 male/ female ratio was noticed in their study. They stated that elongated styloid processes were mostly bilateral. It was found to be bilateral in 75% of cases by Cawich *et al.*³² Bozkir *et al.*³³ claimed that 63% of patients showing elongated process were male and 75% of the cases were bilateral. The incidence of elongated styloid process was estimated at 3.3% for the total sample in the study of Balcioglu *et al.*³⁴ who detected 6 (55%) bilateral cases in PRs, and the male/female ratio as 1:9. Woolery³⁵ in his study also found a female preponderance.

Langlais *et al.*³⁶ classified elongated styloid process and mineralised styloid complexes based on the radiographic appearance and structures as follows: *Type I*: The elongated type pattern represents an interrupted process; *Type II*: Characterized by a single pseudo articulation that seems to be an articulated elongated styloid process; *Type III*: Represents an interrupted process that gives the appearance of multiple pseudo- articulations within the ligament. This type can be nodular or completely calcified. He also observed that Eagle's syndrome occurs mainly in 30-50-year-old patients, because regional ligaments and the soft tissues of the styloid process become less elastic with age and offer more resistance to surrounding hard tissue structures.²⁷

CASEREPORT

A 49 years old male reported with pain on the left side of the throat and tongue for the last four years. He also complained of tinnitus with intermittent otalgia. On examination there was

no odontogenic reason of the pain. A thorough clinical and physical examination revealed that the pain exaggerated with neck movements and swallowing. Intraoral palpation on the left side revealed that there was extreme tenderness in the tonsillar area. The tip of the styloid process was palpable in the tonsillar bed. On radiographic examination, bilateral elongated styloid processes were noted and a diagnosis of Eagle's syndrome was made (Fig. 1). The patient was operated intraorally under general anaesthesia and antibiotic coverage. After tonsillectomy the left styloid process was identified through the bed, all the muscles and other structures attached to it were stripped off and the styloid process with length 42 mm was excised (Fig. 2). Since the patient had no complaints on the right side, the styloid process on right side was not excised. Postsurgical healing was uneventful and the patient was relieved of his symptoms.



Figure 1: OPG showing bilateral elongated styloid process



Figure 2: Excised styloid process

DISCUSSION

The stylohyoid complex is composed of the styloid process, stylohyoid ligaments and the stylomandibular ligament.³⁷ The styloid process develops from the second branchial arch, specifically from the Reichert cartilage. Its muscular attachments include the stylohyoid, styloglossus, and stylopharyngeus muscles. The stylohyoid muscle connects the base of the styloid process to the hyoid bone near its greater horn; it is innervated by cranial nerve VII (Facial). It is

perforated near its insertion by the intermediate tendon of digastric. The muscle may be absent or double. It may lie medial to the external carotid artery. The styloglossus muscle arises from the anterior and lateral surface of the styloid process near the apex and descends forward between the branches of the internal and external carotid arteries. It then divides upon the lateral side of the tongue to blend with the fibers of the longitudinalis inferior linguae muscle and obliquely with the hyoglossus muscle. It is innervated by the hypoglossal nerve. The stylopharyngeus muscle traverses the medial aspect of the styloid process to the lateral wall of the pharynx; it is innervated by the glossopharyngeal nerve. The stylohyoid ligament connects the apex of the styloid process and the lesser horn of the hyoid bone, and the stylomandibular ligament extends from the styloid process to the parotidomasseteric fascia between the mandible and the mastoid process.

Other structures relevant to the operative management of Eagle's syndrome include the external and internal carotid arteries and the internal jugular vein. The styloid process is located between the internal and external carotid arteries and is juxtaposed near cranial nerves VII, IX, X, XI (accessory), and XII. Most cases of elongated styloid process are acquired, often as a result of trauma, but some are congenital.

In case of traumatic fracture of the styloid process there is proliferation of granulation tissue, which places pressure on the surrounding structures. In addition to trauma, inciting events for Eagle's syndrome include infection and early menopause. The symptom of pain in Eagle's syndrome may be multifactorial in origin. As mentioned, pain may arise secondary to compression of various cranial nerves (cranial nerves V, VII, IX, and X), irritation of the pharyngeal mucosa by direct compression or post-tonsillectomy scarring. Frictional irritation leading to chronic inflammation progressing to osteitis, periostitis, and tendinitis may likewise incite pain. Pain may also be caused by irritation of the superior and inferior caroticotympanic nerves (leading to otalgia) and the carotid sympathetic plexus close to an elongated styloid process. Albinas *et al.*³⁸ in their exhaustive study observed patients with spasmodic pain in the tonsils, arches of the palate, the soft palate, the root of tongue and the pharynx.

The diagnosis of Eagle's syndrome is frequently entertained only after a number of other diagnoses have failed to explain a patient's complaints. A complete history and physical examination may elucidate symptoms of a foreign-body sensation in the throat, otalgia and pain in the neck with a change of head position, dysphagia, or shoulder pain. The history may include tonsillectomy or neck trauma, often remote from the presentation of symptoms. The physical examination may reveal a palpable styloid process in the tonsillar fossa. Digital palpation of the styloid process often reproduces pain or a foreign-body sensation.³⁰ A suspected

diagnosis of Eagle syndrome can be confirmed in the office setting; the diagnosis is established if an injection of lidocaine into the tonsillar fossa provides relief of symptoms within minutes. Lateral views of the skull base and cervical spine and orthopantomographic (OPG) x-rays have been used as adjuncts to diagnose Eagle's syndrome. Although not essential, CT of the neck aids in discerning anatomic relationships and may rule out other conditions that produce similar symptoms.

Also, barium swallow studies can show the indentation of the elongated styloid process as a filling defect.³⁹ Despite the valuable information about the anatomy, there are some difficulties in reading the plain radiographs (true lateral, PA view and lateral oblique views of skull) secondary to superimposed anatomical structures. Superimposition of the mandible and the teeth can cause difficulty in viewing the styloid process, especially if it is not very long. Again, calcification of the stylohyoid ligament is difficult to evaluate in plain films. Superimposition of several osseous structures, and distortion and magnifications secondary to angulations are the potential disadvantages of conventional radiographs.⁴⁰ In CT imaging, those drawbacks are eliminated.

3D-CT images reformatted from the raw data obtained with a spiral scanner provide all the information about the styloid process, including its length, direction, and anatomical relations.^{41,42} Another advantage of the 3D-CT images is, of course, three dimensional length measurements, which are impossible in 2D images such as in coronal or axial planes. In cross-sectional imaging, even in coronal plane, most of the time the images are not parallel to the styloid process, which leads to underestimation of the actual length of the styloid process.

Differential diagnosis may include laryngopharyngeal dysesthesia, facial neuralgia^{39,43,44} dental malocclusion, neuralgia of sphenopalatine ganglia, temporomandibular arthritis, glossopharyngeal⁴⁵ and trigeminal neuralgia, chronic tonsillo-pharyngitis, hyoid bursitis, Sluder's syndrome, histamine cephalgia, cluster type headache, esophageal diverticula, temporal arteritis, cervical vertebral arthritis, benign or malignant neoplasms and migraine type headache^{13,46} or sometimes even as impacted molar teeth.⁴⁷

The nonsurgical treatment of Eagle syndrome generally involves pharmacotherapy with anticonvulsants (e.g., gabapentin) or antidepressants, but results are short-lived. Other treatments include steroid injections into the affected tissues with varying result^{48,49}. Long-lasting symptomatic relief requires the surgical removal of the elongated portion of the styloid process. Two surgical approaches have been described intraoral and extraoral. Intra oral approach requires more surgical skill as the chances to damage the adjacent vital structures are more owing to limited access and

visualization. However, it is less time consuming and more aesthetic. Conversely the extra oral approach is easier, but leaves a visible surgical scar⁵⁰⁻⁵².

CONCLUSION

It is crucial for the dentists, otolaryngologists and neurologists to be aware of the elongation of the styloid process and its anatomical basis. If any of the symptoms exist, digital palpation of the styloid process as a simple diagnostic procedure should be routine during the examination. Owing to the fact that styloid process with normal length is not normally palpable, the examination may easily reveal the problem. General dentists also need to be vigilant when viewing OPGs to ensure that they assess all the structures that can be seen and not just the teeth, alveolar bone, and temporomandibular joints.

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