

# Atypical chronic head and neck pain: don't forget Eagle's syndrome

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**Abstract.** – We report a case of an adult woman with an Eagle's Syndrome (ES) treated with medical therapy. ES is characterized by an aspecific orofacial pain secondary to calcification of the stylohyoid ligament or elongated styloid process. In about 4% of general population an elongated styloid process occurs, while only about 4% of these patients are symptomatic. We report a case of a 49-year-old lady with a 1-year history of oro-pharyngeal foreign body sensation localized at the left tonsillar fossa, associated with a dull intermittent pain. A bony projection was palpable with bimanual transoral exploration. A lateral radiograph and a computed tomography scan of head and neck showed an elongated styloid process of 57 mm on the left side and 48 mm on the right one. The patient refused surgical treatment as first choice. She underwent a non-steroidal anti-inflammatory local treatment, with progressive disappearance of symptoms. After 6 months she had no recurrence of symptoms.

In conclusion, a precise differential diagnosis is crucial in order to choose the most adequate treatment, which can be either surgical or non surgical. Medical treatment represents the first choice, followed by surgical styloid process resection, in the case of persistence or aggravance of the complaint.

#### Key Words:

Elongated styloid process, Eagle's syndrome, Head and neck pain.

## Introduction

Aspecific orofacial pain secondary to calcification of the stylohyoid ligament or elongated styloid process has been known as "Eagle's Syn-

drome" (ES) since 1937, when an otolaryngologist of the Duke University, Watt W. Eagle, described the first cases<sup>1-4</sup>. This syndrome was characterized by symptoms typically occurring after pharyngeal trauma or tonsillectomy and presents as a nagging dull, long-term ache in the throat, sometimes radiated to the ipsilateral ear, sensation of foreign body, occasionally odynophagia, dysphonia, increased salivation and headache. Not rarely patients believe that they have not properly healed from their tonsillectomy. The second and lesser-known presentation is constant throbbing pain through either the internal or external carotid artery distributions<sup>5</sup>.

Eagle considered tonsillectomy responsible for the formation of scar tissue around the styloid apex, with consequent compression or stretching of the vascular and nervous structures contained in the retrostyloid compartment (in particular glossopharyngeal nerve and perivascular carotid sympathetic fibres)<sup>1-4</sup>.

In the ensuing years, the term "Styloid Syndrome" was created to describe a cervico-pharyngeal pain related to the styloid process, when no previous history of trauma can be found<sup>6</sup>. Pathophysiologically, the styloid syndrome is related to an irritation of the surrounding nerves, the carotid artery or the pharyngeal mucosa.

The normal length of the styloid process varies greatly, but in the majority of patients it is 20 to 30 mm; it is considered elongated when it is longer than 25 mm<sup>7-9</sup>.

In about 4% of general population an elongated styloid process occurs, while only about 4% of these patients are symptomatic; thus the true incidence is 0.16% with a female predominance of 3:1<sup>5,10</sup>.

We report a case of an adult woman with an ES treated with medical therapy.

### Case Report

A 49-year-old lady presented to the Otolaryngology Clinic of the University Campus Bio-Medico of Rome with a 1-year history of oropharyngeal foreign body sensation localized at the left tonsillar fossa, associated with a dull intermittent pain on the left side of her throat. She didn't reported odynophagia, dysphagia, cervical pain, reflex ipsilateral otalgia.

The patient was detailed interrogated and screened thoroughly, and pain due to other factors, such as temporomandibular, dental, orthopaedic, and pharyngoesophageal causes, was ruled out. Questioning disclosed that she underwent a tonsillectomy and a revision of tonsillectomy respectively 40 and 10 years before. There was no history of neck injury.

The patient underwent detailed clinical ear, nose and throat (ENT) examination that included bimanual transoral palpation of left tonsillar fossa, on which a bony projection was felt; pain was elicited during palpation. The examination of the controlateral tonsillar fossa was normal.

A lateral radiograph of the head was ordered: the film showed remarkably elongated styloid processes (> on left side), both measuring more than one third of the length of the ramus of the mandible. This anomaly was confirmed by head and neck computed tomography scan showing an elongated styloid process of 57 mm on the left side and 48 mm on the right one (Figures 1, 2, 3). No other densitometric alterations were found at the skull base.

A positive lidocaine infiltration test was obtained, this consisting in a temporarily subsiding



Figure 2. CT scan showing a 48 mm right styloid process.

of patient's symptoms after infiltration of tonsillar fossa with 1 ml of 2% lidocaine.

The patient refused surgical treatment as first choice and she underwent a non-steroidal anti-inflammatory local treatment, with benefit. After 6 months she had no recurrence of symptoms.

### Discussion

Although the incidence of the styloid process elongation or mineralization of the stylohyoid complex is not uncommon, only a small percentage of these cases are symptomatic.

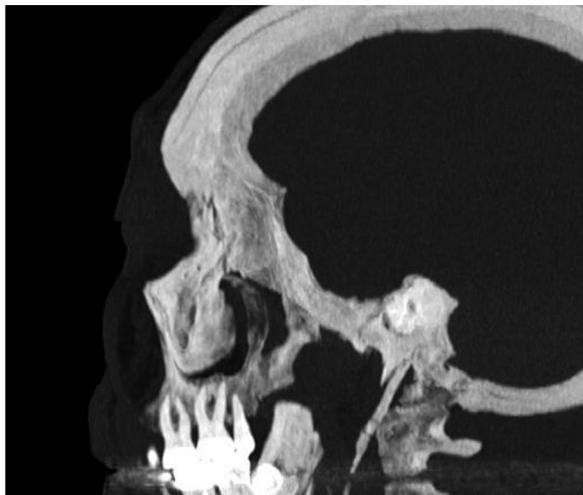


Figure 1. CT scan showing a 57 mm left styloid process.



Figure 3. CT scan showing both styloid processes.

The vagueness of symptoms and the infrequent clinical observation are often misleading. These patients may be seen by a surgeon, a dentist, a neurologist, and a psychiatrist, often receiving a variety of treatments that do not relieve the symptoms and that cloud the clinical picture. Stylalgia is misdiagnosed or overlooked as a possible diagnosis in cases of vague cervicofacial pain.

A variety of head and neck conditions should, however, be considered in the differential diagnosis of ES and cervicopharyngeal pain. These include temporomandibular disorders, glossopharyngeal neuralgia, trigeminal neuralgia, migraine-type headaches, sphenopalatine neuralgia, cervical arthritis, carotidynia, temporal arteritis, otitis, salivary gland disease and possible tumours. Other pathology should be eliminated by a careful medical history, clinical and radiographic examination<sup>5</sup>.

The diagnosis of the condition requires awareness and vigilance. Diagnosis of ES is based on a good medical history and physical examination. Typically the patient refers earache, especially on swallowing with an history of tonsillectomy; more rarely, the patient can also have a foreign body sensation in the pharynx with a persistent dull aching sore throat. Stylalgia is confirmed through palpation of the tonsillar fossa, transpharyngeal injection of long-acting local anaesthetic and /or steroids. With classic presentations of ES, imaging is not necessary for diagnosis; however, especially in doubt cases, plain skull films is enough. CT scan provides precious informations for the surgeon showing a more detailed view and measuring the precise styloid process length.

Once the diagnosis of ES or stylohyoid-related pain is made, surgical or non surgical treatment should be considered. NSA-drugs, transpharyngeal infiltration of steroidal drugs and lidocaine in the tonsillar fossa have been suggested as non surgical treatments<sup>11</sup>.

The surgical approach includes a transoral styloid fracture and/ or a surgical styloid shortening, which can be carried out either transorally or transcervically<sup>12</sup>.

The advantages of the trans-pharyngeal approach are safety, simpleness, shorter time, and no external scar, even though such an approach has been criticised by some authors in view of the possibility of infection of deep neck spaces, the risk of injury to major vessels and the poor visualization of deep planes. The advantages of the external approach are the better visualization and the reduced possibility of deep neck space

infection, while the disadvantages are the external scar, the longer surgical time and the risk of injury to the facial nerve<sup>13</sup>.

In the case of clinical symptoms such as dysphagia, foreign-body sensation and chronic neck or facial pain close to the ear, an ES should be considered in the differential diagnosis.

All patients should be placed on a stepwise therapy plan, which begins with a medical treatment, followed by surgical treatment, in the case of persistence or ingravescence of the complaint.

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