

An Unusual Cause of Recurrent Throat Pain - Calcified Stylohyoid Ligament

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ABSTRACT

Calcified stylohyoid ligament is also called an elongated styloid process and the symptom complex that it produces is called Eagle's syndrome. The symptoms of neck pain, sore throat, foreign body sensation in the throat, dysphagia and otalgia may be confused with other head and neck pain and are often worsened by yawning, opening the mouth wide or turning the head laterally. This is the result of involvement of cranial nerves, carotid plexus and cervical plexus. Carotid artery involvement causes wider symptomatology. Sometimes, it presents as a complication of tonsillectomy procedure. Elongated styloid process is conveniently identified on firm digital examination of tonsillar fossa. Diagnosis is made with appropriate radiological examination. Non-surgical treatment options include re-assurance, analgesia, anti-inflammatory medications and surgical option is shortening of the elongated styloid process by transoral or external approach. The condition is hereby described in a 59 years old male.

Key words: Calcified stylohyoid ligament. Eagle syndrome. Enlarged styloid process. Ossification of stylohyoid ligament.

INTRODUCTION

The Eagle's syndrome is the symptomatic manifestation of an elongated styloid process (or ossification of the stylohyoid ligament) by way of applying pressure in the neighbouring tissue structures. It is most commonly a result of a post-traumatic proliferation of the styloid bone, calcification of the stylohyoid ligament or an unusual elongation of the styloid process by 30 mm or more.¹ The possibility of its being a genetic recessive disorder could also not be excluded.

Cranial nerves which could be involved include the trigeminal, facial, glossopharyngeal, vagus, spinal accessory and hypoglossal nerves. The occasional carotid plexus and cervical plexus involvement can produce a wider symptomatology. It is classified a rare disease by the Office of Rare Diseases (National Institute of Health, USA)² and seen in Pakistani otolaryngology setup once in 5 years. Unless particularly sought after, the anatomical richness of various tissues in this region makes it easy to miss these pathologies on plain radiology. Classical symptoms include ear pain, sore throat, and / or foreign body sensation in the throat.²

This case report describe the syndrome in an elderly male.

CASE REPORT

A 59 years old male attended the Outpatient Department in Sligo General Hospital, Ireland, with a 6 months history of feeling something in his throat. The pain was mainly over the right side of the neck. It was not continuous and would become worse with yawning or turning the head to the right. It was also associated with pain in the right ear (otalgia) on swallowing, that would last for few minutes before it resolved by itself. He also complained of heartburn and reflux from time to time, during the same period. There was no history of dysphagia or hoarseness of voice. He denied any history of vomiting, haematemesis or weight loss. There was no previous history of ear disease and his past medical history was also not significant.

Examination of ear, nose, throat and neck was unremarkable. He was, therefore, booked for some non-invasive and invasive tests including audiogram, radiographic studies, flexible nasoendoscopy and barium swallow.

His audiogram was reported as normal. Flexible endoscopy revealed no abnormality and the result of barium swallow was normal as well. However, soft tissue X-ray of neck showed calcified stylohyoid ligament (Figure 1).

The diagnosis was explained to the patient and treatment options were discussed. The patient refused surgical intervention. Therefore, conservative treatment such as re-assurance and NSAIDs were given. The symptoms were improved after 3 months of treatment.

DISCUSSION

The first reported case of Eagle's syndrome was described by Dr. Watt W. Eagle in 1937 and so the name

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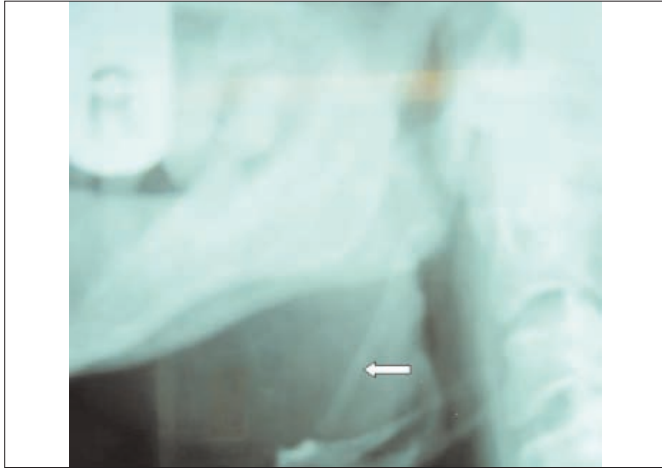


Figure 1: Lateral view plain radiograph of cervical spine shows calcified stylohyoid ligament.

of the symptom complex. It is a multitude of neurological symptoms, which if not correctly diagnosed or suspected by the family physicians, can conveniently leave the patient marked as a malinger or medicine seeker.²

A study by Gokce and colleagues from Turkey, reported a prevalence of styloid process elongation as 7.7% in panoramic radiographs of patients with unrelated symptoms and presenting at the dental department and no statistically significant age disparity in males and females.³ Eagle syndrome is more common in women than in men.⁴

Gokce in another published research hypothesized the role of abnormal calcium, potassium and vitamin D metabolism in patients suffering from end stage renal disease.⁵ He has also proposed the role of dystrophic calcification after mineralization of dead tissue, even with normal serum calcium levels.⁵

Stylohyoid apparatus is composed of the styloid process from temporal bone, the hyoid bone, and the connecting stylohyoid ligament. The literature usually cites 25-30 mm as the usual size of styloid process, varying from 5 to 58 mm and even 82 mm.^{6,7}

The ossified stylohyoid ligament is also reported with cervical spondylosis, anomalies in vertebral arteries, and fracture of the ossified ligament.⁶ Also to be considered for exclusion are oral, dental, and temporomandibular diseases.⁵

The styloid process has an embryological origin from the second branchial arch.⁸ Its elongation is hypothesized as attributable to persistence of the original mesenchymal elements (cartilage) and/or proliferation of osseous tissue at the point where the ligament and the styloid process meet. Ossification of the ligament could lead to its thickness and/or elongation of stylohyoid apparatus.¹

Any one or more of trigeminal, facial, glossopharyngeal, vagus, spinal accessory and hypoglossal nerves which

are present around the stylohyoid apparatus, may get involved. Also present around is the pharyngeal plexus, upper constituent parts of the cervical plexus, and the carotid sheath, the extent of involvement of each defining the resultant presentation.² Pain fibers are carried from the external auditory canal by the facial, the glossopharyngeal and the vagus nerves. Involvement of any one of these can cause ear pain. The same glossopharyngeal nerve is the sensory carrier for posterior pharynx and thus the throat pain and dysphagia. The hypoglossal nerve is the motor nerve carrier for the tongue, and thereby causes the tongue weakness. Involvement of trigeminal understandably can cause facial pain and headaches. The motor component of vagus nerves, which affects phonation and thereby the hoarseness. Neck pain could be a presentation with involvement of the upper components of cervical plexus. Apart from the neurological involvement of the trigeminal causing facial and temporo-mandibular joint pain, the physical obstruction through the elongated styloid process could partly contribute to painful mouth opening.² The styloglossus, stylopharyngeous and stylohyoid muscles; and the stylomandibular and stylohyoid ligaments, are attached to the styloid process.⁷ Pathologies of styloid process and or stylohyoid ligament could directly affect the performance of these muscles and therefore, deglutition, as two of the three assist the swallowing sequence. Gokce and colleagues have also stated that it is not only the size of the styloid process but also its medio-lateral angling, antero-posterior angling and the bending of the styloid process head could also be contributory factors.⁵ The conjoint carotid symptoms could result from pressures caused by an ossified (and/or thickened) stylohyoid ligament, complemented by an elongated styloid process.²

The easiest clinical sign to elicit is an exacerbation of the symptoms (or less often, tenderness) on deep palpation of styloid process in the tonsillar fossa.⁵ The use of panoramic radiography and three dimensional computer assisted tomography (3D-CT) can greatly assist diagnosis as well as surgical planning.^{1,4,5,9}

The condition can be managed surgically or conservatively. Conservative treatment could be use of NSAIDs, steroid injection, massage therapy, re-assurance and even alternative medicine approaches as proposed by various researchers.^{1,2,4,6} Surgical intervention seems to be the treatment of choice except in cases where it may be questionable, contraindicated or rejected by the patient. The two commonest approaches proposed and followed are transpharyngeal and extra oral, each with its own merits and demerits.

A classical study from Turkey, documenting a series of 61 cases operated over 5 years, all approached extraorally, have reported a 93.4% success (n=57),

where success was defined as becoming asymptomatic by the end of 12 months follow-up period.⁴ Three of the remaining 4 cases became asymptomatic after a second bone resection, and one was thought as having tendonitis at the hyoid end. The path of conservative treatment was eventually opted and yielded success. This group also reported a unilateral temporary paresis of marginal mandibular branch of facial nerve, which resolved within a fortnight.

The other surgical approach, which is free of this disadvantage, is trans-oral, also called as transpharyngeal or internal approach. This can be considered as an extension of transpharyngeal tonsillectomy, whereby the styloid process is exposed by removing the tonsils, which is then resected and removed as usual. This approach is simple, and takes less time, but has limited surgical access and poor surgical field. It is also criticized for a liability to deep neck infection. Naik and colleagues used dissection snare cautery under general anaesthesia, followed by palpation of the styloid process, then its dissection, denuding and finally nibbling it off using a bone nibbler.¹⁰ All of their patients were discharged within 7 days and were only prescribed antibiotics and anti-inflammatory medication post-operatively. Of the 15 cases 9 were symptom free in 1 week, 3 in 2 weeks, 2 in 1 month, and one was not symptom-free even after a month. The last one was later treated with carbamazepine for one month.

To conclude, Eagle syndrome is rare, its importance lies in the fact that it is presented as chronic sore throat a common symptom and is refractory to traditional

therapy. A high index of suspicion is essential in diagnosing the disease.

REFERENCES

1. Raina D, Gothi R, Rajan S. Eagle syndrome. *Indian J Radiol Imaging* 2009; **19**:107-8.
2. Dunn-Ryznyk LR, Kelly CW. Eagle syndrome: a rare cause of dysphagia and head and neck pain. *JAAPA* 2010; **23**:28, 31-2, 48.
3. Gokce C, Sisman Y, Ertas ET, Akgunlu F, Ozturk A. Prevalence of styloid process elongation on panoramic radiography in the Turkey population from cappadocia region. *Eur J Dent* 2008; **2**: 18-22.
4. Ceylan A, Köybasioglu A, Celenk F, Yilmaz O, Uslu S. Surgical treatment of elongated styloid process: experience of 61 cases. *Skull Base* 2008; **18**:289-95.
5. Gokce C, Sisman Y, Sipahioğlu M. Styloid process elongation or Eagle's syndrome: is there any role for ectopic calcification? *Eur J Dent* 2008; **2**:224-8.
6. Paraskevas GK, Raikos A, Lazos LM, Kitsoulis P. Unilateral elongated styloid process: a case report. *Cases J* 2009; **2**:9135.
7. Jakhar JK, Khanagwal VP, Aggarwal AD, Paliwal PK, Dhatarwal SK, Sirohiwal BJ, *et al.* Unilateral exceptionally elongated styloid process. *J Punjab Acad Forensic Med Toxicol* 2010; **10**:107-10.
8. Rodríguez-Vázquez JF, Mérida-Velasco JR, Verdugo-López S, Sánchez-Montesinos I, Mérida-Velasco JA. Morphogenesis of the second pharyngeal arch cartilage (Reichert's cartilage) in human embryos. *J Anat* 2006; **208**:179-89.
9. Beder E, Ozgursoy OB, Karatayli-Ozgursoy S, Anadolu Y. Three-dimensional computed tomography and surgical treatment for Eagle's syndrome. *Ear Nose Throat J* 2006; **85**:443-5.
10. Naik SM, Naik SS. Tonsillo-styloidectomy for Eagle's syndrome: a review of 15 cases in KVG Medical College Sullia. *Oman Med J* 2011; **26**:122-6.

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