



Case Series

Eagle eyes on cervicofacial pain: A case series of eagle's syndrome

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Abstract

Background: Eagle's syndrome or elongated styloid syndrome is an unusual condition which often goes misdiagnosed due to its vague presenting symptoms as well as its overlap with other causes of cervicofacial pain. The purpose of this case series is to highlight the importance of keeping eagle's syndrome as a differential diagnosis in patients who present with cervicofacial pain and to create awareness amongst dentists and maxillofacial clinicians about this condition.

Methods: 12 patients ranging in age from 20-66 years reported to the department of oral and maxillofacial surgery with different types of cervicofacial pain that they had for years without any resolution. They were all diagnosed as cases of Eagle's syndrome, some with no etiological factor, few were post-trauma patients and some had a history of tonsillectomy. All the patients were managed conservatively initially, though they were explained about all treatment options including surgical and non-surgical.

Results: All the patients were treated with medications and local anaesthetic infiltration and called for review after a week. Four patients had complete remission of symptoms, five patients had partial remission of symptoms though their pain was well controlled by medications, one patient who had complete remission of symptom was suggested and explained the surgical treatment and two of the patients were lost to follow up.

Conclusions: It is important to be able to differentiate and diagnose this syndrome clinically and radiographically from other similar conditions such as facial neuralgias and temporomandibular disorders so as to avoid patients from going through multiple hospital visits with no definitive treatment. The fact that it often goes undiagnosed is proof of the underestimation of the incidence of this syndrome and hence clinicians and dentists should have a high index of suspicion for Eagle's syndrome in patients with cervicofacial and pharyngeal pain.

Keywords: Elongated styloid process, Cervicofacial pain, Temporomandibular disorders.

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1. Introduction

Eagle's syndrome or Stylohyoid syndrome or stylalgia describes symptoms that are caused due to an elongated styloid process or the mineralisation of part of the entire Stylohyoid ligament.¹ Though this abnormal elongation of the styloid process was first noticed by Pietro Marchetti in 1652, it was Watt Weems Eagle, who in 1937, defined it as an autonomous entity.² Based on the involvement of either neural or vascular symptoms and the subsequent presenting symptoms, Eagle's syndrome has been divided into the classic eagle's syndrome and the vascular form, 'stylo-carotid artery syndrome'.³ Since there is an overlap of symptoms between this syndrome and other causes of cervicofacial pain such as TMJ disorders and neuralgias, there is a necessity to precisely diagnose this condition so as to overcome chances

of misdiagnosis and multiple hospital visits for the patient. Thus, the aim of this study is to report our experience with 12 cases referred to us during a period of two years that were misdiagnosed as other causes of cervicofacial pain. We have also aimed to concise in one place how this syndrome can be differentiated from neuralgias and TMJ disorders so as to prevent misdiagnosis and create awareness among the general dentists and dental students about this condition.

2. Case Description and Results

Twelve patients with eagle's syndrome reported to the department of oral and maxillofacial surgery at Annoor Dental College and hospital, over a period of two years. Age of the patients ranged from 20-66 years with a mean age of 40 years. Nine of the patients were females.

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The patients presented with extremely variable symptoms, for which most of them had undergone multiple visits to dentists and other specialists without any resolution. The usual presenting complaints of the patients were pharyngeal pain, irritation in the ear, a dull ache in the posterior mandibular region and a foreign body sensation in the throat. Initially, the patients underwent examination of the temporomandibular joints, followed by intraoral examination to check for impacted or infected third molars. An OPG was taken as part of the radiographic examination, that showed the presence of elongated styloid process (**Figure 1**). The observation of the elongated styloid apparatus on the radiograph, raised the suspicion of Eagle's syndrome. The patients were then asked for a history of tonsillectomy and any history of trauma. Five of the patients gave a history of tonsillectomy at some point in their life, two of them had a history of trauma following road traffic accidents, of which the radiograph of one of the patients showed pseudocalcified styloid process (**Figure 2**) and five of them had no history of tonsillectomy or trauma. None of the patients gave a complaint of the pain being aggravated on rotation of the head.



Figure 1: OPG showing elongated left styloid process extending to the angle of mandible



Figure 2: OPG showing right side pseudocalcified styloid process following history of trauma

The patients were then subjected to an intraoral digital palpation of the tonsillar fossa which confirmed the diagnosis. They were then explained about the syndrome and the treatment options available. All the patients were given lidocaine infiltration which relieved their symptoms and were also given NSAIDs as a supplement before opting for other treatments. All the patients were called for a review after one week. Five patients had partial remission of symptoms and the pain was well controlled with NSAIDs, one patient had recurrence of symptoms and was given the option for surgical removal of the styloid process, two patients were lost to follow up and four patients had complete remission of symptoms. The data of the patients are given in **Table 1**.

3. Discussion

Eagle's syndrome, an anatomical variation, though first reported by Pietro Marchetti of Padua in 1652, followed by several other clinicians noting this condition, it was Watt Weems Eagle in 1937 who first gave a detailed description of the syndrome. He also identified two different forms of the syndrome.^{4,5} The classic eagle's syndrome or Stylohyoid syndrome was described as those with unilateral or bilateral pharyngeal pain, aggravated by swallowing, Globus pharyngis (sensation of something stuck in the throat), facial and cervical pain.⁶⁻⁸ Eagle described these symptoms as a result of the development of scar tissue around the tip of the styloid process following tonsillectomy.⁹ The vascular form of the syndrome, also known as 'stylo-carotid artery syndrome' is due to impingement of the internal carotid artery by the elongated tip of the styloid process, especially on turning the head which can even result in dissection of the artery or a transient ischaemic stroke.¹⁰

Eagle estimated this syndrome to be a rare condition with only 4% of the population showing elongated styloid process, of which only 4% presented with clinical symptoms.^{3,5,12,13} The greater incidence seen in older patients has been attributed to the deposition of calcium salts into the ligaments and processes.¹⁴

3.1. Brief anatomy

The styloid process is an elongated, slender bony projection of the temporal bone, extending anteromedial to the stylomastoid foramen. The styloglossus, stylopharyngeus and Stylohyoid muscles are attached to the styloid process and insert into the tongue, pharynx and hyoid bone respectively. The Stylohyoid and stylomandibular ligaments also get their attachment from the styloid bone.

Anatomical knowledge of the styloid is important as direction of angulation of the elongated bone may be responsible for the presenting symptoms. Deviation of the tip laterally impinges upon the external carotid artery. Posterior deviation may cause irritation to the last four cranial nerves, internal carotid artery and IJV. Medial deviation can cause impingement into the tonsillar fossa and anterior angulation can result in mucosal irritation and pressure over vital structures in the tonsillar fossa.^{15,16}

The exact cause of eagle's syndrome is still considered a mystery despite the presence of a number of theories. Steinmann proposed three different theories for this anatomical variation.^{17,18} The 'theory of reactive hyperplasia' which states that during the post-traumatic healing period, ossification may take place from the styloid tip to the Stylohyoid ligament. The 'Theory of Reactive Metaplasia' explains that metaplastic changes that occur following a traumatic stimulus may also result in partial ossification. The 'Theory of Anatomic Variation' suggests that the elongation of the process is simply an anatomical aberration and not as a consequence of any traumatic stimulus.

Table 1: Data of 12 patients with Eagle's syndrome with symptoms, radiographic features, history and follow up after a week

Patient	Sex	Age (years)	Symptoms	Radiographic features	History	Follow up
1.	F	37	Right side otalgia and pain at the mandibular angle region	Bilateral elongated styloid process	Tonsillectomy 13 years back	Complete remission of symptoms
2.	F	38	Right side otalgia and pharyngeal pain on palpation, dysphagia	Bilateral elongated styloid process	Trauma 4 years back	Partial remission of symptoms
3.	M	52	Bilateral cervical pain with left side otalgia and pain radiating to left jaw	Bilateral elongation of styloid process	No tonsillectomy or trauma	Complete remission of symptoms
4.	M	43	Right side pharyngeal pain, dysphagia and dull ache at the mandibular angle region	Right side elongated styloid process	No tonsillectomy or trauma	Recurrence of symptoms
5.	F	34	Right side otalgia and pain at the mandibular angle region	Right side elongated styloid process	Tonsillectomy 11 years back	Partial remission of symptoms
6.	F	32	Right side otalgia, dysphagia, neck and mandibular angle pain, foreign body sensation in throat	Pseudocalcified styloid process of right side and elongated styloid process of left side	Trauma 2 years back	Partial remission of symptoms
7.	F	41	Right side otalgia and pharyngeal pain on palpation, dysphagia	Right side elongated styloid process	No tonsillectomy or trauma	Lost to follow up
8.	F	66	Left side otalgia and pain at the mandibular angle region	Left side elongated styloid process	Tonsillectomy done. Patient not sure of age. History of trauma 8 years back.	Lost to follow up
9.	F	42	Right side otalgia and bilateral cervical pain	Bilateral elongated styloid process	Tonsillectomy 17 years back	Complete remission of symptoms
10.	F	20	Pain at the left mandibular angle region and foreign body sensation on palpation	Left side elongated styloid process	No tonsillectomy or trauma	Partial remission of symptoms
11.	M	52	Bilateral cervical pain with dysphagia and dull ache in the jaw	Bilateral elongated styloid process	No tonsillectomy or trauma	Partial remission of symptoms
12.	F	27	Right side otalgia and pain at the mandibular angle region	Right side elongated styloid process	Tonsillectomy 7 years back	Complete remission of symptoms

3.2. Classification

Different types of classifications have been given based on the morphology and calcification patterns. The most commonly followed is the one by Ilguy et al which describes the radiographic appearance of the type of elongation (Type I: elongated, Type II: pseudoarticulated and Type III: segmented); Pattern of calcification (A: Calcified outline, B: partially calcified, C: nodular and D: completely calcified);

Angulation of the styloid process (Narrow: $<65^\circ$, Normal: $65-75^\circ$, Wide: $>75^\circ$).¹⁹

Clinically, on the basis of the location where the styloid tip could be palpated in the tonsillar fossa, Ravinder Verma classified it into three grades. Grade I: Tip of styloid reaches the superior pole of the tonsil, Grade II: Tip of styloid reaching the mid tonsillar fossa and Grade III: Tip reaching the lower end of tonsil or entering into the base of tongue.²⁰

3.3. Diagnosis

The length of the styloid varies greatly in a population, however, it is considered to be normal when measuring between 20-30mm. Any styloid greater than 30mm in length is considered elongated.²¹ According to Monsour and Young, a styloid process is considered to be elongated when the length is more than 40mm, as this is believed to be when the incidence of eagle's syndrome occurs most frequently.²²

The definitive diagnosis of Eagle's syndrome is based on presenting symptoms, digital palpation, radiographic findings and lidocaine infiltration test. Clinical examination involves intraoral digital palpation of the tonsillar fossa which exacerbates the patient's symptoms and a bony structure can be felt in the same region. However, this is not taken as a reliable indicator as studies suggest that the styloid process can only be palpated in the tonsillar fossa if longer than 7.5 cm.²³

Because the symptoms of eagle's syndrome overlap with other causes of cervicofacial pain, radiographic confirmation is imperative especially in cases with vague symptoms. Different types of radiographs can be used to confirm diagnosis of which, lateral views and OPG are significant. In an OPG, the styloid process is considered to be elongated if its length is more than 1/3 the length of the ramus of the mandible.²⁴ To confirm the diagnosis of Eagle's syndrome on radiographs, Goldstein and Scoop suggested certain criterias. If the stylomandibular ligaments region is indistinct, it is said to be not observable. If the radiopacity was found to be less than 1/3rd the length of the ramus, it is arbitrarily considered to be normal and if this length was found to be more than 1/3rd but not down to the angle of the mandible, it was described as 'partially calcified'. If the radiopacity extended upto the level of the angle of mandible, it was described as 'completely calcified'.²⁵

For the lidocaine infiltration test, 1ml of 2% lidocaine is administered to the area where the styloid process has been palpated in the tonsillar fossa. The patient is observed for any relief in the symptoms which considers the result to be positive and the syndrome is confirmed.²⁴

3.4. Differential diagnosis

Due to the vagueness of the presenting symptoms, it is important to know the classical symptoms generally associated with eagle's syndrome and certain similar conditions that may show overlapping symptoms. Eagle in 1948, described that patients who have undergone tonsillectomy and whose symptoms arise during the post-tonsillectomy phase are usually candidates of the typical eagle's syndrome. This is said to occur due to scar tissue formation that stretches the nerve endings and continues to irritate the fifth, seventh, eighth and tenth cranial nerves.⁴

On the other hand, the 'Carotid Artery Syndrome' occurs irrespective of tonsillectomy and is due to impingement of

the carotid artery by the elongated styloid process, especially during turning of the head. The pain extends from just below the eye to the chin on the face, if external carotid artery is impinged.³ In case of impingement of the internal carotid, the pain is usually over the entire head upto the occiput. The nature of these pain is a nagging one but not lancinating or knife like.¹³

The differential diagnosis of Eagle's syndrome includes, but not limited to, glossopharyngeal neuralgia, temporomandibular disorders, idiopathic trigeminal neuralgia and third molar pain.

According to Massey,²⁶ true glossopharyngeal neuralgia usually presents with recurring lancinating pain of short duration stimulated by intake of hot or cold food and beverages and on movements of the tongue as well. However the pain due to Eagle's syndrome are usually nagging in nature but rarely severe.

Another condition of significance for clinicians that needs to be differentiated from the Eagle's syndrome are temporomandibular disorders. Pain originating from TMJ disorders are usually considered chronic in nature, existing and increasing in intensity over a long period of time, worsening as the day progresses with tenderness over the muscles of mastication and may also be accompanied by trismus and clicking or crepitus in the joint itself.²⁷ Eagle suggested that patients who had obvious dental defects such as deranged occlusion, lack of molar support and tenderness on palpation over the joints, could be differentiated as true temporomandibular disorders.

To differentiate Eagle's syndrome from idiopathic trigeminal neuralgia, the latter usually presents with lancinating severe pain in the region of distribution by the three main divisions of the trigeminal nerves, commonly referred to as the trigger zones. And the diagnostic lidocaine infiltration in the region of distribution of these nerves can help differentiate from Eagle's syndrome.²⁸

3.5. Treatment

There exists both conservative and surgical treatment modalities, the preference being dictated by the severity of the symptoms and the etiopathogenesis.

Conservative therapy involves the injection of steroids or local anaesthetics into the tonsillar fossa or the lesser cornu of the hyoid bone to relieve the symptoms.²⁹

Two different surgical approaches can be used, namely the extraoral or transcervical and the intraoral or transpharyngeal approach.

The extraoral approach was described by Loeser and Caldwell in 1942. The possible complications of this approach include an unaesthetic scar, osteomyelitis, possible ligation of the carotid artery and injury to the facial nerve.²

The intraoral approach involves removal of the styloid process through the tonsillar fossa after dissection and reflection of the overlying muscles. Though the intraoral approach leaves no scar, there are more chances of deep cervical infection and hence is only recommended if the surgeon is familiar with the technique.⁹

Irrespective of the treatment modality chosen, the success rate is found to be in the range of 80%.³⁰

4. Conclusion

In this article, we have presented a case series of 12 patients who reported to the department with vague symptoms of cervicofacial pain. On further clinical and radiographic examination they were all diagnosed to be having Eagle's syndrome and were managed conservatively. Radiographic analysis using OPG and a clinical confirmation with local anaesthetic infiltration is considered to be diagnostic. Patients were given the option of conservative and surgical management and were on regular follow up. The article also highlights the overlapping symptoms between this condition and facial neuralgias as well as TMJ disorders and the need to keep Eagle's syndrome as a differential diagnosis in cases presenting with pain of the facial, neck and submandibular regions. The fact that it often goes undiagnosed is proof of the underestimation of the incidence of this syndrome and hence clinicians and dentists should have a high index of suspicion for Eagle's syndrome in patients with cervicofacial and pharyngeal pain.

5. Source of Funding

None.

6. Conflict of Interest

None.

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