

# Eagle's syndrome mimicking dental pain: A case report with a novel surgical approach

### ABSTRACT

PAIN in the craniofacial and neck region can be both intriguing and equally frustrating for the surgeon. This is principally because there is a multitude of related pain syndromes in this region, many of which are lacking in physical signs. Diagnosis then becomes even more dependent on an accurate description of the pain in terms of character, localization, duration, radiation, relieving and exacerbating factors. Familiarity and identification of a more obscure causative factor in a particular case lends itself not only to liberate the patient but also an increased awareness of the practitioner for the need to consider the coinciding minute diagnostic points of otolaryngology, ophthalmology and rhinology besides dentistry and oral surgery. The characteristic elongation of a styloid process may explain some occasions of pharyngeal, ear pain and sometimes headache, which have defied exhaustive diagnostic studies. A large spectrum of signs and symptoms has been mentioned in various reports of Eagle's syndrome. Diagnosis can be made with careful clinical evaluation and confirmed with radiographs showing an elongated styloid process or calcification of the stylohyoid complex. Styloidectomy is the procedural choice for Eagle's syndrome having high success rate. In our case, the intraoral approach for styloidectomy was not the routine one, for which the post-operative outcome was exceptionally good without any complications.

**Keywords:** Eagle's syndrome, orofacial pain, stylohyoid complex, styloid process, styloidectomy

### INTRODUCTION

Eagle's syndrome is an infrequent clinical condition, which habitually presents with recurrent pain in the oropharyngeal and facial region, sensation of a foreign body in the throat, referred otalgia and dysphagia owing to a lengthened styloid process or calcified stylohyoid ligament.<sup>[1]</sup> Its incidence has been reported inconsistently in the literature (1.0%–84.4%).<sup>[2]</sup> Eagle's syndrome is widely accepted to occur when either the overall length of the styloid process is >25 mm or when stylohyoid or stylomandibular ligaments present with ossification. Wide-ranging facial neuralgias and/or oral, dental, and temporomandibular joint (TMJ) diseases can be jumbled with the symptoms ascribed to Eagle's syndrome. Ample history, suitable clinical and radiological examination, and thorough knowledge of imitating pathology can help in diagnosing the syndrome. In this article, Eagle's syndrome presenting as pain of dental origin is described which was treated by a newer and simple intra-oral surgical approach.


In 2011, Raychowdhury proposed an incision line between the anterior faucial pillar and the tonsil.<sup>[3]</sup> A transoral, retromolar, para-tonsillar surgical approach, comparable to the procedure described by Raychowdhury, was implemented in six patients by Scheller *et al.* in 2014.<sup>[4]</sup> Kapoor, in 2015, proposed a simplified newer technique for styloidectomy using an intraoral approach under local anesthesia. The ease of execution, minimal anesthetic complications, and avoidance of extraoral scar commands that this approach could be practiced safely in patients with the enlarged styloid process.<sup>[2]</sup>

**RAVINDER SINGH BEDI, JITENDER KUMAR AURORA, HIMANSHU CHAUHAN, ARPITA KOMAL<sup>1</sup>**

Department of OMFS, Saraswati Dental College and Hospital, Lucknow, Uttar Pradesh, <sup>1</sup>Department of OMFS, Awadh Dental College and Hospital, Jamshedpur, Jharkhand, India

**Address for correspondence:** Dr. Arpita Komal, National Insurance Colony, M.G.Nagar, Kankarbagh, Patna - 800 026, Bihar, India.  
E-mail: arpita28.ak@gmail.com

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## CASE REPORT

A 23-year-old female patient reported to the Department of Oral and Maxillofacial Surgery, Saraswati Dental College, Lucknow, India, with a chief complaint of pain in the right back region of the lower jaw and surrounding area, including the neck, ear, and temporal region of the head of same side for 3 years. Pain was dull and intermittent in nature which was aggravated on masticating and swallowing and was relieved by bending neck on the same side. She visited various dentist and ear, nose, and throat specialist but was not relieved of the pain. The patient also presented with tenderness at the right inferior border of the angle of the mandible. On examination, there was no possible odontogenic reason of the pain, and an elongated styloid process was palpable in the right tonsillar bed. On radiographic examination (orthopantomogram [OPG] and three-dimensional computed tomography [CT]), bilateral elongated styloid processes were noted, of which the right process was elongated more as compared to the left one and a diagnosis of Eagle's syndrome was made.

Informed consent was obtained, and the patient was operated under general anesthesia using a distinctive intraoral approach (modified retromolar approach), in which the

incision was made along the right anterior border of the ascending ramus [Figure 1]. The plane of dissection was between the medial pterygoid muscle and the superior constrictor of the pharynx. The styloid process was then identified, its attachments were stripped off [Figure 2], and 24-mm length of the styloid process was removed [Figure 3].

Since the patient had no complaints on the left side, the styloid process on that side was not excised [Figure 4].

## RESULTS

Postsurgical healing was uneventful, and the patient was relieved of her symptoms who was followed up for 1.5 years. No early or late postoperative complications, including massive bleeding, neurovascular injury or infection, were encountered.

## DISCUSSION

An abnormally long styloid process may occasionally cause mild degrees of pain, restricted to the pharynx in the region of the tonsil. An elongated styloid process occasionally gives rise to a form of neuralgia in the distribution of the glossopharyngeal nerve.



Figure 1: Modified retromolar approach; incision line along the ascending border of the ramus for styloidectomy



Figure 2: The right styloid process exposed after dissection

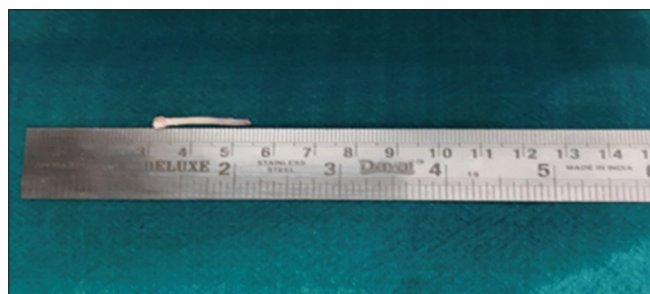


Figure 3: Excised styloid process



Figure 4: Postoperative orthopantomogram

The pain is referred to the malar bone, the tonsillar region, the ear, and the mastoid tip and often radiates downward into the neck and toward the shoulder. Palpation over the tonsillar fossa elicits pain, simulating the neuralgia complained of by the patient. The elongation of the styloid process is visible on the roentgenogram.<sup>[5]</sup>

In summary, it appears that an elongated styloid process could be a source of more obscure head and neck pain. The incidence is thought to be about 4% in the general population but estimates have ranged as high as 28%. Of that group, only about 4% actually exhibit any significant symptoms for which treatment is sought. These complaints may include unilateral pain and/or difficulty in swallowing; vague discomfort associated with normal mandibular motions, referring pain throughout the head-and-neck region including the TMJ; severe headaches, hemicrania, vertigo, tinnitus, syncopic spells, and visual disturbances. The most important diagnostic tools are digital palpation of the tonsillar fauces coupled with a thorough radiographic examination.<sup>[6]</sup>

### Treatment alternatives

#### Nonsurgical treatment

Various nonsurgical procedures suggested for the treatment of Eagle's syndrome include transpharyngeal injection of steroids and lidocaine. Although this is advocated and can be accomplished as an office procedure, its long-term therapeutic effectiveness has not been substantiated. Transpharyngeal manipulation with digital fracturing of the enlarged process can be performed. This is best done when the patient has been sedated and under local anesthetics administered in the pharyngeal region to abate pain during manipulation.

#### Surgical treatment

##### Extraoral approach

An external approach is widely recommended. Several authors have criticized this approach in view of the increased surgical time, morbidity, and complex dissection process because of adjacent vital anatomic structures. In the extraoral procedure, the patient is positioned in supine manner with the ipsilateral shoulder slightly elevated. With neck extended and rotated to the contralateral side, landmarks consistent with a Risdon approach to the mandible or the submandibular approach may be appropriate. After incising the skin and dividing the posterior extension of the platysma muscle, a blend of blunt and sharp dissection is undertaken to divide along the posterior border of the mandible, exposing part of the external carotid artery system which can be retracted forward. Underlying the investing fascia of the external carotid or internal maxillary artery, the styloid process can

be identified and easily palpated. Once the fascia has been detached from the surface of the styloid process, an incision can be made along the periosteum, thereby facilitating the reflection of the periosteum along with its muscular attachments. The styloid process can be excised near its base with dissection of the stylohyoid ligament at a point distal to the calcified portion. The closure of the wound can be done in a conventional manner.

##### Transpharyngeal surgical approach

Glogoff *et al.* recommended an intraoral approach under general anesthesia. If easily palpated, the styloid process in the tonsillar fossa can be used as a guide for placing the incision in the pharyngeal mucosa. The tissue overlying the elongated styloid process can be fixed and pressed onto the process itself to ensure that the bony projection is in proximity to the mucosal surface. A 1-cm incision is sufficient for its exposure. An angled antral curette is used to "lasso" the elongated process and to reflect the adjacent tissues. Once the styloid process is well visualized, the tip can be excised with a rongeur, and the edges are smoothed. A normal styloid process is about 2.5 cm and should be used as a guide to determine the approximate length to be removed.<sup>[7]</sup>

Fini *et al.* favored transoral approach after a detailed study on 10 patients who were followed up for 5 months to 4 years.<sup>[8]</sup> Prasad *et al.* retrospectively reviewed 58 patients who were followed up for 6 months to 5 years using intraoral approach along with tonsillectomy.<sup>[9]</sup> Furthermore, Beder *et al.* used similar technique on 19 patients.<sup>[10]</sup> Significantly, our technique avoids tonsillectomy, thus reducing surgical time and morbidity.

As advocated by Raychowdhury and Kapoor *et al.*, paratonsillar technique varies from the routine intraoral transpharyngeal approach where dissection is reserved lateral-to-superior constrictor, and hence there is no need of tonsillectomy and dissection becomes less time-consuming with reduced surgical morbidity. In our case, we slightly modified the incision which was placed along the anterior border of the ramus (modified retromolar approach). This procedure not only benefits the patient of not having a facial scar but is also comparably simpler as we have a bony guide for the dissection along with greatly reduced surgical time, easy access, and reduced surgical morbidity. Chase *et al.* proposed the usage of the intaoral approach only in cases of easily identifiable and palpable styloid process due to risk of damage to vital structures such as hemorrhage of major blood vessels and possible risk of glossopharyngeal nerve injury. However, with this technique, by careful dissection and restraining to the anterior compartment

of the lateral pharyngeal space which houses no vital structures (vital structures are a part of retrostyloid space), these complications can be avoided.

## CONCLUSION

An accurate case history coupled with appropriate radiographic evaluation (OPG, CT scan etc.) is a must for the diagnosis. The specialist's intuition provides imperative importance for the differential diagnosis regarding several other pharyngo-craniofacial pain disorders. In our opinion, surgical treatment is the first choice and the transoral approach is preferable. This modified retromolar approach avoiding injury to important structures contained in the maxillo-vertebro-pharyngeal space is characterized by a short-operative time, lacking visible scars, and condensed period of hospitalization, thereby being more economical and significantly can be the paradigm shift in surgical intervention for styloidectomy. The study on a large sample size needs to be done to account for the success rate and routinely use this novel surgical approach.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

There are no conflicts of interest.

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