Case Report - Surgical Pathology

Styloid-stylohyoid syndrome

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ABSTRACT

This is a case report of Eagle's syndrome due to osseous metaplasia of the stylohyoid apparatus treated conservatively by injection of a local anesthetic – steroid combination. The incidence, etiopathogenesis, classification, clinical picture and various approaches to treatment of ossified stylohyoid ligament associated with Eagle's syndrome have been discussed. Anterolateral glossodynia, which is a previously unreported finding in Eagle's syndrome, and its possible etiology, has also been discussed.

Keywords: Anterolateral glossodynia, eagle, osseous metaplasia, stylohyoid, styloid injection

INTRODUCTION

Stylalgia associated with abnormal length of the styloid process or mineralization of the styloid ligament complex was first brought to literary light in 1937 by Eagle.[1] The styloid process, which is an elongated conical projection of the temporal bone that lies anterior to the mastoid to the mastoid process, between the internal and external carotid arteries, and laterally the tonsillar fossa, gives origin to three muscles and two ligaments, namely the stylohyoid, styloglossal, and stylopharyngeal muscles, and the stylohyoid and stylomandibular ligaments respectivey. The carotid arteries, the internal jugular vein, the facial, glossopharyngeal, vagus, and hypoglossal nerves are all located in this region.[1]

Eagle primarily described two types of the syndrome associated with such findings in about 4% of the population, and stressed that not all cases of elongation or osseous metaplasia are symptomatic.[1] The classic styloid syndrome is usually seen after tonsillectomy as pharyngodynia localized in the tonsillar fossa, sometimes with dysphagia, odynophagia, hypersalivation, foreign body sensation, and more rarely by temporary voice changes, all of which presumably occur when tightened tonsillectomy scar tissue moves across the tip of the elongated styloid process during functional movements. The stylo-carotid syndromes (Carotidynia and Ernst syndromes) are due to compression of the internal and/or the external carotid arteries and especially their perivascular sympathetic fibers, resulting in a persistent pain radiating to the carotid territory, as headache, chronic neck pain, pain upon turning the head and pain radiating to the eye. Ear pain and vertigo are other possible presenting complaints. Patients with any of these clinical manifestations may thus present to the dental, otorhinolaryngology, ophthalmology or neurosurgeon with a plethora of complaints.[2]

Ossification of the stylohyoid ligament may result in a similar clinical picture, which has also been described as the styloid–stylohyoid syndrome by some authors.[3] The stylohyoid ligament may ossify from its origin at the styloid process to its attachment at the hyoid bone, but is rarely symptomatic.

This article reports a case of elongation and osseous metaplasia of the stylohyoid apparatus in a patient with unilateral anterolateral glossodynia and history of episodes of vague generalized pain (intraoral as well as facial) on the right side of face, managed by a conservative approach involving the injection of a local anesthetic–steroid combination. The incidence, etiopathogenesis, classification, clinical picture and various approaches to treatment of ossified stylohyoid ligament associated Eagle's syndrome have been discussed. The previously unreported clinical feature of anterolateral glossodynia, its possible cause and management are also discussed.

CASE REPORT

This 36 year old female patient presented to the department

with glossodynia of her right tongue, poorly localized moderate to severe pain on the right side of face (including the buccal and temporal regions) and intraorally in the right buccal and oropharyngeal mucosa since about 3 months. These complaints had been earlier diagnosed in various centers as temporomandibular joint pain, atypical facial pain, and pain associated with impacted right mandibular third molar, and treated accordingly with no significant relief. Patient was eventually referred to our institution for removal of the impacted tooth.

The impacted third molar was completely asymptomatic on examination, with no evidence of recent or past pericoronitis. Orthopantomogram (OPG) [Figure 1] showed bilateral impacted third molars. Of more immediate interest were the slightly elongated styloid processes and the bilateral ossified stylohyoid ligaments, which provided an alternative diagnosis for the patient's complaints, that is, Eagle's syndrome. This diagnosis was further reiterated when elective removal of the impacted tooth (the patient being willing) did not relieve the patient's symptoms.

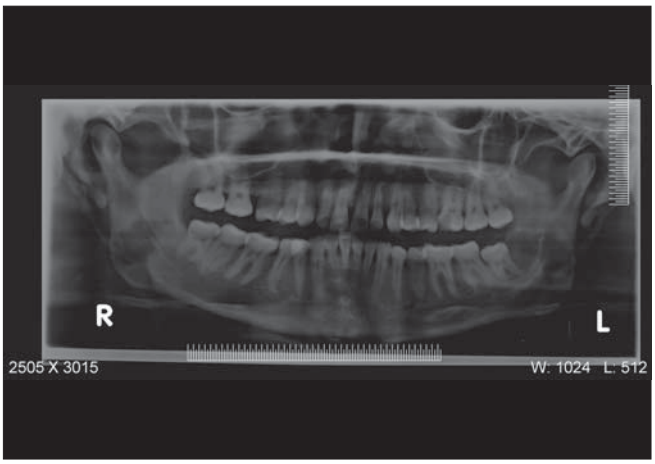

Figure 1: OPG showing osseous metaplasia of stylohyoid ligament

Elongated styloid process was not palpable in the tonsillar fossa.

She was then apprised of the possibility of Eagle's syndrome, and its treatment modalities. Conservative treatment was thought appropriate in her case, because the styloid process itself was not significantly elongated, so its fracture and removal were likely to have little impact. Moreover, since the diagnosis of Eagle's syndrome was provisional, relief with local therapy would serve as a confirmatory test at least.

Palesy *et al*., (2000), based on their cadaveric study in 1992, determined the safest pathway for injection in the styloid region, which helped guide our injection technique.[2] The steroid–local anesthetic combination used was Hydrocortisone (50 mg in 2 ml) and 0.5% Bupivacaine (1 ml).

The ear lobe, Frankfurt horizontal plane (FHP), and attachment of masseter muscle on the ascending ramus served as landmark for the point of insertion of needle [Figure 2]. Patient was instructed to clench teeth, and the postero-superior border of masseter palpated. After antiseptic and local anesthetic application, needle (25-G, 38-mm long), with bevel faced posteriorly (to avoid major vessels), was inserted below the ear lobe and 2-mm distal to posterior border of the ascending ramus at a level corresponding to postero-superior border of masseter, approximately 10º posterior to perpendicular at the skin surface and parallel to FHP. On reaching a penetration depth of 12 mm, after negative aspiration, 0.5 ml of the steroid – local anesthetic combination was injected. The needle was then inserted another 6 mm in the same direction (total depth 18 mm). The syringe was now angled anteriorly, some 30º from the previous position, while keeping it parallel to FHP, and inserted further to a total penetration depth of 24 mm. 1.5 ml of solution was deposited in this position [Figure 3]. In this way, 0.5 ml solution was deposited in the area of stylomandibular ligament insertion at the mandibular angle, followed by 1.5 ml solution deposition around the styloid–stylohyoid apparatus.

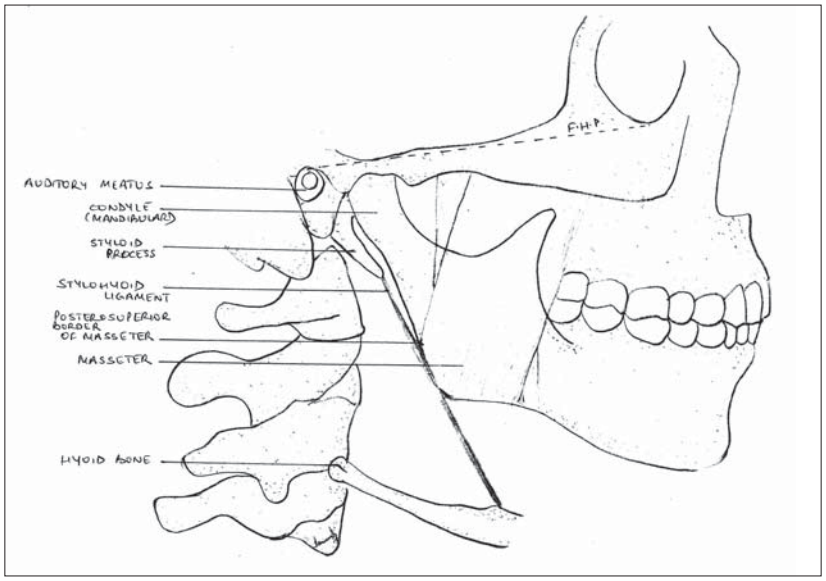

Figure 2: Schematic diagram of landmarks, seen from a lateral view of skull

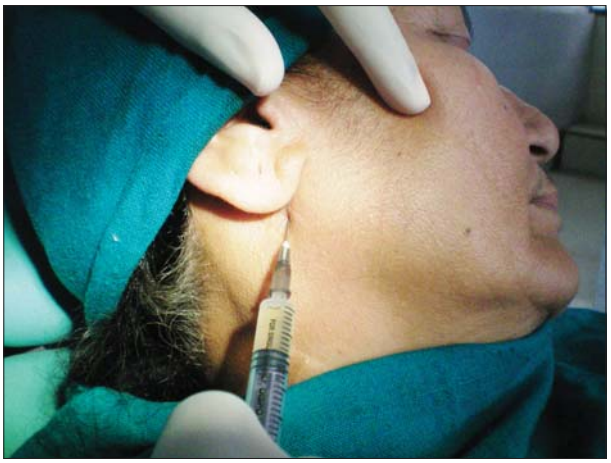

Figure 3: Demonstrating final injection position, in the region of styloid process

Another syringe was used intraorally to administer 0.5 ml of the steroid – local anesthetic combination in the region of the lingual nerve employing the standard technique for inferior alveolar and lingual nerve block administration in the pterygomandibular space.

The patient had immediate relief from pain in all regions, and has remained asymptomatic for the last 1 year since injection, which is a significant relief period. Moreover, the patient can always be readministered the injections if symptoms recur, or taken up for surgical intervention if symptoms worsen.

DISCUSSION

Reported incidence of osseous metaplasia of stylohyoid ligament varies from 1.4 to 88.7% in asymptomatic individuals; Krennmair and Piehslinger (1999) found an incidence of 17.7% when teleradiography, computed tomography and magnetic resonance tomography were used along with panoramic radiography.[4] Symptomatic ossification of the stylohyoid ligament is much rarer.

There have been various attempts at explanation of its pathogenesis, which remains debatable. According to Steinmann, ossified stylohyoid ligament is a congenital anomaly.[5] During development from the facial arch, four distinct segments are present in the stylohyoid apparatus: the tympanohyal, the stylohyal, the ceratohyal and the hypohyal. The ligamentous part has its origin from the ceratohyal cartilage. It extends from the stylohyal to the (lesser) cornu of the hyoid bone. Although the ceratohyal segment becomes the stylohyoid ligament, it retains some persistent embryological cartilage, which has the potential to grow and mature into bone. Potential for ossification of the stylohyoid ligament may thus be explained as an embryological phenomenon.[5]

Osseous metaplasia of residues of Reichart's cartilage (developmental, due to surgical trauma or local chronic irritation) resulting in osteitis and periostitis with reactive ossifying metaplasia, a recessive autosomal inheritance, malformation of styloid apparatus associated with malformation of the atlanto-occipital hinge, mucopolysaccharidoses and diffuse idiopathic skeletal hyperostosis, and endocrine disorders in postmenopausal women have all been implicated as pathogenetic factors.[6] However, ossification of the ligament alone is not sufficient to explain clinical presentation. Postoperative scar tissue, ossified stylohyoid causing contraction of the stylopharyngeal muscle and consequently stretching of adjacent cranial nerves, fracture and medialization of the ossified stylohyoid ligament, with frustrated repair disturbed by the continuous hyoid bone movements, causing excessive proliferation of granulation tissue responsible for compression of the structures nearby, ossification of muscular tendons leading to irritation, abnormal length associated with an abnormal angulation are possible explanations.[6]

Pseudoarticulated or segmented ossified stylohyoid ligaments represent a more progressed form of this phenomenon. Based on various radiological pictures, Langlais *et al*.,[7] classified them into; Type I pattern which is an uninterrupted, elongated styloid process, type II characterized by the styloid process apparently being joined to the stylohyoid ligament by a single pseudoarticulation giving the appearance of an articulated elongated styloid process, and type III consisting of interrupted segments of the mineralized ligament, sometimes creating the appearance of multiple pseudoarticulations.

Glossodynia of the base of tongue has been often described as a clinical feature of this syndrome. Anterolateral glossodynia, however, is extremely rare; in fact, the authors could find no mention of the same in previous literature. An explanation for the same was not easily forthcoming.

Palesy *et al*.,[2] have hypothesized that since the styloid process lies within the lateral pharyngeal space, a chronic soft-tissue injury there may result in oedema resulting in an increase in fascial compartment pressure, thereby affecting neurovascular contents of the space, including the cervical sympathetic chain and glossopharyngeal nerve, through direct mechanoreceptor stimulation. The lingual nerve is intimately related to styloglossus muscle (arising from styloid process) during its course. Ossified ligament may result in stretched styloglossus and resultantly lingual nerve. The lingual nerve and stylohyoid ligament also come in relatively close contact just above the hyoglossus, from where the lingual nerve moves deep and the stylohyoid apparatus carries on superficial to the hyoglossus muscle [Figure 4]. It may be hypothesized that since the stylohoid ligament is intimately related to the lateral wall of oropharynx, in close association with the lateral pharyngeal space, acute or chronic injury – related trauma and edema may result in increased intracompartmental pressure and therefore altered sensation and increased sensitivity to nociceptor stimuli of the lingual nerve. However, this is pure speculation, the authors were unable to find a convincing evidence – based etiopathogenetic relationship.

Differential diagnoses in patients with similar clinical picture include temporomandibular joint diseases, trigeminal, sphenopalatine or glossopharyngeal neuralgias, temporal arteritis, chronic pharyngotonsillitis, otitis media, external otitis, mastoiditis, dental pain, improperly fitting dental prostheses, submandibular sialadenitis or sialolithiasis, true pharyngeal foreign bodies, and tumors of the pharynx or tongue base, etc.[8] Laryngopharyngeal dysesthesia, temporomandibular arthritis,

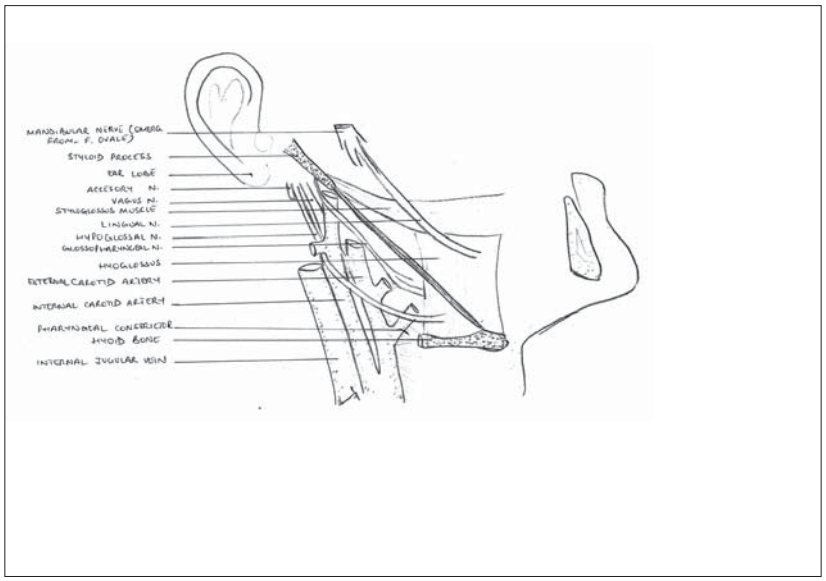

Figure 4: Schematic diagram illustrating the structures around stylohyoid ligament, and the vicinity of lingual nerve to stylohyoid ligament

hyoid bursitis, Sluder's syndrome, histamine cephalgia, cluster type headache, esophageal diverticula, cervical vertebral arthritis, and migraine – type headache should also be considered.[9]

Treatment options in all cases remain similar to that of Eagle's syndrome, with surgical and non-surgical alternatives. Conservative means have included administration of anti-epileptics and Anti-histaminics, vasodilators, neuroleptics, antidepressants and tranquilizers as supportive therapy with variable reported benefits.[10] Surgical removal has been described intraorally and extraorally. However, there are reports of recurrence of symptoms post-operatively, questioning the benefit of surgical technique, which leaves not many secondary options in such cases.

Injections of corticosteroids diluted in local anesthetics close to the styloid process is another alternative alternative.[2] Such injections have been described for diagnostic as well as palliative purposes, via mostly an intraoral approach transpharyngeally, into the region of anterior pillar and deeply into the anterior tonsillar fossa.[8] They reiterate the role of chronic trauma – related injury, oedema, raised intracompartment pressure in fascial spaces, and resultant increased nociception, all of which are relieved by the anti-inflammatory effect of corticosteroids and the anesthetic effect of local anesthetics.

This technique was thought most appropriate for our present patient, especially when an easily reproducible, well guided, anatomically established technique was at hand, as described. A lingual nerve injection was administered in addition to one in the styloid region. It would also serve to manage glossodynia in case it was simply coincidental.

This therapy is a conservative, nonsurgical approach which seems to have significant symptomatic benefit. It has the added advantages of minimal surgical trauma and/or damage, patient convenience and compliance, and the possibility of readministration, or surgical intervention at a later stage if symptoms persist. Advanced imaging techniques can further enhance results by better localization of the injection site. It is, therefore, recommendable, especially in patients presenting for treatment for the first time, and when the symptoms are mild to moderate. The possibility of antero-lateral glossodynia coincidental with, or as a new addition to the clinical spectrum of styloid-stylohyoid syndrome, is further emphasized.

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