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Eagle's Syndrome: A Report Of Three Cases

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Abstract

Eagle's syndrome, also known as elongated styloid process, is a condition that may be the source of craniofacial and cervical pain. It is infrequently reported but is more common than generally considered. The symptoms related to Eagle's syndrome can be confused with those attributed to a wide variety of facial neuralgias and/or oral, dental and TMJ diseases. These symptoms are familiar to dentists and can indicate a broad range of dental conditions also. The simple diagnostic procedure of palpation of the tonsillar fossa should be a routine during oral examination by the dentist also and Eagle's syndrome must be considered as a differential diagnosis of hemifacial pain of obscure causation. This condition can be managed by nonsurgical and surgical means but here we are presenting three case reports of Eagle's syndrome managed successfully by conservative means.

Key Words*: eagle's syndrome, styloid process, facial pain, neuralgia*

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I. INTRODUCTION

Eagle syndrome, also known as stylohyoid syndrome, styloid syndrome, or styloid–carotid artery syndrome, is a rare condition caused by an elongated or disfigured styloid process.[1] It comprises a constellation of symptoms which may include facial pain, ear pain, dysphagia, voice changes, and a globus sensation in the throat that prompts frequent swallowing-that occurs secondary to an elongation of the styloid process.[2] This elongation was first described in 1652 by Italian surgeon Pietro Marchetti, who attributed it to an ossifying process [3]. In 1937, Watt W. Eagle coined the term stylalgia to describe the pain associated with this abnormality.[4] Eagle postulated that there are two types of the syndrome that came to bear his name: the classic type and the carotid artery type:

(A)Classic type: The classic type is characterized by pain secondary to the stimulation of cranial nerves V (trigeminal), VII (facial), IX (glossopharyngeal), and X (vagus), and it is often seen following tonsillectomy. Eagle theorized that these patients develop scarring near the styloid apex that subsequently compresses or stretches nerve structures in the space surrounding the styloid process.[5] (B)Carotid artery type:The carotid artery type occurs when the styloid process becomes involved with the carotid nerve plexus and causes a foreign-body sensation in the pharynx and neck pain on rotation of the head.

Studies have shown that the average length of the styloid process is less than 3 cm, with the normal length ranging from 1.52 to 4.77 cm.[6] Massey[7] reported that only 11 of 2,000 cranial dissections detected a styloid process longer than 4 cm. Harma[8] reported that the incidence of elongated styloid process is 4 to 7%.According to Murtagh et al[9], only 4 to 10.3% of patients with an elongated styloid process experience pain. The length of the styloid process has not been found to be correlated with the severity of pain. The differential diagnosis of the Eagle's syndrome should include all the conditions causing cervicofacial pain. Medical history is the main guide for the diagnosis of Eagle's syndrome; however, palpation of the lateral tonsillar fossa, infiltration of local anesthetics to the tonsillar fossa, and radiologic examination are combined to confirm the diagnosis. Although several types of radiographs have been used since long time including 3-dimensional computed tomography (3DCT), which is the current and advanced technique. OPG is still the mainstay for measuring the definitive length of styloid process and takes the physician straightforward to the exact diagnosis of elongated styloid process [9,10]. Eagle's syndrome can be treated pharmacologically or surgically, or both. The surgical management of elongated styloid process consists of two major procedures: the transoral approach and the extraoral-cervical approach.[11] The choice of treatment usually depends on the experience of the surgeon.The aim of this paper is to present our management and treatment modality for Eagle's syndrome and to discuss the importance of exact

diagnosis of Eagle's syndrome and simplicity of conservative means when the styloid process has elongated especially at threshold level. It is a simple attempt to add to the contemporary knowledge of the treatment of this kind of rare disease commonly misdiagnosed.

II. CASE REPORTS

Case report 1

A 62 year old female presented with a two year history of right neck pain that worsened on flexion and when turning to the right. She also complained of a globus sensation and odynophagia. She did not report hoarseness or changes in her voice. The medical history revealed that she had gone for tonsillectomy 10 yrs back.For her complaint, the patient was initially referred to a general dental practitioner and misdiagnosed and treated for trigeminal neuralgia without any relief. The patient was then referred to Department of Oral & Maxillofacial Surgery. The complete head and neck examination revealed pain on palpation in the right tonsillar pillar. Other clinical findings were within normal limits. Her OPG(Orthopantomogram) examination detected a 35 mm styloid process on the right.1 ml of 2% lidocaine block was given at the site at which styloid process was palpable on the tonsillar fossa, temporary relief of the pain supported the diagnosis of Eagle's syndrome.

The patient was then explained about the nonsurgical and surgical approaches for the management of the problem. But she opted for nonsurgical management being aware that results of this treatment will be short lived. A block of 0.5 cc of steroid(Kenacort) mixed with 0.5cc of local anesthetic agentwas then given deep to the tonsillar fossa and analgesics were prescribed for 5 days.At six month follow up the patient could not elicit any pain by rotating her head down and to the right, and she reported that herglobus sensation was also resolved.

Case report 2

A 32 yearold female presented with a primary complaint of neck pain and stiffness of insidious onset for two months. The pain was more severe on the right side and was exacerbated when she rotated her head on right. In addition, she reported a dull headache occurring atleast two times per week. The headache lasted for a few hours and began with the onset of the neck pain and was worse in the morning with the pain starting in the suboccipital region and then referred to right retro-orbital region. The symptoms were aggravated during the festive season due to overhead work. The previous medical history was unremarkable except for a tonsillectomy 10 years back.Patient was referred to an Orthopedician, as per his physical examination there was a decrease in active range of motion of the cervical spine with pain on the end-range of extension and lateral flexion. Motion palpation revealed multiple joint restrictions with tenderness of the mid to upper cervical spine. The neurological examination was unremarkable. Patient was referred back to Department of Oral & Maxillofacial Surgery. Her radiographic examination (OPG)revealed elongation of styloid process (32mm).

Subsequent to the radiographic findings, re-examination of the patient revealed pain in the right tonsillar fossa on palpation, increasing the patient's neck pain. In this case also, 1 ml of 2% lidocaine block was given at the site at which styloid process was palpable on the tonsillar fossa, temporary relief of the pain supported the diagnosis of Eagle's syndrome. After discussion with Orthopedician, primary diagnosis of vertebrogenic headache in addition to stylalgia was made. The patient was then subjected to a short course of spinal manipulative therapy directed at the involved facet joints as well as manual soft-tissue therapy on the hypertonic cervical musculature and intra-oral tissues surrounding the styloid process. In addition to a block of 0.5 cc of steroid (Kenacort) mixed with 0.5cc of local anesthetic agent was given deep to the tonsillar fossa and analgesics were prescribed for 5 days, the patient was also advised a regimen of upper cervical stretches. In approximately one month of treatment the patient reported remission of the headache and a full and pain-free range of cervical motion.

Case report 3

A female patient aged 65 years with a history of multiple extractions of teeth from a private dental clinic reported with a chief complaint of severe pain on the right side of face radiating to the right temporal and retro orbital region along with globus sensation since 6 month.The history revealed multi-modal medicinal therapy including that for trigeminal neuralgia. The clinical examination revealed tenderness in the right tonsillar fossa. Patient was advised for OPG which showed bilateral elongation(right side 40mm and left side 37mm) of styloid process. Further palpation of both right and left tonsillar fossae confirmed tenderness on the right side but not on the left side. Provisional diagnosis of eagle`s syndrome was made and a diagnostic 0.5 cc lidocaine block on the right tonsillar fossa alleviate the pain. This led to the diagnosis of eagle`s syndrome. The next day 0.5cc of steroid(Kenacort) mixed with 0.5cc of local anesthesia was injected deep in to right tonsillar fossa near the styloid process. Patient was prescribed analgesic for five days. Follow up visits on alternate day

for two week showed excellent result with complete relief to the patient from pain. Six month follow up showed no signs of reoccurrence.

III. DISCUSSION

Eagle's syndrome comprises the symptoms of pharyngealforeign body sensation, recurrent throat pain,dysphagia, referred otalgia, and neck pain with an incidence of 4 to 8 per 10,000 people.[12-14] The misdiagnosedpatients with Eagle's syndrome may undergo unnecessary treatments; some patients mayeven undergo various surgical procedures, includingserial dental extractions, tuberosity reductions, alveoplasties,and temporomandibular arthroscopies.[15-20]Therefore, the extensive differential diagnosis of Eagle'ssyndrome should include any condition that mayresult in cervicofacial pain such as temporomandibularjoint diseases, trigeminal, sphenopalatine or glossopharyngealneuralgias, temporal arteritis, chronicpharyngotonsillitis, otitis media, external otitis, mastoiditis,dental pain, improperly fitting dental prostheses,submandibular sialadenitis or sialolithiasis, true pharyngeal foreign bodies, and tumors of the pharynxor tongue base. [16-20]. Our patients were previouslytreated with vague diagnosis,mostly neuralgias, their symptoms persisted despiteall therapies.

Eagle's syndrome should be suspected in the presenceof persistent throat pain that is triggered orexacerbated by head rotations, lingual movements,swallowing, or chewing. The pain in the throat maybe accompanied by hypersalivation, foreign body sensationon the affected side, and, rarely, bychange of voice lasting for a few minutes. Exacerbationof pain during the palpation of lateral tonsillarfossa should alert the clinician to a possible diagnosisof Eagle's syndrome. Local anesthetic block can beapplied to the tonsillar fossa to localize the site of thepain; relief of the pain may support the diagnosis andconfirm the need for treatment. [18]

Eagle had noticed that 4% of the population hadelongated styloid processes and only 4% of theseindividuals showed symptoms.[l3-16] Gossman and Tarsitano[21]reported the incidence of elongated styloid processas 1.4% by analyzing 4,200 panoramic radiographsof men between 18 and 22 years old and theupper limit was 25 mm. However, taking the radiographicabnormality into account, Keur et al[22] foundthe incidence to be 30% in their clinical and radiologicstudy on 1,135 patients.

The most useful radiographs that can demonstratethe styloid process are panoramic radiograph, posteroanteriorskull view, lateral cephalogram, lateraloblique mandible view, Towne's view, and openmouthodontoid view.[17] The computed tomographyand 3DCT scans are still being frequently used, besides theseradiographs. However, the mere presence of an elongated styloid process or mineralization of the stylohyoid complex radiographically in the presence of cervicopharyngeal pain does not automatically confirm a diagnosis of Eagle's syndrome. The reasons are three-fold. First, many patientswith an ossified stylohyoid complex are asymptomatic,like in our third case report our patient had bilateral elongated styloid process but was symptomatic only on the right side.Second, there does not appear to be any correlation between the severity of pain and the extent of ossification of the stylohyoid complex. Finally, the majority of symptomatic patients have had no recent history of tonsillectomy or any other cervicopharyngeal trauma. Despite this, the literature still categorizes patients into those with a pain pattern following the carotid artery distribution and those with a classical palpable mass in the tonsillar region.

The patients as outlined in our case reports had elongated styloid processesthat was at threshold level andthat made them more risky to be symptomatic. Eagle's syndrome can be treated conservatively orsurgically, or both. A more conservative approach should be attempted to decrease any muscle spasm and scar tissue around the styloid process and mineralized ligaments. However, failing this attempt, surgery remains a viable alternative.

Other treatments have concentrated on steroid injections into the affected tissues with varying results.[23,24]

Manual fracturing of styloid process throughtranspharyngeal manipulation does not usually relieve the symptoms and maycause possible damage to nearby neurovascular structures.Surgical resection has generally been accepted asthe primary treatment modality of Eagle's syndrome. Several transoral and extraoral-cervical approacheshave been described for the surgical managementof elongatedstyloidprocess.[25-28]Transoral resection of the styloid process is relativelyeasy to perform and avoid external scar aswell as extensive fascial dissection. Both operationand recovery times of this procedure are short and can be performed under local anesthesia. But the disadvantages of the transoral approach are the possibilityof a deep cervical infection, the poor visualizationof the surgical field, and the risk of neurovascularinjury, while attempting to leave the shortestresidue of styloid process.[28]

The external approach, on the other hand, provides adequate anatomic exposure of both the styloid process and nearby structures, prevents vascular injury and decreases the risk of bacterial contamination.The major disadvantage of the external approachis the postoperative cosmetic deformity due to scarformation. The other disadvantages are the necessity ofgeneral anesthesia, extensive fascial dissection andparesthesias of cutaneous nerves.[24-28]

We believe that conservative nonsurgical approach is a better alternative treatment of Eagle's syndrome. However, it does not imply that this approachis superior to others.

IV. CONCLUSION

In patients presenting with symptoms in the throat with associated headaches or facial pain, a thorough detailed case history and physical examination of the head and neck are mandatory. The differential diagnosis of neoplasm, unerupted molars, TMJ dysfunction, inner ear problems and neuralgias should be included with an elongated styloid process as sources of head and neck pain.The diagnosis of Eagle's syndrome is made with a history and finding of an elongated styloid process in the tonsillar fossa, of which palpation reproduces the symptomatology. Traditionally, treatment has been surgical excision of the styloid process. However, a more conservativeapproach may be undertaken to decrease any muscle spasm or decrease fibrosis around the styloid process. An awareness of pain syndromes related to the styloid process is important to all medical and dental practitioners involved in the diagnosis and treatment of head and neck pain.

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