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Eagle syndrome: A rare case of chronic cervical pain case report

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Abstract
Each temporal bone consists of four components, squamous, petromastoid and tympanic parts and styloid process. The styloid process is an elongated conical protuberance of the temporal bone that is located anterior to the mastoid process. A 38 year old man came to the outpatient unit complaining of discomfort and frequent pain in the neck region during swallowing, driving and playing outdoor games.Three-dimensional computed tomography reconstruction confirmed bilateral elongated styloid processes, measuring about 5.1 cm in length on the right side and 5.3 cm on the left side, from the level of the skull base to the hyoid, oriented adjacent to each other. A cervicofacial pain and foreign body sensation in pharynx connected with styloid process elongation is called Eagle syndrome. Treatment of Eagle syndrome is by both surgical and nonsurgical conservative approach.

Keywords: styloid process, eagle's syndrome, cervicofacial pain, computerised tomography scan, otalgia

Introduction
Each temporal bone consists of four components namely squamous, petromastoid, tympanic part and styloid process. The styloid process is an elongated conical protuberance of the temporal bone that is located anterior to the mastoid process and lateral to the tonsillar fossa. The stylohyoid complex is made of styloid process of the temporal bone, stylohyoid ligament, and lesser cornu of the hyoid bone. All are derived from Reichert's cartilage of second branchial arch. It runs between the internal and external carotid arteries. Further laterally, facial nerve, hypoglossal nerve, occipital artery and posterior belly of digastric muscles are found. Medially, lingual, facial, superficial, temporal, maxillary and internal carotid arteries, internal jugular vein and sphenomandibular ligament are located. The styloid process provides attachment to the styloid group of muscles [1, 2].

Case Report
A 38 year old man came to the outpatient unit complaining of discomfort and frequent pain in the neck region during swallowing, driving and playing outdoor games. He was referred to department of Ear, Nose and Throat (ENT) for clinical examination of the neck. Then he was directed to the department of radio diagnosis and imaging for diagnostic interpretation.
A detailed medical history and clinical examination of the head and neck were carried out for the patient. Physical examination included careful palpation of the bilateral tonsillar fossa, lateral pharyngeal wall and the area between mastoid apex and mandibular angle in an attempt to precipitate the patient's discomfort. Pain on swallowing was the most common complaint.
The other prominent symptoms were cervicofacial pain and otalgia. There was no history of neck injury. The patient underwent meticulous clinical ENT examination that incorporated bimanual transoral palpation of both tonsillar fossae, on which bony projections was felt. Pain was elicited during palpation on both sides.
Spiral CT scan showed the presence of bilateral elongated styloid processes (Figure-1). Three-dimensional computed tomography reconstruction also confirmed bilateral elongated styloid processes, measuring about 5.1 cm in length on the right side and 5.3 cm on the left side, from the level of the skull base to the hyoid, oriented adjacent to each other (Figure-2).

Discussion
In this case, we present the diagnostic 2D CT scan and 3D scan radiography findings with clinical evidence of bilaterally elongated styloid process. This is a rare finding that frequently goes undetected in the absence of radiographic studies. It should be noted that the normal length of the styloid in an adult is approximately 2.5 cm whereas an elongated styloid process is generally more than 2.5 cm in length [1, 3, 4].
Both radiographic diagnosis and clinical examination confirmed the elongated styloid process. Palpation of the styloid process in the tonsillar fossa is investigative of elongated styloid process. Styloid process with normal length is usually not palpable. Occasionally, palpation of the tip of the styloid will exacerbate existing symptoms [4-6].
As it was highly suspicious for Eagle syndrome on clinical examination, confirmation has been made by radiographic study. Eagle syndrome can occur unilaterally or bilaterally and most frequently results in symptoms of dysphagia, headache, pain during neck movements, pain on protrusion of the tongue, change in voice, and a sensation of hyper salivation. A cervico facial pain and foreign body sensation in pharynx connected with styloid process elongation is called Eagle syndrome [2,7-9].
In 1937, Eagle defined "stylalgia" as an autonomous entity associated with abnormal length of the styloid process or due

to mineralization of the stylohyoid complex. In about 4% of general population an elongated styloid process occurs, while only about 4%of these patients are symptomatic and the real incidence is 0.16% with a female predominance of 3:1.The incidence of Eagle's syndrome varies among population. By and large asymptomatic, it occurs in adult patients [8]. In spite of the valuable information about the anatomy, there are some difficulties in reading the plain radiographs secondary to superimposed anatomical structures. Therefore three-dimensional computed tomography (CT) reconstructions can accurately depict the lengthand angulations of the styloid process [10, 11].

3D-CT is the diagnostic tool preferred to delineate the anatomy, modify thesurgical plan, and offer a detailed explanation to the patients.It is a precious, superior diagnostic imaging tool in patients with Eagle's syndrome. It helps clinicians to evaluate the styloid process in spatial geometry, construct accurate length measurements, and explain the problem in detail to patients [6, 7, 10]

Several theories have been projected to explain the symptoms associated with elongation of the styloid process:

1) Congenital elongation or ossification of the styloid process due to persistence of cartilaginous stylohyal complex.
2) Calcification of the stylohyoid ligament by an anonymous process.
3) Pain is caused by compression of the glossopharyngeal nerve as it passes a styloid process elongated because of anatomical variability.

The first two, the theories of reactive hyperplasia and reactive metaplasia are usually in response to injury. This phenomenon may explain the occurrence of Eagle syndrome in patients after tonsillectomy, as it was originally described by Eagle, [8, 9]

Treatment of Eagle syndrome is both surgical and nonsurgical. That is by conservative approach. Nonsurgical treatments include reassurance, transpharyngeal nonsteroidal anti-inflammatory medications and steroid injections in the tonsillar fossa region, application of heat have been advocated. [10]

Surgical treatment is by one of two methods. Otolaryngologist W. Eagle preferentially used a transpharyngeal approach through which the elongated portion of the styloid process are removed. As there is less external scarring, it has been criticized due to its increased risk of deep space neck infection and poor visualization of the surgical field. Resection of the elongated styloid process is the treatment of choice. Theoretically, the elongated portion can be removed by an extraoral approach. The most significant advantage of an external approach is enhanced exposure of the styloid process and the adjacent structures. It also facilitates the resection of a partially ossified stylohyoid ligament. It should be noted that blind fracture of the styloid process does not usually relieve symptoms and risks damage to nearby neurovascular structures like cranial nerves V, VII, IX, X, carotid arteries etc. Pathophysiologically, the Eagle syndrome is related to an irritation of the surrounding cranial nerves, carotid arteries and or the pharyngeal mucosa [10, 11]

Although both procedures are effective in removing an elongated styloid process, the extraoral approach is thought to be superior for better recovery. However, the main disadvantage is lack of access, due to massive hemorrhage and subsequent spread of deep neck infections.

Fig 1: Two-dimensional computed tomography scan (CT scan) of styloid process. The lengths of the styloid processes are 5.1 cm on right side and 5.3cm on the left side.

Fig 2: Preoperative 3D CT scans of the skull base showing the elongated styloid process on the right and left side from the level of the skull base to the hyoid, oriented adjacent to each other for assessment

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