Eagle's Syndrome: An Unusual Cause of Recurrent Retro mandibular Pain Case Report

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Abstract:

Eagle’s syndrome occurs when an elongated styloid process or calcified stylohyoid ligament causes recurrent throat pain radiating to the ipsilateral ear or retromandibular neck pain. Such symptoms easily confused with those caused by different facial neuralgias. Diagnosis usually made on physical examination by palpation of the styloid process on the tonsillar fossa. Computed tomography (CT) scan provides complementary information to that of plain radiography, including definition and relationship of the elongated styloid process to the surrounding soft tissue structures in the axial plane. Treatment is usually surgical with resection of the elongated part of the styloid process through trans-oral or external approach. We reported a case of 38-year-old male who presented to otolaryngology clinic with physical examination and radiological findings typical for Eagle's syndrome. Operation was done by consultant otolaryngologist who resected the elongated part successfully via trans-oral approach without complications. The patient was completely asymptomatic at follow up visit six months postoperatively. Awareness of Eagle’s syndrome is important to all health practitioners involved in the diagnosis and management of neck and head pain because it can be confused with many other conditions that must be excluded.

Keywords: Eagle's syndrome; Retromandibular ,Neck pain; Elongated styloid process.
Introduction:

Eagle’s syndrome (ES) is characterized by craniofacial or cervical pain due to an elongated styloid process or calcified stylohyoid ligament. Patients with Eagle’s syndrome may present with a sore throat, ear pain, or even with foreign body sensation in the pharynx secondary to pharyngeal and cervical nerve interactions. Watt.W. Eagle American otolryngologist was first who described the clinical findings of this syndrome in 1937 (1).

Since the symptoms are variable and non-specific, patients seek treatment in several different clinics such as otorhinolaryngology, family practice, neurology, neurosurgery, psychiatry, and dentistry (2).

The normal length of the styloid process may vary, but with the majority of the population it is 20-30 mm long (3,4). However, a 30 mm or longer process is considered anomalous and responsible for the so-called Eagle syndrome. ES is a rare entity which is not commonly suspected in clinical practice (5). Approximately 4% of the population is thought to have an elongated styloid process, only a small percentage (between 4% and 10.3%) of this group is thought to actually be symptomatic (6,7).

A diagnosis can usually be made by physical palpation of the styloid process in the tonsillar fossa. In addition, orthopantomography (OPG) or a cervical radiograph using a lateral projection, and computed tomography (axial and three-dimensional CT) are complementary imaging modalities to confirm the diagnosis (8).

Eagle syndrome can be treated both surgically and non-surgically. The surgical approach is styloidectomy when the resection of the elongated part of the styloid process is
performed through trans-oral or extra-oral approach. The trans-oral approach was introduced first by Watt. W. Eagle, while the extra-oral approach, described by Loeser and Caldwell (9). The most significant advantage of an external approach is enhanced exposure of the styloid process and the adjacent structures, and this outweighs all other considerations. It also facilitates the resection of a partially ossified stylohyoid ligament. Transoral resection causes no outside scars, but involves the risk of deep neck space infection (DNSI) and possible neurovascular injury (10-11). Conservative treatment options have included transpharyngeal injection of steroids and lignocaine preparation close to the styloid process(7). This option proved to be not efficient enough since symptoms recurs in most of the patients after 6-12 months(12).

Case Report:

A 38-year-old male with prior history of tonsillectomy two years before his recent presentation to Babakr charitable hospital Otolaryngology clinic, with complaints of persisting right side nagging retromandibular pain, which occasionally radiating to his right ear. He underwent outside tonsillectomy two years back for chronic hypertrophied tonsils with transient relief of his symptoms.

On transoral physical examination he had a palpable, mildly tender bony projection along the course of the styloid process on the right tonsillar fossa; pain was elicited during palpation. The examination of the contralateral tonsillar fossa was normal, supporting the diagnosis of Eagle syndrome.

Lateral cervical radiograph revealed elongation of the bony shadow of the styloid process on the right side. Further confirmatory neck CT scan without intravenous contrast media was obtained and showed elongated right styloid process seen as continuous visualization of the styloid process in the consecutive neck axial CT images up to the level of the oropharynx.
(Figure 1). Further 3D reconstructed coronal image showed the elongated styloid process with an approximate length of 40 mm (Figure 2).

Based on the clinical examination and radiological findings, surgical resection of the elongated styloid process was done by consultant ENT laryngologist under general anesthesia via trans-oral approach who resected the elongated part successfully without complications (Figure 3, 4).

Figure 1: CT scan of the neck showing unilateral elongated styloid process on the right side. (arrow).

Figure 2: 3D CT scan, showing elongated styloid process on the right side.

Figure 3: Showing the site of palpable bony projection on the right tonsillar fossa (arrow).

Figure 4: Intra operative (transoral approach) showing the elongated styloid process (arrow).
The length of the resected part of the styloid process was approximately 12mm (Figure 4).

Figure 5: Surgically excised segment of the styloid process

The patient was prescribed oral 250 mg metronidazole for five days and 4.5 g parenteral cefuroxime was administered for a period of 72 hours, followed by 500 mg cefuroxime oral therapy for two days to prevent the DNSI. The patient had eventless, smooth post operative period and discharged home third day after operation. The operation resulted in complete disappearance of the patient complaints.
Discussion:

Although the incidence of the styloid process elongation or mineralization of the stylohyoid complex is not uncommon, only a small percentage 4% of this group is symptomatic. The vagueness of symptoms and the infrequent clinical observation are often misleading. These patients may be seen by a surgeon, a dentist, a neurologist, and a psychiatrist, often receiving a variety of treatments that do not relieve the symptoms and that cloud the clinical picture\(^6,\!7\). 

Stylalgia is misdiagnosed or overlooked as a possible diagnosis in cases of vague cervicofacial pain. A variety of head and neck conditions should however be considered in the differential diagnosis of ES, these include temporomandibular joint disorders, glosso-pharyngeal neuralgia, trigeminal neuralgia, migraine type of headaches, sphenopalatine neuralgia, cervical arthritis, carotidynia, temporal arteritis, otitis media externa, salivary gland disease and possible tumours. Other pathology should be eliminated by a careful medical history, clinical and radiographic examination\(^13\).

Eagle considered tonsillectomy responsible for the formation of scar tissue around the styloid apex, with consequent compression or stretching of the vascular and nervous structures contained the retrostyloid compartment (in particular glossopharyngeal nerve and perivascular carotid sympathetic fibers)\(^14\).

Diagnosis can be usually made by physical palpation of the styloid process in the tonsillar fossa. In addition OPG or a cranial radiograph using a lateral projection can give a rough idea about the diagnosis. While CT scan confirms the diagnosis and provides precious information for the surgeon showing a more detailed view and measuring the precise styloid process length using three dimensional (3D) reconstruction images.\(^8\).
Eagle's syndrome can be treated surgically and nonsurgically. Surgical treatment has two approaches transoral and external (transcervical) approach. Advantages of the transoral approach are safety, simpleness, shorter time, and no external scar, even though such approach has been criticized by some authors in view of the possibility of DNSI infection of deep neck spaces, the risk of injury to major vessels, nerves and surrounding structures due to the poor visualization of deep planes. While the disadvantages of the external approach are the external scar, wound infection, the longer surgical time and the risk of injury to the terminal branches of facial nerve (10).

**Conclusion:**

Awareness of Eagle syndrome is important to all health practitioners involved in the diagnosis and management of neck and head pain. It can be confused with many other conditions that must be excluded. Diagnosis of ES can be made by physical examination and radiological investigations. Surgical resection of the elongated styloid process is the treatment of choice.
References:

الملخص:
تحدث متلازمة إيجل بسبب استطالة النتوء الإبري العظمي أو تكسرب الرابط اللامي في العنق، مسببة أوجاعًا متكررة في الحلق والأذن أو الشعور بوجود جسم غريب، مع صعوبة البلع. بالإضافة إلى أوجاع في الوجه تتجه أحيانا نحو الأذن أو خلف الحنكة.
على سبيل المثال هذه الأعراض من السهل أن تختلط مع أمراض أخرى تسبب نفس الأعراض والشكوى.
تشخيص هذا المرض عادة ما يتم عبر الفحص السريري بجسم النتوء العظمي داخل حفرة اللوزتين في الحلق. الفحص الشعاعي للركبة عادة ما يظهر النتوء الإبري الطويل ولكن الأشعة المقطعية للركبة تبين تفاصيل تشريحية دقيقة عن طول النتوء العظمي ومجاوراته، ما يساعد الجراح عند إجراء العملية.
علاج المرضى عادة ما يتم جراحياً وذلك باستئصال الجزء الباز من النتوء عن طريق الفم أو عن طريق الجراحة الخارجية. نستعرض هذه التقارير حالة مماثلة لمريض عمره 80 سنة تعرض لأستئصال اللوزتين قبل سنتين من وصوله إلى العيادة الاستشارية لل yatشفى بابكر الخيري بحضوره في مركز ACSIM يميني من الرقبة تتجه أحيانا نحو الأذن اليمنى وتزداد عند تحريكه للركبة إلى الجهة اليسرى.
تبين بالفحص السريري والإشعاعي أن لديه متلازمة إيجل. أجريت للمريض عملية استئصال للجزء المزمن من العظم وأسفرت العملية عن اختفاء الأعراض التي يعاني منها المريض نهائيا.
الوعي بمتلازمة إيجل مهم لكل الأطباء المعنيين بعلاج وتشخيص أمراض وأوجاع الرقبة والحلق نظراً لإمكانية احتلاطها مع جملة من الأمراض التي تسبب نفس الشكوى للمريض.