



Bibliographical Review on the Calcification of the Stylohyoid Ligament, Eagle Syndrome: Contribution of a Clinical Case

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Abstract

The Eagle Syndrome or Stylohyoid Syndrome manifests an association of occlusive, neuropathic and/or vascular symptoms caused by a pathological elongation of the stylohyoid process. Its diagnosis, based on radiographic techniques or palpation of the area, is usually complex due to the multiple syndromes that present with the same or similar symptoms. The differential diagnosis of Eagle Syndrome should include all conditions that present cervicofacial pain. The objective of this study is to approach the origin of the disease and discuss the radiological evidences and clinical signs and symptoms in this syndrome; as well as the review of a clinical case.

Keywords: Calcification; Stylohyoid Ligament; Eagle Syndrome

Introduction

The Eagle Syndrome is characterized by excessive calcification of the styloid process of the temporal bone, increasing its length and exceeding the stipulated average between 25 and 30 millimeters. On the other hand the calcification can be total, affecting the stylohyoid ligament belonging to the hyoid apparatus, creating a small chain of jointed bones. These anatomical modifications give rise to a series of symptoms that we will describe later, which may or may not generate pain. If pain is manifested, it is usually preceded by a tonsillectomy. Diagnostic techniques for this type of pathology is orthopantomography, lateral teleradiography and, of course, a computerized axial tomography. Lateral palpation can also be performed [1-5].

First described in 1652 by an Italian surgeon named Pietro Marchetti, it was definitely schematized by Watt Eagle in the late 1940s or early 1950s [6]. We also found documented cases that manifest this pathology dating from 1872, date in which Weinlecher reported cases with symptoms of ossification of the stylohyoid ligament. In 1896 Sterling presented cases of elongated styloid processes [7-10]. Eagle in 1937 reported 2 cases with pharyngeal and facial symptoms due to irritation of the carotids at the bifurcation of the common carotid [2-4,11,12].

The sore throat is one of the most difficult to diagnose, this occurs when the styloid process (long and cylindrical projection that has a normal length between 20-30 Mm) exceeds this length causing or not symptom [5-7].

The stylohyoid ligament is a component of the hyoid apparatus, which in certain cases, calcifies creating a chain of ossicles with their respective joints, with one end of the styloid process being the temporal and the other the lower horn of the hyoid bone. The origin of the same is in the embryonic stage as referred by Escolan [13]. This device is formed from the 2nd branchial arch, also called hyoid arch or cartilage of Reichert, and a remaining part by the 3rd branchial arch [13].

2nd arch origin: -cartilage of Reichert-Asta minor of the hyoid - stylehioideo ligament -Asshode styloid temporal -Stirrup

3rd arch origin: - Body of the hyoid -The greater the hyoid

Due to a process of calcification in the 2nd arch, the Reichert cartilage is formed, which has at one end the hyoid bone and at the other end the otic capsule of the temporal bone. After 2 months of gestation, it is reabsorbed, leaving the styloid process of the temporal bone in the upper part and the lower horn of the hyoid bone in the lower part [2,13].

The styloid process after its formation is related to surrounding structures, including 3 muscles that provide insertion in the apophysis (stylohyoid, stylopharyngeal and styloglossus), and 2 ligaments (stylohyoid and stylomandibular) [2,10,13]. All together they form the called "Arc of Riolo". The insertion of the stylohyoid ligament occurs in the posterolateral aspect of the styloid process, moving downwards and inwards, crossing or not the tendinous buttonhole of the middle tendon of the digastric muscle, ending its course in the lower horn of the hyoid bone.

The contribution of Marchety 1652, was cited by Ollivier, who in 1923 described the hyoid apparatus in a more detailed way, describing that generally three bones form it:

- a) Styloid or stylial processes.
- b) Stylohyoid (calcified) ligament called ceratohial (in some cases it is double).
- c) Minor horn of the hyoid that receives the name of apóyalo hipohial, soldier to the body of the hyoid (basihial).

Ollivier considers 3 variants:

1. 64% is formed by 3 ossicles (stylial, ceratohial and apohial or hypohial).
2. 12% in this case 4 bony elements are isolated, due to the unfolding of the ceratohial in 2 ossicles, accessory ceratohial.
3. 24% formed only by 2 ossicles due to the fusion of the 2 bone pieces that form the previous cases.

Among the structures adjacent to the styloid process are vessels such as the internal carotid artery, internal jugular vein and nerves such as glossopharyngeal, hypoglossal and pneumogastric (vagus). Laterally we found the external carotid and emerging through the stylomastoid foramen and posterior tearing (jugular) the facial and glossopharyngeal nerves respectively.

As we have described at the beginning of this article, the clinic of the styloid syndrome is very varied, thus making it difficult to diagnose and therefore treat it. In any case, the symptoms that patients may present include headache, otalgias, myalgias, temporomandibular arthralgias, facial pain, pharyngeal pain, carotidynia, dysphagia, dysphonia and can complete the picture with cervical affection, glossopharyngeal neuralgia, migraines and also dental inclusions. third molars [5,7,9-12].

The characteristic pain is due to irritation of contiguous structures, pharyngeal mucosal irritation compromising the trigeminal, glossopharyngeal, pneumogastric nerve.

With regard to involvement of the carotids, it is called a stylo-carotid syndrome, affecting the external and internal symptoms, which causes Horner syndrome, and pain due to irritation in the pericarotid sympathetic sheath [9,12].

Eagle classified the possible manifestations in two: Stylohyoid Syndrome and Stylecarotid Syndrome [15]. Due to the calcification of the elongated styloid process, the latter may be laterally or medially deviated, affecting the carotid vessels by compression, thus termed the carotid syndrome. The symptoms of this syndrome are dull, throbbing, throbbing pain in the lateral region of the neck,

tinnitus, dizziness, headaches, lipothymias and neuralgia due to carotid pressure [11,12]. The involvement of the external carotid artery presents with symptoms such as facial pain distributed in the genital region and below the ocular region [15]. As regards the internal carotid causes cranial pain affecting the ophthalmic artery causing little or no facial pain and below the ocular region.

In the second case, the so-called stylohyoid syndrome, the symptoms are dysphagia, odyphagia, ipsilateral otalgia, foreign body sensation in the throat, altered sense of taste and vertigo.

As previously mentioned, being the Eagle syndrome complex to diagnose, the number of cases in which it is located is quite underestimated in the general population, since in addition the symptoms are varied and quite nonspecific, and patients go to very different specialists. All professionals who are responsible for the area should include this syndrome in the differential diagnosis of atypical pain in the head and neck [16].

The differential diagnosis must be made taking into account the following syndromes:

- Ernest's syndrome (inflammation of the stylomandibular ligament) caused by a macrotrauma, pain extends the temporal region, choroid, eye, ear, back, body of the jaw, which reduces the range of mandibular rotation [12].
- The pseudo-syloid syndrome proposed by CARMADA, given that the stylohyoid ligament hardened, and therefore causes symptoms of stylage.
- We must also take into account TMJ diseases, trigeminal neuralgia, sphenopalatine or glossopharyngeal neuralgia, temporal arteritis, otitis media, otitis externa, mastoiditis, dental pain, submandibular sialadenitis or sialolithiasis, real foreign bodies in the pharyngeal region and tumors of the pharynx or language base [5].

Clinical Case

A 61-year-old man who attends the Alfonso X el Sabio University Clinic at the Department of Oral Surgery, with the intention of performing a restorative treatment based on implants (Figure 1).



Figure 1: Ortopantomography of the patient showing the articulation of the stylohyoid ligament and the styloid process with ossification of the ligament.

In your medical record there is no evidence of any symptoms, only that it is under medical care due to depression, which is not relevant to our investigation.

In the routine exploration we appreciate teeth mobility of the pieces, does not present lymphadenopathies to the palpation and presents mandibular reabsorption.

The main characteristic of this patient in regard to the study is the presence of a chain of jointed bones.

The patient does not present discomfort to the rotation of the head, nor dysphagia, nor otalgia, nor odynophagia, pain when swallowing, foreign body sensation, headaches, dizziness, which are very common symptoms when we have an elongated styloid process.

According to the classification of Ollivier mentioned at the beginning, 64% of cases with this pathology is formed by 3 bones, 24% by two bones and 12% is that in which we take into account 4 bones, due to the stylohyoid ligament called ceratohial is divided into 2; ceratohial, accessory ceratohial.

It is one of the less common variants in which we can observe 3 joints and 4 bones. The presence of this ossification of the stylohyoid ligament can be adjusted to one of the theories proposed by Steiman, The theory of Reactive Hyperplasia. This involves traumas, which condition some section of the stylohyoid ligament to transformation by metaplastic changes leading to intermittent ossification [3].

Technique

The techniques to treat the elongation of the stylohyoid process when presenting symptoms are summarized in; a surgical treatment, which would be intraoral or extraoral resection, which is generally only contemplated if non-invasive therapies have failed or will certainly do so [15], or there are also non-surgical alternatives such as the transferase infiltration of lidocaine or steroids and the manual transpharyngeal fracture of the stylohyoid process [1]. In case of the patient presenting mild symptoms, analgesic treatment can be resorted to.

In the diagnosis of the patient presenting symptoms such as dysphagia, pain when swallowing, foreign body sensation, a palpation is made in the tonsillar fossa. If the styloid process is prominent, exacerbation of symptoms usually precedes this palpation. Among the procedures that are used to diagnose we find steroids, carbamazepines (controversy between authors) and as a choice lidocaine 2%; after which application a remission of symptoms is observed, confirming the diagnosis of the styloid process.

The surgical techniques, the intraoral is performed through an incision in the tonsillar fossa, to reach the mucosa lining the process estilohiideo which is detached from its origin to the muscles and ligaments which form the "posy Riolo" a free time This ligament muscle complex is followed by the removal of the temporal styloid process, after which it is covered with mucosa and sutured with reabsorbable polyglycolic acid or polylactic acid thread.

The advantage of intraoral treatment is: its reliability, since it requires less operative time and avoids scarring. The disadvantages are: bacterial infection, poor visualization and the risk of damaging important vessels such as the carotid and cranial nerves: V, VII and XI.

In the extraoral technique it offers the advantage of a better exposure of the carotid region and as a disadvantage the affection of the facial nerve in addition to leaving a scar [4,5,8].

Other techniques of reception of the styloid process is the fracture of this, but this technique is obsolete.

Discussion

In most of the articles reviewed, it can be observed that all patients present cervical pharyngeal symptoms, very few cases of asymptomatic styloid syndrome.

The main problem of this syndrome is that it still does not have an exact definition, nor why the calcification of said ligament occurs (being able to associate with the reactive metaplastic theory of Steiman, as far as our clinical case is concerned). According to authors like Harman, they think that it can be hereditary [3].

Regarding the average age, there are several opinions, but they agree that the age range is between 30 - 50 years. It has greater predominance in the female sex, as data to review in the mentioned articles. Another important fact is the prevalence of symptoms on the right side, regardless of whether it is unilateral or bilateral.

The study of the calcification of the stylohyoid ligament, styloid process, stylecarotid process found that several authors agree that in the treatment of mild syndromes, it can be treated with analgesia; in severe cases, transoral surgery is used, which has better acceptance for the patient and the professional, since it requires less work time and does not leave a scar.

There is controversy in respect to whether Eagle syndrome and styloid, as they do not always have symptoms despite having a calcification of the ligament itself.

The etiology of Eagle syndrome is due to a previous tonsillectomy or cervical trauma, however, the etiology of the asymptomatic elongated styloid process is not linked to something concrete. Some authors say that the origin can occur in the stage of growth after birth due to small stimuli that condition the calcification of the total or partial ligament, considered within a radiographic classification of Langlais, 1986 that divides it into 3 types: type I (elongated), type II (pseudoarticulate), type III (segmented) [11].

The question to ask is the exact reason for the calcification of this process as well as the own styloid ligament. Among the theories presented was the reactive metaplasia, which can be said to concern the proposed case, but on the other hand we have to mention reactive hyperplasia (where the stylohyoid process is stimulated based on traumas causing an ossification in the temporal portion to costs of the styloid ligament) and the theory of anatomical variation, covers the fact that stylohyoid ligament and the styloid process as ossified structure, is developed already in the first year after, the birth is innate form [17].

Other authors continue to advocate an unknown pathogenesis, in which a traumatic origin is usually admitted, although more recent embryological data suggest a possible genetic origin with individual variations [18]. In any case the pathogenesis is still under discussion, and can finally be classified in a simple way as idiopathic, congenital (due to the persistence of cartilaginous elements precursors of the stylohyoid process) or acquired (due to the proliferation of bone tissue in the insertion of the stylohyoid ligament) [19].

Conclusion

We can summarize that both the lengthening syndrome of the styloid process and the calcification of the stylohyoid ligament are rare, its frequency in the population in general is low, and also depending on its degree, it can be asymptomatic or present with pathology, in which case its diagnosis is complicated to establish. Therefore we should include it as a possibility among the diseases or syndromes that present pain in the head or neck, and take it into account when reviewing the radiographic tests that include that area.

We also conclude that once diagnosed, it should proceed to its surgical approach or not depending on the degree of affectation or severity of the clinic that it generates, and in any case, it will always be the patient's decision to undergo surgery or not.

Bibliography

- Buono U and Michele Mangone G. "Surgical approach to the Stylohyoid. Process in Eagle's Syndrome". *American Association of Oral and Maxillofacial Surgeons* 63.5 (2005): 714-716.
- Diamond L and Cottrell D. "Eagle's Syndrome: A Report of 4 patients treated using a modified extraoral approach". *American Association of Oral and Maxillofacial Surgeons* 59.12 (2001): 1420-1426.
- Ladeira Pereira F and Iwaki Filho L. "Styloid- Stylohyoid Syndrome: Literature Review and Case Report". *American Association of Oral and Maxillofacial Surgeons* 65.7 (2007): 1346-1353.
- Chandra Prasad K and Panduranga Kamath M. "Elongated Styloid Process (Eagle's Syndrome): A clinical Study". *American Association of Oral and Maxillofacial Surgeons* 60.2 (2002): 171-175.
- Beder E and Bagis Ozgursoy O. "Current Diagnosis and Transoral Surgical. Treatment of Eagle's Syndrome". *American Association of Oral and Maxillofacial Surgeons* 63.12 (2005): 1742-1745.
- Cavalcante Gomes de Souza Carvalho A., et al. "Intraoral approach for surgical treatment of Eagle syndrome". *British Journal of Oral Maxillofacial Surgery* 47.3 (2009): 153-154.
- Blythe JN., et al. "Eagle's syndrome alter fracture of the elongated styloid process". *British Journal of Oral Maxillofacial Surgery* 47.3 (2009): 233-235.
- Quereshy F and Gold E. "Eagle's Syndrome in an 11 years old patient". *American Association of Oral and Maxillofacial Surgeons* 59.1 (2001): 94-97.
- Sada E and Mampaso C. "Síndrome de la apófisis estiloides: calcificación del ligamento estilohioideo". *Revista española odontoestomatológica de implantes* 8.4 (2000): 234-236.
- Lacet de Lima J and Ferreira Rocha J. "Síndrome de Eagle: revisión de la literatura". *Acta Odontológica Venezolana* 45.2 (2007).
- Hernández Marcos G and Olay S. "Síndrome de Eagle o de la apófisis estiloides. A propósito de 3 casos". *Revista Europea de Odontoestomatología* 13.5 (2001): 267-271.
- Castillo CE and Mantilla JC. "Síndrome de Eagle: Tomografía del proceso estiloideo elongado". *Acta de otorrinolaringología and Cirugía de cabeza y cuello* 31.3 (2003): 89- 96.
- Tisner Nieto JV and Abad Royo JN. "Calcificación del ligamento estiloideo: estalalgia de Aubien y síndrome de Eagle: aportación de 5 casos clínicos". *ORL Aragon* 6.2 (2003): 5-12.
- Aprile H., et al. "Anatomía odontológica orocervicofacial". "El Ateneo" Quinta edición (1972): 794.
- Elmas F and Shrestha BL. "Eagle's Syndrome". *New England Journal of Medicine* 377.13 (2017): e18.
- Aldakkan A., et al. "Vascular Eagle's Syndrome: Two Cases Illustrating Distinct Mechanisms of Cerebral Ischemia". *Journal of Radiology Case Report* 11.8 (2017): 1-7.
- Badhey A., et al. "Eagle syndrome: A comprehensive review". *Clinical Neurology Neurosurgery* 159 (2017): 34-38.
- Pokharel M., et al. "Clinicoradiologic evaluation of Eagle's syndrome and its management". *Kathmandu University Medicine Journal (KUMJ)* 11.44 (2013): 305-309.
- Costantinides F., et al. "Eagle's syndrome: signs and symptoms". *Cranio* 31.1 (2013): 56-60.

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