

Case Report

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Eagle's Syndrome: Transoral Styloidectomy a Technique Modification Case Report

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Introduction

Eagle's Syndrome is a rare condition with unknown etiology that mainly affects female patients between the third and sixth decade of life. It was first described in 1937 by Dr. Watt W. Eagle, in a study carried out in a group of patients whose main symptom was cervicopharyngeal pain caused by elongation of the styloid process and/or calcification of the stylohyoid ligament [1]. The diagnosis of this pathology is based on the anamnesis and physical examination together with imaging exams of the patients. Cervicofacial pain, palpation of the styloid process in the tonsillar fossa and limitation in neck mobility are the most classic signs and symptoms of this disease [2]. Due to the nonspecific symptoms present in these patients, this disease is usually underdiagnosed and confused with temporomandibular disorders, cervical myalgias, and even being diagnosed as atypical trigeminal neuralgias [3]. For this reason, clinical examination and imaging exams, are indispensable for the correct diagnosis and evaluation of anatomical structures [3].

We present a case of a woman with a history of eagle syndrome which was diagnosed and treated at the Hospital clinico metropolitano El Carmen Santiago, Chile.

Clinical Case

A 63 year old woman with history of dyslipidemia, hypertension and hypothyroidism in treatment attends to the Odontology service at Hospital Clínico Metropolitano El Carmen, Santiago de Chile, with pain in the cervical and tonsillar area referred by Otorhinolaryngology service, who suspected dental origin in relation to the pain described by the patient. During the clinical interview, the patient refers a mild pain in the cervical and tonsillar region, since 10 years. She was treated with NSAID'S on multiple occasions without success. She consulted the Otorhinolaryngology service at the Hospital Clínico Metropolitano El Carmen, Santiago de Chile. After ruling out tonsil and glandular pathology, a referral was made

to the odontology service, to rule out dental origins of the pain. On clinical examination, the patient presented cervical hypomobility in lateralities and flexion, cervical and stabbing pain in the left tonsillar fossa, accompanied by dysphagia and foreign body sensation. During intraoral evaluation, a hard painful tumor in the left tonsillar fossa was evident, which increases in ipsilateral cervical movements.

An Orthopantomography was requested where both elongated styloid processes can be assessed, corroborating the diagnosis of eagle syndrome (Figure 1). Conservative treatment was carried out in first instance during two months, which consisted in the use of NSAID'S and kinesiotherapy. Due to no improvement, it was decided to perform a surgical resection of the styloid process using an intraoral approach. Using a cone beam computed tomography (CBCT) for surgical planning, the following anatomical parameters were considered: (a) anterior border of the mandibular ramus, (b) Posterior border of the mandibular ramus, (c) distopalatine cuspid of the upper second molar, differentiating from the classic approach described by Kent in 2014.



Figure 1: Orthopantomography, elongated styloid processes. 34mm (Right styloid processes and 40mm Left styloid processes

Technique Description

Before resection of the styloid process, a surgical planning of the case was necessary, utilizing a maxillofacial Cone Beam (CBCT) in the axial, sagittal and coronal axis (Figure 3A). The distance that were taken into account were: (a) the tip of the styloid process to the anterior edge of the mandibular ramus, (b) the tip of the styloid process to the posterior edge of the mandibular ramus and finally the tip of the styloid process to the distopalatine cuspid of the left upper second molar (Figure 2). This way we modified the anatomical reference parameters proposed by Kent in 2014 for the conventional intraoral approach through the tonsil, also modifying the incision side from the tonsil to the oral mucosa [4]. The surgical procedure was performed under general anesthesia, utilizing a Kilner-Doughty mouth separator to obtain better view of the surgical field. A 2 cm vertical incision was made in the oral mucosa parallel to the mandibular ramus, at the level of the ipsilateral upper second molar and anterior to the palatoglossal arch, in the pterygomandibular raphe (Figure 3A). Then, through a blunt debridement with Metzembaum dissection scissors and Kelly forceps, the caudal portion of the styloid process is accessed and exposed (Figure 3B). Finally we perform the osteotomy and exeresis of the styloid process (Figure 3C) and synthesis of the approach using Polylactinic Acid 4.0.

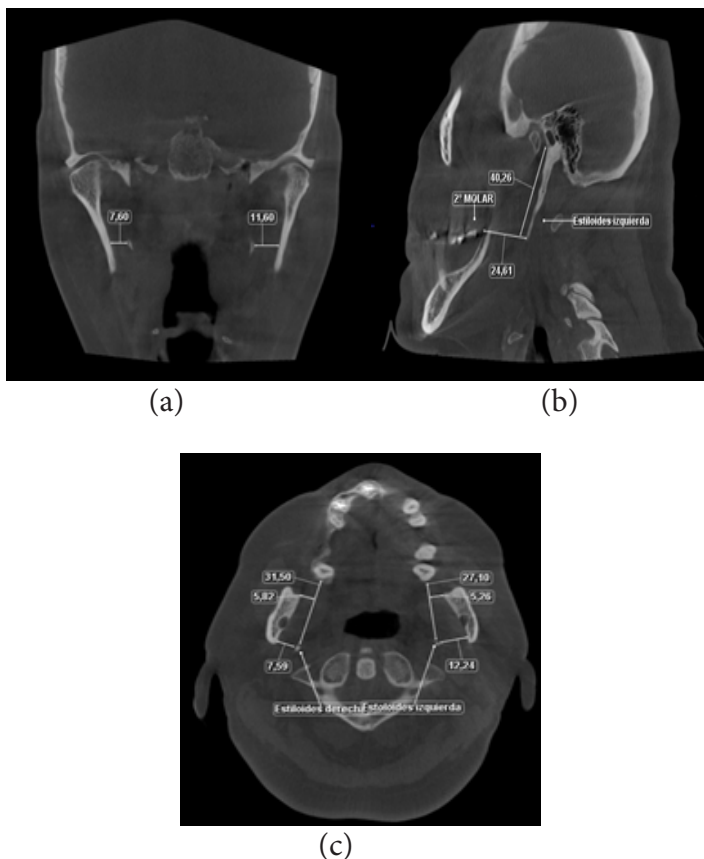


Figure 2: Anatomical parameters and surgical: (A) Coronal axis (B) Sagittal axis (C) Axial

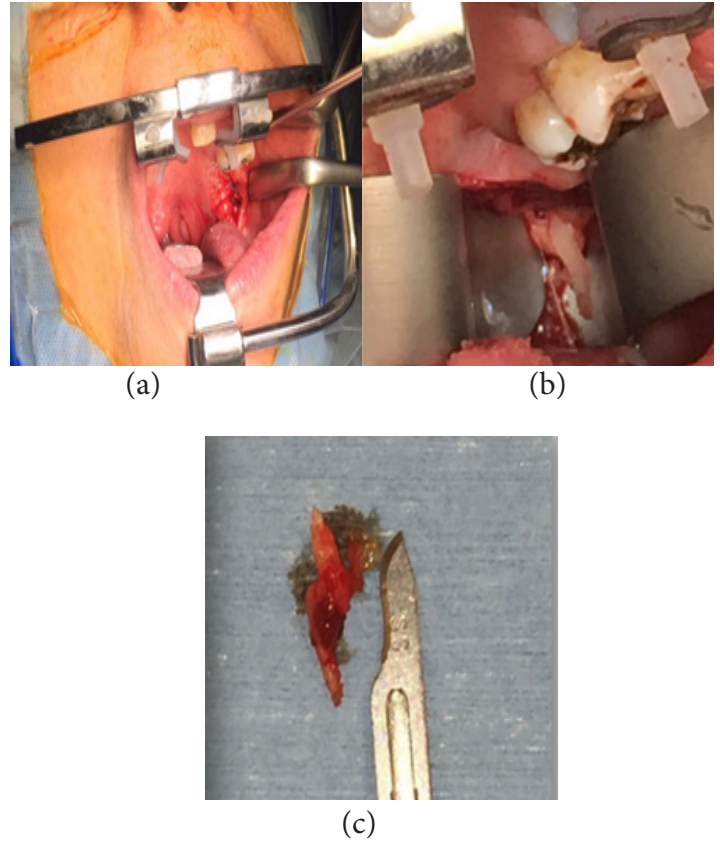


Figure 3: (A) Surgical Field and incision (B) Styloid process exposure (C) Exeresis of the anterior portion of the styloid process

Discussion

Eagle's Syndrome corresponds to a rare entity with unknown etiology. Epidemiologically, its incidence varies between 4 to 28% of the population, mainly affecting women between the third and fourth decade. This syndrome is characterized by an elongation of the styloid process greater than 30 mm or ossification of the stylohyoid ligament. It is associated with diffuse painful symptoms in the cervical and / or tonsillar region, together with a foreign body sensation in the tonsillar region and limitation in cervical movements [1,2]. Eagle in 1937, described two variants of this syndrome; the classic Eagle's syndrome and the carotid artery syndrome. The classic eagle's syndrome is the most common entity, associated in patients after undergoing a tonsillectomy. This surgical injure could cause an inflammatory response that would be the responsible of a hyperplasia of the styloid process and / or calcification of the stylohyoid ligament [1]. Although the hypothesis stated by Eagle has been the subject of studies and preparation of multiple works in this regard, to date there is no clear correlation that explains the association between the inflammatory process after tonsillectomy and the calcification of the stylohyoid ligament or hypertrophy of the styloid process [5]. In the other hand, the carotid artery syndrome is caused when the elongated styloid process compresses

the internal carotid artery, provoking pain in the cervical area, which can cause dizziness, transient loss of vision and syncope [1]. While both entities described by Eagle belong to Eagle's syndrome, the difference between the two of them is found in the structures that will be compressed by the styloid process and the symptoms that the patient may present. Camarda in 1989 described a third type of eagle syndrome, called pseudostyloid syndrome, in which the patient describes the classic symptoms of Eagle's syndrome but there is no evident clinical or imaging presence of any elongation of the process or ossification of the styloid ligament, being this the consequence of aging and loss of the elasticity of the tissues [6].

There are two types of treatment for eagle syndrome. The first corresponds to a conservative treatment, mainly focused on the management of symptoms, using NSAIDS, transpharyngeal infiltrations with corticosteroids, anesthetic infiltration in the tonsillar fossa and exercises that allow strengthening of the cervical musculature [7]. On the other hand, we have surgical treatment where resection of the styloid process or part of it is performed. The surgical approach to access the styloid process can be performed intra or extra orally, depending on the case and the surgeon's experience. Even though the extra oral approaches were widely used during a long period of time to access de surgical area, due to aesthetics compromise and the high risk of damaging noble structures such as the carotid artery and branches of the facial nerve, other surgical techniques were developed being the intraoral approach the most used now a days [7,8]. In the last 10 years, the intraoral transpharyngeal approach has been increasingly used to access the surgical area, accessing the lateropharyngeal space through the tonsillar region and having a direct view of the styloid process utilizing measures described by Kent in 2014 using a CBCT [4]. Although this technique is more direct, simple, with lower surgical time and low aesthetic compromise than the extraoral approach, it requires a high degree of anatomical knowledge in that area, since there's a high risk of damaging any noble anatomical structure [8,9].

In this case, an intraoral surgical approach was performed modifying the anatomical repairs described by Kent in 2014. The distance of the styloid process to the anterior and posterior border of the mandibular ramus and the distance to the distopalatine cuspid of the upper second molar were determined, in contrast of using the distance from the styloid process to the tonsil and the distance from the carotid artery to the styloid process as described by Kent in his study in 2014 [4]. These new parameters modified by the surgical incision made in the oral mucosa, allow a best view of the anatomical parameters throughout the surgical period of time. It also has direct access to the surgical area and a wide surgical field that facilitates the surgeon in the location of the styloid process and its exeresis. The patient's immediate and late postoperative period was satisfactory: prompt symptoms relief, unrestricted cervical movements, pain and swelling relive of the lateral neck region. Finally, she was in outpatient controls until 6 months after surgery, where she was finally discharged after having no limitations in cervical mobility or specific pains.

Conclusion

The intraoral approaches to access the styloid process correspond to a group of techniques that, although they have a great advantage from an aesthetic point of view compared to the extraoral approaches, are not exempt from complications. Due to this, the particular analysis of

each case and alterations in the anatomical features that each patient may have are important to consider before the surgical procedure. On the other hand, although the anatomical parameters used in this case and the incision in the oral mucosa instead of the tonsil have brings certain advantages such as better visibility of the surgical field, a direct and constant view of the anatomical parameters and a direct access to the surgical area, it is important to note that due to the use of the upper second molar as an anatomical parameter, they make this technique very specific for its constant use. Although this approach has an important limitation when choosing it as a surgical technique, it is relevant to consider it when the above mentioned criteria are met, since it allows a more direct and rapid access to the proper surgical field.

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