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Case Report

Paucisymptomatic Bilateral Eagle Syndrome: Seven Years to Diagnosis, Case Report

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ABSTRACT

Eagle syndrome is a radioclinical entity characterized by ossification of the stylohyoid ligament. While cervical pain and dysphagia are among the typical symptoms, patients can present with a wide spectrum of benign and dangerous symptoms. Incidences of abnormal stylohyoid length range from 4% to 7.3%, but only 4% to 10% of these patients present with symptomatology. Palpation of the tonsillar compartments makes it possible to suspect the diagnosis. Standard x-rays generally confirm this, but it is especially CT that allows us to properly explore the calcified ligament and its relationships. The standard treatment is surgery. We present a case of a bilateral eagle syndrome that took seven years to be diagnosed and who was treated in our department by cervicotomy.

KEYWORDS: Eagle syndrome; Seven years; Diagnosis; Surgery.

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INTRODUCTION

Eagle syndrome is defined as a set of clinical signs in the cephalic and cervical regions, resulting from anatomical variations in the styloid process, ossification of the styloid ligament, or both.[1] It was first described by Eagle in 1937 where he described two forms of pain.[2,3] The stylo-hyoid form is characterized by orofacial pain, a sensation of a foreign body in the region of the tonsil space. In most cases, this pain appears after tonsillectomy. The stylo-carotid form results in neck pain associated with irritation of the sympathetic carotid nerve fibers caused by the rubbing of the calcified stylhyoid ligament. In this form, no history of surgery is reported. The non-specificity of clinical symptoms and the rarity of this syndrome explain the diagnostic delay.[2]

CASE PRESENTATION

A 48-year old female, without any particular medical history, presented to our ENT department with a 7-year history of bilateral throat pain accentuated on the left side and globus sensation. The patient denied dysphagia or other associated symptoms. The patient had tried several courses of analgesic drug, anti-inflammatory medication

and antibiotics without sufficient relief. A complete head and neck examination including fiberoptic laryngoscopy was performed, and the only pertinent finding was pain in the intraoral palpation of the left retromolar trigone and left peritonsillar region.

Investigations:

CT imaging with axial, coronal and 3D reconstructions showed 4,5 cm styloid process length in both left and right side (Figure 1). This styloid process was closely related to the left para-pharyngeal wall.

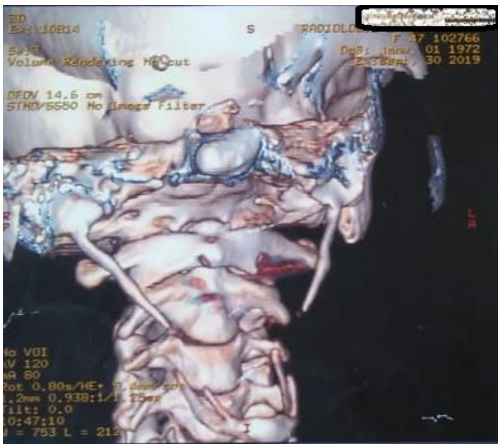

Figure 1: 3D reconstructions CT imaging of the head and neck of the patient showing long styloid processes.

Diagnosis and treatment:

The diagnosis of Eagle syndrome was raised and the indication of an exploratory cervicotomy with resection of the calcified ligament under general anesthesia was proposed. After skin incision, a careful dissection was carried out with respect for the vasculo-nervous structures of the region until the arrival to the styloid process. The dissection is completed with subperiosteal dissection onto the styloid process and its resection (Figure 2). Operative suites were simple. Histological examination of the surgical specimen allowed identifying a much corticalized bone with little bone marrow space.

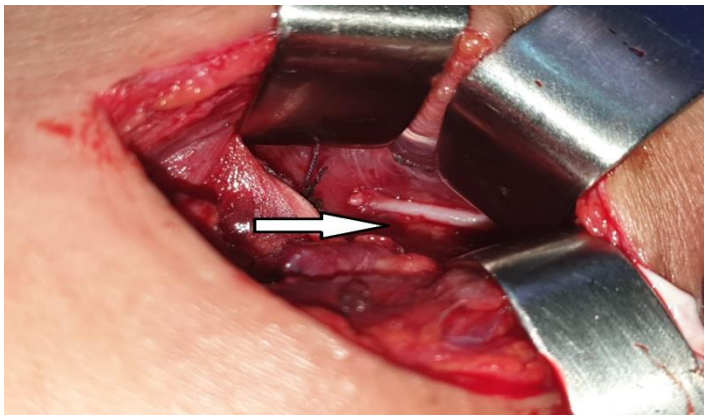

Figure 2: Intraoperative view showing a styloid process of abnormal length.

DISCUSSION

There is variability in determining the epidemiology of Eagle Syndrome due to differences in the diagnostic criteria in radiologic imaging. Some have suggested that the accepted length of the normal styloid process is approximately 2.5 cm, with 3 cm regarded as the upper limit of normal.[4,5] Radiologically, some studies have used stylohyoid length greater than 2.5 cm as abnormal.[4] Other studies have defined length greater than 4.0 cm as abnormal as this length has a higher association with pain.[5] Incidences of abnormal stylohyoid length range from 4% to 7.3%,[6,7] but only 4% to 10% of these patients present with symptomatology.[8] Chez un meme individu, le process us styloide est souvent asymetrique.[9]

The non-specificity of symptoms clinics and the rarity of this syndrome explain the diagnostic delay, this was the case with our patient. The other problem is that of the differential diagnosis at the level of the ENT and maxillofacial sphere. Eagle syndrome remains a diagnosis of elimination.

Standard radiography often shows unilateral or more often bilateral bone processes, prolonging the styloid processes. It sometimes shows bony processes with pseudo-joints, reminiscent of the phalanges of a finger, and extending from the stylid process to the lesser horn of the hyoid bone.

The current radiological exploration of the styloid process is based on CT with axial, coronal acquisitions and 3D reconstructions. It makes it easier to explore the calcified ligament over its entire length and its anatomical relationships with adjacent structures, mainly vascular and nervous.

The management of Eagle syndrome is commonly divided into conservative methods of medical management or more definitive surgical treatment. Basic medical therapy can be further divided into first-line analgesics such as NSAIDs and alternative management consisting of a combination of anticonvulsants, antidepressants, local injections and manipulation.[10] The literature tends to support that surgical treatment results in more definitive treatment and long lasting symptomatic relief.[11] Surgical management is typically divided into the intraoral and cervical approaches.[12] Eagle chose the trans-pharyngeal approach.[13] This technique is simple, allows to avoid skin scars, but visibility of the surgical site is limited and the risk of cellulitis cervical important.[13] Many authors prefer the cervical approach by cervicotomy which allows good visibility of the operative site and decreases the risk of postoperative cellulitis.[14,15] This is the choice that was made in this case in order to have access to the entire stylohyoid complex.

CONCLUSION

Eagle syndrome has a large variety of clinical presentations as evidenced by the multitude of non specific symptoms. It has a many differential diagnoses and remains a diagnosis of elimination. Its management is mainly surgical by exeresis of the styloid process via a transoral or cervical approach.

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