Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Surgery

Eagle Syndrome: A Case Report

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DOI: https://doi.org/10.36347/sjmcr.2025.v13i10.050 | **Received:** 22.05.2025 | **Accepted:** 20.07.2025 | **Published:** 21.10.2025

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Abstract **Case Report**

Eagle syndrome is a set of cervical symptoms resulting from the elongation of the stylohyoid ligament and its subsequent ossification. It is a poorly known syndrome among clinicians. It manifests through cervico-pharyngeal symptoms caused by irritation of anatomical structures in contact with this process. Due to the variability of the symptomatology, this syndrome is often diagnosed late, resulting in therapeutic wandering for patients and a delay in appropriate management. Therapeutic management is surgical, with the indication depending on the extent of associated symptoms. We report a case of bilateral Eagle syndrome, more pronounced on the right side, discovered in a 59-year-old patient with no particular pathological history. He underwent surgical treatment via the laterocervical approach under general anesthesia. The postoperative course was uneventful, and the patient reported a clear improvement in initial symptoms. Through this case report and a literature review, we aim to clarify the clinical, radiological, and therapeutic particularities of Eagle syndrome.

Keywords: Eagle syndrome, Elongated styloid process, Surgical treatment.

INTRODUCTION

Eagle syndrome is a poorly known clinico-radiological entity described by Eagle in 1937 [1,2]. It corresponds to an abnormal hypertrophy of the styloid process and/or calcification of the stylohyoid ligament. The clinical diagnosis is difficult to establish due to the polymorphic nature of its symptomatology. Imaging based on CT scans with 3D reconstruction allows for a positive diagnosis [2]. Through this case report and a literature review, we recall the clinical and radiological characteristics and the different therapeutic options for this entity.

CLINICAL OBSERVATION

We report a case of Eagle syndrome discovered in a 59-year-old patient with no particular pathological history, who presented with bilateral laterocervical pain, more marked on the right, evolving for two years. The pain was continuous, diffuse, sometimes preventing sleep, and not relieved by analgesics. It was aggravated by swallowing and head rotation and associated with right-sided otalgia. Bimanual palpation revealed a hard mass in the homolateral tonsillar region bilaterally. The rest of the ENT examination was unremarkable. Cervical CT scan showed a long bilateral styloid process of type 1 measuring 5.6 cm on the right, with its distal end facing the right lateral wall of the oropharynx, and a lesser elongation of the styloid process on the left measuring 3.2 cm with slight calcification along the left stylohyoid ligament (Figure 1). The patient underwent bilateral surgery via the laterocervical route. The incision was a high vertical one along the anterior border of the sternocleidomastoid muscle. Resection of the calcified process was performed (Figure 2). The postoperative course was uneventful with complete disappearance of the pain.

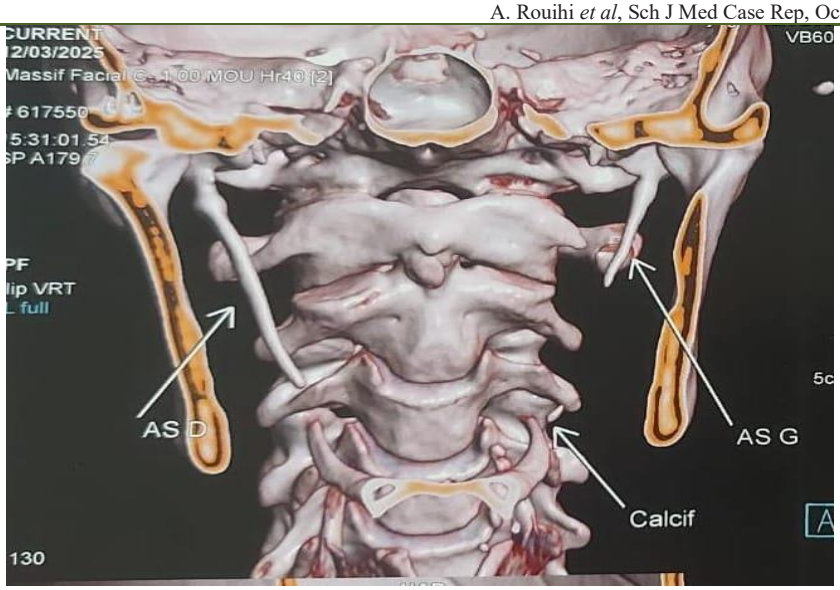

Figure 1: Cervical CT scan with 3D reconstruction showing elongation of both styloid processes composed of a single fragment extending toward the lesser horn of the hyoid bone, indicating ossification of the stylohyoid ligament

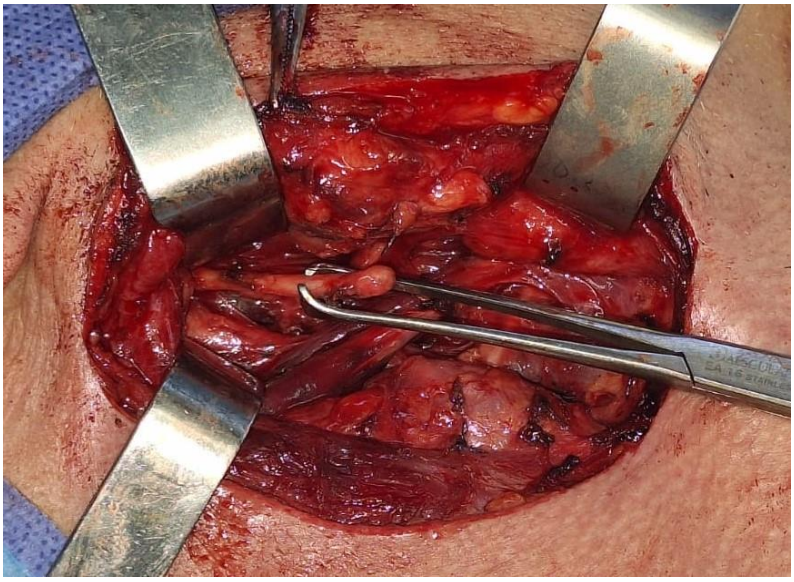

Figure 2: Operative view: approach to the styloid process via the laterocervical route

DISCUSSION

Eagle syndrome, known as elongated styloid process syndrome, stylohyoid syndrome or Garel's styloid angina, is a rare clinical entity described for the first time by Eagle in 1937 [1]. This syndrome is due to an elongated styloid process and/or calcification of the stylohyoid ligament and/or an elongated lesser horn of the hyoid bone. Its frequency is estimated at 4% of the general population, but only 4% of these ossifications are symptomatic. The average age of patients with Eagle syndrome is mostly over 40 years [2], with extremes of 20 and 80 years [2]. The prevalence by sex is highly debated, but some authors find that the syndrome mainly affects women. The elongation of the styloid processes is sometimes bilateral, but the symptoms may only affect one side.

The etiopathogenesis of Eagle syndrome is poorly understood. Several theories attempt to explain it, such as congenital elongation of the styloid process, acquired calcification and ossification of the stylohyoid ligament [1], or trauma to the cervico-pharyngeal region, particularly after tonsillectomy.

The diagnosis of Eagle syndrome is frequently marked by a long period of diagnostic wandering due to its rarity [1,2] and the great variability of its symptomatology, especially the variations in painful topography [1,2,3]. Functional signs are polymorphic and non-specific. Eagle distinguished three groups: the first is the classic syndrome associating ipsilateral cervical pain, otalgia, and pharyngeal discomfort; the second is characterized by pain along the external and internal carotid arteries due to irritation of the pericarotid plexus [3,4], which may present as migraine or cluster headache, aggravated by head rotation; the third is

asymptomatic. Clinical examination, intraoral and extraoral, looks for filling of the tonsillar fossa by a hard mass corresponding to the elongated styloid process. Palpation provokes pain similar to that which led the patients to consult. It must also rule out dental problems (wisdom teeth), tumors, or infections and look for signs suggestive of various pathologies. If the clinical examination remains important for suspecting stylohyoid syndrome, definitive diagnosis relies on imaging. Standard radiography often shows unilateral or more often bilateral bony processes [2,3] extending the styloid processes; they may sometimes show pseudo-articulations, resembling the phalanges of a finger, extending from the styloid process to the lesser horn of the hyoid bone.

CT scan is the examination of choice, having replaced standard radiography; axial, coronal acquisitions and 3D reconstructions (injection and dynamic rotation images) allow for easier exploration of the calcified ligament along its entire length and its anatomical relationships with adjacent structures, mainly vascular and nerve structures; 3D reconstructions allow good evaluation of its length and degree of inclination (Figure 3), which are important for diagnosis and therapeutic decision [4]. They also allow ruling out other cervico-facial pathologies. As there is no correlation between the length of the process, its angulation and the severity of clinical signs, CT measurements have no value outside the clinical context [5,6]. MRI can detect whether the stylohyoid syndrome presents as a transient or established neurological deficit [4-6]. The differential diagnosis mainly includes trigeminal and glossopharyngeal neuralgias, temporomandibular joint pathology or an upper aerodigestive tract tumor; Eagle syndrome remains a diagnosis of exclusion.

Figure 3: Right styloid process after resection

For therapeutic management, there are two possible options: a conservative option and a surgical option. The conservative option is chosen either in cases of mild to moderate symptoms or if the patient refuses surgical treatment [3,6]. The conservative option is mainly based on anti-inflammatories and analgesics, which can improve functional symptoms in the short term; local infiltrations of xylocaine are considered a diagnostic test but not a full-fledged treatment; manual transpharyngeal fracture of the styloid process is associated with frequent recurrence of symptoms in the medium to long term [2,5].

The curative treatment is mainly surgical in cases of disabling clinical symptomatology after radiological confirmation and elimination of other cervico-facial pathologies; according to Eagle, only 3 to 4% of cases require surgical treatment [1,3]. There are two surgical approaches for treating Eagle syndrome: an intraoral approach and an extraoral approach. Two principles must be considered for surgery regardless of the approach used: it is important to resect the styloid process as close as possible to its temporal attachment to prevent recurrence and even if the symptoms are mostly unilateral [1,3], resection must involve both styloid processes if they are elongated [6-8]. The cervical approach is performed under general anesthesia with orotracheal intubation. Cervical hyperextension and head rotation to the opposite side facilitate the approach according to Risdon [6], which is performed via the parotidectomy route [7]; resection of the calcified stylohyoid process is performed after careful dissection. The cervical approach offers good visibility but leaves a visible scar. The transoral approach has the advantage of not requiring a skin incision [8] and can be performed under local anesthesia or, most commonly, under general anesthesia [4,9] with nasotracheal intubation. A tonsillectomy is performed if the tonsils are still present.

The styloid process is generally palpated at the superolateral end of the tonsillar fossa; the overlying mucosa is then incised, and the process is gradually freed from all its ligamentous attachments. It is then resected from its temporal origin. The mucosa and muscle are sutured in two layers, and the average duration of the operation is about one hour [4]; the risk of this approach is nerve or carotid injury; therefore, this intervention must be performed by a trained surgeon. However, postoperative paresthesia of the great auricular nerve is often associated [4,8].

Regardless of the technique used, the disappearance of symptoms can be partial or complete. Between 80 and 90% of patients no longer present any symptoms one year after treatment, which was the case for the patient treated here [4,6].

CONCLUSION

Eagle syndrome is a rare, poorly known, and controversial entity in the literature. It remains a diagnosis of exclusion after ruling out any ENT and neurological, particularly neuralgic (V, IX), pathologies. The ENT surgeon must be familiar with Eagle syndrome, which can present as cervico-facial pain. Given the rarity of this syndrome, therapeutic data are quite limited and mainly rely on styloidectomy.

Declaration of Interests: The authors declare that they have no conflicts of interest in connection with this article.

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