International Journal of Otorhinolaryngology and Head and Neck Surgery
Yuan HY et al. Int J Otorhinolaryngol Head Neck Surg. 2024 Aug;10(4):436-438
http://www.ijorl.com
pISSN 2454-5929 | eISSN 2454-5937

DOI: https://dx.doi.org/10.18203/issn.2454-5929.ijohns20242047

Case Report

Eagle's syndrome: mimicker of submandibular and temporomandibular joint pathology

Ho Yi Yuan[1]*, Vivian Lau Zhi Han[1], Mohammad Nasyatmuddin Yahya[1], Chew Shiun Chuen[1], M. Razif Mohamad Yunos[2]

[1]Department of Otorhinolaryngology, Hospital Sibu, Sarawak, Malaysia
[2]Department of Otorhinolaryngology, Universiti Kebangsaan Malaysia, Malaysia

Received: 09 May 2024
Accepted: 03 July 2024

***Correspondence:**
Dr. Ho Yi Yuan,
E-mail: hoyiyuan@moh.gov.my

ABSTRACT

Eagle's syndrome is characterized by recurrent pain in the oropharynx and face due to an elongated styloid process or calcified stylohyoid ligament. Various theories existed to explain about development of Eagle's syndrome. Symptoms are variable and non-specific, thus making it a challenge and dilemma to recognize and diagnose it clinically, and sometimes it can be missed out. A good history taking is important to recognize the disease with proper physical examination. Radiological intervention has been an important tool to diagnose Eagle's syndrome. A 31 years old lady with presentation of right submandibular pain for one week, radiating to right preauricular region, associated with odynophagia, trismus and reduced oral intake. She was initially misdiagnosed as submandibular sialadenitis and lymphadenitis, and later was referred to dental for temporomandibular joint arthritis. She was treated with antibiotic initially however symptoms were not improved. CT neck done and revealed elongated right styloid process. Excision of right styloid process provided complete pain relief. Even though Eagle's syndrome is not a common disease, in cases of unexplained complain of pain over head and neck region, it should be considered as one of the differential diagnosis. Eagle's syndrome can be managed medically by symptomatic treatment/by surgical intervention.

Keywords: Eagle's syndrome, Elongated, Styloid process

INTRODUCTION

Eagle syndrome also known as stylalgia, is a rare clinical entity involving the oro-maxillo-facial region.[1] It is associated with collection of symptoms due to elongated styloid process or calcification of stylohyoid ligament.

In 1652, An Italian surgeon Pietro Marchetti first describe the elongation as an ossifying process. In 1937, Watt W. Eagle describe the term stylalgia as the pain associated with this abnormality.[2]

Eagle syndrome is commonly seen in woman compared to male with a male:female ratio ~1:2. The average age of the patients are usually 40 years old. In general population, there are estimated 4% of people will be suffered from symptom of elongated styloid process.[3] The complex of symptomatology could lead to diagnosing dilemma like the case we reported.

CASE REPORT

A 31-year-old lady with underlying hyperthyroidism complained of right submandibular pain for one week, radiating to right preauricular region, associated with odynophagia, limited mouth opening and reduced oral intake. Otherwise, there was no infective symptoms, nasal or ear symptoms, snoring, foreign body (FB) ingestion or tuberculosis (TB) contact. She had no prior history of surgery done.

Clinically she was not in respiratory distress with trismus, no dental carries, no raised of floor of mouth. She has tenderness over submandibular region but no swelling

palpable. Otoscopy, nasoendoscopy and laryngoscopic findings was unremarkable.

She was treated as right submandibular sialadenitis and lymphadenitis in the beginning of the disease and symptoms partially improved after completion of 10 days antibiotics.

We sought further opinion from dental colleague for temporomandibular joint arthritis. They noted non tender unerupted molar teeth over right buccal sulcus and right maxillary tuberosity region. Patient was given advisement to be on soft diet, cold compression and to limit mouth opening by not yawning widely.

However, she still complaint of tenderness over preauricular region, hence CT neck was done. CT neck revealed elongated right styloid process measuring up to 4.5 cm. She underwent tonsillectomy and intraoral excision of right styloid process. Symptoms was completely resolved postoperatively.

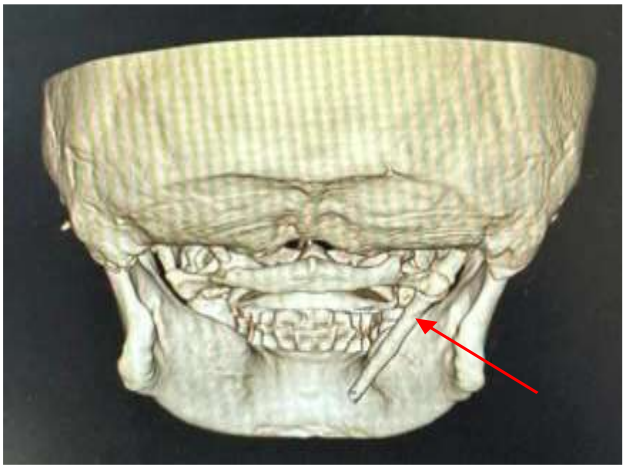

Figure 1: Presurgical 3D reconstruction from CT scan. Arrows indicate the elongated SPs.

Figure 2: Right styloid process around 2.4 cm removed.

DISCUSSION

Styloid process is a cylindric bony outgrowth located in front of the stylomastoid foramen and it is a part of temporal bone. The length of styloid process can be classified to three group by three dimensional (3D) CT: short (<2 cm), long (2-4 cm) and elongated (>4 cm).[9] "Riolan's bouquet" which comprise of 3 muscles and 2 ligaments, is a group of muscles and ligaments that originate from the process. They are stylohyoid muscle, styloglossus muscle, stylopharyngeus muscle, stylomandibular ligament and mandibulo-stylohyoid ligament. The styloid process is located between the internal and external carotid artery and juxtaposed near cranial nerves VII, IX, X, XI and XII.

There are two types of syndromes which is classic styloid syndrome and carotid artery syndrome. Classic type is normally following tonsillectomy. Eagle theorized that the scarring near to the styloid subsequently compress or irritate nerve structures in the space surrounding the styloid process.[4] The pain is secondary to stimulation of cranial nerve V, VII, IX and X. It is characterized by ipsilateral dull and persistent pharyngeal pain, centered in the ipsilateral fossa that can be referred to the ear and exacerbated by rotation of the head to contralateral side. Some patient also can have dysphagia, FB sensation in the throat, otalgia, tinnitus or cervicofacial pain mimicking neuralgic pain.[5]

Carotid artery type is not related to tonsillectomy and occurs when the styloid process is involved with the carotid nerve plexus. It can also cause by compression of internal or external carotid artery. It causes neck pain along the distribution of the artery, during head rotation or compression of the neck, and a FB sensation in the pharynx. In the case of internal carotid artery compression, the pain is generally felt within the lateral region of head or around eye. When the external carotid artery is compressed, pain is at the region of infraorbital and it is related with rotation of the head.[7,9] In our case, the patient had symptoms of the carotid artery type.

In the case of patient who have never underwent tonsillectomy, there are some other theories have been considered: Ossification of the stylohyoid ligament complex that caused stretching of the CNXII and contraction of stylopharyngeal muscle.[4] Irritation of the structure nearby by the ossification of muscular tendons.[6] The abnormal angulation or abnormal length of the styloid process.[7] The traumatic fracture of styloid process, with incomplete repair due to continuous hyoid bone movement and formation of excessive granulation tissue.[8]

Diagnosis is made through clinical symptoms, pharyngeal palpation of the styloid process through the tonsillar fossa and imaging such as orthopantomogram, lateral view radiographs of skull and 3D reconstruction CT scan. A good medical history is important to rule out various others disease with similar presentation such as migraine,

tension headache, atypical facial pain, myofascial pain syndrome, salivary gland disease, tonsillitis and psychosomatic disease. One of the clinical diagnostic test is relieving of the symptoms after injection of 2% lignocaine into the anterior pillar and tonsillar fossa.[10,11] Pharyngeal palpation is approached under local anaesthesia by placing index finger to feel the tonsil from top to bottom and then palpate the tonsillar fossa from lower part upward and behind the tonsil by pushing it outward. There is a bony structure felt but sometimes it can evoke or increase the pain.[1] Gold standard to diagnose Eagle syndrome is a 3D reconstruction CT scan. It is useful to allow us to visualize the elongation of the styloid process and evaluate the exact morphology and adjacent anatomical structure for planning of surgery.[12]

Treatment for Eagle syndrome can be conservative or surgical. Conservative treatment includes adequate analgesia, injection of steroids and long-lasting anaesthetics into the tonsillar fossa.[13] Surgical treatment can be removing of the styloid process through intraoral or external approach. For intraoral approach, it is always accompanied with tonsillectomy. It offers shorter treatment time and better cosmetic effect however has the risk of complications associated with the limited visibility of surgical site.[14] External approach was explained by Chrcanovic et al and Loeser and Cardwel. It provides better surgical access and visualization. The disadvantages are external scar and longer recovery duration.[15] In this case, our patient initially given symptomatic management with analgesia but the symptoms still persistent. Hence, we opted for surgical management with intraoral approach.

In our case, we encounter the longest styloid process so far with length of 4.5 cm. This cause a struggle for us to skeletonize the styloid process intraoperatively. However, we manage to remove big part of it during the operation. Post operatively, patient symptoms resolved remarkably.

CONCLUSION

Eagle's syndrome is rare but can be diagnosed via a detailed history, physical examination and imaging. It can cause diagnosing dilemma due to the complexity and similarities of symptoms. Excision of elongated styloid process is a treatment of choice with good outcomes.

ACKNOWLEDGEMENTS

Authors would like to thank ORL department hospital Sibu for facilitating and participating in this case report.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Montalbetti L, Ferrandi D, Pergami P, Savoldi F. Elongated styloid process and Eagle's syndrome. Sage J Cephalalgia. 1995;15(2):80-93.
2. Kim E, Hansen K, Frizzi J. Eagle syndrome: Case report and review of the literature. Ear Nose Throat J. 2008;87(11):631-3.
3. Branko P, Đorđe R, Vladimir K, Nadežda CS. Styloid syndrome: A review of literature. Srp Arh Celok lek. 2008;136(11-12):667-74.
4. Koshy JM, Narayan M, Narayanan S, Priya BS, Sumathy G. Elongated styloid process: A study. J Pharmacy Bioallied Sci. 2015;7(1):131-3.
5. Monsour PA, Young WG. Variability of the styloid process and stylohyoid ligament in panoramic radiographs. Oral Surg Oral Med Oral Pathol. 1986;61(5):522-6.
6. Kaufman SM, Elzay RP, Irish EF. Styloid process variation. Radiologic and clinical study. Arch Otolaryngol. 1970;91(5):460-3.
7. Baddour HM, McAnear JT, Tilson HB. Eagle's syndrome. Report of a case. Oral Surg Oral Med Oral Pathol. 1978;46(4):486-94.
8. Scavone G, Caltabiano DC, Raciti MV, Calcagno MC, Pennisi M, Musumeci AG, et al. Eagle's syndrome: a case report and CT pictorial review. Radiol Case Rep. 2019;14(2):141-5.
9. Boucher Y, Mularski A, Felizardo R, Tankere F, Dieb M. Chronic oropharyngeal pain and medical nomadism in an Eagle's syndrome patient: a case report. J Med Case Rep. 2022;16:201.
10. Han MK, Kim DW, Yang JY. Non-Surgical Treatment of Eagle's SyndromeA Case Report. Kor J Pain. 2013;26(2):169-72.
11. Joy L, Arabhanvi R, Viswanatha B. Eagle's Syndrome: A Case Report. Res Otolaryngol. 2020;9(1):1-3.
12. Fonseca HB, Da Costa SM, Jamil LC. Eagle syndrome: a case report. Rev Bras Cir Plást. 2019;34(2):287-90.
13. Warrier AS, Nanthini KC, Subadra K, Harini DM. Eagle's Syndrome: A Case Report of a Unilateral Elongated Styloid Process. Cureus. 2019;11(4):e4430.
14. Wacławek M, Pietkiewicz P, Niewiadomski P. Own experience in the diagnosis and treatment of elongated styloid process syndrome. J Olszewski Otolaryngol Pol. 2021;75(2):21-7.
15. Pradhan U, Adhikari TR. Diagnostic and therapeutic dilemma in orofacial pain: A rare case of bilateral Eagle syndrome. SAGE Open Medical Case Reports. 2022;10:2050313X221116950.

Cite this article as: Yuan HY, Han VLZ, Yahya MN, Chuen CS, Yunos MRM. Eagle's syndrome: mimicker of submandibular and temporomandibular joint pathology. Int J Otorhinolaryngol Head Neck Surg 2024;10:436-8.