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Case Report / Olgu Sunumu

Vascular Type Eagle Syndrome: A Case of Styloid Process-Carotid Artery Syndrome with Operation*

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ABSTRACT

Eagle's syndrome (ES), also known as the stylohyoid syndrome, is defined as the elongation of a styloid process by more than 30 mm and the mineralization of the stylohyoid-stylomandibular ligament. Two types of ES are described in the literature: classical stylohyoid syndrome and vascular styloarotid syndrome. While bilateral ES is commonly seen in women, the symptoms are usually unilateral. This case report describes the surgical treatment of a 45-year-old female patient with bilaterally-elongated styloid processes who presented with vascular ES symptoms. A literature review is also included.

Key words: Styloid process, calcification, carotid artery, Eagle's syndrome, panoramic image

ÖZET

Eagle Sendromu (ES) veya Stilohyoid Sendromu, stiloid prosesin 30 mm'den fazla uzaması ve stilohyoid-stilomandibular ligamentin mineralizasyonu olarak tanımlanmaktadır. Literatürde iki ES formu tanımlanmaktadır. Bunlar klasik tip stilohyoid sendrom ve vasküler tip stilokarotid sendromudur. ES sıklıkla kadınlarda ve bilateral olarak görülürken, belirtiler genellikle tek taraflıdır. Bu olguda bilateral olarak uzamış stiloid prosesi olan ve Eagle tipi sendromun vasküler tipini destekleyen semptomlarla başvuran 45 yaşında bir kadın hasta ele alınmış ve ilgili literatür sunulmuştur.

Anahtar kelimeler: Stiloid proses, kalsifikasyon, karotid arter, Eagle sendromu, panoramik görüntü

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INTRODUCTION

Eagle's syndrome (ES), also known as the stylohyoid syndrome, is characterized by a styloid process (SP) that is elongated by more than 30 mm and the presence of calcified stylohyoid ligaments.¹ The SP is an anatomical structure that forms a thin protrusion from the temporal bone. SP elongation and the clinical symptoms associated with structural changes in the stylohyoid ligament were first described by W.W. Eagle, for whom the syndrome is named.²

ES symptoms include a difficulty in swallowing and a foreign-body sensation in the throat as well as craniofacial or cervical pain. A recurrent sore throat or a dull, nagging pain in the throat, otalgia, carotidynia, temporomandibular joint pain, glossopharyngeal neuralgia, facial pain, and headache are the most significant diagnostic findings.³

Two types of ES are described in the literature: classical stylohyoid syndrome and vascular stylocarotid syndrome.⁴ The classical stylohyoid syndrome is caused by post-operative fibrous tissue that occur after tonsillectomy, leading to irritation of the cranial nerve endings in the tonsils. The symptoms occurs following a tonsillectomy. The symptoms include persistent unilateral pharyngeal pain that is aggravated by swallowing and often reverberates in the ear.⁴⁻⁶ Other symptoms may include dysphagia, facial pain, foreign-body sensation, increased salivation, tinnitus, and otalgia. Vascular ES, also referred to as a stylocarotid syndrome, is attributed to extracranial impingement of the internal carotid artery by a deviated and elongated SP.^{7,8} The elongated SP may cause compression when the neck is turned or even a carotid artery dissection that results in a transient ischemic attack or stroke.⁹ The symptoms include syncope, visual symptoms, carotid dissection, and parietal pain.⁵

ES is diagnosed by radiological and physical examinations. The ossification of the stylohyoid ligaments is best seen in lateral cervical and anteroposterior upper cervical radiographs.^{10,11} For symptomatic cases, the preferred treatment is surgical removal of the elongated SP using a transcervical or transoral approach. Ultrasonic osteotomy is another preferred surgical method due to its ability to preserve the anatomical structures close to the resection area.¹² We report a case of bilaterally elongated SPs that presented with symptoms supporting vascular ES. The literature is also reviewed.

CASE REPORT

A 45-year-old female patient was admitted to our dental clinic with a complaint of pain on the right side jaw and neck. She stated that jaw pain increased with chewing, and neck pain occurred during head movements. She had systemic complaints of arrhythmia and hypertension, was using anticoagulant medicines, and had atrial septal defect operation one year ago. The patient stated that she had fallen on her back in 2011, and then had long-term treatment of hemiplegia during this period, the cause of neck, chin and back pain was not found.

When the medical file of the patient was examined, it was learned that head radiographs and brain MR were obtained due to patient's pain, but no reason was found.

It was also learned that an abnormality was detected in the carotid artery when the cause of the hemiplegia was investigated. An artery defect was found on the neck MR, and a stent was placed in the bilateral carotid arteries of the patient due to this defect and possible complications. No abnormal findings were detected in the extraoral and intraoral examination of the patient in our clinic. However, the radiating pain was detected upon percussion applied to the teeth within the painful region. For radiological evaluation, periapical and panoramic images were taken (Figure 1).



Figure 1. Panoramic images demonstrating bilaterally stents in carotid arteries



Figure 2. Panoramic image demonstrating the bilaterally elongated styloid process

After the further investigation of the patient's story it was learned that after the stent surgery, her medical doctors reported to her that one muscle pricked was stuck to the carotid artery during the fall on her back. Based on this information and current complaints, eagle syndrome diagnosis was made and, pre-recorded images of the patient were obtained. Bilateral calcified styloid ligaments were clearly detected in the panoramic image of the patient which was taken four years ago (Figure 2). The styloid process measurement was performed using Image J software (Image J, US National Institutes of Health, Bethesda, MD). On both sides, the styloid process elongated more than 30 mm. The calcified ligaments shapes were ended with sharp tips.

In the light of this information, it is thought that arterial damage was caused by the calcified stylohyoid ligament pricking to carotid artery on at the moment of falling and related paralysis was developed, and since four years there has been a pain because no treatment was administered for calcified stylohyoid ligaments. It was decided that tooth number 15 should be removed to relieve the patient's dental induced pain. In order to remove the symptoms related to Eagle Syndrome, and also considering the systemic condition, the patient was informed and the ear-nose-throat (ENT) consultation was requested.

It has been reported that the stylohyoid ligaments should be shortened surgically in order to relieve the symptoms according to ENT consultation. However, since it is necessary to position the patient's head at a 45-degree angle for a long time during surgical operation, and in this position, stents may cause damage to the arteries again, the stylohyoid ligament surgery couldn't be performed. Further radiographic evaluation with advanced imaging techniques was not considered since it would not provide any benefit to the patient. The clinical follow-up of the patient is ongoing by us and ENT department.

DISCUSSION

The SP is located between the internal and external carotid arteries and is closely related to the tonsillar fossa and the mastoid bone. It is in proximity to the seventh, ninth, tenth, eleventh and twelfth cranial nerves. A normal SP length is about 20–30 mm. ES is defined as the elongation of the SP by more than 30 mm and the mineralization of the stylohyoid-stylomandibular ligament.¹³⁻¹⁷ The primary symptoms include swallowing difficulty, foreign-body sensation in the throat and craniofacial or cervical pain. While ES is often seen in women and bilaterally, the symptoms are usually unilateral.¹⁵

In this case report, the vascular stylocarotid syndrome was not related to any previous surgical intervention, distinguishing it from the classical type of ES. In this syndrome, a prolonged SP exerts pressure on the internal and/or external carotid arteries.¹⁸

The literature has reported complications due to the reduced blood flow caused by the narrowing of the arterial lumen, as well as complications resulting from the mechanical irritation of the sympathetic nerve plexus on the artery.¹⁹ Increased cervical pain can be triggered by rotation and compression of the neck when pressure is applied to the internal carotid artery.¹⁸ In our case, there was a persistent pain in the region of the right face and neck without a history of tonsillectomy. We believe that the patient's syncope occurred due to compression of the carotid artery. If the external carotid artery is irritated, the pain can spread to the infraorbital area, leading to feelings of apathy and drowsiness, along with visual disturbances.

ES diagnosis requires a detailed medical history, a clinical examination and the evaluation of radiological images, including lateral head-and-neck radiography, Towne radiography, panoramic radiography, lateral oblique mandibular radiography, anteroposterior head radiography, cone-beam computed tomography. Diagnosis is complicated by the superpositions, distortions, and magnifications related to the angles used in conventional radiological imaging. While cone-beam and regular computed tomography are not affected by these issues,^{20,21} the risks associated with the high radiation doses needed for these imaging techniques must be considered.

CONCLUSION

ES is easily detected in a panoramic image, which is the preferred imaging method for many dental applications. However, if the evaluation of the radiological images focuses too closely on the patient's physical complaints, significant anomalies may be missed. Informing patients of the anatomical changes, such as ES, that can be definitively diagnosed with dental images can effectively prevent complications that affect their quality of life.

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Conflict of Interest

None declared.

REFERENCES

1. Carter L. Soft tissue calcification and ossification. In: White SC, Pharoah MJ, editors. *Oral radiology, principles and interpretation*. Missouri: Mosby; 2004. pp. 597–614.
2. Eagle WW. Elongated styloid process; further observations and a new syndrome. *Arch Otolaryngol*. 1948; 47(5):630-640.
3. Chebbi R, Chaabani I, Alaya TB, Dhidah M. Elongated styloid process as a cause of facial pain. *Joint Bone Spine*. 2014;81(4):368.
4. Bafaqeeh SA. Eagle syndrome: classic and carotid artery types. *J Otolaryngol*. 2000;29(2):88-94.
5. Diamond LH, Cottrell DA, Hunter MJ, Papageorge M. Eagle's syndrome: a report of 4 patients treated using a modified extraoral approach. *J Oral Maxillofac Surg*. 2001;59(12):1420-6.
6. Sokler K, Sandev S. New classification of the styloid process length--clinical application on the biological base. *Coll Antropol*. 2001;25(2):627-32.
7. Chuang WC, Short JH, McKinney AM, Anker L, Knoll B, McKinney ZJ. Reversible left hemispheric ischemia secondary to carotid compression in Eagle syndrome: surgical and CT angiographic correlation. *AJNR Am J Neuroradiol*. 2007;28 (1): 143-5.
8. Mendelsohn AH, Berke GS, Chhetri DK. Heterogeneity in the clinical presentation of Eagle's syndrome. *Otolaryngol Head Neck Surg*. 2006;134(3):389-93.
9. Farhat HI, Elhammady MS, Ziayee H, Aziz-Sultan MA, Heros RC. Eagle syndrome as a cause of transient ischemic attacks. *J Neurosurg*. 2009;110(1):90-3.
10. Emary PC, Dornink M, Taylor JA. A rare case of Eagle syndrome and diffuse idiopathic skeletal hyperostosis in the cervical spine. *J Can Chiropr Assoc*. 2017;61(2):167-170.
11. Mutlu V, Ogul H. Coincidence of Eagle Syndrome and Diffuse Idiopathic Skeletal Hyperostosis Presenting With Dysphagia as a Result of Compression of the Hypopharynx. *J Craniofac Surg*. 2017;28(2):e129-e130.
12. Hoffmann E, Räder C, Fuhrmann H, Maurer P. Styloid-carotid artery syndrome treated surgically with Piezosurgery: a case report and literature review. *J Craniomaxillofac Surg*. 2013;41(2):162-6.
13. Ramachandra KK, Krishnan SA, Reddy SS. Carotid type of Eagle's syndrome. *J Indian Soc Periodontol*. 2015;19(3):336-8.
14. Ilguy M, Ilguy D, Guler N, Bayirli G. Incidence of the type and calcification patterns in patients with elongated styloid process. *J Int Med Res*. 2005;33:96–102.
15. Prasad KC, Kamath MP, Reddy KJ, Raju K, Agarwal S. Elongated styloid process (Eagle's syndrome): a clinical study. *J Oral Maxillofac Surg*. 2002;60:171–175.
16. Jung T, Tschernitschek H, Hippen H, Schneider B, Borchers L. Elongated styloid process: when is it really elongated? *Dentomaxillofac Radiol*. 2004;33:119–124.
17. Gokce C, Sisman Y, Tarim Ertas E, Akgunlu F, Ozturk A. Prevalence of styloid process elongation on panoramic radiography in the Turkey population from Cappadocia region. *Eur J Dent*. 2008;2:18–22.
18. Fini G, Gasparini G, Filippini F, Becelli R, Marcotullio D. The long styloid process syndrome or Eagle's syndrome. *J Craniomaxillofac Surg*. 2000;28(2):123-7.
19. Keur JJ, Campbell JPS, McCarthy JF, Ralph WJ. The clinical significance of the elongated styloid process. *Oral Surg Oral Med Oral Pathol* 1986; 61: 399-404.
20. Ferrario VF, Sigurtá D, Daddona A, Dalloca L, Miani A, Tafuro F, Sforza C. Calcification of the stylohyoid ligament: incidence and morphoquantitative evaluations. *Oral Surg Oral Med Oral Pathol*. 1990;69(4):524-9 23.
21. Savranlar A, Uzun L, Uğur M. B, Özer T. Three-dimensional CT of Eagle's syndrome. *Diagn Intervent Radiol* 2005; 11:206-209.