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Assessment of stylohyoid ligament in patients with Eagle's syndrome and patients with asymptomatic elongated styloid process: A cone-beam computed tomography study

Adineh Javadian Langaroodi DDS, MSc[1], Seyed Hossein Hoseini Zarch DDS, MSc[2], Amin Rahpeyma DDS, MSc[3], Hamed Ebrahimnejad DDS, MSc[4], Ali Arezoobakhsh DDS[5], Alireza Sanaei DDS, MSc[6]

Original Article

Abstract

BACKGROUND AND AIM: This study was performed to evaluate and compare the calcification patterns of the stylohyoid ligament in Eagle's syndrome (ES) patients, and asymptomatic patients with elongated styloid process (SP) via cone-beam computed tomography (CBCT).

METHODS: A total of 52 CBCT images in two symptomatic (ES) and asymptomatic groups (n = 26 per group) were assessed. The mean length and thickness of the SP, morphology, and pattern of calcification between the two sides in each group and between ES and asymptomatic groups were compared. The t-test was used for comparison. Fisher exact and chi-square tests were used to determine the relationship between different types of calcification pattern and morphology. The level of significance was considered at $P < 0.050$.

RESULTS: The SP was thicker in the ES group than the asymptomatic group. However, the styloid length showed no significant difference among the ES and asymptomatic groups. The most common pattern of calcification in both groups was partially calcified with no significant difference between the two study groups. The most common morphology in the asymptomatic and ES groups was "segmented" and "elongated," respectively.

CONCLUSION: The morphology and thickness of the SP showed a significant difference between the ES and asymptomatic groups. This can be helpful in differential diagnosis of facial, pharyngeal, and tonsillar pain.

KEYWORDS: Eagle Syndrome; Elongated Styloid Process Syndrome; Cone Beam Computed Tomography

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Eagle's syndrome (ES) is characterized by recurrent pain in the oropharynx and face caused by elongation of styloid process (SP) and calcification of stylohyoid ligament.[1] ES may be unilateral or bilateral and can be diagnosed based on the clinical and radiological investigations. Palpation of the SP in the tonsillar fossa is suggestive of the elongation process.[2]

Keur et al.[3] stated that if SP or mineralized stylohyoid ligament length is more than

1- Assistant Professor, Dental Research Center AND Department of Oral and Maxillofacial Radiology, School of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran
2- Associate Professor, Dental Research Center AND Department of Oral and Maxillofacial Radiology, School of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran
3- Associate Professor, Maxillofacial Diseases Research Center AND Department of Oral and Maxillofacial Surgery, School of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran
4- Department of Oral and Maxillofacial Radiology, School of Dentistry, Kerman University of Medical Sciences, Kerman, Iran
5- MSc Student, Department of Prosthodontics, School of Dentistry, Shahed University of Medical Sciences, Tehran, Iran
6- Oral and Maxillofacial Diseases Research Center AND Department of Oral and Maxillofacial Radiology, School of Dentistry, Mashhad University of Medical Sciences, Mashhad, Iran
Correspondence to: Alireza Sanaei DDS, MSc
Email: hsimple11@gmail.com

30 mm in radiography, it should be considered as an elongated SP. In the vicinity of stylohyoid complex (SC), there are critical structures such as cranial nerves and arteries that, in cases of elongation, create symptoms such as sore throat, difficulty in swallowing, foreign-body sensation, and limited head rotation. The pressure that is placed on the critical structures causes these symptoms.[4] The diagnosis is confirmed by radiological examination.[5,6]

Panoramic X-ray is the most common imaging method used for the detection of an elongated SP.[7,8] However, numerous factors such as magnification in different panoramic machines and the angle between the styloid and skull base can affect the measurements.[9,10] Therefore, three-dimensional techniques such as computed tomography (CT) or cone-beam CT (CBCT) are suggested. These methods can eliminate the errors caused by magnification or superimposition.[11,12] CBCT can be utilized to assess the anatomical structures in the craniofacial region accurately. The length and morphology of SP can be clearly shown on CBCT scans.[13,14]

There are different variants of elongated stylohyoid process regarding its length, thickness, angle, ossification degree, and pattern of calcification. Radiographic classification of the condition helps in diagnosis and treatment approaches.[15] Although the clinical assessment of ES has been described extensively in the articles, the radiographic findings have not been adequately described in the radiological literature.[16]

Patients with ES are often referred to a family physician, neurologist, psychiatrist, neurosurgeon or maxillofacial surgeon, but the successful treatment rate is usually low. One possible reason is that the term "ES" is not well known among physicians and thus often misdiagnosed.[17] Therefore, in patients with unexplained refractory head and facial pain, ES should be considered in the differential diagnosis.

Although a number of radiological studies have examined ES and the elongation of the SP, thus far, no research has been performed comparing ES patients and asymptomatic patients with long SP via CBCT. This study was performed with the aim of evaluating and comparing the calcification patterns of the stylohyoid ligament in patients with ES and patients with asymptomatic elongated SP as detected via CBCT images.

Methods

This retrospective study was performed on the patients referred to a private radiology center in Mashhad for CBCT imaging from 2008 to 2014. The patients had no background systemic disorder. The method of sampling was non-probability sampling based on the purpose. Informed consent was acquired. The Ethics Committee of Mashhad University of Medical Sciences approved this study (Code: 920428). CBCT scans were obtained with the ProMax 3D (Planmeca, Helsinki, Finland) (kVp = 64-68 mA = 8-12).

All CBCT images in which SP was completely imaged and its length was longer than 30 mm on both sides were evaluated. 52 CBCT images in two groups of symptomatic cases with ES and asymptomatic (26 in each group) were enrolled.

Two oral and maxillofacial radiologists who were blind to the patients' status evaluated CBCT scans. Divergent interpretations were discussed with a third radiologist, and final agreement was obtained in all cases.

The measurements were made by Romexis® digital imaging software, version 2.9.2 (Planmeca, Helsinki, Finland) on the central sagittal slice (slice thickness = 0.2 mm).[18]

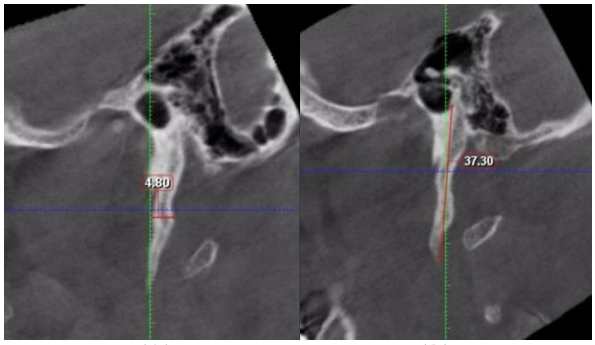
From 52 patients eligible for the study, 26 patients had bilateral long SPs alongside bilateral symptoms such as sore throat, referred otalgia, temporal headache, infraorbital neuralgia, limited head movement/rotation, difficulty in swallowing, and foreign-body sensation in the throat; titled as the ES group (carotid artery syndrome

type). None of these patients had a history of neck trauma or surgery (classic Eagle).

The other 26 cases (asymptomatic group) had CBCT scans for different reasons such as maxillofacial fractures or orthognathic preoperative evaluation. They had bilateral long SP with no symptoms similar to ES.

For each SP, maximum thickness and length, morphology type, and pattern of calcification were recorded based on Langlais et al. radiographic classification.[19] Measurements were repeated at an interval of 2 weeks, and the mean values were registered.

According to Langlais classification, elongated SP has three types. In Type I, elongated process appears as a completely calcified non segmented structure. Type II is a pseudoarticulated type that mineralized stylohyoid ligament is attached to the SP by single-pseudo articulate. In Type III (segmented), the ligament appears as a structure with interrupted mineralized parts. The calcification pattern has also four types including marginal (outline), partial, nodular, and complete calcification. The length of SP was measured from the point that the process is separated from temporal bone up to its tip regardless to its segmentation in sagittal view.[20] SC thickness was measured in the region with the greatest thickness (Figures 1-4).



(A) **(B)**

Figure 1. Measurement of the maximum thickness (A) and height (B) of styloid process (SP)

Finally, the mean length and thickness of the SP, the morphology, and the pattern of calcification were compared between the two groups.

Figure 2. A segmented long styloid process

Good interobserver agreement was obtained between the radiologists (kappa = 0.83). The t-test was used to compare the two groups for variables with normal distribution. Furthermore, the Fisher exact test and chi-square test were used to determine the relationship between different types of calcification pattern and morphology. The level of significance was considered at $P < 0.050$. Statistical calculations were performed with Microsoft Excel 2011 and SPSS (version 16, SPSS Inc., Chicago, IL, USA).

Figure 3. A completely calcified long styloid process (SP)

Results

In our study, the mean age of the patients was 46.11 ± 8.97. It was slightly higher in the symptomatic group than in the asymptomatic

Table 1. Comparison of the length and thickness of the styloid process (SP) according to the side

SP characteristic	Group	N	Mean ± SD	Test result
Length (left)	Asymptomatic	26	38.1700 ± 6.42528	P = 0.314
	ES	26	40.7400 ± 7.61719	
Length (right)	Asymptomatic	26	41.6215 ± 9.16026	P = 0.238
	ES	26	44.62190 ± 11.11813	
Thickness (left)	Asymptomatic	26	3.38080 ± 1.04452	P = 0.001*
	ES	26	4.75420 ± 1.90776	
Thickness (right)	Asymptomatic	26	3.19850 ± 1.12578	P = 0.001*
	ES	26	4.51880 ± 1.64157	
Length (both sides)	Asymptomatic	26	39.89580 ± 7.45124	P = 0.094
	ES	26	42.68100 ± 7.66693	
Thickness (both sides)	Asymptomatic	26	3.28960 ± 0.95292	P = 0.001*
	ES	26	4.63650 ± 1.60358	

*Significant. ES: Eagle's syndrome; SP: styloid process; SD: Standard deviation

group, but the difference between the two groups was not significant (P = 0.760).

Figure 4. A partially calcified long styloid process

The majority of patients in both groups were women. The number of women in the asymptomatic group was 21 (80.8%), and in the symptomatic group, it was 17 (65.4%). The chi-square test showed the same statistical sex distribution in both groups (P = 0.210).

The SP was thicker in the ES group than the asymptomatic group (P = 0.001) (Table 1). The mean length of SP was higher on the right side than the left in the asymptomatic group (P = 0.002) (Table 2). However, the styloid length showed no significant difference among the ES and asymptomatic groups.

The calcification pattern was mostly "partially calcified." A comparison of the two groups in terms of calcification patterns showed no significant difference (P = 0.140). The most common morphology in the asymptomatic and ES groups was "segmented" and "elongated," respectively.

Table 2. Comparison of the length and thickness of the SP according to the study group

Group	N	Mean ± SD	Test result
Asymptomatic			
Length (left)	26	38.17000 ± 6.42528	P = 0.002*
Length (right)	26	41.62150 ± 9.16026	
Thickness (left)	26	3.38080 ± 1.04452	P = 0.381
Thickness (right)	26	3.19850 ± 1.12578	
ES			
Length (left)	26	40.74000 ± 7.61719	P = 0.060
Length (right)	26	44.62190 ± 11.11813	
Thickness (left)	26	4.75420 ± 1.90776	P = 0.282
Thickness (right)	26	4.51880 ± 1.64157	

*Significant. ES: Eagle's syndrome; SD: Standard deviation

Discussion

In this study, the CBCT scans of 52 patients with a mean age of 46 years in two groups-symptomatic (ES) and asymptomatic-were studied. The results of our study revealed that the mean thickness, whether bilaterally or unilaterally, was significantly higher in the ES group than the asymptomatic group.

Oztunc et al.[21] retrospectively studied CBCT scans of 208 patients with neurological symptoms in the maxillofacial region. The structure, length, and medial angulations of SP were measured. Elongated SP was observed at the left side (13%), right side (8%), and bilaterally (33%). Increased prevalence of symptoms was noticed in patients with elongated SP.

In a study of 22 patients with ES, using axial CT, the mean SP length was 41 mm, with no significant difference between the left side and the right side.[22] Similarly, in this study, the mean SP length in the ES group was 42.7 mm with no significant difference at each side.

The most common pattern of calcification was partial with no significant difference between the two groups. In the ES group, the most common morphology pattern on both sides was Type I (elongated or continuous) (50%); whereas, in the asymptomatic group, the segmented type was dominated.

The results of studies performed in this field indicate that the most common type of SP calcification is a matter of debate. A study by Shaik et al.,[23] involved 1162 patients and looked at the prevalence of the elongated SP using panoramic radiography. An elongated SP was observed in 1085 cases, mostly in men and older patients. Type I morphology with outline calcification was reported to be the most common pattern.

In another study,[24] the SP was examined in 20 patients via panoramic and CBCT scans. SP calcification was more prevalent in elderly patients and tended to have an outline calcification pattern, whereas in our study, the most common type of calcification was partial. Another investigation on 207 patients, revealed that that the most common SP morphology observed in panoramic radiography was the continuous type.[9]

A study by More and Asrani,[25] which assessed 500 digital panoramic radiographs, found that Type I morphology and partial calcification were the more common patterns. The results were similar to ours in terms of both morphology and calcification pattern.

A study on 860 panoramic radiographs demonstrated that most of elongated SPs were bilateral and partially or completely calcified.[15]

Interestingly in our study, in patients with ES, a significant difference was observed between the left and the right sides in terms of the morphology and pattern of calcification. One limitation to the present study was that only patients with bilateral long SPs were enrolled; more research is needed to be conducted on other cases.

Conclusions

This study is unique due to a narrow range of the inclusion criteria used such as having the same age and sex distribution in the two groups. It seems that selecting a larger and more comprehensive sample size would help, statistically offering a more valid set of results. The results obtained in this study, which showed a significant difference between the two study groups in terms of morphology type and the thickness of SP, could be useful in differential diagnosis of pain in the face, neck, throat, and tonsils.

Conflict of Interests

Authors have no conflict of interest.

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