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A new viewpoint of Eagle syndrome: The effect of SP medial angulation on symptomatology

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Abstract

Eagle syndrome is an uncommon condition triggered by head movements caused by a long styloid process (SP) or calcification of the stylohyoid ligament, and characterized by symptoms such as sore throat, pain at the root of the tongue, earache, and odynophagia or globus. The aim of this study was to examine the effects of medial angulation of the SP on symptomatology. The study included 49 patients diagnosed with long SP and followed up between January 2018 and January 2020 in the Ear, Nose, and Throat Clinic of Samsun Training and Research Hospital. The medial angle between the SP and the virtual line drawn vertically from the temporal bone attachment point of the SP was measured and recorded. Evaluation was made of 49 patients, comprising 23 (46%) males and 26 (54%) females in the age range of 19-65 years. The SP length ranged from 25mm to 44mm. The presence of at least one symptom was determined in 24 (48%) patients; in 11 (45%) males, and 13 (55%) females. A significant positive correlation was determined between right medial angulation and symptom severity in patients with globus (p:0.01, r:0.5). It should be kept in mind that, in addition to SP length, SP medial angulation could be an additional factor affecting the formation and severity of symptoms in patients with Eagle syndrome.

Keywords: Eagle, styloid process, angulation, severity, globus

1. Introduction

The styloid process (SP) and stylohyoid ligament are thin cylindrical structures, which develop embryologically from the second branchial arch, and are seen inferior and anterior in the lower section of the temporal bone. It is important as muscles and ligaments that play a role in chewing and swallowing are attached to this structure and there is proximity to important anatomic structures (1, 2). The SP generally ossifies between the ages of 5-8 years. However, different degrees of ossification can occur in the stylohyoid ligament and this can cause the development of long SP with calcium deposition at the SP tip (3). Although SP length varies according to individual and ethnic characteristics, normal length has been reported to be 25 mm, and length greater than this is referred to as long SP (4, 5).

Eagle syndrome, first defined by the American otolaryngologist, Watt W. Eagle, is an uncommon condition triggered by head movements caused by calcification of a long SP or stylohyoid ligament, and is characterized by symptoms such as sore throat, pain at the root of the tongue, earache, and odynophagia or globus (6, 7). The reported incidence is 4%, but 4-10% of those with long SP are seen to be symptomatic (3, 8). Opinions have emerged that in addition to SP length at this low rate, medial or anterior angulation could play a role in symptom development, and thus the presence of symptoms with angulation has become a matter of debate. To the best of our knowledge, there is no study in the English literature which has shown a relationship between symptom intensity and the SP angle. The aim of this study was to examine the relationship of SP length and angulation with the presence of symptoms and symptom severity in cases with long SP, and to discuss this in the light of current literature.

2. Material and Methods

The study included 49 patients diagnosed with long SP and followed up between January 2018 and January 2020 in the Ear, Nose, and Throat Clinic of Samsun Training and Research Hospital. Patients were excluded from the study if they were aged <18 years, had any chronic disease, a history of cervical surgery, or any psychiatric disease. ENT examinations and endoscopic nasopharynx and larynx inspections, a complete blood count and routine biochemical analysis including thyroid function tests, were performed on all patients and any patients with any abnormality detected were excluded from the study.

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The tomography images of the patients were examined and the medial angle between the SP and the virtual line drawn vertically from the temporal bone attachment point of the SP was measured and recorded (Fig. 1). Ossified and calcified stylohyoid ligaments were included in the measurements, and the SP length was measured and recorded (Fig. 2).

Fig. 1. Measurement of the SP medial angle: the angle between the SP and the virtual line drawn vertically from the temporal bone attachment point of the SP

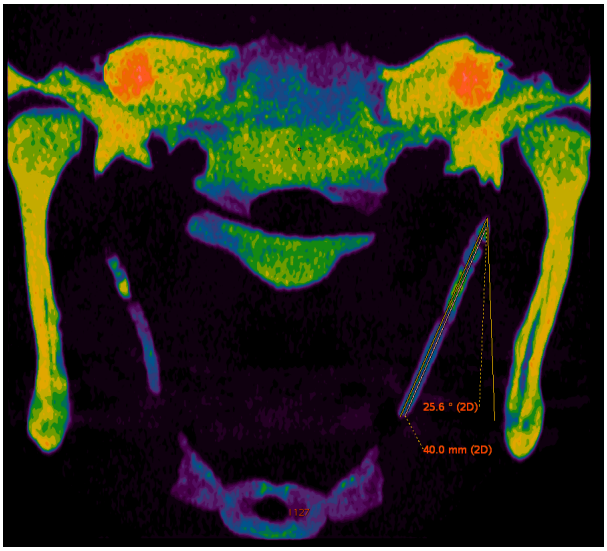

Fig. 2. SP length measurement

The complaints of the patients (earache, sore throat, globus sensation, pain triggered by neck movements) were recorded. The severity of the symptoms was evaluated subjectively using a Visual Analog Scale (VAS) (Fig. 3). The patients were instructed to mark the severity of their symptoms on a line numbered from 0 to 10, where 0 represents no problem and 10, intolerable.

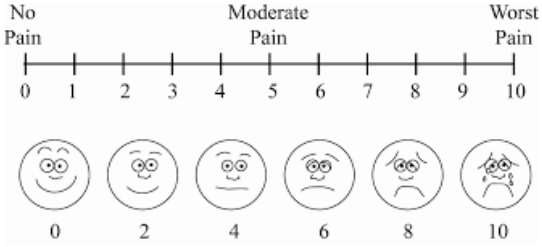

Fig. 3. Visual Analog Scale (VAS): subjective scoring used for the measurement of symptom severity

The patients were separated into two groups according to the presence of symptoms: Group 1 with patients with symptoms, and Group 2 with patients with no symptoms.

2.1. Radiological Imaging

CT scans were obtained in the coronal plane (prone or hanging head position) with 1-mm slice thickness, 0.8-mm reconstruction increment, 16–18-cm FOV, 120 kV, 110 mA and 512×512 matrix size on a Somatom Plus Spiral CT scanner (Siemens AG, Erlangen, Germany). An average of 49 images were obtained from each patient. The images were reconstructed with a real-time 3D (RT3D) interactive volume-rendering module of a Workstation (3D Virtuoso CT/MR Workstation, Siemens AG, Germany), and 3D images were obtained using RT3D render.

2.2. Statistical Analysis

In statistical comparisons between groups, the t-test was used for continuous variables, and the Chi-square test for categorical variables. In all measurements, a value of $p < 0.05$ was considered statistically significant. All statistical analyses were conducted using SPSS 24.0 statistics software (IBM SPSS Statistics for Windows, NY, USA).

3. Results

Evaluation was made of 49 patients, comprising 23 (46%) males and 26 (54%) females in the age range of 19-65 years. The SP length ranged from 25mm to 44mm. The presence of at least one symptom was determined in 24 (48%) patients, 1 (45%) male and 13 (55%) female. The demographic data of the patients and the SP length angulations of the groups are shown in Table 1.

Table 1. The demographic data of patients and the SP length and angulation values of the groups

	Group 1 (symptom + N=24)	Group 2 (symptom- N= 25)	P value
	Mean ±SD	Mean ±SD	
Age	47.208±15.455	41.720±16.789	0.25
Right SP length	32.504±8.585	30.760±5.222	0.704
Left SP lenght	33.533±6.799	28.544±4.331	0.015
Right SP angulation	22.045±4.089	18.364±4.334	0.003
Left SP angulation	21.879±3.292	18.716±4.625	0.014

Mann-Whitney U test

The complaint of earache was reported by 22 (44%) patients. No significant correlation was determined between SP length, SP angulation and symptom severity in the patients with earache.

The complaint of sore throat was reported by 19 (38%) patients. No significant correlation was determined between SP length, SP angulation and symptom severity in the patients with sore throat.

Globus was present in 18 (36%) patients. A significant positive correlation was determined between right medial angulation and symptom severity in the patients with globus (p:0.01, r:0.5). No significant correlation was determined for the other parameters.

The complaint of pain triggered by neck movements was reported by 16 (32%) patients. A significant positive correlation was determined between this complaint and both right and left SP length (right-p:0.01, r:0.5; left-p:0.03, r:0.4). No significant correlation was determined between SP angulation and symptom severity.

4. Discussion

The styloid process provides attachment of the M. stylopharyngeus, M. stylohyoideus, and M. styloglossus, which play a critical role in chewing and swallowing, to the stylohyoid and stylomandibular ligaments (9). It is also in close proximity to important neurovascular structures such as the hypoglossal nerve, carotid artery, and internal jugular vein (10). Originating from pressure on the structures surrounding the long SP, symptoms emerge such as ipsilateral neck pain, and the feeling of a foreign body when swallowing (2, 11). The presence of a long SP together with symptoms, which was first described by Eagle, is now known as Eagle syndrome. Although the incidence shows great variability, it has been reported on average at 4%.

Earache is the most commonly reported complaint (12, 13). In the current study, earache was the most frequently seen complaint, followed by sore throat, globus, and pain triggered by neck movements.

There are studies in the literature showing that there is no correlation between age and gender. SP length and symptomatology (12, 14). In contrast, Pokharelve et al. reported that females have a longer SP than males (15). Bafaqeeh et al. also showed that the SP was longer in females than males and they were more symptomatic (2). In the current study, SP length and angulation were determined to be greater in females than males, and the presence and severity of symptoms were also greater in females.

In the literature, there are various classifications related to SP length and shape. Başekim et al. (16) presented a new classification named LAM (length, angulation, morphology) with the addition of angulation and morphological findings to the previous length and shape-based classifications for SP. In computed tomography (CT) examinations performed for various reasons, 7% of patients have been reported with SP length >4cm, and 77% with SP length 2-4cm. In all the patients in the current study diagnosed with long SP, the SP length was >2.5cm. In 7 (14.2%) patients, the SP length was measured bilaterally >4cm.

The diagnosis of Eagle syndrome is made with the help of radiological examination in addition to symptoms. However, the determination of SP length on direct radiographs is difficult because of the superimposition of anantomic structures. Therefore, 3D CT examination is the gold standard in the measurement of the SP and styloid complex axis, length, and angle (17, 18). It is recommended that CT images be taken in the axial and coronal planes.

Recent studies have shown that the presence of symptoms is associated more with SP angulation than SP length (15, 20). In particular, pain in the throat triggered by neck movements has been associated with irritation of the structures in the parapharyngeal area because of abnormal angulation rather than SP length (4, 20). Yavuz et al. found that the anteroposterior angle had a significant effect in symptomatic patients, whereas no significant difference was observed in mediolateral angulation (21). Some studies have shown that a decrease in medial angulation has a greater effect on the presence of clinical symptoms than anteroposterior angulation (15, 22, 23). Kent et al. reported that the proximity of the SP to the superior constrictor and glossopharyngeal nerve was more important than length and angulation in respect of symptoms (24). In the current study, the relationship was investigated between symptom presence and severity and the SP length and medial angulation. A significant positive correlation was determined between medial angulation and globus sensation, and there was not seen to be any significant correlation between other symptoms and angulation.

The feeling of symptoms unilaterally suggested that the two sides (right and left) of SP angulation could be different. In a study by Kosar et al. (26), a significant difference was determined between the right and left medial angulation in all the patients, whereas Nayak et al. (19) reported that medial angulation was less on the left side. In contrast, Onbaş et al. (27) determined no significant difference between the right and left sides in respect of medial angulation. In the current study, angulation on both sides was found to be greater in the patients with symptoms compared to the patients without symptoms.

The limitations of this study were the relatively low number of patients and the fact that symptoms were evaluated without determination of the side. Therefore, there is a need for further studies with a larger patient population to evaluate symptoms by determining the right and left sides.

In conclusion, it should be kept in mind that SP medial angulation, in addition to SP length, could be an additional factor affecting the formation of symptoms and symptom severity in patients with Eagle syndrome.

Conflict of interest
The authors declared no conflict of interest.

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None to declare.

Authors' contributions
Concept: N.F.T., Design: N.F.T., Data Collection or Processing: N.F.T., U.Y., Analysis or Interpretation: U.Y., Literature Search: U.Y., Writing: N.F.T., U.Y.

Ethical Statement
Approval was obtained from Samsun University Clinical Research Ethics Committee, the study started. The ethics committee decision date is 15/06/2022 and the number of ethical committee decisions is 2022/3/7.

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