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ELONGATION OR ANGULATION OF STYLOID PROCESS: DISCUSSION WITH A CASE REPORT AND REVIEW OF THE LITERATURE

Styloid Çıkıntının Angulasyonu veya Uzaması: Vaka Sunumu eşliğinde Literatür Tartışması

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ABSTRACT

Styloid process of temporal bone is a pointed projection from the petrous part of temporal bone which ranges with an average of 25 mm in length, and any process longer than 30 mm is defined as the "Elongated Styloid Process". In asymptomatic cases, diagnosis is usually made with routine radiological examination, but morphological aspects of styloid process can be evaluated with different imaging modalities. Although digital panoramic radiographs are sufficiently accurate for the diagnosis of elongated styloid process, multislice computed tomography scan with 3D reconstruction can further assist in determining the actual length and correct relationship with the surrounding adjacent anatomical structures.

The aim of this case report is to investigate the morphological characteristics of an unilateral elongated styloid process using both conventional and multidetector computed tomography (MDCT) on a 55 year old asymptomatic male patient and to review the literature.

Keywords: elongated styloid process, multidetector computed tomography, structural deviation

ÖZ

Temporal kemiğin styloid çıkıntısı, temporal kemiğin petroz kısmından başlayan, ortalama 25 mm uzunluğunda olan sivri bir çıkıntıdır ve 30 mm' den uzun olan styloid çıkıntılar "Uzamış Styloid Çıkıntı" olarak tanımlanır. Asemptomatik olgularda genellikle rutin radyolojik inceleme ile tanı konmaktadır, ancak styloid çıkıntının morfolojik özelliklerinin değerlendirilmesi farklı görüntüleme yöntemleri aracılığıyla yapılabilmektedir. Dijital panoramik radyografiler styloid çıkıntının doğru olarak tanılanması için yeterli olmasına rağmen, multidetektör bilgisayarlı tomografi (MDCT) taraması ile 3D rekonstrüksiyon yapılarak styloid çıkıntının gerçek uzunluğu ve komşu anatomik yapılarla ilişkisi daha doğru değerlendirebilir.

Bu olgu sunumunun amacı, 55 yaşında asemptomatik erkek hastada tek taraflı uzamış styloid çıkıntının morfolojik özelliklerini hem konvansiyonel hem de multidetektör bilgisayarlı tomografi (MDCT) kullanılarak araştırmak ve literatürü gözden geçirmektir.

Anahtar kelimeler: Uzamış styloid çıkıntı, multidetektör bilgisayarlı tomografi, yapısal anomali

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INTRODUCTION

The styloid process (SP) of temporal bone is a thin, cylindrical, sharp osseous projection which is located on the inferior aspect of temporal bone, posterior to the mastoid apex, anteriomedial to the stylomastoid foramen and lateral to the foramen jugulare and canalis caroticus. It is a component of human stylohyoid chain, which also includes the stylohyoid ligament and the cornu minus of the hyoid bone.[1,2] SP provides attachment to M. stylopharyngeus, M. stylohyoideus, M. styloglossus, stylohyoid and stylomandibular ligaments which have critical roles in mastication and swallowing.[3] Also, its tip lies in close proximity to important neurovascular structures, including hypoglossal nerve, external-internal carotid artery, and internal juguler vein.[4] Thus, any abnormalities of SP can have clinicopathological outcomes, such as severe pain in head and neck region especially observed as ear and throatache, dysphagia, sense of a foreign body in throat and limitation in mouth opening.[5,6,7]

Even though the exact etiology of SP elongation is still obscure, trauma, surgery, endocrine disorders, embriological mesenchymal remnants, intraligamentary metaplasia, osseous tissue growth and mechanical stress have been suggested among the factors in calcified hyperplasia of the SP.[1,7] Because of cartilaginous content of the ligament itself, the stylohyoid chain may present various degrees of ossification[8], and this calcium deposition on the tip of the process results with the elongation of the SP.[1] After entire ossification of the stylohyoid ligament, a solid stylohyoid chain results with many variations including incomplete ossification, segmentation, and diversities in thickness and/or angle. In some cases, the stylohyoid ligament can be divided into two or more supernumerary bones articulated through fibrous or cartilaginous joints, with more or less articulated osseous chains.[9] According to the type of elongation, Langlais[10] has classified elongated SP as Type I, elongated; Type II, pseudoarticulated; and Type III, segmented. However, other classification methods which evaluate the pattern, length and the angle of the stylohyoid chain complex have been used in literature.[1,7,10-14]

The length of SP can vary depending on individual factors of different populations; but in the literature, the normal length of this anatomical structure has been reported between 20-30 mm and any process longer than 30 mm is defined as the "Elongated Styloid Process (ESP). Although subsequent studies have observed lengths between 15.2 and 50 mm, most authors agree that SPs greater than 30 mm in length should be considered abnormal.[15-17] This abnormality is observed in 4% of the general population and only 4-10% of this group is symptomatic. Rarely, ESP is associated with clinical symptoms of neck and orofacial pain due to compression of surrounding anatomical structures, and the pain is usually referred to the ear, especially during swallowing. Also, vertigo attacks during sudden contralateral head movements, occasional tinnitus, hypersalivation and episodic pain with muscular spasm may be observed[1], and in such cases, the pathology is characterized as “Eagle’s syndrome”. Observation is the treatment of choice for asymptomatic cases, but for symptomatic patients, the treatment plan includes both medical and surgical options.[18] In addition to Eagle’s syndrome, the calcified stylohyoid ligament and the ESP can be associated with other pathologies such as the carotid artery syndrome, stylohyoid syndrome, and pseudostylohyoid syndrome.[19-21]

In asymptomatic cases, ESP is usually detected during routine dental radiographic examination using panoramic radiographs. However, superimposition of the mandibular bone and teeth on SP reduces the quality of the image on conventional radiographs. At this point, multidetector computed tomography

(MDCT) and 3D reconstruction are considered as the best imaging modality in order to accurately assess the location and morphological features of this pathology, especially before any surgical planning or intervention.[22,23] This paper reports an asymptomatic male patient with unilateral ESP and presents the morphological details of the pathology using MDCT.

CASE REPORT

A 56-year-old male patient was referred to the outpatient clinic of Ege University, School of Dentistry, Department of Oral and Maxillofacial Radiology for prosthetic rehabilitation. The patient's medical history was noncontributory. After receiving informed consent; extraoral examination revealed no abnormalities; besides the patient had no complaints on his neck region, such as pain, foreign body sensation, dysphagia or visual impairment. Intraoral examination disclosed missing teeth #14, 15, 17, 22-25, 36, 37, 46, 47, distoproximal caries in #44, and generalized periodontitis. Conventional panoramic radiographic examination revealed a right styloid process with two articulated supernumerary bones protruding to the basal region of mandible, while left styloid process was observed as normal (Figure 1).

Fig. 1: Panoramic view of the patient

In order to accurately assess the location and morphological features of the pathology, further detailed radiographic analysis with MDCT was required. Computerized tomography (CT) scans were obtained with a 64-MSCT scanner (Discovery HD 750 dual energy CT, GE, Milwakee, USA) using 0,625 mm slice thickness, 180 cm FOV, 110 kV, 220 mA scanning parameters. Besides, 3D reformatting of images was also prepared using axial dicom images on recontruction software integrated into the PACS Workstation (syngo, Siemens).

In 3D-CT examination, the length of SP were measured by rotating the image until the long axis of the SP was parallel to the viewing to provide an unobstructed view. According to this technique, the length of the right styloid process from the attachment point of the SP to the temporal bone to the tip of the SP was measured as 80.6 mm while left SP was measured as 27.0 mm in length. Langlais classification concluded that the right styloid process was a Type 3 ESP, while it was normal for the left side (Figure 2).

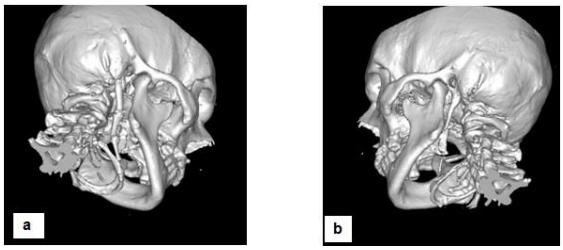

Fig. 2: Multidedector computed tomography images; **a, b.** Three-dimensional (3D) of computed tomography (CT) scan shows length measurement and Type 3 ESP according to Langlais classification.

The medial-lateral angle (MLA) was defined as the angle between the line connecting the base of the SPs and long axis of the SP on the anteroposterior view of 3D-CT images and MLA of elongated SP was measured as *65.95°* (Figure 3a) while it was *59.31°* for the normal SP (Figure 3b).

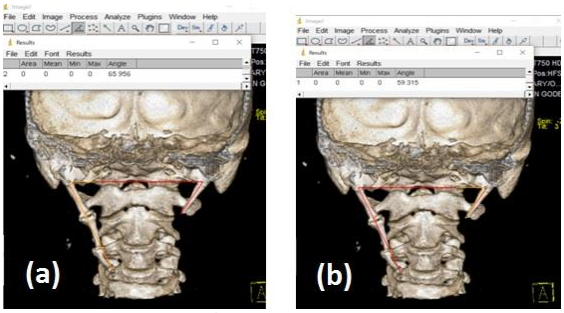

Fig. 3: Coronol view of three dimensions (3D) reconstructions showing MLA of elongated and normal styloid process (a) Medial angulation of the elonge SP (b) Medial angulation of the normal SP

The anterior-posterior angle (APA) was determined as the angle of intersection between a line tangential to the tip of the mastoid process and the axis of the SP on the lateral view of 3D-CT images and was measured as *69.06°* (Figure 4a). APA of normal SP was measured as 80.46 mm (Figure 4b).

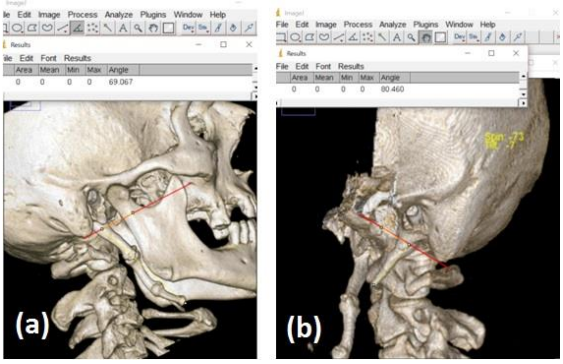

Fig. 4: Sagitall view of three dimensions (3D) reconstructions showing APA of elongated and normal styloid process (a) Anterior angulation of the elongated SP (b) Anterior angulation of the normal SP

All measurements related APA and MLA on right (elongated type) and left side (normal type) were summarized in Table 1. During the evaluation process, the mastoid tip was chosen as a reference landmark because it was reliably visualized in all lateral views.

Table 1: Angle and length measurements of normal and elongated styloid process made on computed tomography

	Right	Left
Length (mm)	80.6 mm	27.0 mm
Anteroposterior angle	69.06°	80.46°
Mediolateral angle	65.95°	59.31°

DISCUSSION

In this report, we describe an unusual case of an unilateral ESP with a length of 80.6 mm. The normal length of the SP ranges from 25 mm to 30 mm.[24] Although most authors agree that SPs greater than 30 cm in length should be considered abnormal[25], other studies claim that a length between 15.2 mm and 47.7 mm could be considered as normal.[26] The variations between the measurements of SPs in abovementioned studies may originate from utilization of various imaging modalities, which fall short to demonstrate the morphological features of SP accurately.

In order to provide an accurate and adequate measurement, the full length of SP must be visualized.[1,27] For this purpose, several imaging modalities including conventional lateral and anteroposterior (AP) views of head and neck radiograph, orthopantomography, CT and more recently, cone beam computerized tomography (CBCT) can be used. Panoramic radiography is frequently selected because it is a regular radiographic examination method requested by dental practitioners to inspect the structures of the maxillofacial complex, including the full length of SP. However, the visualization is not easy on panoramic radiographs due to superimposed neighbouring anatomical structures.[1,8,24] Recent studies showed that Multislice CT scans with 3D reconstruction can overcome the limitation of 2D imaging modalities and provide the most accurate information about the course, morphometric properties (length, angle, etc) of SPs and their relationship with adjacent anatomical structures.[5,28] Furthermore, 3D-CT reconstruction has been advocated as the gold standard for examination of the SP.[29]

In previous studies using CT scans, the length of the SP in millimeters was measured in standard coronal planes of CT.[5,6] However, CBCT images which were rotated sagitally have been utilized in order to investigate the whole structure of SP on a single image.[7] Similarly, in the present study, the image plane was rotated until an angle demonstrating the entire long axis of the SP was obtained to provide an unobstructed view of the anatomically complex SP, and afterwards, the length of the SP in millimeters was measured on 3D-CT images. On the other hand, this methodology may present a limitation: as Kent *et al*.[28], have suggested rotating the image until the long axis of the SP was parallel to the viewing may account for longer SP values than the others reported in the literature.

When ESP is associated with clinical symptoms of neck and orofacial pain, it is referred as "Eagle's syndrome". Although this syndrome is thought to be caused by ESP, the presence of ESP is not a pathognomonic finding, because many patients with incidental findings of ESP are asymptomatic[1], as observed in our case. At this point, another morphometric parameter that requires further evaluation is the angle of the SP.[7,16,30] This concern has been already declared in the literature, and the potential association between the angulation of the SP and clinical symptoms has been emphasized, as presented in Table 2.

Table 2: A summary of studies investigating the angulation and length of the styloid process of the patients with/without symptoms using different imaging modalities.

					Angle (APA)		Angle (MLA)	
Author/year	**Complaints**	**Patient**	**Average of Length (sympt / asympt)**	**Technique**	**Mean value (sympt / asympt)**	**Reference line**	**Mean value (sympt / asympt)**	**Reference line**
Onbaş 2005	asymptomatic	283	26.8 mm	MDCT	93.5°	the skull base line connecting the nasion and the opisthion - axis of the SP	72.7°	the line connecting the base of the SPs-the axis of the SP
Başekim 2005	asymptomatic	138	28.3 mm	3D-CT	-	-	69.4°	the line connecting the base of the SPs-the axis of the SP
Ramadan 2007	asymptomatic	100	27 mm	3D-CT	63.7°	Mc Rae's line-axis of the SP	72.7°	the line connecting the base of the SPs-the axis of the SP
Yavuz 2008	symptomatic+ asymptomatic	30+31	50 mm /27 mm	lateral skull+ Towne's rad.	33.6-36.7 /21.4-18.5	vertical line*-axis of the SP	14-18.1/15-16.3	vertical line-axis of the SP
Okur 2014	symptomatic+ asymptomatic	100+100	40.5 /39 mm	3D-CT	16.5/15	the line passing from cranial base of the SP and axis of the SP	22.60/20	vertikal line-axis of the SP
Kent 2014	symptomatic+ asymptomatic	37+30	48/40 mm	3D-CT	66/66	the line tangential to the mastoid proc.-axis of the SP	62/70	the line connecting the base of the SPs-the axis of the SP
Burulday 2017	symptomatic+ asymptomatic	25+25	40.3/16.8 mm	3D-CT	73.2/74	the line tangential to the mastoid proc.-axis of the SP	69.2/66.5	the line connecting the base of the SPs-the axis of the SP
Kumar 2017	normal+ elongated type	96 skulls	36/14 mm	goniometer	57.7/66.3	frankfurt line-axis of the SP	73.3/74.3	the line connecting the base of the SPs-the axis of the SP
Eraslan 2017	asymptomatic	125	31.3 mm	3D-CTA	57.3	angle of the calcification of the SHC with the sagittal axis of the 3D-CTA image	72.7	the line connecting the base of the SPs-the axis of the SP
Buyuk 2018	asymptomatic	1000	34.5 mm	3D-CBCT	72.2	Skull base- axis of the SP	71.2	the line connecting the base of the SPs-the axis of the SP

Although the normal length and angles of the SP varies considerably according to the different measurement techniques and reference points, most of the studies showed that abnormal angulations of SP rather than elongation are responsible for clinical symptoms. However, previous studies have not reached a consensus on which angulation is the most important.[16,21] Yavuz *et al.*[21] investigated the effect of the angulation of SP on Eagle syndrome (ES) using 2D radiographic techniques and found a significant difference between symptomatic patient and control group in anterior-posterior SP angulation (APA), but there were no significant differences between 2 groups with respect to medial-lateral SP angulation (MLA). However, recent studies using 3D-CT reconstructions of the SP showed that rather than the anterior angulation or the length of SP, the decreased medial angulations of SP might more strongly influence the presence and severity of clinical symptoms.[4,16,29,30] A recent morphological study of SP on human skulls also revealed that the angulations were decreased in ESP as compared with normal type of SP.[4] According to that study, the mean anterior angle of the ESP was 57°, while for the normal type, it was 66°. Burulday *et al.*[30] used 3D CT images to evaluate the importance of MLA in symptomatic eagle syndrome and found that the symptoms are more intense when the angle is smaller. In such cases, the tip of the SP approaches medially in the coronal plane and causes more intense clinical complaints by compressing the surrounding tissues.[7,28,29,31] Kent *et al.*[28] evaluated the SP anatomy in ES and asymptomatic control populations using 3D CT reconstructions. They revealed that the length or angulation of the SP in ES may be

less important than its approximation to the adjacent soft tissue structures, such as the superior constrictor muscle and glossopharyngeal nerve. Rather, the minimum distance from SP tip to the tonsillar fossa was found to have a much greater effect size than any other measured variable.[28] Additionally, a long stylohyoid complex with a narrow anterior sagittal angle and/or a narrow transverse angle may irritate the adjacent anatomical structures and result with clinical complaints due to the compression of the adjacent structures.[7]

Several classification has been reported previously according to the morphology, length and angle of the styloid apparatus.[1,7,10-14] Viela suggested a classification based on the morphogenesis of the skull, while Langlais *et al.* proposed a radiological classification in three types.[10,11] The broadest classification cited in the literature was defined by MacDonald-Jankowski *et al.*[12] who used embryological considerations to describe 12 subtypes according to the ossified segment part of the ligament. Regarding angulation of SP, Buyuk *et al.*[7] classified the transverse angles into three groups; <65° were determined as narrow, 65°-75° as normal, and >75° as wide angles. Similarly, sagittal angles were divided into three groups; <60° were defined as narrow, 60°-70° as normal, and >75° as wide anglesi.[7] However, there is only one study in the literature including both length, angulation and morphological findings of the SP to the classification, and this classification was termed as "LAM "(length, angulation and morphology).[1] According to LAM classification; assessment ranges related length, angulation and morphology are yielded in the below.[1]

L: Length of the SP

1. Short (<2.00 cm)
2. Long (2.00–4.00 cm)
3. Elongated (>4.00 cm)

A: Angulation of the SP

1. Narrow (<65.0°)
2. Normal (65.0–75.0°)
3. Wide (>75.0°)

M: Morphology of the SP

0. Absence of SP
1. Normal appearance of SP
2. Other morphological findings (absence of the proximal part of the SP, duplication of the proximal part of the SP, bent SP, segmented SP, pseudoarticulated SP, etc.)

We think that this classification seems clinically handier and more pertinent to make a comprehensive evaluation, and according to this classification, right SP of our case was L3, A12, M2; while it was L2, A1, M1 for the left side.

In our case, an asymptomatic unilateral ESP was reported. The length of the right SP was measured as 80.6 mm, while left SP was measured as 27.0 cm in length. Besides, the angulations of anterior-posterior and medial-lateral ESP were 69.06° and 65.95° respectively and were within the normal range reported in the literature. Although the length of the SP was extremely longer than the mean levels, our patient presented no clinical symptoms. On the other hand, the angulations of the processes were narrower in our case as compared with the symptomatic ones reported in the literature, and this may be considered as the reason of the absence of clinical complaints.

CONCLUSION

Up to date, severe clinical consequences have been associated with ESP. However, our case approved the results of previous reports and suggested that the angulations rather than the length of ESP may be responsible for patients' clinical complaints. In order to appropriately evaluate the length, angulations and other morphological characteristics of SP, MDCT may be offered as an effective radio diagnostic method.

Compliance with ethical standards

Conflict of Interest: The authors declare that they have no conflict of interest.

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All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1964 and later versions. Informed consent was obtained from all patients for being included in the study. Informed consent was obtained from the patient for being included in the study.

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