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Morphological and morphometric study of elongation of the styloid process with calcification of the stylohyoid ligament in a human skull: a case report

Estudo morfológico e morfométrico do alongamento do processo estiloide com calcificação do ligamento estilo-hioideo em crânio humano: um relato de caso

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Abstract

Elongation of the styloid process (i.e., Eagle syndrome) can compress nearby structures, causing varied symptoms. Studies on the morphological and morphometric aspects of the elongation of the styloid process are scarce, especially in the Brazilian population. Therefore, the present study reports a case of elongation of the styloid process with calcification of the stylohyoid ligament bilaterally. These structures on both sides were two-fold longer than needed to meet the anatomical criterion of the syndrome. Morphological and morphometric aspects and the clinical implications of the structures were also discussed.

Keywords: Anatomy; Temporal bone; Surgery; Measurement equipment; Anatomic variation.

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Resumo

O alongamento do processo estiloide pode comprimir estruturas próximas causando um conjunto de sintomas que caracterizam a Síndrome de Eagle. Nota-se uma escassez de estudos, em especial na população brasileira, sobre os aspectos morfológicos e morfométricos do alongamento do processo estiloide. Sendo assim, o presente estudo relata um caso de alongamento do processo estiloide com calcificação do ligamento estilo-hioideo bilateralmente. O comprimento dessas estruturas em ambos os lados excedeu o dobro do necessário para atender ao critério anatômico de existência da síndrome, bem como detalhou os aspectos morfológicos e morfométricos e as implicações clínicas das estruturas.

Palavras chaves: Anatomia; Osso temporal; Cirurgia; Equipamentos de medição; Variação anatômica.

INTRODUCTION

The temporal bone is characterized by bony projections, depressions, foramina, and bony processes. One of the main bony projections is the styloid process (SP). Derived from the Greek “stylosoide” it is named for its spearhead or stake-like shape[1].

The SP is positioned between the posterior edge of the neck of the mandibular condyle and the anterior edge of the mastoid, and projects caudally, anteriorly, and medially from the base of the skull[2]. The normal length usually ranges between 25 mm and 30 mm; however, this length can vary among individuals and even between the two sides of the same individual. Moreover, the SP is considered elongated when extensions exceed this mean length[3]. Due to its peculiar anatomical configuration, the SP supports three muscles and two ligaments that keep the airway passage in the pharynx[2]. Elongation of the SP can result from ossification of the stylohyoid ligament or excessive SP growth (or both); the former can be triggered by factors that stimulate osteogenesis, such as cervicopharyngeal trauma[3].

Computed tomography of the head and neck is considered the gold standard for identifying elongation in living individuals, which can differentiate the excessive growth of the SP from the calcification of the stylohyoid ligament[3]. However, the identification in post-mortem studies can be achieved by observing bony thickening at the insertion point of the calcified stylohyoid ligament.

Eagle syndrome (ES), or elongated SP syndrome, was described in the literature in 1937 by an otolaryngologist named Watt Weems Eagle[2]. The author described, in a case report, a female individual experiencing throat and neck pain caused by the elongation of the SP and calcification of the stylohyoid ligament[2].

Thus, ES comprises a set of rare symptoms triggered by the compression of structures near the growth of the SP or calcification of the stylohyoid ligament (or both). The impaired functioning of these structures causes pain in the pharyngeal and cervical regions[1]. Moreover, the internal carotid artery influences the SP anatomy, especially due to the proximity between these

structures. Thus, their association may cause a rare vascular variant of ES (i.e., stylocarotid syndrome)[2].

From this perspective, the present study aimed to report a case of elongated SP with bilateral calcification of the stylohyoid ligament, describing its morphological and morphometric aspects and reviewing the key characteristics of ES.

CASE REPORT

The present study was conducted at the Laboratory of Anthropology and Forensic Osteology of a Higher Education Institution. A bilateral elongation of the SP and calcification of the stylohyoid ligament were observed in a skull during the routine washing and drying process of the skeleton collection for storage (Figures 1A and 1B). The studied skeletal belongs to an 84-year-old male individual.

Figure 1. Elongated styloid process (SP) with bilateral calcification of the stylohyoid ligament. **A.** left lateral view; **B.** right lateral view; **C.** length of the SP with ligament calcification.

Regarding morphometry, the following measurements were taken (Figures 1 and 2): the length of the SF and calcified stylohyoid ligament (distance between the bases and tips of the elongated SP) using a digital caliper; distance between the bases of the SP using a digital caliper; distance between the tips of the SP using a digital caliper; measurement of the anterior angulation (between the Frankfurt line and the longitudinal axis of the SP, crossing the tip of the SP) using ImageJ; measurement of the medial angulation (line crossing the bases and the longitudinal axis that cross the tip of the SP) using ImageJ; measurement of the thickness (anteroposterior and horizontal dimensions) at the base, middle region, and tip of the SP using a digital caliper.

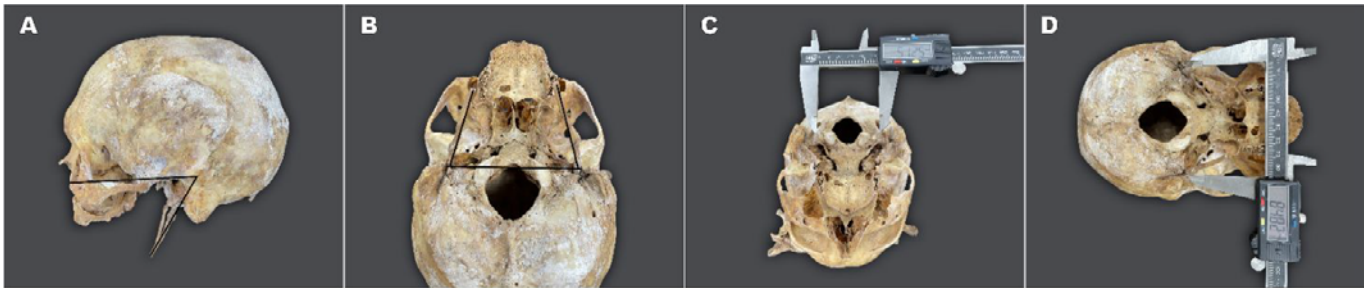

Figure 2. Elongated styloid process (SP) with bilateral calcification of the stylohyoid ligament **A:** measurement of the anterior angulation (between the Frankfurt line and the longitudinal axis crossing the tip of the SP); **B:** measurement of the medial angulation (line crossing the bases), and the longitudinal axis (crossing the tip of the SP); **C:** distance between the bases of the SP; **D:** distance between the tips of the SP.

The measurement of the elongated SP length was 61.91 mm (left) and 66.36 mm (right). The measurement from the base to the bony thickening was 30.11 mm (left) and 20.37 mm (right), while the measurement from the bony thickening to the tip was 41.54 mm (left) and 36.25 mm (right). The distance between the bases of the SP was 84.78 mm, and the distance between the tips of the SP was 46.55 mm. The measurement for the anterior angulation was 55.53°; the medial angulation was 73.56° (right) and 76.18° (left). The thickness of the SP at the base, middle, and tip were 3.68 mm, 5.25 mm, and 2.68 mm on the right (respectively), and 4.31 mm, 7.13 mm, and 2.88 mm on the left (respectively) (Figures 1 and 2).

Regarding the SP, the elongation on both sides was two-fold longer (over 60 mm) than the normal length (25 mm to 30 mm). Additionally, the elongation of SP originated from different processes: the calcification of the stylohyoid ligament (right side), and an elongation caused by abnormal growths and the calcification of the stylohyoid ligament (left side).

DISCUSSION

First described by surgeon Pietro Marchetti in 1652, the SP is a cylindrical and thin projection whose ossification starts at the end of gestation and continues until the first eight years of life[2]. Additionally, the SP is in the anteromedial region to the mastoid process of the temporal bone, from which three muscles (styloglossus, stylohyoid, and stylopharyngeus) and two ligaments (stylohyoid and stylomandibular) originate[4]. Furthermore, the SP relates caudally, medially, and anteriorly to the carotid arteries, internal jugular vein, and cranial nerves VII, IX, X, and XII, respectively. The elongation and compression of these structures cause ES[5].

Elongated SP has an incidence ranging from 4% to 28% of the population and is more predominant in female individuals (3:1 ratio), especially in those aged from 30 to 50 years[4,5]. However, symptoms are prevalent in only 0.16% of individuals with this anatomical variation[6].

In 1937, Eagle proposed two syndromes associated with elongated SP: the classic and stylocarotid syndrome[7]. The classic syndrome can be triggered by post-tonsillectomy scarring or trauma in the pharyngeal region. This process can lead to reactive hyperplasia and metaplasia, overgrowth of the elongated SP, or post-trauma ossification of the stylohyoid ligament (mainly in the mesenchymal tissue), compressing the cranial nerves (especially the glossopharyngeal nerve)[1]. Histological analysis of the SP may reveal deep staining reversal lines along its length. This alteration is associated with multiple recurrent microfractures due to trauma or traction, the differentiation process of mesenchymal tissue into bone, and dystrophic bone deposition[7,8]. On the other hand, stylocarotid syndrome is characterized by direct compression of the carotid arteries by the elongated SP, arterial dissection, or thromboembolism (secondary to injury to the arterial wall)[9]. Stenosis of these arteries, especially the internal carotid artery, is associated with transient ischemic attacks and pseudoaneurysms due to repetitive traumas[6].

The ES is commonly associated with compression of the cranial nerves, resulting in neuropathic pain in the pharyngeal region (ipsilateral tonsillar fossa and cervical area) or the ear[9]. This pain is reported unilaterally or bilateral (rare) and can be exacerbated by head movement, swallowing, and yawning and normally does not improve with medication[1,7]. Other symptoms may include a feeling of a foreign body in the hypopharyngeal region (in 55% of cases), odynophagia, dysphagia, headache, and tinnitus[1,7]. Conversely, the stylocarotid syndrome, leads to syncope, affects perivascular sympathetic fibers, and results in Horner syndrome[4,10].

Furthermore, ES may present as facial paralysis on rare occasions. Facial paralysis occurs due to the compression of the facial nerve (VII cranial nerve), potentially damaging different regions. Consequently, damage to the supranuclear region can cause central facial paralysis, while infranuclear damage is characteristic of peripheral paralysis[11]. However, due to the compression of the extratemporal part, taste and the functions of the sublingual, submandibular, and lacrimal glands remain preserved[11].

From the analysis of ES, differential diagnoses are essential to consider, especially during conditions that cause cervicofacial pain. Among these, differential diagnoses include temporomandibular joint dysfunction, trigeminal neuralgia, cervical masses, poorly fitted dental prostheses, esophageal diverticula, otitis media, and externa, mastoiditis, glossopharyngeal neuralgia, foreign bodies in the pharynx, and tumors in the pharynx or at the base of the tongue[1,8].

Besides the clinical picture, radiological findings and their potential causes can establish the ES diagnosis; they can classify ES as an etiology to be investigated among secondary headaches. The syndrome can also be associated with a clinical history of prior tonsillectomy; however, this association is not mandatory for diagnosis[12]. The gold standard examination for assessing ES is a three-dimensional non-contrast computed tomography of the head and neck, aiming to analyze the parameters of length and thickness of the SP[7].

Furthermore, a reliable diagnostic test can be requested by infusing 3 mL of 2% lidocaine. SE is characterized when the pain is reduced; therefore, this test predicts a good response to styloidectomy[13].

Conservative treatment for pain in SE is the first-line approach and typically precedes surgery, especially when the individual refuses it[13]. The initial preferred drugs are anticonvulsants (e.g., carbamazepine and gabapentin), which may be combined with selective serotonin reuptake inhibitors, nonsteroidal anti-inflammatory drugs, or opioids, depending on the pain intensity[14].

However, styloidectomy via transcervical or transoral is the most effective surgery in individuals with persistent symptoms, vascular involvement, and neurological sequelae[13,14]. The transoral is safer since the other has low surgical exposure, lack of control over the main vessels in the region, and possible bacteriological contamination of deep spaces[15]. In addition, a study of 103 individuals with ES found that this procedure was curative in 46% of cases, effective in 34%, and ineffective in 20%[15].

Last, despite the extensive literature on the clinical aspects of SE, the morphology and morphometry of the SP addressed in this study should be further studied, particularly in the Brazilian population. Considering the importance of this topic in surgeries and imaging, future studies must consider the Brazilian population and include variables, such as age, biological sex, ancestry, and height. Morphological and morphometric aspects of the SP are important because they may vary across different populations and regions within the country, depending on the ethnic group. Therefore, future studies are needed to comprehensively understand the anatomy and its variations in the Brazilian population.

CONFLICTS OF INTEREST

The authors declared no conflicts of interest.

AUTHOR CONTRIBUTIONS

Pacífico FA: conceptualization, data curation, investigation, methodology, project administration, resources, supervision, writing of the original draft, and review and of the original draft. **Saad BS**: writing of the original draft, and review and of the original draft. **Valente TJMBSV**: writing of the original draft, and review and of the original draft. **Galvão IFG**: writing of the original draft, and review and of the original draft. **Lages DB**: writing of the original draft, and review and of the original draft. **Albuquerque LCA**: writing of the original draft, and review and of the original draft. **Campina RCF**: resources, supervision, and writing (review).

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