Case Report

Ossified stylohyoid ligament: A case report

Dr. Prakash Babu B^{1*}, Dr. Huban Thomas², Henry T³, Dr. Punnose³

¹Associate Professor, ²Senior grade Lecturer, ³Post graduate

Anatomy Department, Kasturba Medical College, Manipal University, Manipal, Karnataka, India ^{*}Corresponding author email: **billakantibabu@yahoo.co.in**

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Abstract

Stylohyoid ligament, lesser cornu of hyoid bone and styloid process form stylohyoid complex derived from cartilage of 2nd pharyngeal arch. The anatomy of the ossified stylohyoid ligament and styloid process has immense embryological and clinical importance. The present report is of a fully ossified stylohyoid ligament on right side in an adult male cadaver. On the left side the stylohyoid ligament was remain unossified. Stylohyoid ligament in adults may retain its embryonic cartilage and thus has the potential of ossification. Abnormalities of stylohyoid complex may compress nearby neurovascular structures leading to mild to severe symptoms, such as foreign body sensation and pain in throat, etc. Dentists, otolaryngologists and anesthetists should be aware of the natural variations of the styloid process.

Key words

Styloid process, Stylohyoid ligament, Ossification, Variations.

Introduction

The styloid process is a pointed piece of bone below the ear [1]. Lesser horn of hyoid, stylohyoid ligament (SL) and styloid process (SP) of temporal bone form stylohyoid complex (SC) and are developed from cartilage of second pharyngeal arch [2, 6]. According to Holinshed the SL attached to lesser horn of hyoid represents unossified intermediate part of cartilage of 2nd arch [3]. Neurovascular structures that lie in close proximity to the SL are internal jugular carotid facial, vein, arteries; and glossopharyngeal, vagus, accessory and hypoglossal nerves [1, 2, 4, 5]. Abnormal or excessive ossification of components of SC might result in many variations which may be in the form of incomplete ossification, thickness variations, segmentation [7, 8]. These abnormalities may compress neighboring blood vessels and nerves and is responsible for the

stylohyoid syndrome [9-11]. When the whole SL ossifies solid SC occurs, occurrence of which is rare [12]. Symptoms produced by elongated SP and structural changes in SL include foreign body sensation in the neck, hypersalivation, difficulty in deglutation as reported by Eagle in 1937 as 'Eagle's syndrome' [2]. Elongation of SP on either side was noted in only 50% of patients having Eagle's Syndrome and only half of them had bilateral symptoms as reported by Harma [13]. Keur, et al. [14], assessed 1135 edentulous patients clinically and radiologically to establish relationship between the cause and symptoms. Bafaqueeh SA [15] described two different groups of patients according to symptoms. First one 'classical type' with pain in pharynx and another one 'carotid artery type' with repeated syncopal attack and pain in the same side of the neck. Keur, et al. [14], reported a case in close proximity of laryngopharynx due to irritation of glossopharyngeal nerve. The anatomy of the ossified SL and SP has immense embryological and clinical importance.

Case report

During routine dissection for undergraduate medical students, we observed a case of the fully ossified unilateral stylohyoid complex in a male cadaver on the right side (**Figure - 1**). The length of the stylohyoid complex (styloid process with the ossified stylohyoid ligament) from base of styloid process to lesser cornu of hyoid bone was 43 mm. On left side, the stylohyoid ligament remained unossified.

Discussion

Variations in the length of the SP and SL are of immense importance anatomically and clinically. Extending from base of skull to lesser horn of hyoid is the SC which has four distinct segments: tympanohyal, stylohyal, ceratohyal and hypohyal and are developed from cartilage of 2nd arch [16]. The tympanohyal and stylohyal segments develop into SP which generally fuses at puberty whereas hypophyal segment develops into lesser horn of the hyoid. Ceratohyal segment connects these two structures, which normally comprises

of dense fibrous tissue in adults but may retain its embryonic cartilage and thus have the potential for ossification which causes variations [7-9, 17, 18]. Ossified SL parts may merge or leave gaps in between them. This ossification may commence where the SL attached to tip of SP due to unknown reasons, such as bony growth or trauma during tonsillectomy [6]. Pathogenesis of elongation and mineralization of the SC is poorly understood as cited in literature [14]. Some authors are of opinion that elongation of SC may be due degenerative changes occurring along with the spondylosis of cervical vertebra [19], which was contradicted by others as a non-age dependent degeneration [20].

<u>Figure – 1</u>: Ossified stylohyoid ligament. PB: Posterior belly of digastric; OSL: Ossified stylohyoid ligament; SP: Styloid process; SMG: Submandibular salivary gland; HN: Hypoglossal nerve.



According to Rodriguez-Vazquez, et al. [9], ossified SC extending from skull base on either side may compress a number of important structures close to it, which in turn may produce inflammatory changes like continuous chronic pain in region of pharynx, foreign body sensation, radiating otalgia and dysphagia. Lipschuitz reported a rare abnormality of solid bar of bone extending base of skull to hyoid bone in a young person which possibly might be due to ossification of embryonic cartilage [21]. Normal range of length of the SP differs among the studies in the available literature. It is about 2.5 cm according to Eagle and according to others is 3 cm [2]. It has been reported that SP probably becomes symptomatic when its length

exceeds 4 cm [22]. There are also reports in the literature such as absence of SP, localized or diffuse stylohyoid ossification [7, 22]. Basekim C, et al. [23], evaluated SP in 138 patients by three dimensional computed tomography (CT) but did not find diffuse SL. Diffuse ossification of SC is one of the rare variations. It differs from other variations as it affects the entire SC which may restrict head and neck movements or fracture of ossified SC during movement or trauma [24, 25]. Three dimensional computer tomography (3D-CT) can provide detailed information of SC variation as well as its possible relationship to adjacent structure like carotid artery [23].

Langlais, et al. [8] classified of elongated and mineralized stylohyoid complexes as three types by their radiographic appearance--Type I, interrupted, elongated; Type II, SP being joined to SL by a single pseudoarticulation giving the appearance of an articulated elongated SP; and Type III, Uninterrupted segments of mineralized SL, sometimes showing the appearance of multiple pseudoarticulations. Actual causes of SP elongation is poorly understood although several theories have been proposed like congenital elongation due to persistence of a cartilaginous analogue of SL, calcification of the SL by an unknown process and growth of bony tissue at the attachment of the SL [26].

For diagnosis of Eagle's syndrome several imaging modalities have been used. Conventional radiographs include Town radiograph, lateral head and neck radiograph, A-P head radiograph, panoramic radiograph, lateral-oblique mandible plain film. Potential disadvantages of conventional radiographs are superimposition of several bony structures and distortion and magnifications secondary to angulations. The most advanced diagnostic imaging technique to evaluate the SC is 3D spiral CT in spatial geometry, with accurate measurements in length. The detailed knowledge about the course and relations of the SC and its relationship with adjacent structures also allows planning before surgery; thereby decreasing potential for iatrogenic intra-operative injury [27]. Pain is relieved by the injection of local anesthetic into the tonsillar fossa. Surgery is the main treatment, where an elongated SP can be shortened by trans-tonsillar or by external approach. Steroid injection is advised into lower tonsillar fossa for patients not suitable for surgery. Other procedures described initially are tonsillo-styloidectomy by the intraoral route, where a trans-pharyngeal approach was used to extract the SP after tonsillectomy [26].

Conclusion

In the present case, on the right side we have observed ossification of SL. Diffuse ossification of SC is very rare and it may remain asymptomatic throughout life. But care should be taken to rule out this entity, especially in older patients as it may restrict the neck movements and difficulty may arise during intubation. Awareness of the elongated SP and partial or complete ossification of SL is extremely important for anatomists, dentist, otolaryngologist, surgeon, and radiologists.

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