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Ossification of the stylohyoid ligament and 'Eagle's' syndrome

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SUMMARY

'Eagle's' syndrome is a rare condition in which an ossified stylohyoid ligament may endanger neural and vascular structures in the neck (Eagle, 1949, 1962; Rechtweg et al., 1998; Rodriguez-Vazquez et al., 2006; Chuang et al., 2007). Compression of one or other of these structures may give rise to symptoms including unilateral neck pain, dysphagia, otalgia and tinnitus (Kaufman et al., 1970; Omnell et al., 1998; Basekim et al., 2005; Ramadan et al., 2007; Yagci et al., 2007; Ramadan et al., 2010. Computer Tomography (CT) scans of the head and neck of 431 deceased persons (235 males and 196 females) between the ages of 1 day and 100 years (mean age 35.93 ± 24.15) who were admitted for medico-legal death investigation to the Victorian Institute of Forensic Medicine (VIFM) in Melbourne, Australia were examined. Ossification of the ligament was observed to occur in parts, the superior part at its attachment to the styloid process of the temporal bone being the most common. Complete ossification was not seen. Ligament ossification increased with age although there were no sex differences. In some cases there appeared to be a pseudoarthrosis along the course of the ligament. While the study did not examine the anatomical relationship to neurovascular structures it is apparent that the stylohyoid ligament may undergo almost complete ossification and could compress vital neck structures.

Key words: Stylohyoid ligament – Ossification – Anomalies – Eagle syndrome

INTRODUCTION

The stylohyoid ligament extends inferiorly from the tip of the styloid process of the temporal bone to the lesser cornu of the hyoid. Previous reports indicate that parts of the ligament may become ossified and that this may be an age-related phenomenon.

Embryologically, the stylohyoid ligament is derived from the second pharyngeal arch, or Reichert's cartilage (Arey, 1974), and may be divided into four parts (Omnell et al., 1998; Ramadan et al., 2007; Paigkou et al., 2009; Ramadan et al., 2010):

1. Tympanohyal, base of the styloid process of the temporal bone.
2. Stylohyal, the main body of the styloid process (joined tympanohyal and stylohyal parts).
3. Ceratohyal, stylohyoid ligament.

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4. Hypohyal, lesser cornu of the hyoid bone.

The stylohyoid ligament is intimately related to the lateral wall of the oropharynx and is in close proximity to a number of neurovascular structures. Medial to it is the internal carotid artery with sympathetic chain; internal jugular vein; and cranial nerves VII, IX, X, XI and XII (Moore et al., 2006; Piagkou et al., 2009; Ramadan et al., 2010). In addition, part of the middle pharyngeal constrictor and some fibres of the styloglossus attach to the ligament (Moore et al., 2006). One or more of these structures may be impacted adversely as the ligament undergoes ossification along all or part of its length (Kaufman et al., 1970; Omnell et al., 1998; Basekim et al., 2005; Ramadan et al., 2007; Yagci et al., 2007, Ramadan et al., 2010). Previous reports suggest that ossification of the ligament may give rise to symptoms including unilateral neck pain, dysphagia, otalgia and tinnitus, which have been referred to as 'Eagle's' syndrome, after Eagle (1949, 1962). A vascular form of the condition is said to result from compression of the extra-cranial portion of the internal carotid artery and result in a transient ischaemic event, dissection, or even stroke (Eagle, 1949, 1962; Rechtweg et al., 1998; Chuang et al., 2007). Pain has also been reported in the vicinity of the laryngopharynx which has been attributed to irritation of the glossopharangeal nerve (Loeser and Cardwell, 1942; Frommer, 1974).

This study examined development of the stylohyoid ligament in a sample of deceased individuals ranging in age from newborn to old age to determine the pattern of ossification of its parts and whether or not these events are age-and/or sex related. Both plain radiographic and CT studies (Kaufman et al., 1970; Gozil et al., 2001; Basekin et al., 2005; Onbas et al., 2005; Ramadan et al., 2007; Yagki et al., 2008; Ramadan et al., 2010) have been performed on the ligament in the past; however none of these have alluded to ossification occurring in distinct sections. Ethical approval to conduct this study was provided by the Victorian Institute of Forensic Medicine.

Materials and Methods

Computer Tomography (CT) scans of 431 deceased persons (235 males and 196 females) between the ages of newborn to 100 years (mean age 35.93 ± 24.15) who were admitted for medico-legal death investigation to the Victorian Institute of Forensic Medicine (VIFM) in Melbourne, Australia between 01/01/2006 and 01/09/2010, were examined. Images were obtained using a 16 channel MDCT scanner (Aquilion16®, Toshiba Medical Systems, Minato-ku, Tokyo, Japan). Raw data acquisition consisted of helical 0.5mm slices using 120kVp and 300mA exposure factors which were reconstructed into 1.0mm slices with 0.2mm overlap. Two convolutional kernels (reconstruction algorithms) were applied to the overlapping data set. The first was specifically designed to show soft tissues, but also allows for maximum clarity in volume-rendered three dimensional reconstructed images. The second was an edge enhancement (bone) algorithm that allows for maximum resolution in two dimensional multiplanar reformation, producing views of the stylohyoid ligament in the axial, coronal and sagittal planes.

Images were examined for the presence of an ossified stylohyoid ligament and were viewed using a bone algorithm two-dimensional image that was manipulated using oblique multiplanar reformation (MPR) and a Ray Sum algorithm of 4x slice (i.e. 3.2 mm) to reduce image noise. The axes of the nominal axial, coronal and sagittal images were adjusted using MPR until the stylohyoid ligament could be viewed in the anatomical plane. All images were viewed and measurements obtained using an AquariusNet (AqNet) (TeraRecon Inc, San Mateo, California, USA) thin client workstation. The existence of ossified ligaments was determined, the length of each section measured and the presence of any anomalies, such as intervening joint articulation, noted.

The stylohyoid ligament was found to ossify in five sections or parts, which were defined as follows (Fig. 1):

Styloid –extending from (and attaching to) the styloid process.

Superior –commencing at the styloid process but not attached to it.

Middle –commencing between the styloid process and lesser cornu.

Inferior - concluding at the lesser cornu but not attached to it.

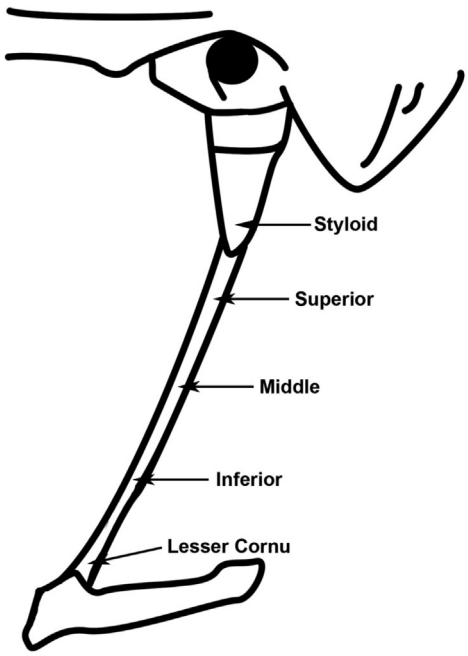

Fig. 1. Parts of stylohyoid ligament.

Lesser cornu –extending and attaching to the lesser cornu.

Data were compiled into pre-determined age groups: infants (0 - 2 years) n= 32; children (3 - 12 years) n= 42; adolescents (13 - 18 years) n= 31; young adults (19 - 24 years) n= 39; adults (25 - 65 years) n= 206; elderly (66+ years) n= 56. Data were analysed using ANOVA and unpaired Student's t-tests, as appropriate (Minitab 16.1.1, Minitab Inc., State College, PA, USA). ANOVA were followed by Tukey's post-hoc range test. Significance was set at $p<0.05$.

Results

Ossification of the stylohyoid ligament

There was large variation in the extent and sites of ossification of the stylohyoid ligament, as well as the age at which ossification was observed. Of 862 ligaments examined, 140 (16%) showed no ossification at all; 420 (48%) were ossified in one part only; 195 (22%) in two parts; and 107 (12%) in three or more (Table 1). The superior section of the ligament, without connection to the styloid process (Table 2), was the most frequently ossified part (479 ligaments or 55.5%). Of these, 61 were ossified unilaterally while the remainder were ossified on both sides. The most common patterns involved the superior part extending all the way to the lesser cornu (N=158 sides) and the superior part attached to the styloid process (N=144 sides).

A number of anomalous stylohyoid ligaments were observed with what appeared to be 'joint spaces' between successive parts. These primarily presented as a separation of successive segments (Fig. 2). However, at nine sites they comprised a fused nodule of bone (Fig. 3). The majority of 'joints' were at the junction of styloid process with the superior part of the stylohyoid ligament (Table 3).

Fig. 2. Pseudoarthrosis between styloid and superior parts of stylohyoid ligament.

Table 1. Segmentation of ossified stylohyoid ligament.

	Left	Right	Both
0 parts	70	70	140
1 part	205	215	420
2 parts	101	94	195
3 parts	46	43	89
4 parts	9	9	18
>4 parts	0	0	0
Total	431	431	**862**

Table 2. Frequency of partial ossification of stylohyoid ligament.

		Styloid process	Superior	Middle	Inferior	Lesser cornua
Left		174	247	62	11	87
Right		175	232	64	12	85
Bilateral		162	209	39	9	81
Unilateral	**Left**	12	38	23	2	6
	Right	13	23	25	3	4

Table 3. Site and frequency of 'joints' between parts of stylohyoid ligament.

Site of Joint	Total number of joints
Styloid process & superior part	60
Superior & middle parts	22
Middle & inferior parts	2
Inferior part & lesser cornu	1

Fig. 3. Nodule of bone between styloid and superior parts of stylohyoid ligament.

Age and ossification

The youngest individual displaying ligament ossification was an infant aged just one year (3.26 mm of the left and 3.64 mm the right lesser cornu section, respectively). Lack of ossification was more common in the young, although some individuals in the age range 20 to 60 years as well as one 96 years of age showed no evidence of ossification.

The styloid process and superior part of the ligament of the adults and the elderly were significantly different from all other age groups ($p<0.05$). The remaining parts did not show significant variation with age, although in general the children had lower mean values in comparison to all other age groups, while the adults and the elderly had significantly larger means than the other age groups. There was no variation between the sexes.

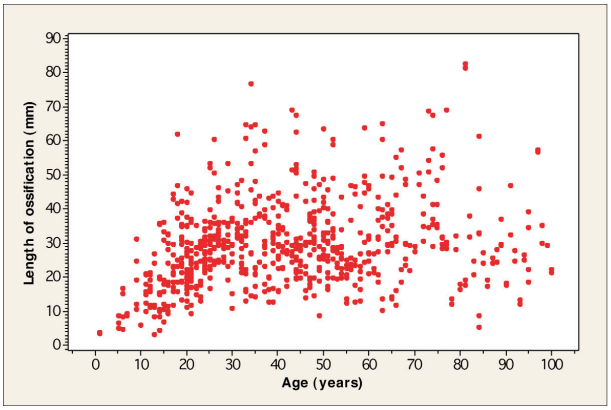

Fig. 4. Scatter plot showing variation of length of ossified ligament related to age.

A scatter plot of individual values (Fig. 4) shows the age-related variation.

Discussion

The onset of ossification of the stylohyoid ligament was variable; sometimes commencing during childhood or adolescence, and in other age groups not at all. Ossification varied greatly with age and, not surprisingly, was most pronounced when comparing ligaments in childhood with those in late adolescence, between which there was a linear increase in the degree of ossification, while age groups thereafter had a much lower rate of growth. The mean length of total stylohyoid ligament ossification observed in the current study was

31.36 ± 11.32 mm for adults and 33.37 ± 15.22 mm for elderly. These values are slightly, but not significantly, higher than those reported in previous studies (Omnell et al., 1998; Gozil et al., 2001; Basekim et al., 2005; Ramadan et al., 2007; Onbas et al., 2005).The incidence of complete ossification of the ligament has been reported to be 0.09% based on 1,215 autopsies (Vougiouklakis et al., 2006), although complete ossification was not observed in this CT study (the greatest degree was 96% in an 81 year old male).

Segmentation of stylohyoid ligament ossification was observed. The majority of ligaments had only one part ossified. However two parts were involved in 22.6% of cases; three in 10.3%; and four in 2%. Ramadan et al. (2007) reported two- or three-part ossification in 24.5% of 200 stylohyoid ligaments. However, those authors made no mention of its separation into a fourth part. Most segmentation appears as a clear division between successive parts of the ligament, although nine individuals presented with a nodule of bone between the styloid process and the superior portion (Fig. 5). The reasons for this remain unclear but it may be due to the fusion of two successive segments of ossified ligament and has even been reported as a callus formed after the healing of a fracture. Yagci et al. (2008) suggested that formation of the stylohyoid

Fig. 5. Almost complete ossification of stylohyoid ligament.

'joints' may be related to a pseudoarthrosis secondary to fractures due to neck movement. Some of these fractures may not heal and present as segmentation on CT images.

Ossification of the stylohyoid ligament has been associated with a condition referred to as 'Eagle's' syndrome, in which patients complain of a variety of symptoms, including unilateral neck pain, dysphagia, otalgia and tinnitus as well as pain on deglutition (Eagle, 1949, 1962; Rechtweg et al., 1998; Chuang et al., 2007). Although a correlation between the symptoms and degree of ossification of the stylohyoid ligament is yet to be confirmed (Prasad et al., 2002; Paigkou et al., 2009), this study indicates that ossification does occur to a variable degree across all age groups, and adjacent structures in which ossification is pronounced could be compromised.

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