ORIGINAL ARTICLE

Cleft Lip and Palate Subjects Prevalence of Abnormal Stylohyoid Complex and Tonsilloliths on Cone Beam Computed Tomography

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Keywords
Cone-beam computed tomography; Incidental findings; Physiological calcification; Cleft lip; Cleft palate

Abstract
Introduction and objectives: Tonsilloliths and abnormal stylohyoid complex may have similar symptoms to others of different aetiology. Individuals with cleft lip and palate describe similar symptoms because of the anatomical implications that are peculiar to this anomaly. The aim of this study was to determine the prevalence of abnormal stylohyoid complex and tonsilloliths on cone beam computed tomography in individuals with cleft lip and palate.
Methods: According to the inclusion and exclusion criteria, 66 CT scans out of 2794 were analysed, on i-Cat® vision software with 0.8 index Kappa intra-examiner.
Results: The total prevalence of ossification of the incomplete stylohyoid complex in individuals with cleft lip and palate was 66.6%; the prevalence of these findings in females was 75% and 61.9% in males. The total prevalence of tonsilloliths was 7.5%.
Conclusion: It is important to ascertain calcification of the stylohyoid complex and tonsilloliths in the radiological report, due to the anatomical proximity and similar symptomatology to other orofacial impairments in individuals with cleft lip and palate, focusing on females with oral cleft formation, patients with incisive trans foramen cleft and incisive post foramen cleft because they are more prevalent. Greater knowledge of the anatomical morphometry of individuals with cleft lip and palate greatly contributes towards the selection of clinical behaviours and the quality of life of these patients, since cleft lip and palates one of the most common anomalies.
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INTRODUCCIÓN Y OBJETIVOS: Los tonsilolitidos y alteraciones en el complejo estilohioideo pueden presentar similar sintomatología a otras de diferente etiología. Los individuos con fisura labiopalatina describen similares síntomas en razón de las repercusiones anatômicas propias de esta anomalía. El objetivo de este estudio fue determinar la prevalencia de alteraciones del complejo estilohioideo y tonsilolitidos en exámenes de tomografía computarizada de haz cónico en individuos con fisura labiopalatina.

MÉTODOS: Según criterios de inclusión y exclusión fueron analizadas 66 tomografías de 2.794 tomografías, en el software i-Cat visión con índice Kappa 0,8 intraexaminador.

RESULTADOS: La prevalencia total de osificación del complejo estilohioideo incompleto en individuos con fisura labiopalatina fue de 66,6%, la prevalencia de estos hallazgos en el género femenino fue de 75% y 61,9% en el género masculino. La prevalencia total de tonsilolitidos fue de 7,5%.

CONCLUSIÓN: Es de relevancia constatar en el informe radiológico la presencia de la calcificación del complejo estilohioideo y tonsilolitidos. Debido a la proximidad anatómica y similar sintomatología clínica con otras alteraciones orofaciales presentes en los individuos con fisura labiopalatina, haciendo énfasis en individuos con fisura labiopalatina del género femenino, pacientes con fisura tipo transformen incisivo y posforamen incisivo por presentar mayor prevalencia. Conocer más sobre la morfometría anatómica de individuos con fisura labiopalatina coadyuva relevantemente en la elección de conductas clínicas y calidad de vida de estos pacientes, teniendo presente que la fisura labiopalatina es una de las anomalías más comunes. © 2017 Sociedad Española de Otorrinolaringología y Cirugía de Cabeza y Cuello. Publicado por Elsevier España, S.L.U. Todos los derechos reservados.

Introduction

The stylohyoid complex comprises the styloid process of the temporal bone, stylohyoid ligament and lesser cornu of the hyoid bone.1

The aetiology of abnormal stylohyoid complex has not been established. Currently, the most accepted theory is that it is due to pathological mineralisation of the stylohyoid complex, which we consider calcification of the stylohyoid complex. Steinmann considers it to result from a hyperplastic/metaplastic reaction.2,3

The clinical manifestations of stylohyoid complex calcification are described as the classical Eagle syndrome which refers to the proximity to various anatomical structures that can result in the following symptoms: dysphagia, tinnitus, otalgia, cervicofacial pain, trismus, foreign body sensation in the neck.1 These symptoms can be accompanied by changes in the voice and hypersalivation. There are case reports where calcification of the stylohyoid complex manifests as: difficulty in endotracheal intubation, cerebral ischaemia, pseudoaneurysms, the latter two caused by compression of the carotid artery, due to a calcified stylohyoid complex or changes in its angle.4-6 In other cases it can irritate the sympathetic nerve plexus.1

The tonsils are lymphoid nodules in the tonsillar fossa in the oropharyngeal wall. They have an immunological function and their calcification can cause tonsilloliths.7

It is thought that tonsilloliths occur through a phenomenon termed dystrophic calcification, together with other aggregates of inflammatory debris.2,7

Clinically tonsilloliths present with chronic halitosis, irritable cough, odinophagia and like calcification of the stylohyoid complex: dysphagia, otalgia, and foreign body sensation. Calcified stylohyoid complex and tonsilloliths can be asymptomatic disorders and found incidentally.5,8

Cleft lip and palate is defined as a solution of continuity. It is considered one of the most common congenital anomalies, with an overall incidence of 1 in 700 people.9-10

Individuals with cleft lip and palate are more vulnerable to recurrent inflammatory symptoms, adenoid hypertrophy, and Eustachian tube dysfunction that can result in hearing loss, and variants at the level of the tensor veli palatini muscle, oroantral communications and repeated episodes of otitis media.11 All these impairments present with similar clinical symptoms in patients with calcified stylohyoid complex and tonsilloliths.

Radiological visualisation of calcified stylohyoid complex and tonsilloliths is difficult on two-dimensional imaging, because they are flat figures. There are also other limitations such as magnification, distortion and superimposing structures.12 This technique is limited for complementary or final diagnoses, especially for people with cleft lip and palate.13

All these problems are overcome with the three-dimensional imaging of cone beam computed tomography
Materials and Methods

According to the inclusion and exclusion criteria, we analysed 66 images of 2794 CBCT scans performed in the period from 2011 to 2015.

The inclusion criteria were: CBCT images with a field of view covering regions from the base of the temporal bone to the body of the hyoid bone and nasopharyngeal and oropharyngeal air space, in a group of individuals with cleft lip and palate aged from 20 to 30 years, and classifying the cleft lip and palate according to Spina (1972).

The exclusion criteria were: Individuals with cleft lip and palate associated with a syndrome, evidence of the presence of tumors, metal devices after surgery, disorders affecting visualisation and artefacts.

The calibration was performed using 30 CBCT by a single examiner (intraobserver), undertaken twice with an interval of 15 days, showing a Kappa index of 0.8.

The analysis was divided into four stages, based on Centurion’s method,15 which shows the presence or absence of calcification of the stylohyoid complex, differentiating it from tonsilloliths. We were able to apply it to people with cleft lip and palate. The CBCT images were analysed on a 20-in. Eizo monitor, visualising the right and left side on multiplanar reconstructions.

Later, i-CAT Vision® software tools were used to measure the course of the stylohyoid complex, from the origin of the temporal stylloid process to the lesser cornu of the hyoid bone. Thus a medial area was delimited on the coronal plane and an anterior area on the sagittal plane in relation to the tracing made, which followed the course of the stylohyoid complex on the hyperdense images located in these areas that were considered tonsilloliths. Based on Ramadan et al.,13 we considered a stylloid process longer than 30 mm to be calcification of the incomplete stylohyoid complex, and a hyperdense image following the complete course of the stylohyoid complex to be calcification of the complete stylohyoid complex.

The project was approved beforehand by the hospital’s ethics committee.

Results

Sixty-six images of 2794 CBCT scans were analysed according to the inclusion and exclusion criteria; of 24 females and 42 males.

The total prevalence of calcifications of the incomplete stylohyoid complex in individuals with cleft lip and palate was 66.6% (Fig. 1). There were no cases of calcification of the complete stylohyoid complex (Table 1).

The total prevalence of tonsilloliths was 7.5% (Fig. 2 and Table 2).

The prevalence of findings according to the type of cleft was greater in people with incisive transforamen clefts, incisive postforamen cleft (Tables 3 and 4).

Discussion

This study assessed the stylohyoid complex and tonsilloliths in individuals with cleft lip and palate not associated with a syndrome, based on archived CBCT images, which had been taken for various dental and medical purposes. CBCT enables the assessor to make an objective diagnosis of the anatomical structures on axial, coronal and sagittal planes, without all the disadvantages of conventional radiography.1,16

Stylohyoid Complex

Calcification of the stylohyoid complex is considered a relatively common incidental finding on radiological reports of people with cleft palate.2

Eagle (1948) reports a 4% prevalence of stylohyoid complex calcification in individuals without cleft lip and palate. Studies performed on edentulous individuals report a 30% prevalence of stylohyoid complex calcification. Recent studies on CBCT show a prevalence of 1.4%–30% of stylohyoid complex calcification in individuals without cleft lip and palate. Khan (2008) found 35% soft tissue calcifications on CBCT scans of individuals without cleft lip and palate, of which 90% were in the area of the head and neck.17-19

There are few studies in English and (PubMed) on incidental CBCT findings in people with cleft lip and palate. Kuijipers (2013) performed a study on individuals with cleft lip and palate showing a prevalence of 95.1% of incidental findings, excluding: caries, dental agenesis and supernumerary teeth, mentioning a prevalence of 84.5%, and moreover did not refer to stylohyoid complex calcification or tonsilloliths. The results of our study show a total prevalence of 66.6% of I SHCC, which is a considerable percentage, in individuals between 20 and 30 years of age with cleft lip and palate.20

Studies performed on CBCT scans on stylohyoid complex calcification in individuals without cleft lip and palate according to gender show no statistically significant differences between females and males. Unlike our finding of a greater prevalence in females with cleft lip and palate with 75% (SHCO I) on CBCT images.15

There is scant literature on the prevalence of stylohyoid complex calcification on CBCT images of individuals without cleft lip and palate and according to location. Centurion (2013) states that 55% of I SHCC findings in individuals without cleft lip and palate would be bilateral, 3% of (I SHCC)
Figure 1  Cone beam computed tomography, sagittal (A), coronal (B) and axial (C).

Figure 2  Cone beam computed tomography, sagittal (A), axial (B) and coronal (C).
Table 1 Prevalence of I SHCC, According to Gender in Individuals With Cleft Lip and Palate.

<table>
<thead>
<tr>
<th>Gender</th>
<th>I SHCC Yes</th>
<th>I SHCC No</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>F</td>
<td>18</td>
<td>6</td>
<td>24</td>
</tr>
<tr>
<td>Total %</td>
<td>75%</td>
<td>25%</td>
<td></td>
</tr>
<tr>
<td>M</td>
<td>26</td>
<td>16</td>
<td>42</td>
</tr>
<tr>
<td>Total %</td>
<td>61.9%</td>
<td>38.1%</td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>44</td>
<td>22</td>
<td>66</td>
</tr>
</tbody>
</table>

I SHCC: incomplete stylohyoid complex calcification; F: female; M: male.
Fisher’s exact test P=.416.

Table 2 Prevalence of Tonsilloliths, According to Gender in Individuals With Cleft Lip and Palate.

<table>
<thead>
<tr>
<th>Gender</th>
<th>Tonsilloliths No</th>
<th>Tonsilloliths Yes</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>F</td>
<td>22</td>
<td>2</td>
<td>24</td>
</tr>
<tr>
<td>Total %</td>
<td>91.67%</td>
<td>8.33%</td>
<td></td>
</tr>
<tr>
<td>M</td>
<td>39</td>
<td>3</td>
<td>42</td>
</tr>
<tr>
<td>Total %</td>
<td>92.86%</td>
<td>7.14%</td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>61</td>
<td>5</td>
<td>66</td>
</tr>
</tbody>
</table>

F: female; M: male.
Fisher’s exact test P=1.000.

located on the left side and 5% of (I SHCC) located on the right side. Our study on people with cleft lip and palate found a prevalence of 36.6% (I SHCC) bilaterally located, 16% (I SHCC) located on the right side and 3.6% (I SHCC) located on the left side.5,15

Tonsilloliths

Studies on tonsilloliths are rarely case reports from 1920 to 2003. Findings of tonsilloliths on CT scans of individuals without cleft lip and palate were 16%.2,17,20 Our study shows a lower prevalence of 7.5% of individuals with cleft lip and palate.

The reports show there is no marked prevalence with regard to the unilateral or bilateral location of tonsilloliths.8,17 They coincide with our findings in that the prevalence of tonsilloliths was not significant at 4.5% tonsilloliths on the right side and 3% tonsilloliths on the left side.

Cleft Lip and Palate

Using Spina’s classification (1972) for people with cleft lip and palate, our study showed a marked prevalence of I SHCC in individuals with incisive post foramen cleft at 80%, and a prevalence of 67.9% of (I SHCO) in individuals with incisive transforamen cleft. The literature in English, PubMed, has no data concerning stylohyoid complex calcification and tonsilloliths in individuals with cleft lip and palate, or data on the prevalence of these anomalies according to cleft type, which makes it difficult to make a comparison with our findings.9

It is difficult to visualise stylohyoid complex calcification and tonsilloliths radiologically using two-dimensional imaging, since they are flat figures, and there are other limitations such as magnification, distortion and superimpositions.18 Using this technique often restricts complementary or final diagnosis, even more so in individuals with cleft lip and palate.21,22

Conclusion

Our research study suggests that the prevalence of incomplete stylohyoid complex calcification is 66.6%, 36.6% more prevalent in individuals without cleft lip and palate. This is noteworthy considering that cleft lip and palate is one of the commonest congenital anomalies. These data are relevant in the areas of radiology, dentistry, ENT, neurology and neurosurgery.

Radiographic and imaging interpretation and reaching a complementary and differential diagnosis in people with cleft lip and palate pose a professional challenge.

We consider that the practitioner who evaluates CBCT images should have a solid knowledge of the anatomical structures and of the impairments in people with cleft lip and palate.

It is important to highlight that CBCT imaging is justified after patient history taking, clinical examination and other imaging tests, based on the ALARA principle (As Low
As Reasonably Achievable) and on the European cone beam computed tomography protocol recommendations of SEDENTEX (Safety and Efficacy of a New and Emerging Dental x-ray Modality). CBCT can thus be used to add new reports of relevance and to assess all the areas in the field of view that should be confirmed in the radiological report.

Conflict of Interest

The authors have no conflict of interests to declare.

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