THREE-DIMENSIONAL ASSESSMENT OF THE PHARYNGEAL AIRWAY AND MAXILLARY SINUS VOLUMES IN INDIVIDUALS WITH NON-SYNDROMIC CLEFT LIP AND PALATE

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Abstract

Introduction: Children with cleft lip and palate (CLP) are known to have airway problems. Introduction of Cone-Beam CT (CBCT) and imaging software has facilitated generation of 3D images for assessing the volume of maxillary sinuses and pharyngeal airway. Consequently, the present study aimed at evaluating and comparing the maxillary sinus and pharyngeal airway volume of patients with cleft lip and palate in healthy patients, using cone beam computed tomography (CBCT) images.

Materials and method: The sample group included 27 individuals (15 with cleft lip and palate subjects and 12 healthy subjects). The pharyngeal airway and each maxillary sinus were three-dimensionally assessed, segmented and their volume was calculated. A comparison between the right and left sinus was performed by Student t-test, and the differences between the control and cleft groups were calculated using ANOVA.

Results: No statistically significant differences were found when the maxillary sinuses volumes from each side were compared (p >0.05). The unilateral CLP patients presented the lowest sinus volume. Individuals with CLP did not exhibit a total airway volume smaller than the non-CLP controls.

Conclusions: 3D imaging using CBCT and Romexis software is reliable for assessing maxillary sinus and pharyngeal airway volume. The present study showed that the pharyngeal airway is not compromised in CLP individuals. The unilateral CLP individuals present maxillary sinuses with smaller volumes, no differences being recorded between the cleft and non-cleft side.

Keywords: cone beam CT, pharyngeal airway and maxillary sinus volumes, cleft lip and palate

1. INTRODUCTION

Patients with cleft lip and palate (CLP) present several anatomical variations, apart from the gap between the nasal and maxillary processes. For example, alteration in midface structures is a common finding in affected individuals.

Anatomical abnormalities associated with CLP increase the risk of airway complications [1]. CLP are frequently associated with nasal abnormalities, such as septal deviation, nostril atresia, turbinate hypertrophy, maxillary constriction, vomerine spurs and alar constriction [2-6].

In part, these abnormalities are attributed to the congenital defect itself and also to the surgeries done to repair the orofacial defect [7,8]. Collectively, nasal abnormalities tend to reduce the dimensions of the nasal cavity and airway functions [4,9]. These differences may lead to numerous impairments, potentially affecting the health condition of the ear, nose and throat. Sinus disease, mainly sinusitis, is frequently noted in CLP patients [10-12]; this highlights the importance of the maxillary sinus, as a major component of the midface, and the need of a better understanding of its role both in the developmental process and in the manifestation of the disease.

Previous studies evaluated the area of maxillary sinuses of CLP patients using different imaging modalities, such as conventional radiographs and multi-slice computed tomography (MSCT) [11, 13–17]. However, area estimation is a two-dimensional (2D) assessment of a three-dimensional (3D) structure, therefore potentially limited and prone to error. In this respect, a 3D volumetric evaluation of the sinuses appears as a more accurate option to better understand the complexity of these structures. Moreover, CBCT is also replacing several MSCT studies, due to its lower cost and lower radiation dose [18].
With the increased application of cone beam computed tomography (CBCT) in dental practice, more patients with CLP are undergoing CBCT scanning prior to alveolar bone grafting procedures. The CBCT image allows visualization of structures in all three planes, providing a 3D view of both defects and structures.

Airway patency has been evaluated by two-dimensional (2D) radiographic imaging, such as lateral and anterior-posterior cephalometric films and functional studies, e.g., rhinomanometry and plethysmography [19-22].

Previous studies have shown that 3D imaging using CBCT is a simple and effective method to accurately analyze the airway [23,24]. Recently, numerous studies have been performed using CBCT, for assessing the airway in relation with facial morphology in individuals with sleep apnea [25,26]. At present, few CBCT studies devoted to the airway of individuals with CLP have been published, despite the frequently occurring airway problems in such subjects [2-5].

The present study compares the volume of the maxillary sinuses and pharyngeal airway in patients with cleft lip and palate to patients without cleft, by means of CBCT images, and assesses the differences between the right and left sides, and also between the cleft and non-cleft sides.

2. MATERIALS AND METHOD

A retrospective study was developed on subjects recruited from the Plastic Surgery Clinic, “St. Mary” Emergency Hospital for Children, Iasi. The experimental group included 27 individuals (15 with cleft lip and palate and 12 healthy subjects).

The exclusion criteria were: 1) history of treatment for sleep-disordered breathing, including tonsillectomy, adenoidectomy, or recurrent tonsillitis; 2) frequent colds (6 or more per year); 3) history of dysphagia and continuous positive airway pressure therapy. A further exclusion criterion for the control groups included any type of syndrome. All individuals with CLP had no maxillary expansion and alveolar bone grafting.

Patients’ skeletal maturity was determined by the cervical vertebral maturation method developed by Baccetti et al. [23]. The juvenile subjects were in cervical stages 2 and 3, and the adolescent subjects - in stages 4 and 5. The juvenile group was formed of 9 individuals with CLP and 7 individuals without CLP; the adolescent one included 6 individuals with CLP and 5 without CLP (Table 1).

Seven patients in the juvenile group with CLP had unilateral CLP, and 2 had bilateral CLP. In the adolescent group with CLP, 6 patients had unilateral CLP. Lip closure by Millard-type lip repair had been performed in most patients, at a mean age of 3.8 months (SD, 1.2 months), and closure of the palate had been performed by pushback palatoplasty and the Furlow method, at a mean age of 8.6 months (SD, 3.9 months). Bone grafting had been done at a mean age of 10.2 years (SD, 1.4 years). No patient had undergone pharyngeal flap surgery.

Table 1. Subjects and their ages in the 4 groups

<table>
<thead>
<tr>
<th>Group</th>
<th>Juvenile (CS 2 and 3)</th>
<th>Adolescent (CS 4 and 5)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>CLP</td>
<td>Control</td>
</tr>
<tr>
<td>N</td>
<td>9</td>
<td>7</td>
</tr>
<tr>
<td>Age (year)</td>
<td>9.6±1.2</td>
<td>10.8±1.8</td>
</tr>
</tbody>
</table>

CS, Cervical stage of cervical vertebral maturation

All control subjects had normal craniofacial morphology with no jaw deformities. The control group included 19 individuals who were gender, age, and Sella-Nasion (SN) matched with the experimental group. The inclusion criteria referred to Angle Class I malocclusion with beginning CBCT records for starting orthodontic treatment and no prior orthodontic treatment, no habitual mouth breathing recorded and no prior adenoidectomy and/or tonsillectomy.
The protocol of the investigation was reviewed and approved by the Ethics Committee of the Faculty of Dentistry, “Gr. T. Popa” University of Medicine and Pharmacy, Iasi, Romania. Before the CBCT scan, patients’ parents were fully informed on the purpose of this study and on the risks associated with CBCT.

The equipment used was PlanmecaPromax 3D CBCT Mid (Planmeca OY, Helsinki, Finland). Scanning was performed by selecting a 200 x 170 mm view field and the following exposure parameters: 90 kV, 12 mA, 13.8 sec and 0.4 x 0, 4x 0, 4 mm voxel size. DICOM files were imported into Romexis 3.0.1 (Planmeca OY, Helsinki, Finland), a software capable of volume rendering. To achieve the axial, coronal and sagittal sections, the CBCT reconstructions were established with a 1 mm thickness, at a distance of 1 mm.

First, a CBCT technician de-identified the files, removing the name, sex and date of birth from the DICOM, after which the files were reoriented to a standardized view. Next, in a secluded room with dim light, a well-calibrated oral radiologist with experience in tomographic appraisal performed the assessment of images.

The maxillary sinuses were evaluated separately (right/left and cleft/non-cleft). The threshold was defined with the Measure Ellipsoid Tool, to include the sinus space and to remove any artifact and background. After threshold selection, a three-dimensional editing was used to obtain refined surfaces of the segmentation, resulting in a VOI subsequently rendered into a shaded surface mesh, and each segmented volume (cm³) was calculated (Figs. 1, 2).

The pharyngeal airway was segmented and the volume was rendered in a multiplanar reconstruction (MPR) mode. The threshold tool was used to select the pharyngeal airway space. The threshold was defined by the Measure Cube Tool, to include the pharyngeal airway. The 3D

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**Fig. 1.** Multiplanar reconstruction of CBCT images demonstrating the contours of the segmented sinuses: sagittal (A), coronal (B) and axial (C) views

**Fig. 2.** Antero-posterior (A) and sagittal (B) views of 3D CBCT images superimposed with the 3D segmentation of the maxillary sinuses and the isolated 3D volume (C)
Region Growing function was used to create a 3D display of the pharyngeal airway space from each region of interest. The volume (cm³) was determined from the total airway segmentation. The airway was further segmented into three sections: nasopharynx, oropharynx and hypopharynx. The superior border of the nasopharynx was defined as the posterior choana and inferiorly as to the horizontal line along the hard and soft palates. The oropharynx was defined superiorly by the soft palate and inferiorly by the vallecula. The hypopharynx was defined superiorly by the hyoid bone and vallecula and inferiorly by the junction of the larynx and oesophagus (Fig. 3).

The linear measurement tool in the Romexis 3.0.1 software was used to determine SN, to properly match the experimental and control groups.

![Fig. 3. 2D sagittal (A) and 3D sagittal (B) views of CBCT images superimposed with the 3D segmentation of the pharyngeal airway and the isolated 3D volume (C)](image)

![Fig. 4. Two-dimensional axial slice images of the pharyngeal airway in the 3 defined planes: A - nasal floor plane; B - soft palate plane; C - epiglottal plane](image)

All volumetric values obtained were tabulated, and Student t-test was performed to compare the maxillary sinus volumes between the sides, the right and left part of the control and BCLP groups, the cleft and non-cleft sides of UCLP group, the pharyngeal airway volume between the cleft and the non-cleft group.

The average volume of the sinuses and pharyngeal airway was calculated, resulting in an average value for each patient, and ANOVA was used to determine the differences among the volumes of UCLP, BCLP and control (non-cleft) groups. Statistical analyses were performed with a SAS software version 9.4 (Cary, NC, USA). The level of significance was set at $p < 0.05$.

3. RESULTS

The final sample consisted of 27 CBCT images, including 12 control subjects and 15 individuals.
with cleft lip and palate (13 UCLP and 2 BCLP). As a result, a total number of 54 maxillary sinuses and 27 pharyngeal airways were evaluated.

The first statistical analysis was performed to compare the volumes of the maxillary sinuses between the sides in the juvenile or adolescent control and the CLP group. To this end, the applied Student t-test showed no statistical significant differences in any group, among the right and left sides in the juvenile or adolescent control group, or among the cleft and non-cleft sides in the CLP group (p > 0.05).

The average volume of the sinuses was then calculated, individual values being obtained for each patient in part. As the distribution of data was not normal, the log of these values was calculated for statistical evaluation. Next, ANOVA was performed to evaluate the volumes among the groups: juvenile or adolescent control, UCLP and BCLP. All comparisons gave significant differences, with p < 0.0001, when the juvenile or adolescent control group was compared to the UCLP, and p = 0.0009, comparatively with BCLP. The lowest volume of the sinus was recorded in the UCLP group.

The airway volumes were not significantly different in the juvenile or adolescent control and CLP groups. The nasopharynx showed an average volume of 5.00 cm$^3$ with a standard deviation of 1.5 cm$^3$ in the juvenile CLP group, and an average volume of 8.57 cm$^3$ with a standard deviation of 2.1 cm$^3$, respectively, in the juvenile control group (Fig. 2). The oropharynx showed an average volume of 6.19 cm$^3$ with a standard deviation of 1.9 cm$^3$ in the adolescent CLP group and an average volume of 10.72 cm$^3$ with a standard deviation of 2.3 cm$^3$, respectively, in the adolescent control group (Fig. 4).

More specifically, the CLP and control groups were assessed based on age. The juvenile males with clefts had an average airway volume of 5.4 cm$^3$ with a standard deviation of 2.1 cm$^3$, whereas the juvenile control males had an average volume of 6.1 cm$^3$, with a standard deviation of 4.1 cm$^3$. The juvenile females with clefts had an average airway volume of 5.6 cm$^3$ with a standard deviation of 2.1 mm, while the juvenile control females had an average airway volume of 6.3 cm$^3$, with a standard deviation of 2.3 mm.

The adolescent males with clefts had an average airway volume of 8.4 cm$^3$ with a standard deviation of 1.1 cm$^3$, and the adolescent control males showed an average volume of 9.1 cm$^3$ with a standard deviation of 2.3 cm$^3$. The adolescent females with clefts had an average airway volume of 8.6 cm$^3$ with a standard deviation of 1.1 mm, and the adolescent control females had an average airway volume of 9.3 cm$^3$, with a standard deviation of 1.3 mm.
4. DISCUSSION

The goal of the study was to develop a reliable 3-D analysis to measure certain characteristics of the pharyngeal airway in children with CLP, and to compare the findings with a non-CLP control group, matched as to age and sex.

The present study attempted at elucidating the volumetric characteristics of the subjects, as well as at comparing these findings with previous reports that had used different methodologies to assess this anatomic structure.

In the current study, significant differences were found between the control and the CLP groups, disagreeing with previous studies, that demonstrated no differences in the structure and development of maxillary sinuses between the cleft palate patients and the age-matched controls [13,15,27]. A possible explanation for the altered volume is the fact that the maxillary sinus develops embryonically in a different way in cleft palate patients, comparatively with patients with normal palate [18].

Equally, no significant differences were recorded between the sides, or between the right and left sides, in the control and CLP groups. This result differs from that obtained by Hiosaka et al., who reported larger maxillary sinuses on the non-cleft side in patients with UCLP [17].

On the other hand, our results agree with those of Suzuki et al. and Lee and You, who also observed similar volumes between sides [14,27].

The hypothesis on which the present study is based is that children with CLP had smaller pharyngeal airways, compared to those from the non-CLP control group. Our data evidenced no significant difference in pharyngeal airway volume in CLP children comparatively with non-CLP children.

However, this was only a retrospective study with sample size limited to a small number of previously obtained scans of children with CLP. The respiratory cycle was not controlled during the scans. Respiration is a dynamic action that may not be accurately depicted in a static 3-D image.

The images were taken before maxillary expansion, so that the smaller airway volume found in CLP children could be the result of the absence of expansion. Future studies should compare the volume in different regions of the pharynx (naso-, oro- and hypopharynx) to elucidate precisely where the airway is larger, and should also explore the airway length differences between CLP and control.

5. CONCLUSIONS

Among the patients without CLP, the oropharyngeal airway was larger in the adolescent control than in the juvenile control group, but no significant differences were observed between the CLP groups. Thus, the narrow pharyngeal airway in patients with CLP might result in a functional impairment of breathing in adolescent, rather than in juvenile patients. Subjects with CLP present maxillary sinuses with smaller volumes than the age-matched control subjects.

Cone beam computed tomography can be regarded as equivalent to CT with regard to the diagnostic information provided for CLP patients, yet with a considerably shorter investigation time and lower purchase costs, as well as lower radiation exposure. Further investigations are necessary to clarify the relationship between the pharyngeal structure and airway function, and maxillary sinus expansion in patients with CLP.

References

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