MAXILLARY SINUS VOLUME IN CLEFT LIP AND PALATE PATIENTS WITH AND WITHOUT AN ORONASAL FISTULA USING CBCT

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Cone-Beam Computed Tomography; Orofacial Cleft; Fistula; Maxillary Sinus.

ABSTRACT

Introduction Formulation of oronasal fistula is a common complication after surgical closure of cleft lip and palate (CLP). This study aimed to compare the maxillary sinus volume in 9-12-year-old CLP patients with and without an oronasal fistula who had undergone surgical closure of the cleft at 1 year of age, using cone-beam computed tomography (CBCT).

Methodology This descriptive, cross-sectional, analytical study was conducted on CBCT scans of 50 patients with unilateral CLP who were between 9-12 years and had undergone surgical closure of the cleft at 1 year of age in two groups with and without an oronasal fistula (n=50). The patients were selected among those presenting to a private orthodontic office between 2001-2009 and already had CBCT scans taken for orthodontic treatment. The 3D CBCT scans were reconstructed with Mimics software, and the volume of the maxillary sinuses was measured on the images. Data were compared using t-test.

Results The maxillary sinus volume was significantly smaller in patients with oronasal fistula compared with those without it (9510.7±492 mm³ vs. 10278.2±512 mm³, P<0.000). The maxillary sinus of the affected side was smaller than that of the unaffected side in both groups of patients with and without an oronasal fistula (P<0.05).

Conclusion Immature patients with unilateral CLP and oronasal fistula have a smaller maxillary sinus than unilateral CLP patients without an oronasal fistula, and may be at higher risk of respiratory infections.

KEYWORDS

Cone-Beam Computed Tomography; Orofacial Cleft; Fistula; Maxillary Sinus.

1. INTRODUCTION

The failed fusion of the medial nasal and maxillary processes would result in the occurrence of cleft lip while failed fusion of the palatine prominences would lead to the formation of a cleft palate [1,2]. Cleft lip and palate (CLP) has a prevalence of 1 per 500 live births [3]. Surgical management of CLP was first performed approximately 150 years ago [4]. At present, CLP patients often undergo surgery before the 1st year of age. However, an oronasal fistula may develop postoperatively due to the infection of the palate or tension of the flap, and cause problems for the patient [5]. The oronasal fistula is a common complication of surgical management of CLP with a prevalence rate of 9-50%. The rate of recurrence of the fistula after surgery is as high as 35-70% [6,7].

Development of an oronasal fistula indicates failure of the surgical closure of the palate to obstruct the communication between the oral and the nasal cavity. According to the classification by Pittsburg, seven types of fistula are present [8], which can be symptomatic or asymptomatic. Symptomatic fistula can cause several complications such as leakage of foods and drinks from the oral cavity into the nasal cavity, bad odor, rhinitis, impaired hearing, hypernasality, infection, and speech problems [7,9,10].

CLP patients often suffer from decreased maxillary sinus volume and significant esthetic impairments due to the maxillary deficiency at the midface, where the maxillary sinuses are located. These patients often develop recurrent sinusitis for no clear reason.
Also, they have smaller sinuses due to a different developmental process during the embryonic stage, surgical scars, recurrence of fistula, leakage of foods and liquids into the nasal cavity, frequent infections, and different pattern of air circulation in the nose and sinuses [1,8,11-14]. Several studies have been conducted on CLP patients using 2D lateral cephalometry, which has high diagnostic accuracy for clinical applications, despite simplicity and low cost [15-20]. However, cone-beam computed tomography (CBCT) has become increasingly popular in the recent years due to its higher accuracy at a comparable cost.

Controversy exists regarding the maxillary sinus volume in CLP patients such that some studies reported a significantly smaller volume of the maxillary sinuses in CLP patients compared with normal individuals [15,21-23] while some others found no significant difference in this respect [8,11,24-26]. Considering the existing controversy in this respect, and the gap of information regarding the maxillary sinus volume in patients who developed an oronasal fistula after surgery compared with those who did not, this study aimed to compare the maxillary sinus volume in 9-12-year-old CLP patients with and without an oronasal fistula who had undergone the surgical closure of the cleft at 1 year of age using CBCT. The null hypothesis was that no significant difference would be found in the maxillary sinus volume between CLP patients with and without oronasal fistula.

2. MATERIALS AND METHODS

This descriptive, cross-sectional, analytical study was conducted on 50 unilateral CLP patients between 9-12 years who had undergone surgical closure of the cleft at 1 year of age in two groups with and without oronasal fistula (n=50). The patients were selected from among those presenting to a private orthodontic office in Tehran between 2010-2019 and who already had CBCT scans taken for orthodontic treatment. The study was approved by the ethics committee of School of Dentistry (IR.IAU.DENTAL.REC.1399.19).

The sample size was calculated to be 25 for each group according to the results of a pilot study on 10 patients from each group considering α=0.05, β=0.2, mean maxillary sinus volume of 10671 mm3 and 10081 mm3 in the two groups and standard deviation of 715 mm3 using two-sample t-test, assuming equal variances in PASS 15.

The CBCT scans of unilateral CLP patients between 9-12 years who had undergone surgical closure of the cleft at 1 year of age were retrieved from the archives of a private orthodontic office from 2010-2019 by convenience sampling, and assigned to two groups with and without oronasal fistula.

The inclusion criteria were age between 9-12 years, history of surgical closure of the cleft before 1 year of age, and cervical vertebral stage (CVS) 2 or 3 (on sagittal CBCT scans). The CVS of each patient was determined by two examiners after reaching a consensus.

The exclusion criteria were history of previous orthodontic treatment, orthognathic surgery, trauma, syndromes, frequent colds (more than 6 times in 1 year), medication intake at the time of CBCT, inflammatory diseases of the upper airways at the time of CBCT, and systemic conditions.

All CBCT images had been obtained in standard upright position with maximum intercuspation. Also, all images had been taken with NewTom 5G CBCT scanner (NEWTOM | CEFLA S.C., Imola, Italy) with a total scanning time of 14-18 s, 3.4 s exposure time, and 0.3 mm3 voxel size. Three-dimensional reconstruction of images was performed according to the Demirtas method [11].

After standardization of images, the maxillary sinus volume was quantified. For this purpose, first the skeletal borders adjacent to the sinus structure were traced.

Figure 1. Quantification of the maxillary sinus volume on axial, sagittal and coronal CBCT sections using the Mimics software.
Next, the sinus area between the bones and the area between the infundibulum and the uncinate process was measured (Figs 1 and 2). Then, a 3D model was prepared to assess the sinus volume. After image reconstruction and standardization of orientation in axial, coronal and sagittal planes, the Mimics software suite-20 (Materialise, 3001 Leuven, Belgium) was used for the measurements.

The data were analyzed by SPSS version 22 using t-test. All measurements were repeated on 20 randomly selected CBCT scans after a 2-week interval by another examiner, and the reliability of the measurements was ensured by test-retest reliability. Since R was found to be >0.8, the results were found to be adequately reliable.

3. RESULTS

This study evaluated 50 patients including 25 with and 25 without oronasal fistula. The group with oronasal fistula included 17 females (66%) and 8 males (34%) with a mean age of 10±1 years. Of all, 30 patients (80%) were in CVS II (15 from each group) and 10 (20%) were in CVS III (5 from each group). The control groups were matched with the test group in terms of age, CVS and gender.

Table 1 presents the mean maxillary sinus volume in the two groups of patients. As shown, the maxillary sinus volume at the cleft side of patients with oronasal fistula was significantly lower than that at the cleft side of patients without fistula (P<0.000). The maxillary sinus volume at the non-cleft side of patients with oronasal fistula was also significantly lower than that at the non-cleft side of patients without fistula (P<0.000). The maxillary sinus volume at the cleft side was significantly smaller than that at the non-cleft side in patients with (P<0.000) and without (P<0.000) oronasal fistula.

4. DISCUSSION

This study compared the maxillary sinus volume in 9-12-year-old CLP patients with and without oronasal fistula who had undergone surgical closure of the cleft at 1 year of age using CBCT. The null hypothesis was that no significant difference would be found in the maxillary sinus volume between CLP patients with and without oronasal fistula. The results showed that the maxillary sinus volume was significantly smaller in patients with oronasal fistula compared with those without it. Also, the maxillary sinus volume in the cleft side was significantly smaller than that in the non-cleft side in both groups. Thus, the null hypothesis of the study was rejected. The smaller size of the maxillary sinus at the cleft side can be due to different developmental processes during the embryonic stage, maxillary deficiency, surgical scars, recurrence of fistula, leakage of foods and liquids into the nasal cavity and frequent infections, and different patterns of air circulation in the nose and sinuses. The present results were in agreement with those of Demirtas et al [11]. Our methodology was also similar to that of Demirtas et al, [11] although they did not assess the effect of the presence of oronasal fistula on the maxillary sinus volume; however, they assessed patients with a mean age of 13.5 years while we evaluated patients between 9-12 years. The assessment of patients in this age range was an advantage since evidence shows airway growth and development in two periods of 6-9 and 12-15 years, with an interval between 9-12 years [27]. Also, CBCT images are not often obtained from patients younger than 9 years of age (CBCT is often first requested for grafting prior to canine eruption). Moreover, the soft tissue becomes more stable after 9 years of age. Erdura et al. [8] evaluated the maxillary sinus volume of

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<th>Table 1. Mean maxillary sinus volume in the two groups of patients with and without oronasal fistula.</th>
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<td><strong>Maxillary sinus volume</strong></td>
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unilateral CLP patients of approximately 13 years of age. They found no significant difference in the maxillary sinus volume between the cleft and non-cleft sides, which was different from the present results, and may be attributed to the different age range of patients. Hikosaka et al. [18] measured the maxillary sinus volume in patients with CLP. They assessed the computed tomography (CT) scans of 109 CLP and 100 non-CLP patients and found no significant difference between the two groups, which was in contrast to our findings. Also, they found no significant difference in the maxillary sinus volume of the cleft and non-cleft sides, which was different from the present results. The difference between the two studies may be due to the use of different imaging modalities (CBCT vs. CT). Also, they did not specify the patients’ age. Agarwal et al. [16] evaluated the differences in maxillary sinuses of the cleft and non-cleft sides. They made the measurements on the maxilla using a combination of reconstructed axial, coronal and lateral CT images. The maxillary sinus volume was calculated on 3D reconstructed images. They found significant reduction of length, width, height, depth and volume of the maxillary sinuses at the cleft side, which supported the present results. However, they did not mention the patients’ age range.

The assessment of patients between 9-12 years was a strength of this study since this age range is ideal for assessment of the size of maxillary sinuses [27]. The use of CBCT was another strength of this study since it provides highly accurate data regarding the dimensions of anatomical structures, and has high measurement accuracy. Also, the effect of the presence of the fistula on the maxillary sinus dimensions was evaluated in this study, which has not been addressed before. Not evaluating bilateral CLP patients and small sample size were among the limitations of this study. Future studies with a larger sample size are required on bilateral CLP patients. Another limitation was that, patients evaluated in this study had been operated by different surgeons at 1 year of age, and different techniques had been used for cleft closure, which could have different effects. Due to the small number of patients, they could not be standardized in this respect. Future studies should address this topic and preferably enroll patients operated by the same surgeon and with the same surgical technique for cleft closure. Moreover, the effects of the surgical technique and time of surgery on the recurrence of the fistula are important topics that need to be scrutinized in further studies.

5. CONCLUSION

Immature patients with unilateral CLP and oronasal fistula have a smaller maxillary sinus than unilateral CLP patients without an oronasal fistula, and may be at higher risk of respiratory infections. Also, the maxillary sinus at the cleft side is smaller than that at the non-cleft side in unilateral CLP patients.

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AUTHOR CONTRIBUTIONS

AJ: Study concept and design; critical revision of the manuscript for important intellectual content; administrative, technical, and material support; study supervision. LH: Acquisition of data. BK: AJ: Analysis and interpretation of data. FA: Drafting of the manuscript. MK: Statistical analysis.

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COMPETING INTERESTS

The authors declare that they have no competing interests.

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Maxillary Sinus Volume in Cleft Lip and Palate

Dr. Kiaee received her DDS (2013) and MSc in Orthodontics (2017) from Tehran University in Tehran, Iran. Since 2017, she has been working as an Assistant Professor at the Department of Orthodontics within the Faculty of Dentistry of Tehran University. Her research areas of interest are mostly clinical projects.
Questions

1. Which item was among the exclusion criteria stated in this article?
   - a. History of surgical closure of the cleft before 1 year of age;
   - b. Orthognathic surgery;
   - c. Cervical vertebral stage (CVS) 2 or 3;
   - d. Age between 9-12 years.

2. Which statement is NOT TRUE based on the findings of this article?
   - a. The maxillary sinus volume at the cleft side of patients with oronasal fistula was significantly lower than that at the cleft side of patients without fistula;
   - b. The maxillary sinus volume at the non-cleft side of patients with oronasal fistula was significantly lower than that at the non-cleft side of patients without fistula;
   - c. The maxillary sinus volume at the cleft side was significantly smaller than that at the non-cleft side in patients with and without oronasal fistula;
   - d. The maxillary sinus volume at the cleft side of patients without oronasal fistula was significantly lower than that at the cleft side of patients with fistula.

3. Which of the following statements is True based on the findings of this article?
   - a. Immature patients with unilateral CLP and oronasal fistula have a smaller maxillary sinus than unilateral CLP patients without an oronasal fistula;
   - b. Immature patients with unilateral CLP and oronasal fistula may be at lower risk of respiratory infections;
   - c. The maxillary sinus at the non-cleft side is smaller than that at the cleft side in unilateral CLP patients;
   - d. Adult patients with bilateral CLP and oronasal fistula have a smaller maxillary sinus than unilateral CLP patients without an oronasal fistula.

4. Which one is the prevalence of cleft lip and palate patients based on the present study?
   - a. 1 per 700 live births;
   - b. 1 per 1000 live births;
   - c. 1 per 500 live births;
   - d. 2.3 per 1000 live births.