Pharyngeal Airway Dimensions in Non-Syndromic Unilateral and Bilateral Cleft Lip and Palate Patients Compared With Class I Subjects

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Abstract

Objective: To analyze and compare the pharyngeal airway dimensions of Bilateral Complete Cleft Lip and Palate (BCCLP), Unilateral Complete Cleft Lip and Palate (UCCLP) and Class I patients in different age groups.
Design: Cross-sectional study of cephalometric data.
Materials and Methods: The lateral cephalometric films of 76 patients were divided into two main groups according to chronological age; Group 1 was consisted of patients between the ages 8 and 13 years, Group 2 was consisted of patients older than 13 years. Group 1 and 2 were divided into three subgroups as UCCLP, BCCLP and control groups. Airway dimensions were evaluated with area measurements using planimeter. SNA, SNB, ANB and GoGnSN angles were measured in all cephalograms by the same examiner. The values were compared within subgroups in Group 1 and 2 seperately.
Results: There was no statistically significant difference in SNA, SNB, GoGnSN measurements between subgroups in each group. ANB differed significantly between three subgroups in Group 1 ($p<0.001$) and Group 2 ($p<0.01$). In Group 1, the volumetric area of nasopharynx ($p<0.05$), in Group 2 nasopharynx ($p<0.001$) and oropharynx ($p<0.01$) differed significantly between UCCLP, BCCLP and Control groups.
Conclusion: Both UCCLP and BCCLP had decreased airway when compared with Class I subjects. However BCCLP patients were affected most adversely in nasopharyngeal and oropharyngeal airway areas.

Key Words: Airway, Cleft lip and palate

Introduction

Cleft Lip and Palate (CLP) is a congenital malformation with an incidence of 0.69-2.51 per 1000 births [1,2]. The defect results from disruption to the development of the facial and airway structures. This results in abnormalities in the final conformation and function of the upper airway [3]. Complaints like mouth breathing, snoring, and hypopnea during sleep is usually observed in patients with CLP [4,5].

Both anatomical and functional changes associated with CLP increase the risk of sleep disordered breathing especillally Obstructive Sleep Apnea (OSA) [3]. There are similarities in the reduced sagittal maxillary and mandibular development, larger vertical dimensions between cleft patients suffering from nasal airway obstruction [6-8] and patients with OSA [9]. According to Oosterkamp et al. craniofacial, craniocervical and pharyngeal morphology of OSA and Bilateral Complete Cleft Lip and Palate (BCCLP) patients demonstrate similarities [10]. More retrognathic mandible in BCCLP patients leads more retruded position of the tongue and reduction of the pharynx size [11].

Evaluation of pharyngeal airway is important in patients with CLP. Individual reports reveal that infants with CLP may have life-threatening airway obstruction, but they can also suffer from mild to severe sleep disordered breathing [3]. The airway has been studied in some studies in a group of Unilateral Complete Cleft Lip and Palate (UCCLP) [12,13] patients or BCCLP patients [10,14] or researchers have used complex groups [15,16] containing both types of clefts. The results of these studies differ in evaluation of the airway according to the cleft groups used; because, the type of the cleft influences the type of the airway and respiratory related problems [3]. According to Kadowaki et al., 40% of newborn mice with cleft lip and palate had cyanosis, which they concluded was related to airway obstruction. The mice with cyanosis had more severe cleft abnormalities (BCCLP) [17].

The current literature does not permit an accurate determination of the airway in different types of CLP. CLP patients might have different characteristics in the upper airway. The purpose of this study was to analyse and compare size of the pharyngeal airway among UCCLP, BCCLP and control groups and demonstrate a relationship between the type of the cleft and changes in the pharyngeal anatomy / volume between the UCCLP, BCCLP and control groups. It may be hypothesized that more severe cleft abnormalities cause more severe airway problems.

Materials and Methods

Subjects

This retrospective study was done on lateral cephalometric films from the department archives according to the Ethics Committee of Ankara University. The pre-treatment radiographs of the subjects were selected from University of Ankara, Faculty of Dentistry, Department of Orthodontics' archieve. The patient radiographs were selected among those applied for orthodontic therapy between the years 1998-2006. The lateral cephalometric films of 76 patients were divided into two main groups according to chronological age; Group 1 was consisted of patients between the ages 8 and 13 years, Group

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2 was consisted of patients older than 13 years. Group 1 and 2 were divided into three subgroups as UCCLP, BCCLP and control groups. The number and gender distribution, mean ages and minimum-maximum ages of the subjects in each group are shown on *Table 1*.

The inclusion criteria for CLP and control groups included good quality lateral cephalograms taken from patients after swallowing. The exclusion criteria were history of treatment for sleep disordered breathing, including tonsillectomy, adenoidectomy; previous orthodontic therapy. All of the CLP patients were non-syndromic and did not have bone-grafting before. All control subjects had Class I skeletal and dental relationship with normal craniofacial morphology.

Radiographs

The lateral cephalometric radiographs were taken by the same technician on the same machine. Cephalograms were obtained under standardized conditions in natural head position with the mandible in centric relation; patients were informed not to swallow during radiography.

Lateral cephalograms were traced by one examiner and cephalometric reference points were determined by using acetate paper. The skeletal landmarks were digitized and calculated with the help of PorDios (Purpose on Request Digitizer Input Out-put System, trademark of the Institute of Orthodontic Computer Science, Aarhus, Denmark) cephalometric analysis program. Four main skeletal measurements were used (*Figure 1*). The pharyngeal airway area measurements were chosen similar to the investigations done before (*Figure 2*) [18,19]. Pharyngeal area measurements were made on the acetate paper by using planimeter (Ushikata X-Plan380Dııı/460Dııı, Tokyo, Japan) (*Fıgure 3*).

Reliability

Cephalometric landmarks of the radiographs were digitized twice and area measurements were repeated three times by the same investigator, and the avarage values of three pharyngeal measurements were calculated to eliminate the errors in measurements.

Statistical Analysis

The statistical analysis of the study was performed by using 'Variance Analysis'. The mean values and standard error of the means were calculated. Variance Analysis was used to compare the measurements between UCCLP, BCCLP and control groups in both Group 1 and 2 seperately.

Results

There was no statistically significant difference in SNA, SNB, GoGnSN measurements between subgroups in each group (*Table 2*). ANB differed significantly between three subgroups in Group 1 ($p<0.001$) and Group 2 ($p<0.01$). In

***Table1**. The number and gender distribution, mean ages and minimum-maximum ages of the subjects in each group.*

Parameters	GROUP 1			GROUP 2		
	UCCLP	BCCLP	CONTROL	UCCLP	BCCLP	CONTROL
Age (minimum-maximum)	8.75-13.08	8.00-12.67	8.58-13.00	13.92-22.08	14.00-22.08	13.75-21.17
Mean Age (X ± Sx)	10.53 ± 1.47	10.43 ± 1.65	10.96 ± 1.43	16.67 ± 2.31	17.77 ± 2.72	17.37 ± 3.19
Male number	6	5	8	6	5	8
Female numberer	5	5	8	7	5	8
Total number	11	10	16	13	10	16

Group 1 indicates patients between 8 and 13 years; Group 2, patients older than 13 years; UCCLP, unilateral complete cleft lip and palate; BCCLP, bilateral complete cleft lip and palate.

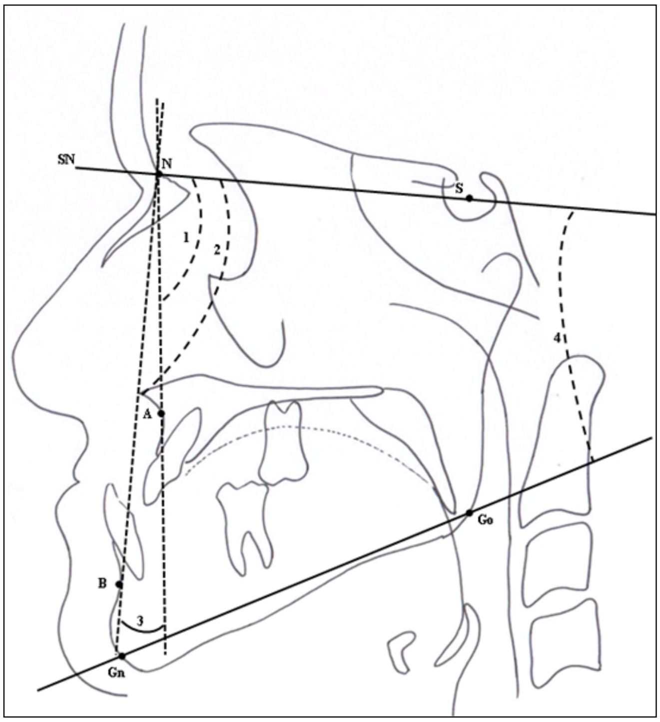

***Figure 1**.Skeletal measurements. (1) SNA. (2) SNB. (3) ANB.(4) GoGnSN.*

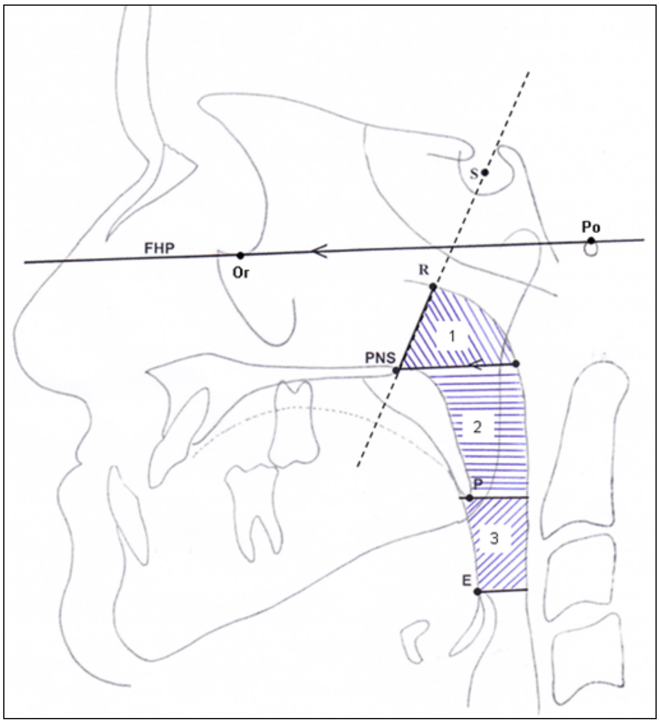

***Figure 2.** Pharyngeal area measurements. (1) Nasopharynx. (2) Oropharynx. (3) Hypopharynx.*

***Table 2**. Means and standard error of the means of the parameters according to groups.*

Parameters	GROUP 1				GROUP 2			
	UCCLP (n=11) X ± Sx	BCCLP (n=10) X ± Sx	CONTROL (n=16) X ± Sx	P	UCCLP (n=13) X ± Sx	BCCLP (n=10) X ± Sx	CONTROL (n=16) X ± Sx	P
SNA, degree	76.75 ± 1.35	80.27 ± 1.62	78.17 ± 1.06	NS	76.26 ± 1.33	79.70 ± 3.05	80.24 ± 1.02	NS
SNB, degree	75.46 ± 1.19	72.36 ± 1.08	75.74 ± 0.99	NS	76.40 ± 0.76	74.02 ± 2.51	77.20 ± 1.03	NS
ANB, degree	1.29 ± 1.42	7.91 ± 1.60	2.42 ± 0.55	***	-0.14 ± 1.34	5.68 ± 1.68	3.04 ± 0.44	**
GoGn/SN, degree	36.92 ± 1.37	39.74 ± 2.15	37.65 ± 1.28	NS	36.91 ± 1.17	38.14 ± 1.89	33.44 ± 1.28	NS
Nasopharynx, mm^2	151.80 ± 37.40	95.50 ± 16.20	183.50 ± 15.80	*	204.80 ± 25.30	130.80 ± 22.40	273.90 ± 21.40	***
Oropharynx, mm^2	235.40 ± 21.90	199.60 ± 24.80	261.90 ± 18.10	NS	331.90 ± 17.90	252.20 ± 26.70	348.70 ± 22.00	*
Hypopharynx, mm^2	290.00 ± 29.40	193.40 ± 20.60	223.40 ± 19.40	NS	311.40 ± 32.10	310.00 ± 31.00	264.10 ± 18.80	NS

Group 1 indicates patients between 8 and 13 years; Group 2, patients older than 13 years; UCCLP, unilateral complete cleft lip and palate; BCCLP, bilateral complete cleft lip and palate; n, number; NS, not significant; *, p<0.05; **, p<0.01; ***, p<0.001.

***Figure 3**a) Digital planimeter b) measurement of the areas using digital planimeter.*

Group 1, the volumetric area of nasopharynx ($p<0.05$), in Group 2 nasopharynx ($p<0.001$) and oropharynx ($p<0.01$) differed significantly between UCCLP, BCCLP and control groups.

Discussion

We used lateral cephalograms in our study and evaluated posterior pharyngeal airway space in area measurements using planimeter. In our study we seperated our subjects into two main groups because there is a marked age dependent difference in upper airway length in growing children [20,21]. Especially with puberty with the help hormones a change in pharyngeal airway occurs [20]. The dimensions of the nasopharyngeal airway typically continue to grow rapidly until around 13 years of age [22,23], and the growth rate decreases thereafter [24]. According to Yoshihara et al., significant age related changes occur in the pharyngeal airway of CLP patients and controls [14]. Besides, lymphoid tissues such as the palatine and pharyngeal tonsils reach their maximum sizes before the adolesence and then gradually atrophy, thus affecting the airway volume [25,26].

The position of the maxilla and mandible is directly related with the position of soft palate, base of tongue; thus nasopharyngeal and oropharyngeal airway volumes. Surgically treated CLP patients have reduced sagittal maxillary and mandibular development and larger vertical dimensions [10,12,27,28]. In our study although not significant, there was a tendency of mandibular retrusion and higher vertical dimensions in BCCLP and UCCLP groups. However the decreases in nasopharyngeal airway in Group 1 ($p<0.05$) and in nasopharyngeal ($p<0.001$) and oropharyngeal airway ($p<0.05$) in Group 2 were significant.

In our study BCCLP patients were affected most adversely in nasopharyngeal and oropharyngeal airway areas. This result support our hypothesis that ‘more severe cleft causes more severe airway problems’. However, we could not detect decrease in hypopharyngeal area in UCCLP in Group 1 and both cleft groups in Group 2. This result might be because of a tendency to longitudinal growth in hypopharyngeal area for increasing the volumetric area which can be attributed to adaptation for breathing in CLP patients.

In CLP patients morphological anatomic alterations mostly occur at the pharyngeal area. Those anatomic alterations can self-correct because of structural changes within the upper airway like the change in orientation of the pharynx, with growth to more horizontal to the more vertical type [29]. The more vertical type of growth can explain the adaptation of the airway for breathing.

It is difficult to compare our findings with the previous studies since no study evaluated pharyngeal airway area in different types of clefts. Çelikoğlu et al. evaluated airway in BCCLP-control groups, UCCLP-control groups in two different studies with three-dimensional computed tomography. They conluded that BCCLP and UCCLP patients had an insignificant decrease in nasopharynx and a significant decrease in oropharynx when compared with control groups [13,14]. Yoshihara et al. also evaluated airway in a complex group of CLP patients with a three-dimensional study [14]. They did not evaluate nasopharynx and found insignificant decrease in oropharyngeal area in adolescent CLP patients.

The children are more prone to sleep disordered breathing because the airway anatomy and function differ between children and adults. Growth of craniofacial skeleton and development of respiratory neuromuscular system is affected with cleft palate deformity. More mature respiratory neuromuscular system in older patients theoretically makes them less predisposed to moderate to severe upper airway obstruction during sleep when compared with younger ones [20]. From this point of view orthodontists should be aware of decreased airway and possible sleep disordered breathing in CLP children, orthopedic growth modification therapy might be applied for those children for the increase of the airway volume.

The present study presents valuable information about the patients affected by CLP, however the poor number of subjects can be considered as a limitation of our study. Therefore, further investigation including more patients are needed to discuss the present findings. The doctors should be aware of the decreased airway volume in CLP patients and define the best treatment choice.

Conclusions

Both UCCLP and BCCLP patients had decreased airway when compared with Class I subjects. However BCCLP patients were affected most adversely in nasopharyngeal and oropharyngeal airway areas.

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