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Evaluation of Pharyngeal Airway Dimensions in a Sample of Jordanian Preadolescent Cleft Lip and Palate Patients before Orthognathic Surgery

Ahmad M AI Tarawneh[1]
Abdalmawla A. Ali[2]*
Kholoud F. Alazm[3]

[1]Department of Orthodontics, Royal Medical Services, Amman, Jordan
[2]Department of Orthodontics, Faculty of Dentistry, Sirte University, Sirte, Libya
[3]Consultant in Orthodontics, Riyadh Colleges of Dentistry and Pharmacy, Riyadh KSA

Abstract

Objective: To exam the pharyngeal airway dimensions in cleft lip and palate subjects and to compare them with controls before orthognathic surgery and to assess the need for orthognathic surgery for these cases.

Material and methods: Pre-orthodontic cephalograms of two hundred patients were used in this study. Twenty one landmarks for sagittal pharyngeal airway and soft tissue measurements had been identified for each cephalogram. Patients were selected according to the specific inclusion criteria and divided into 2 groups according to the presence of cleft lip and palate. The first group was Jordanian cleft lip and palate and consisted of 100 subjects (50 females and 50 males, age ranged between 5 and 12 years, mean age was 8.4 ± 2.4 years), and the second group was 100 controls without cleft lip and palate (50 females and 50 males, age ranged between 5 and 12 years, mean age was 8.7 ± 2.2 years). Independent t-test was used to detect differences between the two groups.

Results: Subject's pharyngeal airway dimensions were significantly narrower, in the lower airway thickness (PNS - AD1) (P= 0.000), upper airway thickness (PNS - AD2) (P= 0.000), total lower sagittal depth of the bony nasopharynx (PNS - Ba) (P= 0.000), posterior sagittal depth of the bony nasopharynx (Ptm - Ba) (P= 0.000), total upper airway thickness (PNS - H) (P= 0.000) and McNamara's lower pharynx dimension (P= 0.007) when compared to control. On the other hand, control group showed significantly longer soft palate (PNS–P) (P= 0.000), and wider in both thickness of tongue (TGH) and soft palate (MPT) with (P= 0.000). The Tongue Length (TGL) and hyoid bone position did not record any significant difference when compared to subject group.

Conclusions: Subjects had narrower pharyngeal airway dimensions when compared with the controls and this was not due to soft tissue hypertrophy but attributed to the smaller bony nasopharyngeal framework and skeletal deformations. Orthognathic surgery is indicated in cleft lip and palate population to correct skeletal discrepancies.

Keywords

Cleft lip and palate; Pharyngeal airway; Cephalometrics

Introduction

Clefting of Lip and Palate (CLP) is the most common orofacial congenital malformation found among live births [1]. Prevalence rates range from as low as 0.43 to 2.45 per 1000 births [2]. The prevalence varies according to race/ethnicity, sex, and cleft type [3].

Anatomical abnormalities associated with CLP causing impaired patency, function and size of airway apparatus [4]. Early surgical interventions involve lip and nose repairs before 6 months, and pharyngeal and palatal flaps surgery before 15 months [5]. Both patients with repaired and unrepaired CLP might present with reduced size of airway dimension, nasal obstruction, speech impairment, Velophyngeal Insufficiency (VPI) and even reported Obstructive Sleep Apnea (OSA). Authors found that surgical intervention involved in CLP treatment tend to further compromise nasal airway and breathing [6-9].

Pharyngeal airway studies have gained an interest for the researchers since nasopharyngeal and oropharyngeal airway dimensions have a central role in the growth and development of oro-dental and craniofacial anatomy. Sheng et al. reported that during growth, developmental changes in airway dimensions takes place and continue into young adulthood. This results in differences in airway dimensions between young and adult subjects. Sexual dimorphism in the airway dimensions, especially in lower pharyngeal airway, has also been documented. Furthermore, skeletal classification does affect airway dimension, class III malocclusion patient usually have larger airway dimension in comparison with their class I and class II malocclusion counterparts. As the ANB angle

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*Corresponding author:

Ali AA
Department of Orthodontics
Faculty of Dentistry
Sirte University, Sirte, Libya
Tel: 00218922661344
Fax number 00218545260361
E-mail: abdelmola_alhossin@yahoo.com

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increases, the inferior pharyngeal space is reduced [10].

Many studies have compared airway dimension in CLP and non CLP adults, but only few studies compared airway spaces before orthognathic surgery. Therefore, it is of interest to evaluate whether children with CLP also differ from children without CLP with regard to specific cephalometric variables.

The impetus for the present study was the clinical finding of airway dimensions differences between Jordanian patients with and without non-syndromic CLP in term of pharyngeal airway dimensions before orthognathic surgery (which usually needed in CLP patients) and it is considered as the first research to assess pharyngeal airway measurement differences and identify the reason of these differences as well as the degree of requirement of orthognathic surgery.

Material and Methods

Study design

This was cross-sectional retrospective study carried out on the available pre-orthodontic treatment records of two groups of patients who had their orthodontic treatment at Jordanian medical royal services. The first group comprised Jordanian subjects with CLP and the second comprised Jordanian controls without CLP patients. Control subjects were recruited from a population seeking orthodontic consultation at the Orthodontic Department of the Royal Jordanian Rehabilitation Center. Patient consents and an ethical approval for conduction of this study were obtained from the Institution of Research Board (IRB).

Subjects and selection criteria

Two hundred patients were included in this study; they comprised two groups according to the presence of CLP: 100 subjects (50 females and 50 males, age ranged between 5 and 12 years, mean age was 8.4 ± 2.4 years) these patients underwent primary repair of a cleft of the secondary palate by one surgeon, using double-opposing Z-plasty (Furlow) technique and 100 controls without CLP (50 females and 50 males, age ranged between 5 and 12 years, mean age was 8.7 ± 2.2 years). All patients included in this study were selected according to the following criteria:

1. Patients with medical history of pharyngeal pathology and/ or nasal obstruction, snoring, OSA, adenoidectomy, and tonsillectomy were excluded.
2. All patients had repair CLP and before orthognathic surgery if needed.
3. Asyndromic CLP subjects.
4. Patient's age between 5 – 12 years.
5. All subjects were nose breathers.
6. Patients with poor quality, distributed or unclear pre-orthodontic treatment lateral cephalograms radiograph were excluded.

Jordanian controls group was selected to match the non-syndromic CLP subjects in terms of age, sex and skeletal classifications.

Record Analysis

Cephalometric Records: Pre-orthodontic treatment lateral cephalograms for each participant were taken with a Siemens Orthophos-5 machine (Siemens AG, Munich, Federal Republic of Germany) using a standardized technique with teeth in light intercuspation. All cephalograms were analyzed and hand traced by one investigator (Abdalmawla Alhussin Ali) on acetate tracing paper attached to radiographs. During tracing, the room was darkened and the viewing screen was blanked off showing only the radiograph. Twenty one landmarks for sagittal pharyngeal airway and soft tissue measurements had been identified for each cephalogram. The definitions of different landmarks and measurements are shown in Table 1 and Figure 1.

The measurements were performed manually using a ruler to the nearest ± 1mm to measure linear distance and protractor to the nearest ± 1º to measure the angular measurement.

Method Error: In order to calculate the error of the method, Dahlberg's formula ($M.E=\sqrt{\Sigma d^2/2N}$) was applied. Twenty lateral cephalograms (10% of total sample) were selected randomly and reanalyzed after one month interval by the same examiner [11]. For intra-examiner reliability, Houston's coefficient of reliability was calculated [12].

Statistical analysis

Statistical analysis was performed using the Statistical Package for Social Science (SPSS) computer software (SPSS 17.0, SPSS Inc., Chicago, USA).

Descriptive statistics (means and standard deviation) were calculated for all the measured variables. Independent t-test was conducted to detect differences between the two groups. Level of significance was set at 0.05 level.

Results

Error testing

Dahlberg's error ranged from 0.006 mm for distance between hyoid and C3 (C3H) to 0.125 mm for lower adenoid thickness (AD1–Ba), indicating that there were no significant differences between any of the measured variables at two different time points. Houston coefficient of reliability was calculated and was above 90% for all measured variables.

Pharyngeal airway cephalometric measurements

The means, standard deviations, mean differences and significance level of pharyngeal airway cephalometric measurements in the Subjects with CLP and Controls groups are shown in Table 2.

Significant differences were recorded between subjects with CLP and controls groups in terms of lower airway thickness (PNS - AD1) (P= 0.000), upper airway thickness (PNS - AD2) (*P= 0.000*), total

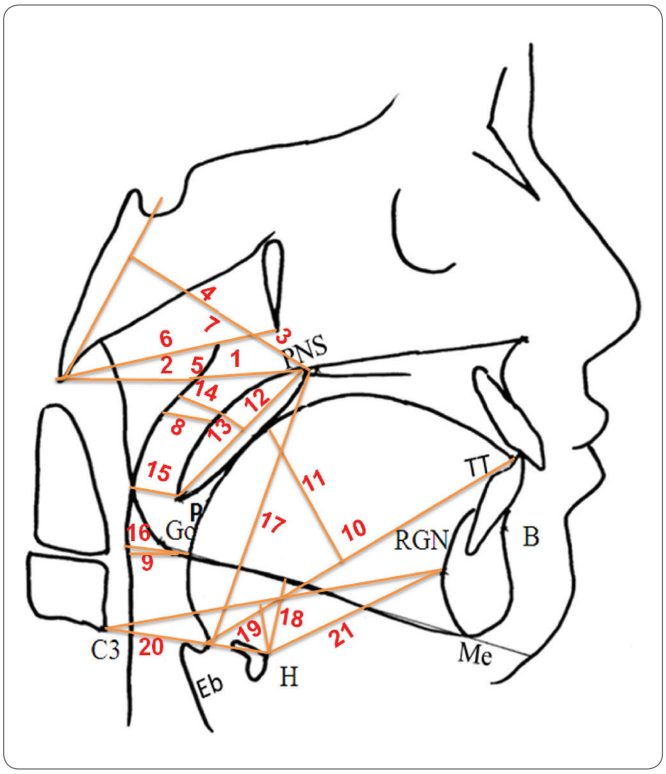

Figure 1: Cephalometric points and measurements used for airway analysis 112x125mm (150 x 150 DPI)

Cephalometric points		
Landmark		**Abbreviations**
TT		Tongue tip
Eb		Base of epiglottis
P		Tip of soft palate
PNS		Posterior nasal spine
Me		Menton
Go		Gonion
B		Point B
RGN (retrognathion)		The most posterior point of symphysis
H (hyoidale)		The most superior and anterior point on the body of the hyoid bone
C3		Anteroinferior limit of third cervical vertebra
Airway dimension measurements as numbered in Figure 1.		
1	**PNS-AD1**	Lower airway thickness; distance between PNS and the nearest adenoid tissue measured through the PNS-Ba line (AD1).
2	**AD1-Ba**	Lower adenoid thickness; defined as the soft-tissue thickness at the posterior nasopharynx wall through the PNS-Ba line.
3	**PNS-AD2**	Upper airway thickness; distance between PNS and the nearest adenoid tissue measured through a perpendicular line to S-Ba from PNS (AD2).
4	**AD2-H**	Upper adenoid thickness; defined as the soft-tissue thickness at the posterior nasopharynx wall through the PNS-H line (H, hormion, point located at the intersection between the perpendicular line to S-Ba from PNS and the cranial base).
5	**PNS-Ba**	Total lower sagittal depth of the bony nasopharynx.
6	**Ptm-Ba**	Posterior sagittal depth of the bony nasopharynx.
7	**PNS-H**	Total upper airway thickness.
8	**McNamara's upper pharynx dimension**	Minimum distance between the soft palate and the nearest point on the posterior pharynx wall.
9	**McNamara's lower pharynx dimension**	Minimum distance between the point, where the posterior tongue contour crosses the mandible, and the nearest point on the posterior pharynx wall.
10	**TGL**	Tongue length (Eb-TT).
11	**TGH**	Tongue height (maximum height of tongue along perpendicular line of Eb-TT line to tongue dorsum).
12	**PNSP**	Soft palate length (PNS-P).
13	**MPT**	Soft palate thickness (maximum thickness of soft palate measured on line perpendicular to PNS-P line).
14	**SPAS**	Superior posterior airway space (width of airway behind soft palate along parallel line to Go-B line).
15	**MAS**	Middle airway space (width of airway along parallel line to Go-B line through P).
16	**IAS**	Inferior airway space (width of airway space along Go-B line).
17	**VAL**	Vertical airway length (distance between PNS and Eb).
18	**MPH**	Perpendicular distance from hyoid bone to mandibular plane.
19	**HH1**	Perpendicular distance from hyoid bone to the line connecting C3 and RGN.
20	**C3H**	Distance between hyoid and C3.
21	**HRGN**	Distance between hyoid bone and RGN.

Table 1: Definitions of the airway points and measurements

lower sagittal depth of the bony nasopharynx (PNS - Ba) (*P= 0.000*), posterior sagittal depth of the bony nasopharynx (Ptm-Ba) (*P= 0.000*), total upper airway thickness (PNS - H) (*P= 0.000*) and McNamara's lower pharynx dimension (*P= 0.007*) were significantly wider in controls group compared to subjects one. On the other hand, the results showed that no statistically significant differences between the two different groups in upper and lower adenoid thickness.

Soft tissue cephalometric measurements

The means, standard deviations, mean differences and significance level of soft tissue cephalometric measurements in the subjects with CLP and controls groups are shown in Table 3.

Statistically significant differences were found between subjects with CLP and controls in which controls had longer soft palate length (PNS - P) *(P= 0.000)* and wider in both thickness of tongue (TGH) and soft palate (MPT) with (P= 0.000) and (P= 0.000) respectively. The tongue length (TGL) did not record any significant difference.

Hyoid bone position

The means, standard deviations, mean differences and significance level of Hyoid bone position cephalometric measurements in the Subjects with CLP and controls groups are shown in Table 4.

No significant differences among two groups in hyoid bone position measurements e.g. distances between hyoid bone to C3 (C3H) and to RGN (HRGN), perpendicular distance from hyoid bone to mandibular plane (MPH) and to the line connecting C3 and RGN (HH1).

Discussion

CLP almost associated with reduction in dimensions of pharynx that increases the risk of airway complications. Many factors contribute to the narrowing pharyngeal airway and lack of its patency such as, adenotonsillar hypertrophy, maxillary hypoplasia or surgery in the palate and the velopharyngeal region. This study revealed an overall significant reduction in airway dimensions in patients with CLP. The results showed that statistically significant differences between the two different groups in all pharyngeal airway dimensions e.g lower airway thickness (PNS - AD1), upper airway thickness (PNS - AD2), total lower sagittal depth of the bony nasopharynx (PNS - Ba), posterior sagittal depth of the bony nasopharynx (Ptm - Ba), total upper airway thickness (PNS - H) and McNamara's lower pharynx dimension. This result was in agreement with the findings reported by previous study who concluded that the subjects characterized by reduction of the pharyngeal airway width [13]. Other study revealed similar results, where patients with CLP had reduction of the nasopharyngeal bony framework when compared with controls. They attributed that to posterior position and decrease the posterior height of maxilla [14].

On the other hand, vertical length of the pharyngeal airway (VAL) in subjects was increased but not reached to significant degree as compensatory to narrowing in the airway passage width. This result was in agreement with the findings suggested by another studies [15,16].

This was a linear analysis of pharyngeal airway dimensions and sample was assessed for any patient with CLP. None of the subjects suffered from such as medical history of pharyngeal pathology and/or nasal obstruction, snoring, obstructive sleep apnea, adenoidectomy, and tonsillectomy were excluded.

Adenoid hypertrophy was evaluated via e.g. upper (AD2-H) and lower (AD1-Ba). Adenoid thickness showed non significant results, where subjects had thicker upper and lower adenoid when compared to controls but not reached to significant degree. This was in agreement with previous study which concluded that the adenoid hypertrophy in cleft patients did not occur more frequently than normal individuals [14,17]. Although they demonstrated reduction in the pharyngeal airway dimensions when compared with controls.

Also according to a studies carried out by Lowe et al. the cephalometric characteristics showed that adenotonsillar hypertrophy is not dominant criteria in cleft patients [18,19]. In another study, Rose et al. showed that the size of adenoid and tonsil in subjects and controls was found to be equal [13].

However, in another study performed by Brader concluded that there was association between the pharyngeal airway width and the adenoid tissue [20]. He reported that the adenoidal tissue hypertrophy in various types of cleft subjects cause nasopharyngeal size reduction.

Regarding the soft tissue measurements, surrounding the upper airway wall and when control and subject groups were compared, control sample showed a significant increase in the soft palate (MPT) and tongue (TGH) thickness as well as in soft palate length (PNS - P) and this increase confirms and correlates well to the variations in the airway dimensions between the two different groups. Soft palate length in cleft patients was investigated by Coccaro et al. and Mazaheri et al. who reported that soft palate length was shorter in subjects than controls [21,22]. Smahel and Mullerova reported that it became identical length after surgical repair. This present study was showed no statistical difference in tongue length (TGL) in both samples [14].

Regarding hyoid bone position, the perpendicular distance from hyoid bone to mandibular plane (MPH) and to the line connecting C3 and RGN (HH1), distances between hyoid to C3 (C3H) and to RGN (HRGN) were statistically no significant between the two groups. According to Deljo et al. who suggested that the position of hyoid bone is dependent on the antero-posterior position of maxillary and mandibular arches and two different samples were matched skeletally so, the distances from hyoid bone to the antero-inferior limit of the third cervical vertebra (HC3) and to the most posterior point of the symphysis (HRGN) are expected to be in the same position antero-posteriorly in both groups [23].

The first limitation of this study was the alveolar cleft size which did not considered. Second, study sample was small. It is, however, one of the largest studies done on Jordanian preadolescent subjects with UCLP and comparable normal (non-cleft) controls. We recommended

Measurement	Control	Subject	Mean Difference	Significance P – value
PNS – AD1	25.32(±5.40)	20.74(±5.13)	4.59	0.000***
AD1 – Ba	21.69 (±5.66)	22.54(±4.70)	-0.86	0.245
PNS - AD2	19.19 (±4.96)	15.76(±4.19)	3.43	0.000***
AD2 – H	10.84(±3.51)	10.89(±3.66)	-0.05	0.922
PNS – Ba	46.83(±8.43)	43.17(±5.13)	3.66	0.000***
Ptm – Ba	43.57(±7.50)	39.74(±5.40)	3.83	0.000***
PNS – H	29.94(±5.67)	26.50(±4.94)	3.44	0.000***
McNamara 's Upper Pharyngeal dimension	8.31(±2.86)	8.09(±2.94)	0.21	0.605
McNamara 's Lower Pharyngeal dimension	11.36(±3.60)	10.00(±3.44)	1.37	0.007**
SPAS	11.46(±3.51)	10.93 (±3.38)	0.53	0.276
MAS	9.77(±2.83)	9.45(±3.03)	0.31	0.452
IAS	12.21(±3.62)	11.12(±3.80)	1.09	0.039
VAL	57.07(±10.22)	59.27(±9.26)	1.81	0.129

Tables 2: The means, standard deviations, mean differences and significance level of pharyngeal airway cephalometric measurements in the Subjects with cleft lip and palate and Controls groups.
Where, n= 200, *P ≤ 0.05, **P< 0.01, ***P < 0.001,

Measurement	Control	Subject	Mean difference	Significance P – value
TGL	67.37(±13.05)	65.38 (±13.05)	1.99	0.192
TGH	29.72(±6.03)	25.79(±5.31)	3.93	0.000***
PNS – P	34.86 (±6.70)	29.46(±4.48)	5.40	0.000***
MPT	5.22(±1.52)	4.40(±1.25)	0.82	0.000***

Table 3: The means, standard deviations, mean differences and significance level of Soft tissue cephalometric measurements in the Subjects with cleft lip and palate and control groups

Where, n= 200, *P ≤ 0.05, **P< 0.01, ***P < 0.001,

Measurement	Control	Subject	Mean Difference	Significance P – value
M PLANE-H	13.49(±5.74)	14.26(±5.97)	-0.76	0.359
HH	5.75(±4.32)	6.94(±4.32)	-1.19	0.059
C3H	32.11(±6.25)	31.63(±5.48)	0.47	0.569
H RGN	37.09(±9.04)	35.41(±6.26)	1.69	0.126

Table 4: The means, standard deviations, mean differences and significance level of Hyoid bone position cephalometric measurements in the subjects with cleft lip and palate and Controls groups.

Where, n= 200, *P ≤ 0.05, **P< 0.01, ***P < 0.001,

further researches regarding pharyngeal airway dimensions in CLP subjects with larger sample and considering the cleft size.

Conclusion

Jordanian preadolescent CLP characterized by narrowing in the pharyngeal airway dimensions which attributed to skeletal deformations and smaller bony nasopharyngeal not due to soft tissue hypertrophy. Orthognathic surgery is indicated in CLP population to correct skeletal discrepancies and further enhance improve the pharyngeal airway dimensions.

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