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Research Article

PHARYNGEAL AIRWAY DIMENSIONAL CHANGES IN CLEFT LIP AND PALATE PATIENTS FOLLOWING INTRA-ORAL MAXILLARY DISTRACTION-A RETROSPECTIVE CEPHALOMETRIC STUDY

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ABSTRACT

Oro-facial clefts(OFC) are amongst the most commonly occurring craniofacial anomalies(CFA). The occurrence of OFC is of two main types, CL/P and CP. The maxillary hypoplasia is the most commonly encountered secondary deformity in patients with CP. The maxillary hypoplasia often results in compromised mastication, speech and nasal pharyngeal airway insufficiency and finally a weak outlook on the life due to a disturbed self-perception. The patients with such syndromic conditions require early functional rehabilitation along with aesthetic enhancements. The treatment of maxillary hypoplasia often involves maxillary surgical advancements or distraction osteogenesis. A major disadvantage of surgical maxillary advancement is a deterioration of velopharyngeal function as soft tissue moves forward. But, in case of distraction osteogenesis involving anterior maxilla there has been very few articles assessing the pharyngeal airway. The aim of the study was therefore to evaluate the impact of AMD using intra-oral distractors on Pharyngeal airway dimensions in Cleft lip and palate patients. The study consists airway and skeletal parameter assessment of cephalogram evaluation of 15 cleft lip and palate patients, with maxillary hypoplasia who had undergone anterior maxillary distraction osteogenesis with tooth borne internal distractors. The results shows an increase in the entire Velo-pharyngeal dimensions in all the five parameters. Post distraction statistically significant changes in the airway parameters are seen only in PPS and SPSS, also certain statistically significant changes seen in skeletal parameters support the increase in pharyngeal airway as a result of distraction. Thus, Anterior maxillary distraction osteogenesis in cleft lip and palate patients with retrognathic maxilla is proved to be a viable treatment option which not only improves the facial profile and esthetics but also enhances the functional characteristics of the airway by increasing its dimension.

INTRODUCTION

Oro-facial clefts(OFC) are amongst the most commonly occurring craniofacial anomalies(CFA), Their overall prevalence is estimated to be approximately 1 in 700 live births. The occurrence of OFC is of two main types, CL/P and CP, these account for nearly one half of all CFA [1, 2]. As per the reports by the World Health Organization (WHO) in 2001, the prevalence at birth of OFC varies worldwide, ranging from 3.4–22.9 per 10,000 births for CL/P[3]. The Prevalence has been found to vary based on ancestry, with the highest incidence rates observed amongst Asian populations (0.82–4.04 per 1000 live births)[1, 4]. Furthermore, in patients with cleft lip and palate, the most commonly encountered secondary deformity is Maxillary hypoplasia with a reported incidence of about 15-50%[5]. The Maxillary hypoplasia in cleft individuals is partly due to the intrinsic deformity, partly due to genetic inheritance of facial growth pattern, and partly the result of scar from the multiple surgical interventions.[6,7]. The clinical manifestations of maxillary hypoplasia are usually severe concave profile due to midface deficiency and Class III skeletal and dental malocclusion. The maxillary hypoplasia often results in compromised mastication, speech and nasal pharyngeal airway insufficiency and finally a weak outlook on the life due to a disturbed self-perception.

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The patients with such syndromic conditions require early functional rehabilitation along with aesthetic enhancements. The treatment of maxillary hypoplasia often involves maxillary surgical advancements or distraction osteogenesis. With surgical advancement in case of cleft lip and palate relapse, there is often relapse noted in few studies which range from 22 % to 40 % in a horizontal plane and 28-30 from 19 % to 70 % in vertical plane[8]. Another major disadvantage is that maxillary advancement may predispose cleft lip and palate patients to velopharyngeal function deterioration as soft tissue moves forward[9,10]. Since Dolanmaz D *et al*. in 2003[11] reported the first successful clinical application of AMD using a tooth-borne distractor, the technique has shown to produce excellent results in cleft patients with maxillary hypoplasia in terms of facial balance, aesthetics and stable occlusion; however; its effects on speech and Pharyngeal airway outcomes in patients with cleft maxillary hypoplasia after AMD still remains unassessed. The aim of the study was therefore to evaluate the impact of AMD using intra-oral distractors on Pharyngeal airway dimensions in Cleft lip and palate patients.

MATERIALS & METHODS

This study was carried on at Meenakshi Ammal Dental College and Hospital under the approval from Institutional Review Board (2016). The retrospective study enrolled subjects from Meenakshi Cleft and Craniofacial Centre, Chennai treated between 2010-2015. The study consists of 15 cleft lip and palate patients, six males and nine female subjects with an age range of 15–25 years and mean age of 18.2 years with maxillary hypoplasia who had undergone anterior maxillary distraction osteogenesis with tooth borne internal distractors. The inclusion criteria were patients with a unilateral or bilateral cleft, negative overjet of 6mm or above and normal mandibular morphology, The patients with persistent palatal fistulas, syndromic, systemic diseases were excluded.

Each of the patients whose records deemed complete with Pre-treatment (T0) and Post-treatment (T1) Lateral cephalograms are included in the study. The lateral cephalograms were taken using PLANMECA (PROMAX Oy 00880 Finland). The cephalometric tracings were done using the Ilexis FACAD AB-2014 Version 3.8.0.0 software. The Upper-airway and skeletal morphological changes were evaluated using computed cephalometric radiography registered at the intercuspal position with the head parallel to the FH plane (natural head position). Following parameters were assessed at T0 and T1.

Skeletal Parameters

SNA (degrees) - Angle between the anterior cranial base and point A
ANB (degrees) - Angle between lines NA & NB
PP-SN (degrees) - Angle between the palatal plane and anterior cranial base
MP-SN(degrees) - Angle between the mandibular plane (Go-Gn) and anterior cranial base

ANS-PNS (millimetres) - Distance between the anterior nasal spine and posterior nasal spine
N perpendicular – Point A (millimetres) - Linear distance from Nasion perpendicular to point A

Airway Parameters

The palatal pharyngeal space (PPS) - Anteroposterior depth of the pharynx measured between and The posterior nasal spine (PNS) to the posterior pharyngeal wall

The superior posterior pharyngeal space (SPPS) - Anteroposterior depth of the pharynx measured between the dorsum of the soft palate to the posterior pharyngeal wall.

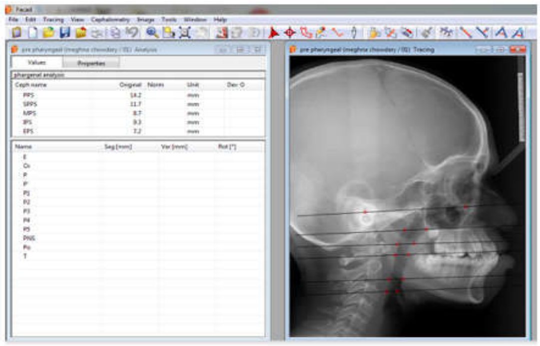


FIGURE 1 – PRE-DISTRACTION AIRWAY ANALYSIS

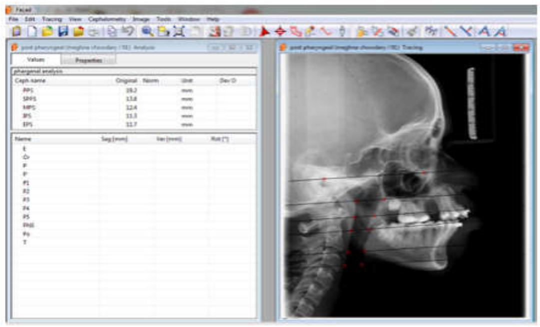


FIGURE 2 – POST-DISTRACTION AIRWAY ANALYSIS

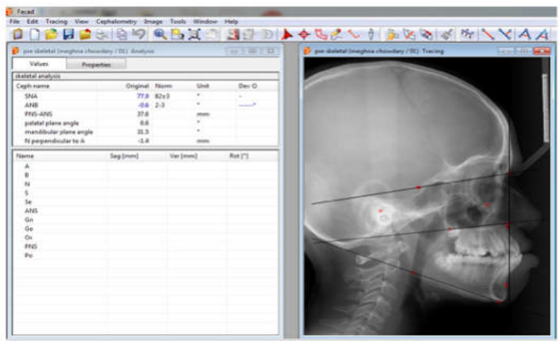


FIGURE 3 – PRE-DISTRACTION CRANIOFACIAL SKELETAL MORPHOLOGY

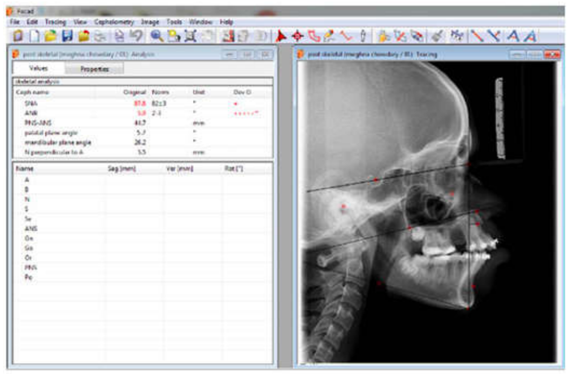


FIGURE 4 – POST-DISTRACTION CRANIOFACIAL SKELETAL MORPHOLOGY

The middle pharyngeal space (MPS) - Anteroposterior depth of the pharynx measured between the dorsum of the tongue to the posterior pharyngeal wall

The inferior pharyngeal space (IPS) - Anteroposterior depth of the pharynx measured between the surface of the tongue to the posterior pharyngeal wall.

The epiglottic pharyngeal space (EPS) - Anteroposterior depth of the pharynx measured between the surface of the tongue to the posterior pharyngeal wall below IPS

Statistical Analysis

The measurements were repeatedly performed at 3-week intervals to estimate the reliability of a single measure. The descriptive statistics including the mean, standard deviation, and minimum/maximum values were calculated for each cephalometric variable for each group using SPSS (version 20.0). To compare the mean values between the pre-surgical and post-surgical pharyngeal airway space volume paired t-test was applied. Spearman correlation was performed to check the associations between maxillary skeletal and pharyngeal variables. A p-value < 0.05 level of significance was used for the test.

RESULTS

All 15 patients treated by intra-oral tooth borne distractors demonstrated marked advancement of the maxilla. The descriptive statistics as summarised in Table-I shows the changes in the cephalometric measurements of the skeletal parameters taken before and after anterior maxillary distraction osteogenesis in cleft lip and palate patients (n=15).

TABLE I: VARIABLES DESCRIBING THE AIRWAY MORPHOLOGY OF THE SAMPLES BOTH PRE-DISTRACTION AND POST-DISTRACTION

PATIENTS	PPS (mm)		SPSS (mm)		MPS (mm)		IPS (mm)		EPS (mm)	
	T_0	T_1	T_0	T_1	T_0	T_1	T_0	T_1	T_0	T_1
PATIENT 1	21.3	21.5	15	14.1	12.7	12.6	16.7	16.5	18	18.6
PATIENT 2	24.5	24.7	22.2	21.3	21	21.6	16.9	17	16.3	17.3
PATIENT 3	20.1	20.2	15.5	15.8	11.7	13.2	9.2	9.8	10.8	11.1
PATIENT 4	21.5	21.5	14.8	15.6	14.1	12	16.3	13.2	14.4	13.2
PATIENT 5	17.4	17.5	12.7	13.2	11.7	11.3	9.1	9.9	12.7	12.6
PATIENT 6	18	19	13.1	13.9	10.5	11.3	7.1	7.8	8.3	8.9
PATIENT 7	28.5	28.1	14.9	14.7	12.5	12.4	10	10.4	14.5	14.1
PATIENT 8	25.4	25.8	21.4	22.2	21.3	21.8	14.1	15.5	17.8	17.3
PATIENT 9	14.9	14.6	11.2	11.8	10.5	10.7	8.7	9.1	10.4	10.6
PATIENT10	12.5	12.8	9.3	9.2	8.2	8.5	6.2	6.5	9.3	9.2
PATIENT 11	15.9	16	10	9.6	10.9	10.7	6.6	6.9	6.7	7.1
PATIENT 12	23	23.5	8.5	8.2	10.5	10	6.4	5.3	10.8	10.6
PATIENT 13	21.5	22.1	17.5	17.2	16.8	15.6	10.7	11.1	9.2	19.4
PATIENT 14	16.9	17	13	13.2	10.9	11.1	5.5	6.4	10.9	10.9
PATIENT 15	16.9	17.1	10.7	11.8	10.7	11.4	5	5.8	12.6	13.1
MEAN	**19.8**	**20.09**	**13.98**	**14.12**	**12.93**	**12.94**	**9.90**	**10.08**	**12.18**	**12.93**

The descriptive statistics in Table II presents the Post distraction(T1) velopharyngeal dimensional changes as compared with Pre-treatment (T0). The cephalometric evaluation showed an increase in the entire Velo-pharyngeal dimensions in all the five parameters. The paired t-test results tabulated in Table.III highlights that palatal pharyngeal space(PPS) showed a statistically significant increase (t=2.346, p=0.034 < 0.05). Also, there exists a greater amount of changes in epiglottic pharyngeal space (EPS) post distraction but not statistically significant. Furthermore, Spearman correlation shows an increase in the PPS before and after Anterior Maxillary Distraction by 0.21mm with With a mean maxillary advancement of 1.1 mm. Also, SPSS is shows a statistically significant increase post distraction.

In this study there exists statistically significant increase in the following craniofacial parameters: SNA, ANB, ANS-PNS and N⊥r – POINT A, and this proved that there were an evident maxillary advancement and upward movement which brought about a positive change in the facial morphology (TABLE IV). Although the palatal plane and the mandibular plane did not show a statistically significant value, clinically significant differences are noted. The palatal plane was tipped upwards in counter-clockwise direction whereas the mandibular plane rotated clockwise.

TABLE II: VARIABLES DESCRIBING THE CRANIO-FACIAL MORPHOLOGY OF THE SAMPLES BOTH PRE-DISTRACTION AND POST-DISTRACTION

PATIENTS	SNA °		ANB °		PP-SN °		MP-SN °		ANS – PNS (mm)		N⊥r – POINT A (mm)	
	T_0	T_1	T_0	T_1	T_0	T_1	T_0	T_1	T_0	T_1	T_0	T_1
PATIENT 1	76.6	78.9	-6.2	-2.5	15.9	34.7	39.2	39.3	26.9	40.7	-1	5
PATIENT 2	81	91.8	2	11.5	9.8	5.1	34.5	33.3	42.7	51.7	3.2	15.7
PATIENT 3	82.7	94.9	-2.3	9.2	5.9	3.2	23.9	25.2	33.6	50.6	-3.2	6.1
PATIENT 4	74.5	82.5	-8.5	-2.6	3.9	3.2	32.3	32.8	36.1	39.9	-14.2	-4.9
PATIENT 5	80.7	93.7	0.7	9.1	1.3	7.7	29.9	23.2	35.1	43.5	0.3	8.7
PATIENT 6	73.4	76.4	-5.9	-2.3	12	6.6	26.6	27.3	29.2	39.4	-10.2	-5
PATIENT 7	79.6	83.7	-1.6	3.5	8.6	3.1	24.2	26.9	43.6	51.6	0.7	1.3
PATIENT 8	83.1	85.4	-3.1	-0.5	5.2	7.1	30.5	31	44.5	45.4	-0.8	2.8
PATIENT 9	80.8	89.8	-2.7	6.9	6.2	1.9	30.9	34.9	40.3	55.4	-2.1	1.7
PATIENT 10	76.2	81.2	-1.1	1.3	5.8	2.9	36.3	35.7	41.7	48.1	-6.5	-4.1
PATIENT 11	78.9	79.7	-3.4	-1.9	6	2.1	28.6	29.1	42.4	48.7	-10.7	-6.5
PATIENT 12	77.4	82.7	-3.9	2.6	9	1.3	28.7	31	43.3	46.7	-7.7	-3.8
PATIENT 13	77.1	75.5	-0.2	1.3	12.1	4.7	30.9	35.4	41	46.5	-4.3	-1.4
PATIENT 14	79.2	81.6	-3.7	-1.2	2.4	1.5	37.8	37.5	37.1	39.1	-3.9	2.1
PATIENT 15	80.9	82.6	5.7	2.4	9.5	3.7	50.9	49.8	43.7	45.6	-3.8	-1
MEAN	**78.80**	**84.02**	**-2.28**	**2.45**	**7.57**	**5.92**	**32.34**	**32.82**	**38.74**	**46.1**	**-4.28**	**1.11**

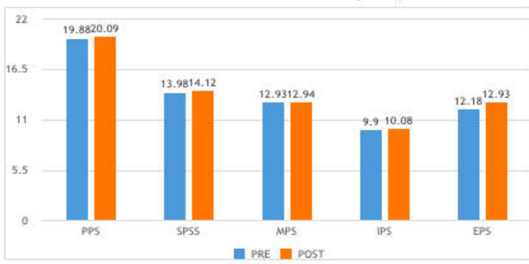
TABLE III: PAIRED DIFFERENCES & TEST RESULTS OF THE AIRWAY PARAMETERS

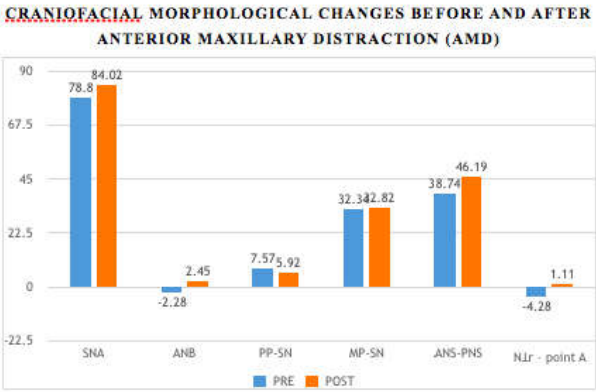
AIRWAY CHANGES	MEAN PAIRED DIFFERENCE	t - Value	p - Value
PPS (mm)	0.2066	2.346	0.034*
SPSS (mm)	0.1333	0.816	0.428
MPS (mm)	0.0133	0.060	0.953
IPS (mm)	0.1800	0.653	0.524
EPS (mm)	0.7533	1.093	0.293

TABLE IV: PAIRED DIFFERENCES & TEST RESULTS OF THE SKELETAL PARAMETERS

CRANIOFACIAL MORPHOLOGICAL CHANGES	MEAN PAIRED DIFFERENCE	t - Value	p - Value
SNA°	5.220	4.586	0.000**
ANB°	4.733	4.700	0.000**
PP-SN°	-1.653	-0.949	0.359
MP-SN°	0.483	1.041	0.316
ANS-PNS (mm)	7.446	5.847	0.000**
N⊥r – POINT A (mm)	5.393	6.488	0.001**

PHARYNGEAL AIRWAY SPACE CHANGES BEFORE AND AFTER ANTERIOR MAXILLARY DISTRACTION (AMD)





DISCUSSION

Cleft lip and palate is a developmental anomaly of the craniofacial structures characterised by maxillary hypoplasia. In such CLP patients with severe maxillary retrusion often present with a constriction of the upper airway. It has been reported that CLP patients have a reduced nasal airway compared with healthy subjects[12] therefore, the airway is impaired and often suffer from nasal airway obstruction[13] and obstructive sleep apnea (OSA)[14].

Past studies have investigated the effect of surgical treatment techniques such as Le Fort I osteotomy, sagittal split ramus osteotomy, or combined bimaxillary surgery on the airway in maxillary hypoplasia in cleft patients[15-18]. Most of these patients are often treated with a mandibular setback surgery to compensate for the severity of maxillary hypoplasia [19]. The drawback here is that most cleft patients have a customarily developed mandible with respect to size and position before surgery which has to be conventionally sacrificed. The Lefort I maxillary advancement procedure results in a reduction of the pharyngeal airway volume [20,21]. This leads to velopharyngeal incompetence, increase in hypernasality, nasal emission and worsening of speech due to the increased distance between the posterior pharyngeal and the palatal musculature which is gets stretched post surgery. But since the inception of maxillary anterior segmental distraction osteogenesis[22,23] as a new surgical-orthodontic treatment to produce an ideal maxillary dental arch form in these patients this procedure can solve problems in the small and retruded maxilla without the risk of deterioration of velopharyngeal function and successful results were obtained in both aesthetic and functional aspects[22]. This nature of enchantment of deteriorated pharyngeal airway is proven from the statistical point of view in our present study, maxillary advancement using intra-oral distraction osteogenesis has lead to an appreciable amount of changes in the upper airway. There exists an increase in palatal pharyngeal space (PPS) and superior posterior pharyngeal space (SPPS) post-distraction. This increase in the PPS and SPSS reported in our study is positively correlated with the statistically significant changes in skeletal parameters of SNA, ANS-PNS and ANB which indicates the relative change in the length of the maxilla. These positive changes reported in our study are in accordance with the study by Mochida *et al.* in 2004 which reports a close link between the change in PNS position and increase in Upper Pharyngeal airway[24].

In the present study, evidential changes are also present in the lower airway dimensions with a meagre increase in the middle pharyngeal space (MPS) and the inferior pharyngeal space (IPS). However, these changes were only evident in patients that presented initially with a steep mandibular plane angle due to a posterior vertical maxillary excess that seemed to lock the mandible in an inferior-posterior position. With the maxillary distraction, the autorotation of mandible into a more anterior position leading to an increase in the lower airway dimensions. These findings also fall in line with results of Mochida *et al.*[24]. A particular study by Harada *et al.*[25] points out the decrease in pharyngeal airway post distraction exceed 15mm, by in case of our present study the mean maxillary distraction was 7 mm such a VPI has not been noted as our study being a cephalometric. Also, a clinically evident increase reported in our study, correlated with a study by Abunizada *et al.* in 2016[26] the anteroposterior distance of the superior velopharynx (PPS) and middle velopharynx (SPPS), and with less increase in the inferior velopharynx (MPS) and the oropharynx (IPS, EPS).

Drawbacks of our study are that we evaluated the airway changes on lateral cephalometric radiographs. These measurements are only two dimensional, however, the upper airway in a three-dimensional space. The measurements of the upper airway volume on a three-dimensional nature would give a more accurate picture of these changes. A recent study by, Xu *et al.* measured the pharyngeal airway volume changes in syndromic patients who underwent midface distraction osteogenesis and found a 64% enlargement after a mean of 20 mm advancement[27]. Further, a 13% to 21% increase in the nasopharyngeal airway dimensions with maxillary advancement was reported by Jakobson *et al.* [28]. Future studies to evaluate upper airway changes after maxillary distraction using cone beam CT would give valuable answers to valuable questions like how maxillary DO change the upper airway in CLP patients. Although all patients reported clinical improvement in breathing during sleep, future studies to evaluate upper airway changes with volumetric measures using cone beam CT would help unveil the mystery of airway changes in CLP patients after maxillary DO.

CONCLUSION

The following conclusions were obtained from the present study Anterior maxillary distraction osteogenesis increased the Palatal Pharyngeal Space (PPS) in cleft lip and palate patients with retrognathic maxilla.

With a mean maxillary advancement of 1.1 mm, the palatal pharyngeal space showed about 1% (i.e.) 0.21 mm of improvement which was statistically significant. There was a positive correlation between the amount of maxillary advancement and the increase in pharyngeal space.
Anterior maxillary distraction osteogenesis improved the midfacial skeletal and soft tissue profile with remarkable improvement in the facial aesthetics and occlusion.

Anterior maxillary distraction osteogenesis in cleft lip and palate patients with retrognathic maxilla is proved to be a viable treatment option which not only improves the facial profile and esthetics but also enhances the functional characteristics of the airway by increasing its dimension.

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