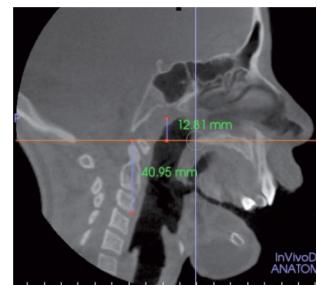


# Evaluation of Airway Volume Changes in Patients Treated with Distraction Osteogenesis: A Pilot Study



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## ABSTRACT

*The use of distraction osteogenesis to improve the airway volume of individuals affected by craniofacial anomalies is common practice today. The methods of assessing the outcomes of such surgical procedures have changed over the last several years.*

**Objective :** The objective of the present pilot study was to determine if the CBCT imaging modality may serve as a simple and reliable method for craniofacial practitioners to evaluate airway volume changes following distraction osteogenesis.

**Materials and Methods :** Twelve patients who had previously undergone distraction osteogenesis for the primary purpose of improving their airways were enrolled in the current study. Pre (T0) and post-surgical (T1) CBCT's volumes were analyzed to measure the nasopharyngeal (NP) and oropharyngeal (OP) volume changes. InVivoDental 4.0 (Anatomage Inc., San Jose, CA) (IVD) program was used to visualize and render the oropharyngeal (OP) and nasal passage (NP) volumes, separately. Means and standard deviations were calculated.

**Results :** Of the 7 males and 5 females in the study, 4 patients underwent mandibular distraction (MandDO) and 8 underwent maxillary distraction (MaxDO). Four in the MaxDO group were treated with internal distractors and 4 were treated with external distractors. Individuals who underwent internal

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*MaxDO greatly improved their NP volumes ( $1605 \pm 1736$  to  $3273 \pm 3130 \text{ mm}^3$ ) as did patients who underwent external MaxDO ( $5763 \pm 5077$  to  $9243 \pm 9442 \text{ mm}^3$ ). In comparison, the MandDO group's NP volume showed modest gains ( $3519 \pm 1944$  to  $3894 \pm 2516 \text{ mm}^3$ ). With regard to the OP volumes, the MandDO group gained substantially ( $4906 \pm 2347$  to  $11385.8 \pm 6393.5 \text{ mm}^3$ ) while the MaxDO groups showed humble increases; internal MaxDO group ( $1779 \pm 273.8$  to  $2639.5 \pm 898.4 \text{ mm}^3$ ) and the external MaxDO group ( $8959 \pm 5311$  to  $9734.13 \pm 7124.7 \text{ mm}^3$ ).*

**Conclusions :** *In the present preliminary study of assessing airway volume changes using CBCT on patients who have undergone different DO techniques, MandDO greatly increases OP volumes and MaxDO tends to increase NP volumes. The CBCT imaging modality holds great promise for craniofacial practitioners in its application to assess airways of individuals affected by craniofacial anomalies.*

#### KEYWORDS

*Airway volume  
Distraction osteogenesis  
CBCT imaging.*

## INTRODUCTION

Craniofacial anomalies involving the midface, such as craniofacial synostoses (Crouzon, Apert and Pfeiffer syndromes), anomalies involving primarily the mandible (Nager, Sticklers and Pierre Robin sequence) or anomalies associated with both the maxilla and mandible (Treacher Collins syndrome, hemifacial microsomia) can lead to a decrease in volume of the oropharyngeal and nasopharyngeal airway. The altered size and position of the maxilla and mandible typically cause a retro displacement of the tongue which results in a reduction of the oropharyngeal airway and may lead to upper airway obstruction. Affected

patients often suffer from Obstructive Sleep Apnea (OSA) and in severe cases are treated by early tracheal intubation and tracheostomies<sup>1</sup>.

Formerly, a variety of surgical procedures were indicated for these patients in order to increase airway volume or allow for easier respiration. Distraction Osteogenesis (DO) has become an accepted method of treatment for patients requiring reconstruction of hypoplastic mandibles/maxillae and achieve lengthening of the jaw without the need for bone grafts<sup>1</sup>. DO is based on manipulation of a healing bone and stretching an osteotomized area before calcification has occurred,

to generate additional bone and investing soft tissue<sup>2</sup>. Such procedures aim to lengthen, modify and reposition the affected mandible/maxilla by performing one or more osteotomies, manipulating the resulting bone fragments into a more desirable configuration, and “growing” the mandible through daily distraction<sup>3</sup>. The goal of the procedure is to reposition the bones in a more anterior position through the process of distraction and thereby open up the airway.

The advent of cone-beam computerized tomography (CBCT) in this decade, allows us to quantitatively evaluate the cross-sectional area and the 3-dimensional volume of the airway utilizing a lower radiation levels

than traditional medical CT's, with a rapid (< 20 seconds) scan that is non-invasive<sup>4</sup>.

The information within the (3D) CBCT image data includes hard and soft tissue structures, allowing visualization of the pharyngeal airway without any obstruction of bony structures.<sup>5</sup> Segmentation of the nasopharyngeal and oropharyngeal airways, allows for these compartments to be independently evaluated.

The purpose of this retrospective, descriptive, pilot study was to use CBCT images of patients affected by craniofacial anomalies, who had undergone DO, and to compare the pre-surgical to post-surgical changes in

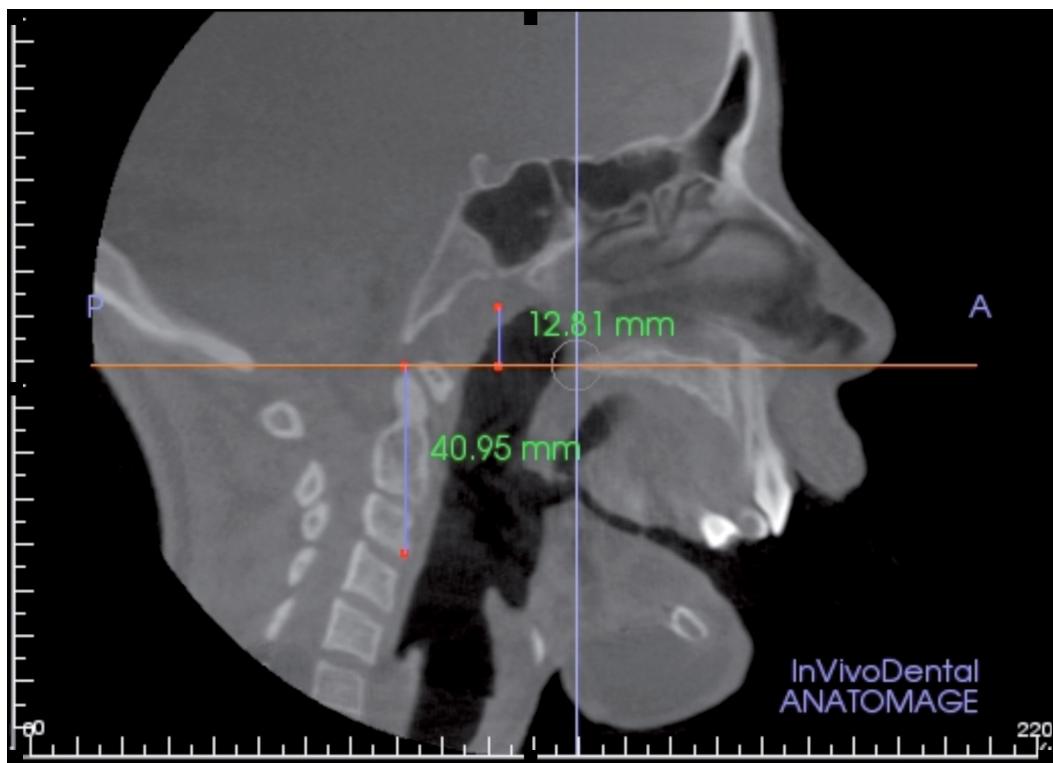
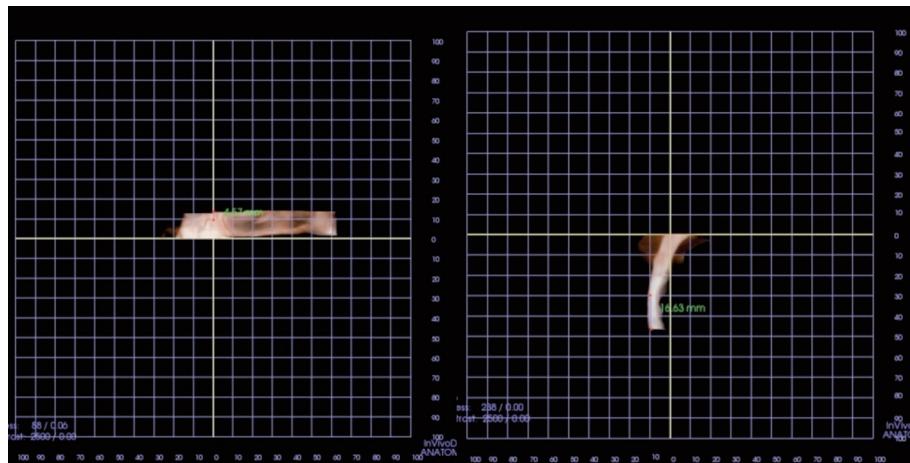


Figure 1

Illustrates the superior and inferior borders of the Nasopharyngeal (NP) and oropharyngeal (OP) volumes respectively.



*Figures 2 and 3  
Segmented NP and OP volumes.*

nasopharyngeal (NP) and oropharyngeal (OP) airway volumes. Specifically, we intended to identify if the CBCT

imaging modality could detect changes in the airway of patients treated with DO in this pilot study.

## MATERIALS AND METHODS

The experimental protocol used in this study was approved by the Case Western Reserve University Institutional Review Board. Seventeen patients affected by different craniofacial anomalies and previously treated with DO were selected from the pool of patients in the Department of Orthodontics. Due to extensive previous histories of DO, unavailability of pre or post operative CBCT scans, and/or unidentifiable landmarks, five patients were excluded from the study. The final sample consisted of 12 individuals (7 males and 5 females) and ranged in age from 3 years 8 months to 17 years 2 months. All included patients had initial (T0) and post-operative (T1) CBCT scans. Of the 12 patients, 8 were treated with maxillary distraction (MaxDO) and

four with mandibular distraction (MandDO). 4 of the MaxDO patients were treated with internal distractors and 4 with external distractors. In order to blind the operator, subject volumes were de-identified and the volume sets were not separated based on the procedure (MaxDO or MandDO). Similar DO protocol was used for the subjects. Distraction commenced at a rate of 1mm/day after a 5-7 day latency period. Duration of distraction was based on the improvement of the maxillary/mandibular position and improvement in airway. Anterior crossbite/overjet and was recorded clinically. In most cases, intentional overcorrection was achieved to negate a tendency to relapse and to account for continued growth.

The InVivoDental 4.0 (Anatomage Inc., San Jose, CA) (IVD) program was used to visualize and render the oropharyngeal (OP) and nasal passage (NP) volumes, separately. Patients were first viewed in the sagittal plane. In order to find the midsagittal point, images were scanned until the incisal foramen was readily visible as well as the anterior nasal spine and posterior nasal spine. Once this position was identified, images were reoriented so that their horizontal plane aligned through the anterior nasal spine and posterior nasal spine. OP volume was defined as the volume of pharynx between the palatal plane (ANS-PNS) extending to the posterior wall of the pharynx and the plane that is parallel to the palatal plane which passes from the most anteroinferior point of the third cervical vertebrae (Fig. 1). The inferior limit of the NP airway was defined as the superior limit of the OP airway and the superior limit was defined as the last slice before the nasal septum fused with the posterior wall of the pharynx. In order to better

view this fusion, the superior border of the NP was defined on the axial slice first and then reflected to the sagittal plane. The NP volume was rendered as a whole structure and included nasopharynx, turbinates and nares. The described airway volumes were rendered with the IVD program, according to the manufacturer's recommendations<sup>6</sup>. The IVD program allows the user to "sculpt out" the desired airway volume from the rest of the 3D structure, and by adjusting the brightness and opacity values one can clean out the unwanted voxels before calculating the final airway volume (Fig. 2 and 3). The program also allows the user change the threshold values in order to obtain a solid airway volume. Operator reliability was tested by repeating all measurements 2 weeks apart and using the Spearman's rho correlation coefficient. The pre-distraction and post-distraction Spearman's = 0.965 and 0.984 respectively. Both were deemed highly significant.

## RESULTS

The descriptive demographics of our data are provided in Table I. The mean age of the total sample in the beginning of treatment (T0) was  $10.28 \pm 5.27$  years.

Age at T0 ranged from 3.67 years to 20.83 years. Mean age at the postoperative stage (T1) was  $12.12 \pm 5.34$  years and ranged from 4.08 years to 21.25 years.

Table II and III present the individual cases with the means  $\pm$  standard

deviations grouped by the type of DO performed.

Of the 7 males and 5 females in the study, 4 patients underwent mandibular distraction (MandDO) and 8 underwent maxillary distraction (MaxDO). 4 in the MaxDO group were treated with internal distractors and 4 were treated with external distractors.

**The most prominent change in OP airway volume was observed for**

Type of DO	Gender	Age at Initial Examination	Days from T0-T1	Mean # of days from T0-T1
<b>Mandibular Distraction Patients</b>	M	11 Years, 0 Months	855	958.25 ± 157
	M	4 Years, 6 Months	911	
	F	11 Years, 8 Months	875	
	F	11 Years, 6 Months	1192	
<b>Maxillary Distraction Patients (Internal Distractor)</b>	M	13 Years, 10 Months	372	344.75 ± 218
	F	3 Years, 8 Months	144	
	F	8 Years, 10 Months	641	
	M	4 Years, 0 Months	222	
<b>Maxillary Distraction Patients (External Distractor)</b>	M	9 Years, 2 Months	503	458.25 ± 291
	F	7 Years, 2 Months	841	
	M	20 Years, 10 Months	160	
	M	17 Years, 2 Months	329	

Table I:

Demographics and procedures performed on all 12 patients with time points in days from T0-T1.

Type of DO	Patient ID	Gender	NP volume T0 (mm <sup>3</sup> )	NP volume T1 (mm <sup>3</sup> )	NP% Change
<b>MandDO</b>	Case ID 1	M	1316	1045	-20.59
	Case ID 2	M	4994	4827	-3.34
	Case ID 3	F	4441	4712	6.10
	Case ID 4	F	4246	5811	36.86
<b>Means ± stdev</b>			<b>3519 ± 1944</b>	<b>3894 ± 2516</b>	
<b>MaxDO (Internal Distractor)</b>	Case ID 5	M	3744	6423	71.55
	Case ID 6	F	153	288	88.24
	Case ID 7	F	2289	5487	139.71
	Case ID 8	M	232	892	284.48
<b>Means ± stdev</b>			<b>1605 ± 1736</b>	<b>3273 ± 3130</b>	
<b>MaxDO (External Distractor)</b>	Case ID 9	M	2215	2606	17.65
	Case ID 10	F	678	1307	92.77
	Case ID 11	M	10980	21737	97.97
	Case ID 12	M	9170	11323	23.36
<b>Means ± stdev</b>			<b>5763 ± 5077</b>	<b>9243 ± 9442</b>	

Table II:

NP volume changes denoted in means and standard deviations, from T0 to T1, for the three different distractors used.

Type of DO	Patient	Gender	OP volume T0 (mm <sup>3</sup> )	OP volume T1 (mm <sup>3</sup> )	OP% Change
<b>MandDO</b>	Case ID 1	M	5783	7924.50	37.03
	Case ID 2	M	2246	4760.00	111.93
	Case ID 3	F	2906.5	19246.50	562.19
	Case ID 4	F	6688	13612.50	103.54
<b>Means ± stdev</b>			<b>4906 ± 2347</b>	<b>11385.88 ± 6393.5</b>	
<b>MaxDO (internal Distractor)</b>	Case ID 5	M	1962	2520.50	28.47
	Case ID 6	F	1388	3760.50	170.93
	Case ID 7	F	1790	2708.00	51.83
	Case ID 8	M	1975	1569.00	-20.56
<b>Means ± stdev</b>			<b>1779 ± 273.8</b>	<b>2639.5 ± 898.4</b>	
<b>MaxDO (internal Distractor)</b>	Case ID 9	M	3736	1966.00	-47.38
	Case ID 10	F	5320	5535.50	4.06
	Case ID 11	M	14944	16882.00	12.97
	Case ID 12	M	11837	14553.00	22.95
<b>Means ± stdev</b>			<b>8959 ± 5311</b>	<b>9734.13 ± 7124.7</b>	

Table III:

OP volume changes denoted in means and standard deviations, from T0 to T1, for the three different distractors used.

**MandDO patients.** The volume difference between T0 and T1 time intervals in MandDO subjects ( $4906 \pm 2347$  to  $11385.8 \pm 6393.5$  mm<sup>3</sup>) was substantially greater than the changes noted in the internal ( $1779 \pm 273.8$  to  $2639.5 \pm 898.4$  mm<sup>3</sup>) and external ( $8959 \pm 5311$  to  $9734.13 \pm 7124.7$  mm<sup>3</sup>) MaxDO subjects.

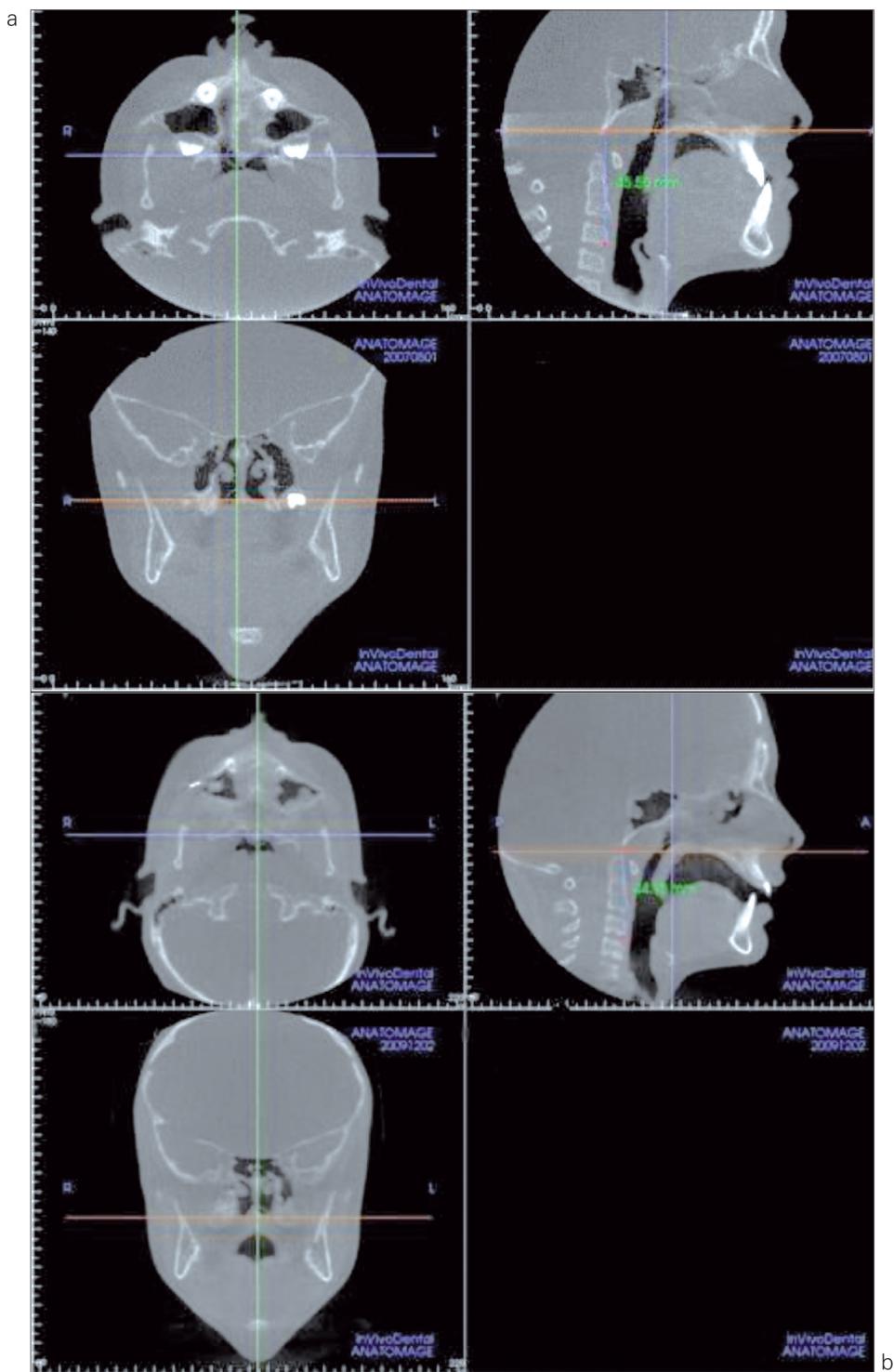
**On the contrary, Individuals who underwent internal MaxDO greatly improved their NP volumes** ( $1605 \pm 1736$  to  $3273 \pm 3130$  mm<sup>3</sup>) as did patients who underwent external MaxDO ( $5763 \pm 5077$  to  $9243 \pm 9442$  mm<sup>3</sup>). In comparison, the MandDO group's NP volume showed modest gains ( $3519 \pm 1944$  to  $3894 \pm 2516$  mm<sup>3</sup>).

## DISCUSSION

Recent advances in CBCT technology makes it a popular static imaging modality to evaluate the airway. The reason for this interest is mainly due to the considerably lower radiation

doses of CBCT devices when compared to helical CTs<sup>7,8</sup>.

There is also a significant improvement in soft tissue contrast when compared to X-ray cephalometry<sup>9</sup>.



Figures 4 and 5

Noticeable pre to post distraction changes on an individual who underwent external distraction of the nasomaxillary complex.

The use of commercially available or manufacturer supplied segmentation programs allows precise measurements of cross-sectional areas at different levels as well as three-dimensional reconstruction and volumetric assessment. In a previous study, the intraoperator reliability of the IVD program, an automatic segmentation program, was found high<sup>10</sup>. The same methodology was used as in the previous study and one calibrated operator (T.E.M.) rendered and measured all of the airway volumes using the same software package. Therefore, when proper technique and protocol are followed, the validity of this method may be considered high. Most of the patients with craniofacial anomalies require airway management ranging from prone positioning to tracheotomy and DO<sup>11</sup>. DO is accepted as a reliable surgical technique and an alternative to tracheotomy for long-term management of the airway<sup>11,12,13</sup>. It has been observed that DO reduces the need for tracheotomy especially in pediatric patients and allows for early decannulation in patients who previously underwent tracheotomy<sup>14,15</sup>. In addition, oral feeding is significantly improved after DO<sup>16,17</sup>. Surgical intervention using DO in early childhood is a common approach. However, children with less obvious craniofacial anomalies and mild airway competence problems may seek treatment later in adolescence, even in their adult years.<sup>18</sup> DO treatment is also performed successfully in the treatment of patients with craniofacial anomalies during their adolescent and adult years as noted in our sample.

Different types of distraction devices are used for the treatment of

craniofacial anomalies. These are mainly classified as external and internal distraction devices and they have been used extensively for the maxillary and mandibular region according to the patients' needs. In the current study, subjects with mandibular retrusion were treated with internal and external mandibular distraction devices. However, given the small sample and the limited objective of the study; to evaluate airway changes using the CBCT modality, no effort was made to create sub-groups. Subjects with maxillary hypoplasia were treated with external and internal devices. Internal devices require a second surgery for removal and a greater degree of difficulty has been associated with controlling the distraction vector<sup>19</sup>. In order to eliminate the need for surgical removal at the end of treatment, several resorbable distraction devices have also been introduced more recently<sup>20,21</sup>. Patients tend to tolerate the internal devices better. Perhaps the main disadvantages of external distraction devices are their bulky construction and the psychosocial impact on the patients' life<sup>22</sup>. Yet, they appear to provide more extended bone osteogenesis advancement when compared to internal devices and the distraction vectors are under better operator control<sup>23,24,25</sup>. This may explain why a bigger NP airway volume change has been obtained in external MaxDO subjects when compared to the internal MandDO subjects (Fig. 4 and 5). It was an interesting finding that the changes mainly occurred in the NP region in the MaxDO patients and in the OP region for the MandDO patients. Xu *et al.* conducted a CT study on 11 patients with severe

midface retrusion in young patients who underwent a LeFort III osteotomy with DO and concluded that DO can significantly improve the upper airway space<sup>26</sup>. However, they noted that the enlargement in airway space was especially above the uvula and posterior nasal spine level (nasal cavity region) which was in agreement with the current study. They were unable to show any statistically significant difference for the airway volume at the level of epiglottis and esophagus. On the other hand, hyoid bone is known to have an important effect over the upper airway volume, especially the OP region. When the anatomic relationship of the hyoid bone to the soft tissues of the mandible and upper airway are taken into account, any displacement of the hyoid bone away from the posterior pharyngeal wall is believed to aid in increasing the airway size<sup>27,28</sup>. Furthermore, the small size of the mandible and its retruded position, cause a corresponding retrodisplacement of the tongue and reduction in the OP airway. In such conditions mandibular DO has been proposed as a useful method to resolve OP airway obstruction<sup>29,30,31</sup>. The mean OP volume at T1 was approximately 2.5 times the T0 time

interval. This finding supports the aforementioned studies in terms of effectiveness of mandibular DO treatment over the OP airway.

There were limitations to the study that must be addressed.

- The limited sample size does not allow us to make definite conclusions.
- We did not have a homogeneous sample in terms of age/gender/specific anomaly. However, we feel comfortable demonstrating, as many others referenced from the past, that DO is a valuable surgical technique in improving airway volume. Importantly, to our knowledge, this is the first CBCT study, albeit a pilot, that demonstrates the use of CBCT technology to effectively evaluate the effects of DO on NP and OP airway volume.
- The average number of days that elapsed before the T1 CBCT volumes may also come in to question. However, this is not an issue easily controlled as the resolution of post surgical edema shows large individual variations.

Further studies are needed to validate trends identified in this pilot effort.

## CONCLUSIONS

The current study indicates that distraction osteogenesis enhances the airway and is an extremely useful surgical technique in managing the airway of individuals affected by

craniofacial anomalies. Additionally, the study suggests that CBCT imaging modality is a beneficial and reliable method of assessing the changes in the airway.

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