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Effects of maxillary distraction osteogenesis on the upper-airway size and nasal resistance in subjects with cleft lip and palate

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Structured Abstract

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Objectives – To investigate the short- and long-term effects of maxillary distraction osteogenesis (DOG) on the upper-airway size and nasal resistance in nine patients with cleft lip and palate (CLP).

Study design – Changes in the upper-airway size were measured by using lateral cephalometric radiographs taken immediately before and after DOG, and 1 year later. Nasal resistance was measured with a rhinomanometer. An analysis of variance was used to establish statistical significance. Spearman correlation coefficient was used to evaluate the relationship between changes in the cross-sectional area of the upper airway and nasal resistance in association with DOG.

Results – Immediately after DOG, the anteroposterior dimension of the superior part of the upper airway was significantly increased ($p < 0.01$) and nasal resistance was significantly decreased ($p < 0.05$). Moreover, the cross-sectional area of the total upper airway was significantly increased ($p < 0.01$). There was a significant correlation between the increase in the upper-airway cross-sectional area and the reduction in nasal resistance ($p < 0.05$). The upper-airway size was significantly augmented ($p < 0.05$) and nasal resistance was significantly reduced ($p < 0.05$) at 1 year after DOG compared with immediately before DOG.

Conclusion – An increase in the upper-airway size and a reduction in nasal resistance occurred after maxillary DOG in patients with CLP, and these changes were stable after 1 year.

Key words: cleft lip and palate; maxillary distraction; upper airway

Introduction

Scar tissue in both the orofacial skin and mucous membrane has been regarded as a crucial factor that affects the stability of the maxilla after conventional orthognathic surgery in subjects with cleft lip and palate (CLP) (1). Distraction osteogenesis (DOG), the gradual formation of new bone by progressive lengthening (2,3), has widely been applied in the orofacial region to overcome the drawbacks of so-called 'one-step' orthognathic surgery such as Le Fort I osteotomy. Recently, this technique has been widely used in the mid-face region in subjects with CLP who exhibit a retruded maxilla.

There are at least two advantages in maxillary DOG over conventional orthognathic surgery in subjects with CLP. First, the soft tissue is remodelled in harmony with the hard tissue. This may counteract relapse of the maxillary bone more effectively than conventional surgical intervention. Second, the maxilla can be moved forward to a greater extent. The greater anterior displacement of the maxilla may induce more dramatic changes in the upper-airway structure and function, including an enlargement of the upper-airway caliber and a reduction in airway resistance. However, only a few studies have investigated morphological and functional changes in the upper airway longitudinally in subjects with CLP who underwent maxillary DOG (4,5). In these previous studies, the authors demonstrated longitudinal changes in velopharyngeal function by measuring cephalometric parameters at the level of the palate (4,5). Neither respiratory function nor cephalometric upper-airway variables below the soft palate were evaluated in these studies (4,5).

The present study was performed 1) to define the short-term effects of maxillary DOG on upper-airway size and nasal resistance in subjects with CLP, and 2) to determine whether these changes were stable after 1 year.

Materials and methods

The study was carried out in nine subjects (five males and four females) with CLP. They consisted of six subjects with unilateral CLP, two with bilateral CLP and one with isolated cleft palate (Table 1). All of the subjects were treated at Tokyo Medical and Dental University Dental Hospital. Prior to the study, all of the subjects gave their informed consent after receiving a full explanation of the aim and design of this study. The age at the time of operation, i.e. bone division by a high Le Fort I osteotomy, was 17.6 ± 4.6 (mean ± SD) years. A rigid external distraction (RED) device (6,7) for maxillary DOG was fitted on the same day of bone division. After a latency period of 2–6 days, i.e. the time between bone division and the initiation of traction force, lengthening was started at a rate of 0.5–2.0 mm a day for a total of 20.9 ± 7.7 days.

Table 1. **Characteristics and demographic variables for nine subjects with cleft lip and palate**

Subject no.	Gender	Cleft type	Hypernasality	Age at operation (years)	Duration of advancement (days)	Amount of advancement (mm)*
1	F	UCLP	N	11.9	30	10.5
2	M	UCLP	Y	15.6	20	17.0
3	M	BCLP	N	12.7	22	10.0
4	F	UCLP	Y	19.3	20	14.0
5	M	UCLP	N	25.9	35	14.5
6	M	CP	N	18.4	11	14.5
7	F	UCLP	Y	14.1	21	9.0
8	F	UCLP	Y	18.3	16	12.0
9	M	BCLP	N	22.0	13	10.0

*The amount of advancement was measured as displacement of the A point between immediately before and after maxillary distraction osteogenesis.

F, female; M, male; UCLP, unilateral cleft lip and palate; BCLP, bilateral cleft lip and plate; CP, isolated cleft plate; Y, presence of hypernasality before study; N, absence of hypernasality before study.

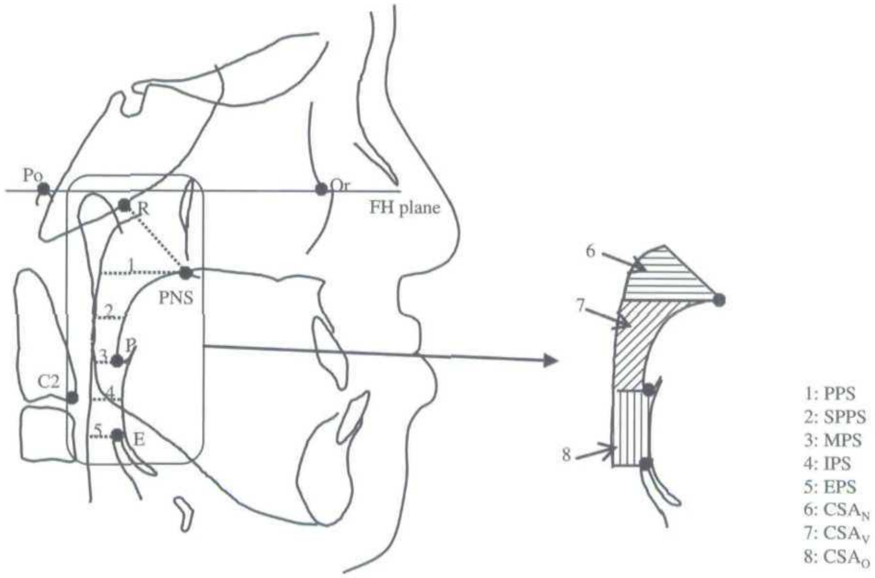

Fig. 1. Diagram of anatomic points and lines used to identify cephalometric variables, and definitions of the area variables. *P*, tip of the soft palate; *E*, tip of the epiglottis; *R* (roof of the pharynx), the point on the posterior pharyngeal wall constructed by the line from the PNS to the intersection of the cranial base and the lateral pterygoid plate; *C2*, the most anteroinferior point on the body of the second cervical vertebra. Abbreviations for figures 1–3. 1: PPS, palatal pharyngeal space. The anteroposterior depth of the pharynx measured between the posterior pharyngeal wall and the PNS on a line parallel to the FH plane that runs through the PNS; 2: SPPS, superior posterior pharyngeal space. The anteroposterior depth of the pharynx measured between the posterior pharyngeal wall and the dorsum of the soft palate on a line parallel to the FH plane that runs through the middle of the line from the PNS to P; 3: MPS, middle pharyngeal space. The anteroposterior depth of the pharynx measured between the posterior pharyngeal wall and the dorsum of the tongue on a line parallel to the FH plane that runs through P; 4: IPS, inferior pharyngeal space. The anteroposterior depth of the pharynx measured between the posterior pharyngeal wall and the surface of the tongue on a line parallel to the FH plane that runs through C2; 5: EPS, epiglottic pharyngeal space. The anteroposterior depth of the pharynx measured between the posterior pharyngeal wall and the surface of the tongue on a line parallel to the FH plane that runs through the tip of the epiglottis; 6: CSA_N (sagittal cross-sectional area of the nasopharynx), the area surrounded by a line between R and the PNS, the posterior pharyngeal wall and the PPS; 7: CSA_V (sagittal cross-sectional area of the velopharynx), the area surrounded by the PPS, MPS, posterior pharyngeal wall and the dorsum of the soft palate; 8: CSA_O (sagittal cross-sectional area of the oropharynx), the area surrounded by the MPS, EPS, posterior pharyngeal wall and the dorsum of the tongue.

Dentoskeletal and upper-airway morphological changes were evaluated by computed cephalometric radiography (FUJIX FCR7000; Fujifilm, Tokyo, Japan) registered at the intercuspal position with the head parallel to the FH plane. Lateral cephalometric radiographs were taken using a cephalostat (HD-150B-30; Shimadzu, Kyoto, Japan) in each subject immediately before (17.6 ± 4.5 years; T1) and after (17.8 ± 4.6 years; T2) DOG, and 1 year after DOG (18.7 ± 4.6 years; T3). The landmarks and contours that are commonly used in orthodontic analysis and additional variables for the upper airway (8,9) were defined (Fig. 1). The superior level of the velopharynx was defined as a line that passed through the PNS parallel to the FH plane, whereas the inferior margin of the velopharynx and superior margin of the oropharynx passed through P (tip of the soft palate) parallel to the FH plane. The inferior part of the oropharynx was defined as a line that passed through E (tip of the epiglottis) parallel to the FH line. The displacement of the PNS was evaluated according to x–y coordinates; the x-axis was parallel to the FH plane, and the y-axis was perpendicular to the x-axis. The same investigator (MM) traced all cephalometric radiographs, and the method error (10,11) for each parameter was calculated by comparing duplicate tracings. The method error was calculated to assure the reproducibility of measurement: $(d^2/2n)^{1/2}$, where d represents the difference between the first and second measurements, and n denotes the sample size.

We measured nasal resistance five times each at T1, T2 and T3 by the anterior-nozzle method (12) with a rhinomanometer (MPR3100; Nihon Kohden, Tokyo, Japan). The mean of these five values was calculated for each patient. An analysis of variance was performed to test whether there were significant differences in the mean value of each subject at each time-point. A Spearman correlation coefficient by rank was used to evaluate the relationships between measured values. A *post hoc* power analysis was used to confirm the statistical power of the study.

Table 2. **Changes in cephalometric variables in association with maxillary distraction osteogenesis in nine subjects with cleft lip and palate**

	SNA (°)			SNB (°)			Mandibular plane angle (°)		
Subject no.	T1	T2	T3	T1	T2	T3	T1	T2	T3
1	82.0	91.0	88.0	82.0	82.0	82.0	33.5	33.5	34.5
2	63.5	75.0	76.0	71.0	67.5	71.0	43.0	50.0	46.0
3	82.0	90.0	88.0	76.5	77.5	77.0	45.0	45.5	45.0
4	70.0	80.0	78.0	81.0	77.5	77.0	33.0	37.5	37.5
5	73.5	85.0	82.0	77.0	76.0	77.0	47.0	47.0	47.0
6	67.5	80.0	79.0	86.0	84.0	84.5	33.5	37.5	35.5
7	74.0	82.5	82.0	76.0	75.0	77.0	30.0	31.5	30.0
8	73.0	85.0	82.5	79.0	77.0	78.5	32.0	36.0	32.0
9	70.5	78.5	76.0	73.0	73.0	72.0	35.0	36.5	35.0

T1, immediately before maxillary distraction osteogenesis (DOG); T2, immediately after DOG; T3, 1 year after DOG.

Results

The method error for each variable of the upper airway ranged from 0.16 to 0.87 mm (mean: 0.46 mm), which is comparable with the results in previous studies (10,11). The power for comparisons of upper-airway size was 0.99 for palatal pharyngeal space (PPS), 0.50 for superior posterior pharyngeal space (SPPS), 0.29 for middle pharyngeal space (MPS), 0.14 for inferior pharyngeal space (IPS) and 0.36 for epiglottic pharyngeal space (EPS). Changes in cephalometric variables associated with maxillary DOG in individual subjects with CLP are provided in Table 2. SNA consistently increased from T1 to T2, however, it decreased to various degrees from T2 to T3 in eight (89%) subjects. On the other hand, SNB showed little or no change from T1 to T2, or from T2 to T3. The mandibular plane showed clockwise rotation in most (7/9, 78%) subjects from T1 to T2. The mandibular plane angle decreased in most of these seven subjects (6/7, 86%) from T2 to T3, and the remaining subject showed no change.

Immediately after maxillary DOG, a significant increase ($p < 0.01$) in the anteroposterior distance was seen in the superior (PPS) and middle (SPPS) velopharynx (Fig. 2). The PPS then significantly decreased ($p < 0.01$), while the SPPS showed no significant changes between T2 and T3. However, there were significant increases in both the PPS ($p < 0.01$) and SPPS ($p < 0.05$) between T1 and T3. On the other hand, variables that reflected the anteroposterior distances of the inferior velopharynx (MPS) and oropharynx (IPS and EPS) showed no significant changes between T1 and T2, T2 and T3, or T1 and T3. The sagittal cross-sectional area (CSA) of the nasopharynx (CSA_N) showed a significant increase ($p < 0.01$) immediately after DOG (Fig. 3). This increase leveled off and showed no significant change between T2 and T3. Overall, there was a significant increase ($p < 0.01$) in CSA_N between T1 and T3. On the other hand, the CSA of both the velopharynx (CSA_V) and oropharynx (CSA_O) showed no significant changes between T1 and T2, T2 and T3, or T1 and T3. The summed sagittal CSA of the nasopharynx, velopharynx and oropharynx (CSA_{N+V+O}) showed a significant increase ($p < 0.01$) immediately after DOG, and then showed a significant decrease ($p < 0.05$) at 1 year after DOG. However, there was an overall significant increase ($p < 0.01$) in CSA_{N+V+O} between T1 and T3.

Nasal resistance showed a significant decrease ($p < 0.05$) at T2 compared with T1, while there was no significant change between T2 and T3 (Fig. 4). Thus, there was a significant decrease ($p < 0.05$) in nasal resistance between T1 and T3. The change in the total CSA of the upper airway, i.e. CSA_{N+V+O}, and the change in nasal resistance between T1 and T2 were significantly correlated ($p = 0.03$, $r = -0.60$, Fig. 5).

The amount of advancement by DOG was 12.4 ± 2.7 mm (range: 9.0–17.0 mm), whereas the total displacement of the PNS was 11.3 ± 2.0 mm, i.e. anterior displacement along the x-axis was 10.7 ± 1.7 mm and inferior displacement was 2.3 ± 2.8 mm from T1 to

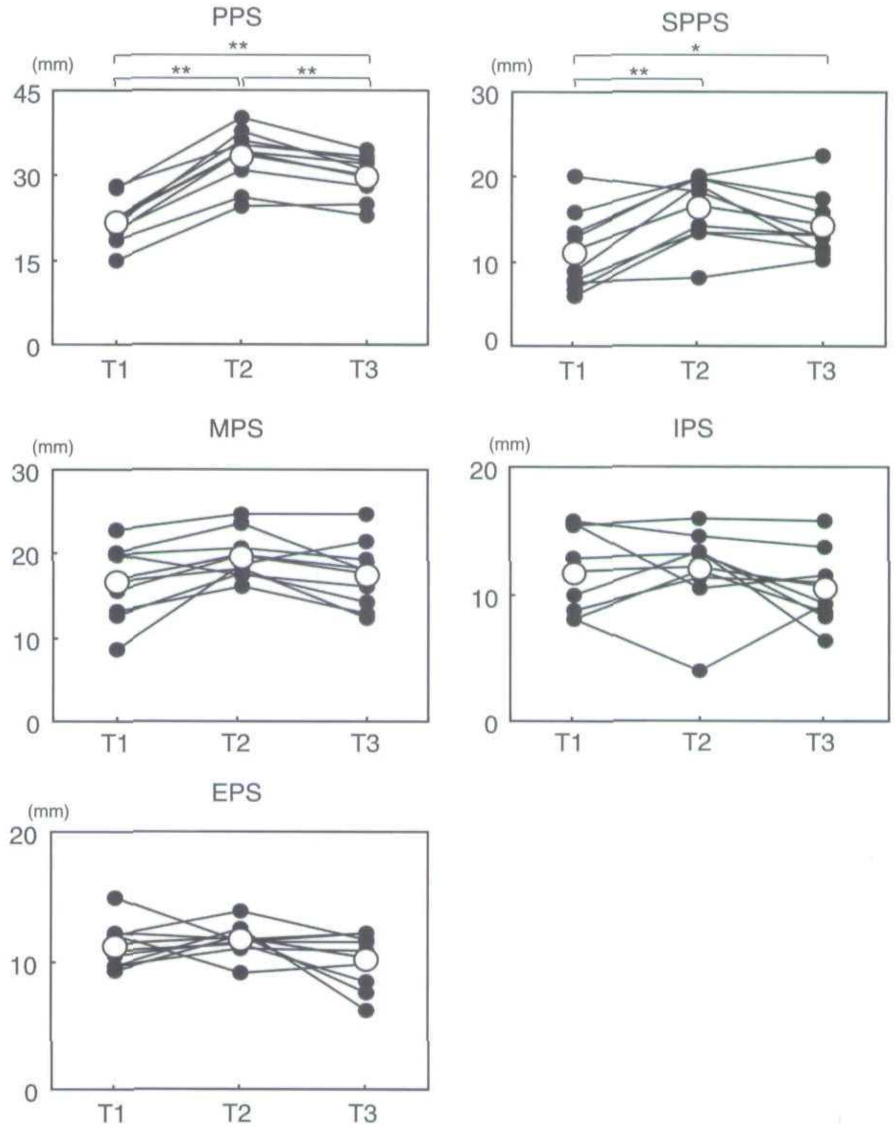

Fig. 2. Longitudinal changes in the anteroposterior distance in the upper airway. Individual (filled circles) and mean (open circles) values are plotted. T1, immediately before DOG; T2, immediately after DOG; T3, 1 year after DOG. $^*p < 0.05$; $^{**}p < 0.01$.

T2. Subsequently, the PNS moved -2.4 ± 0.6 mm, i.e. -2.3 ± 1.7 mm posteriorly and 0.6 ± 2.0 mm inferiorly, from T2 to T3. Small p-values that tended to suggest some relationship were seen between the summed ($p = 0.06$) and orthogonal (x-axis: $p = 0.07$, y-axis: $p = 0.07$) displacement of the PNS and the change in nasal resistance from T1 to T2 (Fig. 6). On the other hand, there were no significant correlations between the summed and orthogonal displacement of the PNS and the change in nasal resistance from T2 to T3.

Discussion

The present findings suggest that the anteroposterior dimension of the superior part of the upper airway increased and nasal resistance decreased in association with maxillary DOG. To the best of our knowledge, only a few studies have compared the structure of the craniofacial skeleton, distracted length and articulation in association with DOG (4,5,13). It has been shown that subjects with upper-airway obstruction gained airway sufficiency after DOG in the mandible (14–16). Nevertheless, there is little information available on how maxillary DOG alters the upper-airway structure. Warren and associates (17) reported that nasal resistance in subjects with CLP is 20–30% higher than that in age-matched non-CLP subjects. Hairfield and Warren (18) reported that there are dimensional and physiologic differences in the nasal airway between surgically-repaired subjects with CLP and individuals without CLP. In our study, we found that the decrease in nasal

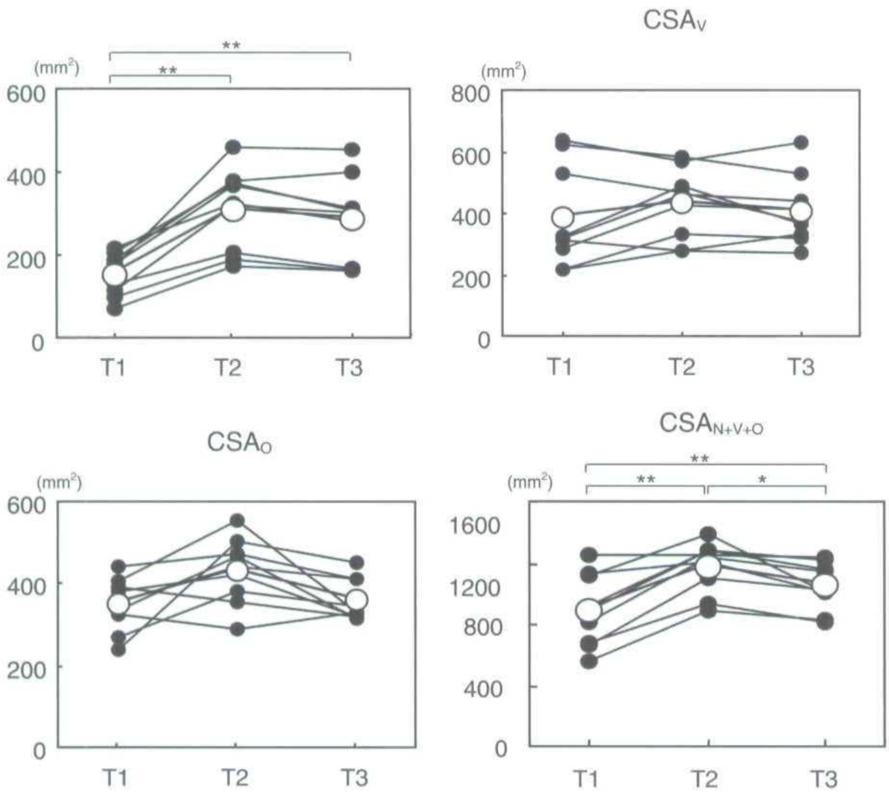

Fig. 3. Longitudinal changes in the sagittal cross-sectional area of the upper airway. Individual (filled circles) and mean (open circles) values are plotted. T1, immediately before DOG; T2, immediately after DOG; T3, 1 year after DOG. $^{*}p < 0.05$; $^{**}p < 0.01$.

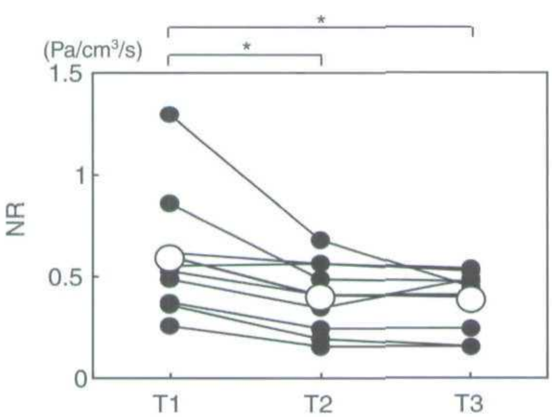

Fig. 4. Longitudinal changes in nasal resistance. NR, nasal resistance; T1, immediately before DOG; T2, immediately after DOG; T3, 1 year after DOG; Pa, pascal. $^{*}p < 0.05$.

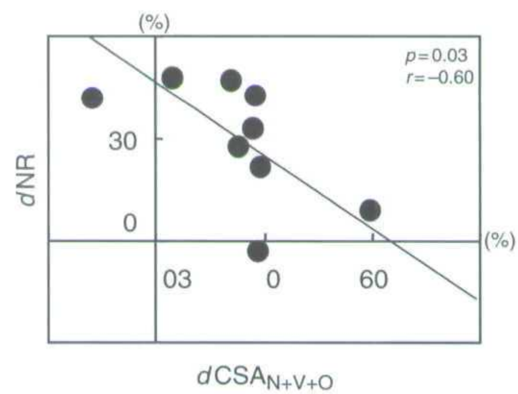

Fig. 5. Relationship between changes in the sagittal cross-sectional area of the upper airway and nasal resistance. $dCSA_{N+V+O}$; the percentage change in the total cross-sectional area of the upper airway (i.e. CSA_N, CSA_V and CSA_O) between immediately after and before DOG $[(T2-T1) \times 100/T1]$. dNR, the percentage change in nasal resistance between immediately before and after DOG $[(T1-T2) \times 100/T1]$.

resistance in subjects with CLP who underwent maxillary DOG significantly correlated with the increase in the upper-airway CSA. This suggests that there is a critical relationship between the gradual change in upper-airway structure and function, as was noted in the acute change that followed orthognathic repositioning of the maxilla (19,20). We found that the increase in the upper-airway dimension and the reduction in nasal resistance were still significant at 1 year after DOG. This is the first study to rationalize the application of DOG with regard to morphological and functional stability of the upper airway.

Muscular and soft tissue traction force may play a role in maxillary relapse. Thus, it is difficult for the musculature and soft tissue to adapt immediately to maxillary displacement by orthognathic surgery. With a gradual bone distraction procedure, the surrounding

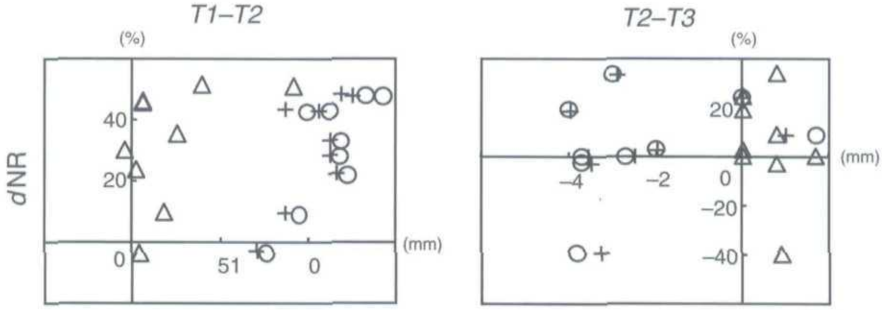

Fig. 6. Relationship between displacement of the PNS and changes in nasal resistance during DOG (T1–T2) and during the consolidation period after DOG (T2–T3). The total two-dimensional displacement of the PNS (circles) was subdivided into horizontal (crosses) and vertical (triangles) components. T1, immediately before DOG; T2, immediately after DOG; T3, 1 year after DOG. dNR, the percentage change in nasal resistance between immediately before and after DOG [left, (T1–T2) × 100/T1] and between immediately after and 1 year after DOG [right, (T2–T3) × 100/T2]. The *p*-values for total displacement of the PNS (circles), and horizontal (crosses) and vertical (triangles) components for T1–T2 were 0.06, 0.07 and 0.07, respectively. Likewise, those for total displacement of the PNS (circles) and horizontal (crosses) and vertical (triangles) components for T2–T3 were 0.3, 0.3 and 0.2, respectively.

soft tissues may have a better chance of accommodating the structural change than with an acute change elicited by other conventional surgical interventions such as Le Fort I osteotomy. It is known that there is a greater risk of hypernasal speech after maxillary advancement surgery (21,22). In our study, DOG increased the anteroposterior upper-airway dimension in association with forward movement of the maxillary bone. This procedure induces widening of the velopharyngeal orifice, which may sometimes cause incomplete velopharyngeal closure, and thereby deteriorate speech function (14). Fortunately, none of our subjects with CLP newly developed hypernasality, although two subjects (nos 4 and 8) who had shown hypernasality before the study reported an aggravation of speech function after DOG. On the other hand, four subjects (nos 2, 5, 7 and 8) reported that they could more easily breathe through the nose after DOG. Therefore, the interaction between the amelioration of respiratory function and the deterioration of articulatory function by DOG should be critically evaluated in a future study in a comprehensive manner.

It is generally accepted that subjects who undergo DOG adapt to maxillary advancement by approximately 1 mm a day. During the course of DOG, the anterior velopharyngeal wall (i.e. soft palate) moves anteriorly, while the posterior wall of the velopharynx is stable. This mutual relationship elicits an increase in the velopharyngeal depth (Fig. 2). In this study, all (100%) of the subjects with CLP showed an increase in the PPS, 7 (78%) showed an increase in the SPPS, and 8 (89%) showed an increase in the MPS. The PNS, which is located on the bony structure, was moved forward by DOG. However, the PNS moved backward after DOG with relapse of the maxillary bone. We believe that this is reflected in the significant increase in the PPS by DOG, and is also reflected in the significant decrease in the PPS during the consolidation period after DOG. Indeed, the change in the position of the PNS and nasal resistance during DOG were closely linked (Fig. 6). On the other hand, both the SPPS and MPS are defined as the length of lines constructed by two landmarks on the soft tissue, which are far from the PNS. Therefore, it is plausible that both the SPPS and MPS did not necessarily follow the movement of the bony structure with DOG. It appears that there is no significant relationship between the change in the position of the PNS and nasal resistance at 1 year after DOG. In contrast to the short time-frame between T1 and T2, many biological events (e.g. residual growth, adaptation of soft tissues and occlusal changes) must have occurred between T2 and T3. Presumably, this complexity obscured the relationship between changes in the position of the PNS and nasal resistance during T2 and T3 (Fig. 6). It is plausible that there is a relationship between the CSA and volumetric change in the upper airway. According to a previous study (23), it has been demonstrated that the CSA_N is significantly correlated with volume. Thus, the significant changes in the CSA_N appear to be accompanied by a volumetric change in the nasopharynx. With regard to the significant correlation between changes in the CSA_{N+V+O} and nasal resistance, Fig. 5 shows that nasal resistance decreases with an increase in CSA_{N+V+O}. Since there was a significant ($p = 0.028$, $r^2 = 0.61$) negative correlation between CSA_{N+V+O} and nasal resistance before DOG, maxillary advancement by DOG at an identical rate must have different effects on individuals with different CSA_{N+V+O} and nasal resistance before DOG. To be precise, maxillary DOG at an identical rate should increase CSA_{N+V+O} more in subjects with smaller CSA_{N+V+O} and higher nasal resistance than in those with larger CSA_{N+V+O} and lower nasal resistance. Therefore, as shown in Fig. 6, the rate of the decrease in nasal resistance in subjects who showed a greater increase CSA_{N+V+O} was less than that in those who showed less of an increase in the CSA_{N+V+O}. This assumption may be verified in a future study using a larger sample size.

There are several limitations in this study. First, two-dimensional analysis was used to evaluate the three-dimensional structures. Since we used computed cephalometric radiography, both accuracy and sensitivity of cephalometric measurements have significantly improved compared with those of the conventional cephalometric radiography. However, the phase of respiration has not been standardized when the cephalometric film was taken. It has been shown that there are significant changes in both position and shape of the oropharyngeal soft and hard tissues in association with the respiratory phase (24), one must be careful when interpreting the upper-airway measurement. Second, the sample size was small and it was a mixed sample of uni- and bilateral CLP, with a variation in age. It has been reported in Caucasian (25) and Japanese (26) children that nasal resistance decreases with age and levels off during young adulthood. In this study, we could not segregate the effect of age on upper-airway changes in teenagers after 1 year. It is assumed that the anteroposterior distance of the upper airway increases with age in children with unilateral CLP as well as in the age-matched controls (27). However, it is unknown whether this also happens to those with bilateral CLP. Therefore, further controlled prospective study is needed with homogenous subjects.

Conclusions

The total CSA of the upper airway, i.e. the nasopharynx, the velopharynx and the oropharynx, significantly increased after maxillary DOG in subjects with CLP. There was a significant correlation between the changes in the total CSA of the upper airway and nasal resistance between immediately before and after DOG. This increase in the upper-airway size and the reduction in nasal resistance were stable after 1 year. These results suggest that DOG may have the potential to improve respiratory function in association with morphological changes in the upper airway of subjects with CLP.

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