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Short-term changes in temporomandibular joint function in subjects with cleft lip and palate treated with maxillary distraction osteogenesis

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Structured Abstract

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Objectives – To investigate the short-term effects of maxillary distraction osteogenesis (DO) on temporomandibular joint (TMJ) function in 21 subjects with cleft lip and palate (CLP).

Design – Morphological changes in the maxillofacial region were measured using lateral cephalometric radiographs taken immediately before (pre-DO) and after DO (post-DO) and 1 year after DO (1-year follow-up). A questionnaire was evaluated using a visual analog scale. A chi-square test was used to compare the prevalence of TMJ symptoms between pre-DO and 1-year follow-up. The Spearman correlation coefficient was used to determine the correlation between changes in cephalometric variables and TMJ symptoms in association with maxillary DO. Statistical significance was set at $p < 0.05$.

Results – The ANB (anteroposterior relationship of the maxilla with the mandible) angle and the mandibular plane angle at pre-DO, post-DO, and 1-year follow-up were −4.3°, +5.8°, +4.3° and 32.1°, 33.5°, 33.6°, respectively. The average amounts of anterior and downward movement of the maxilla at post-DO and 1-year follow-up were 8.3, −1.3 and 0.9, 1.1 mm, respectively. The prevalence of TMJ symptoms showed no significant increase in association with maxillary DO. Moreover, there was no significant correlation between changes in cephalometric variables and TMJ symptoms.

Conclusion – These results suggest that there was no short-term (i.e., up to 1 year after DO) effect of maxillary DO on TMJ function in subjects with CLP.

Key words: cleft lip and palate; osteogenesis distraction; maxilla; temporomandibular joint disorders

Introduction

Maxillary distraction osteogenesis (DO), the gradual formation of new bone by progressive lengthening (1, 2) has been widely applied in the orofacial region. Recently, this technique has been used to lengthen the hypoplastic mid-face in subjects with cleft lip and palate (CLP). Orthodontic treatment planning and orthognathic surgery are more complex in subjects with CLP than in non-CLP subjects, as a large maxillary advancement is generally required. Hence, conventional orthognathic surgery, such as Le Fort I osteotomy is more difficult to perform in subjects with CLP than in non-CLP subjects, and this often contributes to skeletal relapse. Consequently, the choice between orthognathic surgery and DO depends largely on the predicted amount of advancement of the maxilla (3). DO is associated with changes in the maxillofacial morphology, including the maxilla and mandible, and gives skeletal stability after maxillary advancement in subjects with CLP, as supported by cephalometric analysis (4).

Subjects with CLP show higher frequencies of dysfunction in temporomandibular joint (TMJ) than those without CLP (5). On the other hand, it has been reported that subjects with CLP exhibited a significantly reduced jaw-opening pattern compared to controls; however, the overall jaw function was not significantly different (6). Maxillary advancement by Le Fort I osteotomy does not influence mandibular mobility permanently and the impact on the TMJ is minimum (7, 8). However, no studies have investigated the relationship between changes in the maxillofacial morphology and TMJ function in subjects with CLP in association with maxillary DO. As maxillary DO induces a larger forward maxillary displacement than other types of orthognathic surgery, such as Le Fort I, its impact on TMJ function may differ. The purpose of this study was to investigate the short-term effects of maxillary DO on TMJ function in subjects with CLP.

Materials and methods

The present study was carried out in 21 subjects (14 males and 7 females, mean age: 16.5 years) with non-syndromic CLP. Eighteen subjects had a unilateral CLP and three had a bilateral CLP. All subjects were treated with a full edgewise appliance at Tokyo Medical and Dental University Dental Hospital. Prior to the study, all subjects gave their informed consent after receiving a full explanation of the aim and design of this study. The age immediately before DO (pre-DO) was 16.5 ± 3.6 (mean ± SD) years. A rigid external distraction (RED) device (4, 9, 10) for maxillary DO was fitted on the same day of the bone surgery. Maxillofacial changes were evaluated using computed cephalometric radiography (FUJIX FCR7000; Fuji Film, Tokyo, Japan) registered at the intercuspal position with the head parallel to the Frankfort horizontal (FH) plane. A set of lateral cephalometric radiographs was taken using a cephalostat (HD-150B-30; Shimadzu, Kyoto, Japan) in each subject at pre-DO and after DO (post-DO; 16.9 ± 3.6 years) and 1 year after DO (1-year follow-up; 17.9 ± 3.6 years) (Table 1). In addition, subjects who were treated by additional types of orthognathic surgery, such as sagittal split ramus osteotomy from pre-DO to 1-year follow-up were excluded from this study.

Table 1. **Characteristics and demographic variables of 21 subjects with cleft lip and palate**

No.	Gender	Type of cleft	Age at pre-DO (years)	Age at post-DO (years)	Age at 1-year follow-up (years)
1	M	LCLP	15.6	15.8	17.3
2	M	BCLP	12.6	12.8	13.8
3	F	LCLP	19.3	19.6	20.4
4	M	RCLP	11.8	12.0	13.2
5	M	RCLP	15.8	16.3	16.8
6	M	RCLP	20.3	20.8	21.5
7	F	LCLP	21.0	21.2	22.2
8	F	RCLP	14.0	14.2	15.1
9	F	LCLP	18.0	19.1	19.9
10	M	LCLP	15.1	15.2	18.2
11	M	LCLP	23.3	23.8	24.9
12	M	BCLP	17.5	18.1	19.0
13	M	RCLP	21.0	21.1	21.6
14	M	LCLP	12.7	13.0	14.0
15	M	LCLP	18.8	19.0	20.0
16	F	BCLP	17.0	17.1	18.9
17	M	LCLP	17.5	17.6	18.5
18	M	LCLP	20.9	21.0	21.9
19	M	LCLP	13.1	13.5	14.4
20	F	LCLP	11.0	11.8	12.2
21	F	LCLP	11.2	11.3	12.2

Abbreviations: F, female; M, male; LCLP, left unilateral cleft lip and palate; RCLP, right unilateral cleft lip and palate; BCLP, bilateral cleft lip and palate; pre-DO, immediately before distraction osteogenesis; post-DO, immediately after distraction osteogenesis.

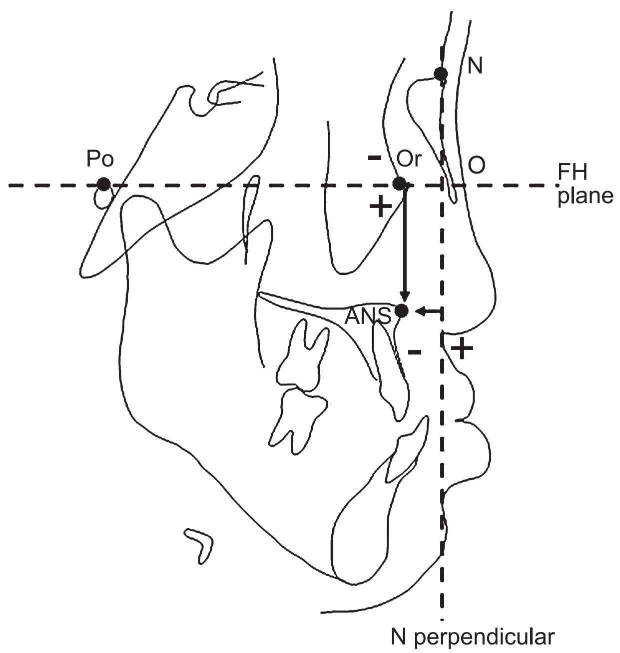

Fig. 1. Anatomic points and lines used to identify cephalometric variables. These included the SNA angle to determine the anteroposterior position of the maxilla relative to the anterior cranial base, the SNB angle to determine the anteroposterior position of the mandible relative to the anterior cranial base, the ANB angle to determine the relationship between the maxilla and the mandible and the mandibular plane (MP) angle to determine the in the vertical position of the mandible relative to the anterior cranial base for angular analysis. A vertical line that passes through the Nasion was drawn to the Frankfurt horizontal (FH) plane (Nasion perpendicular). The horizontal and vertical positions of the maxilla were measured by the horizontal and vertical positions of the anterior nasal supine (ANS). The horizontal ANS and vertical ANS positions are measured from the N perpendicular line (+: behind the line, −: ahead of the line) and the FH plane (+: below the line, −: above of the line), respectively. Abbreviations: Po, Porion; Or, Orbitale; N, Nasion; ANS, anterior nasal spine; O, intersection made by the FH plane and N perpendicular.

The landmarks and contours that are commonly used in cephalometric analysis and additional variables for the maxillary region were defined (Fig. 1). Friedman's chi-square *r*-test and the Student–Newman–Keuls test were used to determine the statistical significance ($p < 0.05$) of differences in cephalometric variables among pre-DO, post-DO and 1-year follow-up. The same investigator traced all cephalometric radiographs, and the method error (11, 12) for each parameter was calculated by comparing duplicate tracings at an interval of at least 2 weeks. Errors for a single measurement of linear and angular variables were calculated with the formula, method error = $(\sum d^2/2n)^{1/2}$, where d is the difference between measured pairs and n is the number of pairs.

Changes in TMJ function/dysfunction were investigated using a questionnaire regarding symptoms, such as noise, pain and trismus taken at pre-DO and at 1-year follow-up. The degree of TMJ symptoms was evaluated using the visual analog scale (VAS), which was formed from a standard 10-cm VAS with anchors at '0' and '100' (6, 13). The subject was asked to grade symptoms on an arbitrary 'point' scale, with a minimum of 0 and a maximum of 100. The prevalence of TMJ symptoms was calculated by dividing the number of subjects with symptoms by the total number of subjects. A chi-square test ($p < 0.05$) was used to compare the prevalence of TMJ symptoms between pre-DO and 1-year follow-up. The Spearman correlation coefficient ($p < 0.05$) was used to determine the correlation between changes in cephalometric variables and VAS scores that indicated the degree of TMJ symptoms associated with maxillary DO.

Results

Means and SDs for all cephalometric variables are shown in Table 2. Although the SNA (anteroposterior position of the maxilla relative to the anterior cranial base) significantly increased from pre-DO to post-DO, it significantly decreased by various degrees from post-DO to 1-year follow-up. On the other hand, the SNB (anteroposterior position of the mandible relative to the anterior cranial base) showed no significant change from pre-DO to post-DO, or from post-DO to 1-year follow-up. The ANB significantly increased from pre-DO to post-DO (10.1 ± 4.4°), however, it significantly decreased from post-DO to 1-year follow-up (−1.4 ± 2.5 degrees). The mean mandibular plane (MP) angle showed no significant change from pre-DO to post-DO, but inter-individual differences were noted: an increase in the MP angle (clockwise rotation; CR) was observed in 12 subjects (57%) and a decrease in the MP angle (counter-clockwise rotation; CCR) was observed in five subjects (24%). The mean MP angle remained stable from post-DO to 1-year follow-up, but again inter-individual differences were noted: the CR was observed in 10 subjects (47%) and the CCR was observed in nine (43%). The anterior nasal spine (ANS) moved significantly forward (8.3 ±2.5 mm) in association with maxillary DO, whereas it moved significantly backward (−1.3 ± 1.9 mm) suggesting a certain amount of relapse from post-DO to 1-year follow-up. The method errors for SNA, SNB, ANB, MP angle, horizontal ANS (hANS)

***Table 2.* Changes in cephalometric variables in association with maxillary distraction osteogenesis**

Variables	Timings	Mean	SD
SNA (°)[†]	pre-DO	74.1	4.6
	post-DO	83.2	4.1
	1-year follow-up	82.0	4.3
SNB (°)*	pre-DO	78.4	3.9
	post-DO	77.5	3.1
	1-year follow-up	77.7	3.8
ANB (°)[†]	pre-DO	−4.3	4.6
	post-DO	5.8	2.9
	1-year follow-up	4.3	3.6
MP (°)*	pre-DO	32.1	5.6
	post-DO	33.5	5.5
	1-year follow-up	33.6	5.7
hANS (mm)[†]	pre-DO	−3.4	3.0
	post-DO	4.9	3.3
	1-year follow-up	3.6	2.6
vANS (mm)*	pre-DO	27.1	3.8
	post-DO	28.0	4.5
	1-year follow-up	29.1	4.6

SNA, anteroposterior position of the maxilla relative to the anterior cranial base; SNB, anteroposterior position of the mandible relative to the anterior cranial base; ANB, anteroposterior relationship of the maxilla with the mandible; MP, mandibular plane; hANS, horizontal position of the anterior nasal spine; vANS vertical position of the anterior nasal spine; pre-DO, immediately before distraction osteogenesis (DO); post-DO, immediately after distraction osteogenesis.
*Not significant between any pair of pre-DO, post-DO and 1-year follow-up.
[†]Significant between any pair of pre-DO, post-DO and 1-year follow-up.

and vertical ANS (vANS) were 0.64°, 0.49°, 0.66°, 0.30°, 0.79 mm and 0.54 mm, respectively.

The prevalence of TMJ symptoms showed no significant increase in association with maxillary DO (Table 3). Figure 2 and Table 4 show subjects who revealed marked morphological changes and/or TMJ symptoms.

Figure 3 shows changes in isolated TMJ noise in association with maxillary DO. Of 11 TMJs that showed noise at pre-DO, seven showed little or no change, two deteriorated, and two disappeared from pre-DO to 1-year follow-up (Fig. 3A, B). Moreover, noise was observed at the 1-year follow-up in a TMJ that showed no noise at pre-DO. There were no significant correlations between changes in the MP angle and the VAS score regarding TMJ noise (Fig. 3C). Furthermore, there were no significant correlations between changes in other cephalometric variables and TMJ noise.

***Table 3.* Changes in prevalence of temporomandibular joint symptoms in association with maxillary distraction osteogenesis**

Variables	Timings	Prevalence	Probabilities
Noise	pre-DO	43	
	1-year follow-up	43	NS
Pain	pre-DO	10	
	1-year follow-up	10	NS
Trismus	pre-DO	0	
	1-year follow-up	10	NS

Prevalence was depicted by percentages.
Pre-DO, immediately before distraction osteogenesis; NS, not significant.

Figure 4 shows changes in isolated TMJ pain in association with maxillary DO. Of three TMJs that showed pain at pre-DO, one showed no change, one ameliorated, and one deteriorated from pre-DO to the 1-year follow-up (Fig. 4A, B). There were no significant correlations between changes in the MP angle and the VAS score regarding TMJ pain (Fig. 4C). Furthermore, there were no significant correlations between changes in other cephalometric variables and TMJ pain.

Figure 5 shows the change in trismus in association with maxillary DO. In two subjects, trismus appeared at the 1-year follow-up (Fig. 5A). There were no significant correlations between changes in the MP angle and the VAS score regarding trismus (Fig. 5B). Furthermore, there were no significant correlations between changes in other cephalometric variables and trismus.

Discussion

The RED device (9, 10) is highly effective for correcting maxillary hypoplasia in subjects with CLP. The long-term morphological changes after the correction of sagittal maxillary deformities using the RED system in both adult and paediatric subjects have been stable (4). In this study, although the maxilla was moved significantly forward with DO, it moved significantly backwards within 1 year after DO. A previous study argued that a decrease in SNA after maxillary DO was not related to skeletal relapse but rather growth, as there was a significant downward movement of ANS after maxillary DO (4). Our sample consisted of juvenile and adult subjects, and their amounts of residual growth might vary. Although changes in SNA and hANS appear to indicate skeletal relapse, a further study with

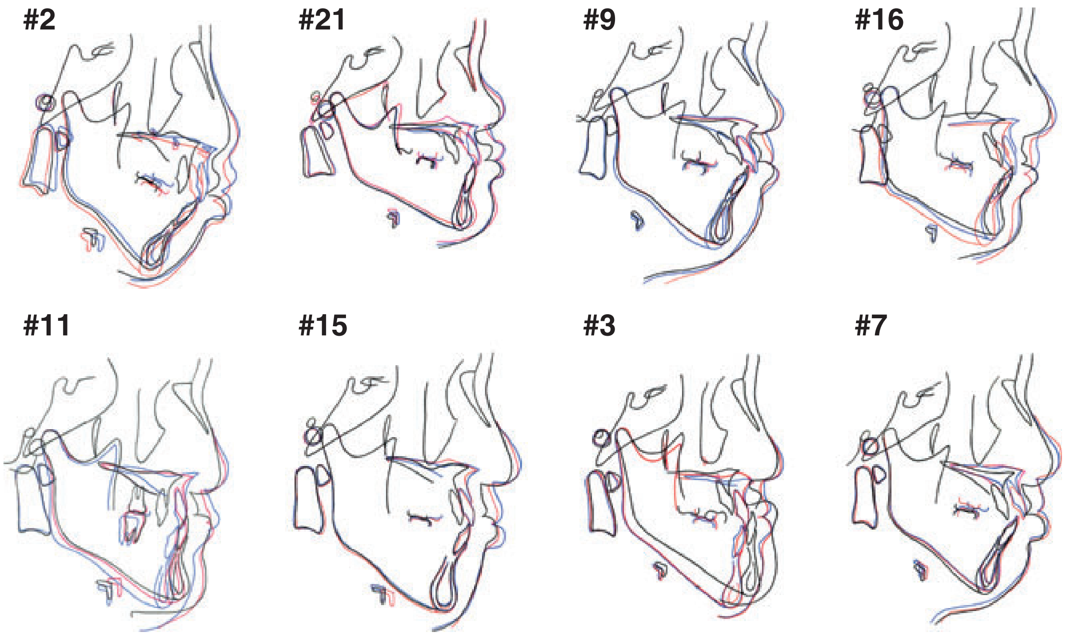

Fig. 2. Cephalometric tracings of morphological changes in typical subjects. Black, blue and red lines show tracings at pre-DO, post-DO and 1-year follow-up, respectively. The subjects' numbers are identical to those in Tables 1 and 4. pre-DO, immediately before distraction osteogenesis; post-DO, immediately after distraction osteogenesis

Table 4. **Changes in cephalometric variables and temporomandibular joint symptoms in Fig. 2**

					Δ VAS score					
					Noise		Pain		Trismus	
No.	ΔMP (°)	ΔANB (°)	ΔhANS (mm)	ΔvANS (mm)	R	L	R	L	R	L
2	1.0	8.0	6.5	7.0	0	10	0	0	0	0
21	−1.0	8.5	7.5	0	−24	−24	0	0	0	0
9	0.5	11.5	10.5	1.5	0	0	−60	−60	0	0
16	2.0	10.0	4.3	2.0	−5	−5	0	0	0	0
11	2.0	10.0	4.0	−1.0	0	0	0	0	0	15
15	0.5	8.0	4.5	2.5	29	0	0	0	0	1
3	6.0	12.0	7.5	6.5	0	0	0	0	0	0
7	−2.0	8.0	7.5	0.5	0	0	0	0	0	0

ANB, anteroposterior relationship of the maxilla with the mandible; R, right; L left; MP, mandibular plane; VAS, visual analog scale; hANS, horizontal anterior nasal spine; vANS, vertical anterior nasal spine; pre-DO, immediately before distraction osteogenesis (DO); °, angles in degrees.
The difference (Δ) in cephalometric variables and the VAS score was calculated by subtracting values at pre-DO from those at 1-year follow-up. The subjects' numbers are identical to those in Table 1 and Figure 2.

homogeneous samples is necessary to determine the cause of changes in cephalometric variables in association with maxillary DO.

With regard to the prevalence of TMJ noises, Agerberg and Carlsson (14) found that the frequency of TMJ noises was 39% in a randomly selected population. Solberg et al. (15) also reported a 28.3% prevalence of TMJ noises in young adults. Sadowsky et al. (16) showed that 20.6% and 25% of patients were aware of TMJ noises before and after orthodontic treatment, respectively. They found that there were no significant differences between patients treated orthodontically and untreated controls (16). Other studies showed similar percentages in subjects who underwent orthodontic treatment, but there were no significant differences between before and after treatment (17–22). Our

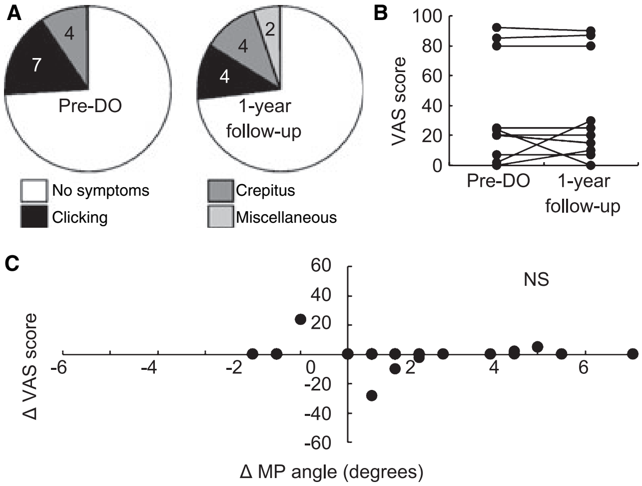

Fig. 3. Changes in isolated temporomandibular joint (TMJ) noise in association with maxillary distraction osteogenesis. A, Disaggregated data of TMJ noise. Number of symptomatic TMJ is shown in the inset. B, Changes in the visual analog scale (VAS) score. C, Scatter plot of the relationship between changes in the mandibular plane (MP) angle (*x*-axis) and the VAS score regarding TMJ noise (*y*-axis). The difference (Δ) in the MP angle and the VAS score was calculated by subtracting values at pre-DO from those at 1-year follow-up. NS, not significant; pre-DO, immediately before distraction osteogenesis.

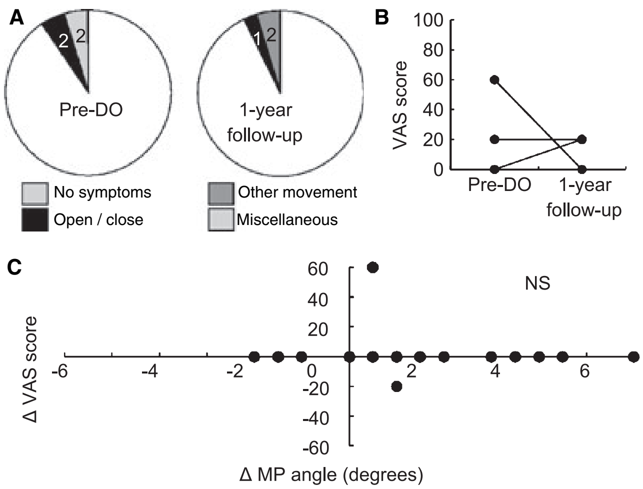

Fig. 4. Changes in isolated temporomandibular joint (TMJ) pain in association with maxillary distraction osteogenesis. A, Disaggregated data of TMJ pain. Number of symptomatic TMJ is shown in the inset. B, Changes in the visual analog scale (VAS) score. C, Scatter plot of the relationship between changes in the mandibular plane (MP) angle (*x*-axis) and the VAS score regarding TMJ pain (*y*-axis). The differences (Δ) in the MP angle and the VAS score were calculated by subtracting values at pre-DO from those at 1-year follow-up. NS, not significant; pre-DO, immediately before distraction osteogenesis.

samples showed a higher percentage, but there were no significant differences between before and after DO. TMJ noises are classified as clicking or crepitus. Clicking is assumed to be related to disc displacement, while crepitus is because of a degenerative change in the TMJ (23, 24). In adolescents, the prevalence of self-reported

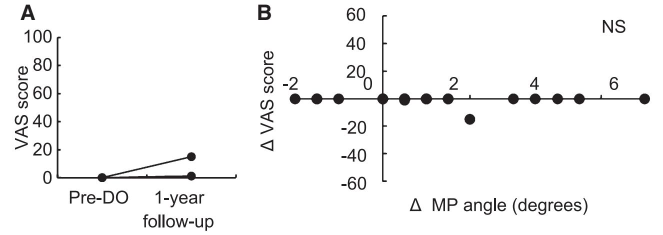

Fig. 5. Changes in trismus in association with maxillary distraction osteogenesis. A, Changes in the visual analog scale (VAS) score. B, Scatter plot of the relationship between changes in the mandibular plane (MP) angle (*x*-axis) and the VAS score regarding temporomandibular joint trismus (*y*-axis). The differences (Δ) in the MP angle and the VAS score were calculated by subtracting values at pre-DO from those at 1-year follow-up. NS, not significant; pre-DO, immediately before distraction osteogenesis.

clicking at ages 14, 15, 18, and 23 was 11, 12, 25 and 31%, respectively (25). Another study reported that the prevalence of clicking and crepitus was 32% and 8% in subjects without CLP, and 17% and 14% in subjects with CLP, respectively (6). The frequencies of clicking (6/21, 29%) and crepitus (3/21, 14%) in our study were comparable to those in the previous study (6).

Epidemiological studies have reported that the prevalence of pain in the masticatory system is approximately 10% in healthy subjects (26, 27). On the other hand, in subjects with CLP, the prevalence of pain in the face is 14%, which does not significantly differ from that in controls (6). Our study showed that the prevalence of TMJ pain was 10% at both pre-DO and 1-year follow-up. This is supported by previous studies that have shown no significant differences in the prevalence of TMJ pain between before and after orthodontic treatment (19, 21, 28).

The prevalence of TMJ lock increases from 12 to 15 years of age, especially in girls, and continues to increase up to 19 years (29). In this study, two subjects, who showed trismus after DO were 23.8 and 19.0 years old, and both were males. Conversely, the distraction devices may have been uncomfortable or may have made it difficult for the subject to talk or eat, or led to social embarrassment. These stresses might induce trismus.

With regard to the cephalometric variables, the change in the MP angle is related to mandibular movement in association with maxillary DO. Previous studies have demonstrated that there were significant correlations between the mandibular position and TMJ symptoms (30, 31). We investigated the possible relationship between the change in TMJ symptoms and the change in the MP angle was based on the assumption that maxillary DO may result in a change in the mandibular position. However, there were no significant correlations between changes in TMJ symptoms and the change in the MP angle in this study. It has been reported that conventional Le Fort I osteotomy for maxillary advancement does not cause any significant change in the TMJ (32, 33). Maxillary DO also does not have an unfavourable effect on the TMJ.

This study has several limitations. First, the ANS was difficult to determine at post-DO. Although the average method error was equivalent to that in previous studies (11, 12), the method error for hANS at post-DO was 1.17 mm, which was significantly greater than those at other stages. Second, only a subjective, and not an objective, method for evaluating TMJ symptoms was used. Lastly, this study evaluated short-term effects of DO. Despite these limitations, there was no previous study on the relationship between TMJ symptoms and skeletal changes induced by maxillary DO. As DO is a developing alternative treatment for surgical correction of CLP, further careful studies regarding its effects and side effects are needed.

Conclusions

The prevalence of TMJ symptoms showed no significant increase in association with maxillary DO. Moreover, there were no significant correlations between changes in cephalometric variables and TMJ symptoms. These results suggest that there was no short-term effect of maxillary DO on TMJ function in subjects with CLP.

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