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Original Article

Anterior maxillary segmental distraction in the treatment of severe maxillary hypoplasia secondary to cleft lip and palate

Hongliang Li*, Jiewen Dai*, Jiawen Si, Jianfei Zhang, Minjiao Wang, Steve Guofang Shen, Hongbo Yu

Department of Oral & Cranio-maxillofacial Surgery, Shanghai Ninth People's Hospital, Shanghai Jiao Tong University School of Medicine, Shanghai Key Laboratory of Stomatology, Shanghai, China. *Equal contributors.*

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Abstract: Anterior maxillary segmental distraction (AMSD) is an effective surgical procedure in the treatment of maxillary hypoplasia secondary to cleft lip and palate. Its unique advantage of preserving velopharyngeal function makes this procedure widely applied. In this study, the application of AMSD was described and its long-term stability was explored. Eight patients with severe maxillary hypoplasia secondary to CLP were included in this study. They were treated with AMSD using rigid external distraction (RED) device. Cephalometric analysis was performed twice at three time points for evaluation: before surgery (T1), after distraction (T2), and 2 years after treatment (T3). One-way analysis of variance was used to assess the differences statistically. All the distractions completed smoothly, and maxilla was distracted efficiently. The value of SNA, NA-FH, Ptm-A, U1-PP, overjet and PP (ANS-PNS) increased significantly after the AMSD procedure ($P < 0.05$), with the mean overjet increased by 14.28 mm. However, comparison of cephalometric analysis between T2 and T3 showed no significant difference ($P > 0.05$). Changes of palatopharyngeal depth and soft palatal length were insignificant. AMSD with RED device provided an effective way to correct maxillary hypoplasia secondary to CLP, extended the palatal and arch length, avoided damage on velopharyngeal closure function and reduced the relapse rate. It is a promising and valuable technique in this potentially complicated procedure.

Keywords: AMSD, maxillary hypoplasia, CLP, VPI, stability

Introduction

Maxillary hypoplasia is a common deformity secondary to cleft lip and palate (CLP). Correction of this deformity presents a great challenge for oral and maxillofacial surgeons. Although surgical techniques and methods have been greatly improved in the last decades [1, 2], maxillary advancement in excess of 6 mm is difficult to achieve due to maxillary scars, meanwhile, postoperative stability is discounted. Furthermore, even if the mandible is in the normal position, it is often essential to setback the mandible with the advancement of maxilla to correct 6 mm or greater anterior cross-bite and achieve a functional dental occlusion [3, 4].

Maxillary distraction osteotomy (DO) was proposed in 1997 using a rigid external distraction (RED) device [5]. It was beneficial in the treatment of CLP patients because of more stability and fewer limitations of the amount and direction of advancement [6].

While in some cases, velopharyngeal insufficiency (VPI) could be deteriorated due to the increase of nasopharyngeal distance. Since the extensive application of maxillary DO with RED, numerous studies [7-9] have reported that the risk of velopharyngeal insufficiency following maxillary distraction was similar to that observed in Le Fort I maxillary advancement.

Anterior maxillary segmental distraction (AMSD), first reported by Karakasis, marked the premier application for the correction of maxillary hypoplasia secondary to CLP. Compared with the traditional maxillary distraction osteogenesis [6], AMSD showed less negative effects on velopharyngeal closure [10-13]. To our best knowledge, no long-term follow-up of maxillary

Table 1. Case characteristics

Pt. NO	Gender	Age (yr)	Osteotomy position	CLP Classification	Reverse Overjet (mm)	Distraction period (d)	Follow-up (mo)
1	M	23	45,56	B	6	10	26
2	M	26	56,56	L	15	24	18
3	M	24	46,46	L	5	11	18
4	M	21	67,67	L	12	20	20
5	M	31	56,56	L	7	14	24
6	M	23	56,56	R	10	15	30
7	M	20	56,56	B	15	24	19
8	M	19	67,67	L	12	20	28
Mean ± STD		23.38 ± 3.81			10.25 ± 3.92	17.25 ± 5.52	22.88 ± 4.76

Abbreviations: Pt. NO, patient number; CLP, cleft lip and palate; M, male; B, bilateral; L, left; R, right; d, day; mo, month. Osteotomy position: 45 means osteotomy line is between the first and second premolar; 56 means osteotomy line is between the second premolar and first molar; 67 means osteotomy line is between the first molar and second molar.

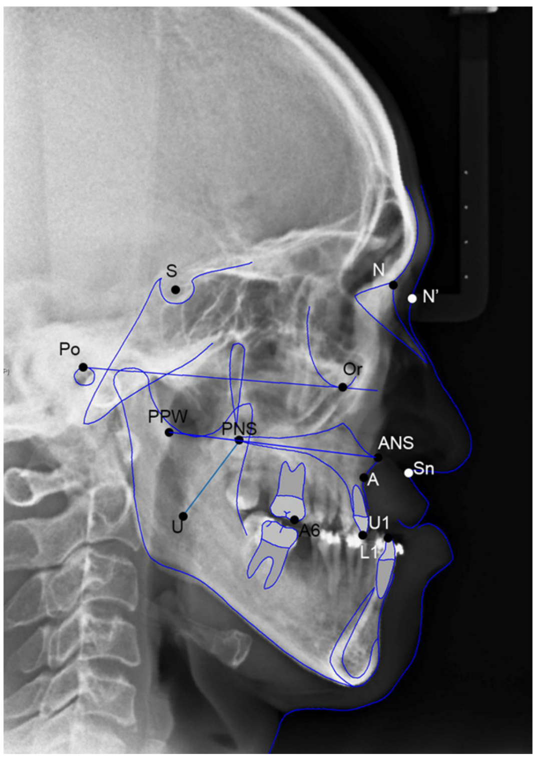

Figure 1. Landmarks for cephalometric analysis.

advancement with AMSD was reported before. The purpose of this study was to evaluate the post-surgical stability of AMSD in the treatment of severe maxillary hypoplasia secondary to CLP.

Materials and methods

This study was approved by the ethics committee of Ninth People's Hospital Shanghai Jiao Tong University School of Medicine. Eight patients (all males) were enrolled in the study (**Table 1**), of which 6 patients were unilateral CLP and 2 were bilateral CLP. All of them showed severe maxillary hypoplasia secondary to CLP. The mean age was 23.38 (ranging from 19 to 31), and average reverse overjet was 10.25 mm (ranging from 6 to 15). They underwent alveolar bone grafts and orthodontic treatment before and after surgery. All patients were diagnosed with maxillary hypoplasia and almost normal mandibular development (sella-nasion-point B angles were less than 84°), indicating that only maxillary advancement was necessary. However in order to establish a functional dental occlusion, setting back the mandible was usually needed. Tooth-borne distractors were placed preoperatively for orthodontic treatment.

Anterior maxillary segmental osteotomies were performed under general anesthesia, the only difference between Le Fort I osteotomy and anterior maxillary segmental osteotomy was that the posterior maxillary osteotomy was performed between molars or between the first molar and the second premolar as described before [14]. After a 5-day latency period, the maxillary distraction was initiated at a rate of 0.4 mm twice a day to correct horizontal and vertical maxillary hypoplasia. According to the requisite forward distance of establishing a functional occlusal relationship and correcting the reverse overjet, the duration of the distraction ranged from 18 to 30 days. What's more, a prolonged distraction of three more days was essential for overcorrection of the overjet.

Table 2. Definitions and implications of cephalometric landmarks

Landmarks	Definition/full name	Implication
SNA (°)	sella-nasion-point A angle	Maxillary prominence
NA-FH (°)	nasion-point A-Frankfort horizontal plane angle	Maxillary prominence
Ptm-S (mm)	pterygomaxillary fissure-sella distance	Posterior maxillary edge to skull
Ptm-A (mm)	pterygomaxillary fissure-A distance	Maxillary length
U1-PP (mm)	upper incisor to palatal plane distance	Anterior maxillary height
A6-PP (mm)	upper first molar to palatal plane distance	Posterior maxillary height
0-meridian to Sn (mm)	0-meridian to subnasale distance	Maxillary prominence
PNS-PPW (mm)	posterior nasal spine to posterior pharyngeal wall distance	Palatopharyngeal depth
PNS-U (mm)	posterior nasal spine to uvula distance	Soft palatal length
PP (ANS-PNS) (mm)	anterior nasal spine to posterior nasal spine distance	Palatal length

Velopharyngeal closure examination was performed with nasopharyngeal fiberscopy and speech function was examined by speech clinicians. Before and after AMSD, cephalometric analysis was performed with Nemoceph analysis system (Nemotec, Madrid, Spain) (**Figure 1**). Tracing was performed twice at different time points by the same investigator. SNA, NA-FH, Ptm-S, Ptm-A, U1-NA, U1-PP, A6-PP, overjet, 0-meridian to Sn, PNS-PPW, PNS-U, and PP (ANS-PNS) (**Table 2**) were included in the cephalometric analysis.

Statistical analysis

All data were presented as mean ± standard deviation. Data were analyzed with SPSS (SPSS, Chicago, IL), and the significance level was set at $P < 0.05$. The results of cephalometric evaluation were compared among three time points with one-way analysis of variance

Results

All the distractions were completed smoothly with midfacial deformities efficiently corrected, and no insufficiency of velopharyngeal closure or speech function deterioration was found (**Figure 2**). The cephalometric analysis showed that there's pretty large improvement for maxilla, as the mean overjet was increased by 14.28 mm. Comparison of cephalometric analysis among the three time points-before surgery (T1), after distraction (T2), 2 years after treatment (T3), including SNA, NA-FH, Ptm-A, U1-PP, overjet and PP (ANS-PNS)-showed statistically significant ($P < 0.05$) improvement between T1, T2 and T1, T3, while no significant difference was found between T2 and T3. On the other hand, Ptm-S, A6-PP, 0-meridian to Sn, PNS-PPW, PNS-U showed no significant increase (**Table 3**).

Comparative evaluation between T1 and T2 showed that the mean value of SNA was improved by 10.73°, NA-FH by 10.64°, and Ptm-A by 13.91 mm ($P = 0.001$). This data indicated that the maxilla had been significantly moved forward with AMSD for correcting midfacial deformities. Improvement of U1-PP demonstrated that the anterior height of maxilla was increased by 6.34 mm. Evaluation of maxillary sagittal depth by PP showed a great increase of 10.02 mm. What's more, palatopharyngeal depth and soft palatal length had no significant changes by contrastive results of the PNS-PPW and PNS-U. Similar results were found by comparing these measurements between T1 and T3. However, comparative evaluation between T2 and T3 showed no significant change. Relapse tendency was easy to find but showed no statistical significance ($P > 0.05$), as the horizontal overjet decreased to 3.02 mm by a loss of 1.01 mm. SNA diminished to 84.63° by a loss of 0.53°, NA-FH diminished to 91.86° by a loss of 1.26°, Ptm-A reduced to 50.18 mm by a loss of 1.57 mm, and PP diminished to 51.63 by a loss of 0.29 mm.

Discussion

Patients with cleft lip and palate are always accompanied with maxillary hypoplasia in three dimensions. Many CLP patients show sagittal hypoplasia of maxilla, speech disorders and VPI [15]. Various reasons would be liable for maxillary hypoplasia, such as tension of scars, teeth agenesis and poorly reconstructed nasolabial muscles [16-18]. Maxillary advancement is essential to improve the aesthetic profile and

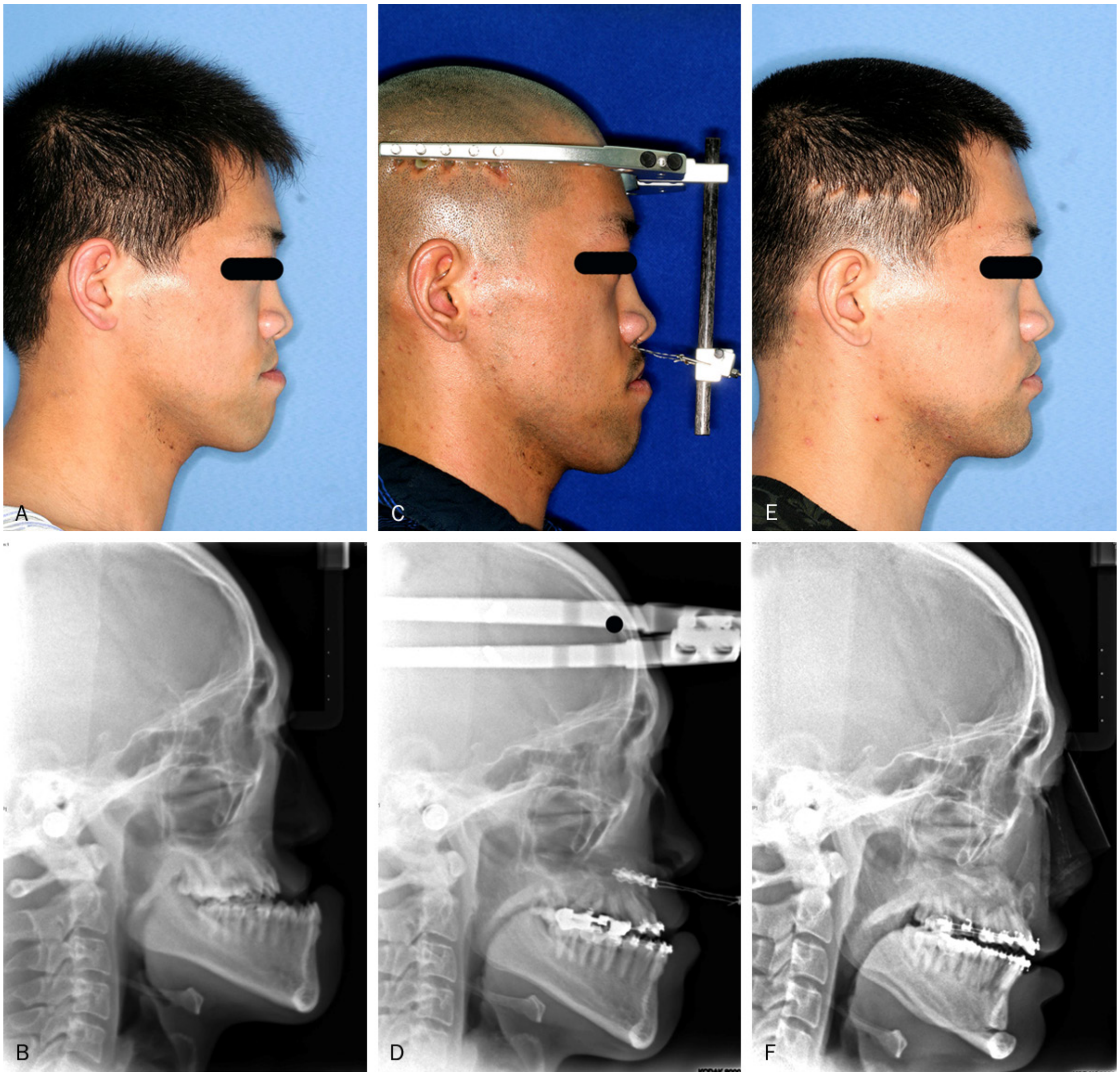

Figure 2. Lateral profiles, cephalograms at three time points: A, B before surgery; C, D after distraction; E, F 2 years after treatment.

functional occlusal relationship. Le Fort I osteotomy and entire maxillary distraction osteogenesis have been widely used to deal with maxillary retrusion and dental crossbite [19, 20]. Maxillary distraction osteotomy increases the amount of maxilla and induces mucosa extension, which in turn make DO a better technique for the correction of maxillary hypoplasia. However, velopharyngeal closure function can be damaged by this procedure [21]. On the other hand, anterior maxillary segmental distraction (AMSD) can avoid the situation mentioned above and provide distraction space that can be used for orthodontic alignment of teeth or dental implants [22-24].

No matter Le Fort I osteotomy or distraction osteogenesis, relapse is difficult to avoid, especially for CLP patients. A number of studies indicated that relapse occurred after Le Fort I osteotomy for correcting midfacial hypoplasia in CLP patients. What's more, with the popular application of distraction, more and more relapses after DO were also reported. Figueroa [6] demonstrated a long-term decrease of SNA in a 3-year follow-up. Krimmel [25] evaluated the stability of maxillary advancement and discovered that maxilla was stable in the first year, but the deterioration of ANB occurred after longer follow-up. Besides, Suzuki [9] reported an obvious relapse in unilateral CLP patients just 6

Table 3. Cephalometric changes amoung different time points

Group	T1	T2	T3	P1 (T2-T1)	P2 (T3-T1)	P3 (T3-T2)
SNA (°)	74.43 ± 4.99	85.16 ± 3.16	84.63 ± 4.38	0.001	0.001	0.965
NA-FH (°)	82.48 ± 4.37	93.12 ± 1.97	91.86 ± 3.17	0.001	0.001	0.731
Ptm-S (mm)	17.60 ± 1.79	17.97 ± 1.06	19.11 ± 4.34	0.696	0.532	0.961
Ptm-A (mm)	37.84 ± 3.40	51.75 ± 8.14	50.18 ± 3.35	0.001	0.001	0.836
U1-PP (mm)	19.56 ± 7.16	25.90 ± 1.92	28.95 ± 4.57	0.05	0.003	0.459
A6-PP (mm)	20.93 ± 2.04	21.11 ± 1.02	22.23 ± 2.63	0.995	0.623	0.614
Overjet (mm)	-10.25±3.92	4.03 ± 1.49	3.02 ± 3.29	0.001	0.001	0.838
0-meridian to Sn (mm)	6.12 ± 1.35	6.77 ± 1.30	6.52 ± 1.89	0.68	0.863	0.994
PNS-PPW (mm)	22.57 ± 2.88	23.19 ± 3.04	24.01 ± 4.06	0.929	0.675	0.878
PNS-U (mm)	25.08 ± 4.22	24.15 ± 3.59	22.61 ± 3.27	0.864	0.372	0.673
PP (ANS-PNS) (mm)	41.90 ± 0.96	51.92 ± 2.97	51.63 ± 1.58	0.001	0.001	0.954

T1: cephalometric analysis before surgery; T2: cephalometric analysis after distraction; T3: cephalometric analysis 2 years after treatment.

months after surgery. Investigation of this phenomenon showed that muscle and soft tissue stretching could affect the long-term outcome of DO. All patients in this study had achieved satisfactory result, as SNA and NA-FH increased significantly and showed no significant relapse. As described before, anterior maxillary distraction osteotomy was generally performed between the premolar and the first molar in the rate of 0.8 mm, allowing new bone regeneration in the distraction gap, which benefited a lot for mucosa extension. Furthermore, this kind of gradual process might reduce the risk of relapse and obtain more stable advancement at the same time. With new bone formation, the gap would offer orthodontists greater simplicity for leveling and alignment of teeth, as well as coordinating of the dental arches.

In this study, the posterior segmental maxilla was kept in the original position, while anterior maxillary segment was distracted in two dimensions: horizontal and vertical. U1-PP, which represented the height of anterior maxilla, was increased, indicating that distraction in the vertical dimension was viable to attain. Meanwhile, further increase of U1-PP and A6-PP after surgery was possibly due to the post-surgical orthodontic treatment. The greatest advantage of keeping the posterior position of maxilla was minimizing the effect on post-maxillary space. The palatopharyngeal depth (PNS-PPW) and soft palatal length (PNS-U) were affected by AMSD more or less, but no significant difference was observed among the three time points, which meant the least worsening on VPI when using AMSD procedure. The two landmarks, Ptm-A and PP (ANS-PNS), which indicated the sagittal length of palate, were significantly increased and no relapse was found.

In conclusion, AMSD is a good therapeutic procedure to realize aesthetic improvement and establish a good occlusal relationship with little risk of deterioration in velopharyngeal function. Furthermore, this study demonstrated that AMSD had good postoperative stability. Due to the ability of increasing the palatal and arch length, avoiding changes in palatopharyngeal depth, preserving palatopharyngeal closure function, and reducing the relapse rate, anterior maxillary segmental distraction has great value in the treatment of maxillary hypoplasia secondary to CLP. It is a promising and valuable technique in this potentially complicated procedure.

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Disclosure of conflict of interest

None.

Address correspondence to: Hongbo Yu and Steve Guofang Shen, Department of Oral & Cranio-Maxillofacial Surgery, Shanghai Ninth People's

Hospital, Shanghai Jiao Tong University School of Medicine, No. 639, Zhizaoju Road, Shanghai 200011, People's Republic of China. Tel: +86-21-23271207; Fax: +86-21-63136856; E-mail: yhb-3508@163.com

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