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Two-stage maxillary distraction osteogenesis using a modified external device: clinical outcome and complications

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Abstract

Rigid external distraction is currently used to correct severe maxillary hypoplasia. The purpose of this retrospective study was to present the clinical results and complications of a two-stage surgical approach using a modified external distraction system that consists of maxillary distraction and then maxillary fixation. We treated eight patients with cleft lip and palate in this way from 2016 to 2018. Lateral cephalograms taken before the first operation, after distraction, two weeks after the second operation, and one year after treatment were used to examine maxillofacial morphology. Velopharyngeal function was evaluated by a speech therapist. The mean movements of the maxilla forwards and downwards at Point A were 12.0 mm and 8.0 mm at the completion of distraction and those at Point B were 5.0 mm backwards and 9.7 mm downwards. Mouth opening was limited at this time, and was relieved after maxillary fixation. The mean relapse one year postoperatively was 24.3% horizontally and 52.5% vertically. Velopharyngeal function was unchanged by the operation. We conclude that the method has advantages that include the short duration of wearing distractors and increased acceptance by patients. The modified external device advanced the midface sufficiently.

Keywords: cleft lip and palate; Le Fort I osteotomy; distraction osteogenesis

Introduction

Midfacial hypoplasia is a common dentofacial malformation associated with cleft lip and palate, and orthognathic surgery is required for its correction when severe. In 1997 Polley and Figueroa[1] first introduced the use of a rigid external distractor to treat severe maxillary hypoplasia using distraction osteogenesis. This consisted of a halo frame attached to the skull, which exerted traction on the cut maxilla through a tooth-borne intraoral splint, and it has been shown to be effective. After distraction, the distractors are usually left in place for a consolidation period to ensure formation of new bone and to prevent relapse. It has a considerable influence on patients' normal lives and may lead to physical and psychosocial problems, so we developed a two-stage surgical approach that consists of maxillary distraction and internal rigid fixation immediately after distraction.

In previous studies our team developed a modified external distraction system with intranasal bone-borne traction hooks[2–4] to overcome the dental compensation and deformation[5,6]. The aim of the present study was to present the clinical results and complications of this two-stage approach using a modified external distraction system.

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Material and methods

We reviewed the patients who were treated in this way at the Center of Cleft Lip and Palate, Plastic Surgery Hospital, Chinese Academy of Medical Sciences and Peking Union Medical College, from May 2016 to September 2018.

Ethics

The Plastic Surgery Hospital, Chinese Academy of Medical Sciences, and Peking Union Medical College ethics committee approved the study, which followed the principles of the Declaration of Helsinki. Written informed consent was obtained from the patients or from the guardians of those younger than 18 years of age. All identifying images were authorised for publication by the patients or guardians.

Surgical technique

The system consisted of a rigid external distractor (Cibei Medical Treatment Appliance Co. Ltd), a nickel–titanium “shape memory alloy” spring, and nickel–titanium bone-borne traction hooks (diameter: 1.5 mm; GEE Co). The spring generated a continuous force of roughly 250 g/mm.[4]

We made maxillary vestibular incisions, and moved the buccal tissue to expose the pyriform rim. A thick fissure burr was then used to drill a hole about 1 cm outside the lateral pyriform rim and 5 mm above the apices of the teeth through the lateral nasal wall. We then made a Le Fort I osteotomy. The traction hooks were introduced through the hole, and the caudal ends extended from the base of the nostril. The cranial frame of the extraction device was placed 20°–30° upwards from the Frankfort horizontal plane, with the vertical rod 10–12 cm anterior to the base of the nostril. The spring was then usually used to connect the hooks to the device.

Distraction was started immediately after the operation, with an initial force of 250 g on each side and a direction of 20° off the Frankfort horizontal plane. A period of adaptation of 3–5 days followed, and then the distraction was initiated with a variable rate of 1–2 mm in the change of springs each day. The maxilla moved slowly with the continuous elastic distraction, which usually took 2–3 weeks. The end of the distraction was decided by clinical judgement and cephalometry.

Immediately after distraction, and after a short consolidation period (normally 3–7 days), we did the second operation. First the distractors were removed, and then the pyriform rim and the zygomatic buttress were exposed. After intermaxillary fixation using an occlusal guide plate, the L-shaped titanium plates (1.0 mm thick) were placed and secured with four crews (2.0 mm in diameter and 6 mm long). The rigid internal fixation was away from the osteotomy line to avoid the influence of granulation tissue. A maxillary protractor was used in three cases after treatment.

Imaging analysis

A single examiner who was unaware of the treatment made assessments based on manual cephalometric analysis. Lateral cephalograms were taken just before the first operation (T0), immediately after distraction (T1), two weeks after the second operation (T2), and a year later (T3). The cephalometric analysis had an *x*-axis and *y*-axis coordinate system that originated at the sella, with the x-axis established by rotating the sella-nasion (SN) line downward by 7° around the sella. The *y*-axis was constructed perpendicular to the x-axis through the sella. The bony landmarks used for analysis of distance included the subspinale A-point (A), and the supramental B-point (B). The following angular measurements were assessed: sella-nasion-point A (SNA), sella-nasion-point B (SNB), and A point-nasion-B point (ANB) for the positions of the maxilla and mandible. The palatal-plane angle (palatal plane-Frankfort horizontal (PP-FH)) and the mandibular plane angle (mandibular plane-Frankfort horizontal (MP-FH)) were assessed for rotational movement of the maxilla and mandible. All the lateral cephalographs were traced and superimposed using the cranial base points.

Assessment of velopharyngeal function

Velopharyngeal function was examined by a speech therapist preoperatively, and four weeks and one year postoperatively. Hypernasality was recorded. The spontaneous speech sample was rated for hypernasality using an equal interval scale (1 = normal, 2 = mild, 3 = mild to moderate, 4 = moderate, 5 = moderate to severe, or 6 = severe hypernasality).

Statistical analysis

The statistics were analysed with the aid of IBM SPSS Statistics for Windows (version 20.0, IBM Corp). The same process was repeated a week later to assess accuracy of measurement. The significance of the differences between the two sets of tracing measurements were compared using a paired *t* test ($p < 0.05$ or less was accepted as significant), and there was no significant difference between them. The paired *t* test was also used to analyse the significance of differences between the changes; again, $p < 0.05$ was accepted as significant.

Results

Maxillary and mandibular positions (point A, point B)

The mean (SD) movements and changes are shown in Table 1, together with the significance of the differences. The postoperative relapse rates one year postoperatively are shown in Table 2.

Gradual antero-upwards movement continued during postoperative orthodontic treatment,

Table 1
Mean (SD) value for each variable, and change.

Variable	Mean (SD) value				Mean (SD) change			p values		
	T0	T1	T2	T3	T1-T0	T2-T1	T3-T2	T1-T0	T2-T1	T3-T2
SNA (°)	73.4(4.5)	84.5(4.9)	81.7(3.9)	80.5(3.2)	11.1(1.7)	−2.8(1.5)	−1.2(1.1)	0.000	0.002	0.017
SNB (°)	77.5(5.1)	74.9(5.1)	75.8(4.7)	76.8(4.5)	−2.7(1.5)	1.0(0.9)	1.0(0.9)	0.002	0.026	0.025
ANB (°)	−4.2(2.3)	9.6(2.8)	5.9(2.8)	3.7(2.5)	13.8(1.6)	−3.8(1.3)	−2.2(0.9)	0.000	0.000	0.000
MP-FH(°)	31.7(8.1)	38.8(7.1)	35.9(7.5)	34.6(7.9)	7.1(3.0)	−2.9(1.4)	−1.3(1.2)	0.000	0.001	0.022
PP-FH (°)	9.8(5.0)	19.7(6.8)	16.0(7.1)	12.3(7.1)	9.9(3.1)	−3.7(2.3)	−3.7(2.4)	0.000	0.004	0.005
A- *x* (mm)	53.6(2.8)	61.5(3.8)	57.5(4.9)	55.5(3.5)	8.0(2.6)	−4.07(2.5)	−2.0(1.5)	0.000	0.004	0.020
A- *y* (mm)	55.5(7.1)	67.5(7.4)	65.0(7.1)	62.7(5.9)	12.0(1.4)	−2.5(1.0)	−2.3(1.5)	0.000	0.000	0.006
B- *x* (mm)	93.5(5.2)	102.7(4.3)	98.8(5.3)	97.4(5.4)	9.2(1.8)	−3.9(2.7)	−1.4(1.4)	0.000	0.006	0.033
B- *y* (mm)	57.0(9.5)	51.9(9.9)	54.5(9.7)	53.8(9.7)	−5.0(2.4)	2.6(2.0)	0.7(1.8)	0.001	0.013	0.321

SNA = sella-nasion-point A angle; SNB = sella-nasion-point B angle; ANB = point A-nasion-point B angle; MP-FH = mandibular plane-Frankfort horizontal plane angle; PP-FH = palatal plane-Frankfort horizontal plane angle; B- *x* = distance from *x*-axis to point B; B- *y* = distance from *y*-axis to point B; A- *x* = distance from *x*-axis to point A; A- *y* = distance from *y*-axis to point A. Paired *t* test, $p < 0.05$.

Table 2
Relapse of point A.

Variable	Mean (SD) value		Relapse	p values	
	T2-T0	T3-T2	Rate (%)	T2-T0	T3-T2
A- *x* (mm)	3.9(2.8)	−2.0(1.8)	52.5(25.5)	0.008	0.020
A- *y* (mm)	9.5(1.7)	−2.3(1.5)	24.3(16.3)	0.000	0.006

A- *x* = distance from *x*-axis to point A; A- *y* = distance from *y*-axis to point A. Paired *t* test, $p < 0.05$.

Maxillary and mandibular positions relative to the cranium (SNA, SNB) and maxillomandibular relations (ANB)

Changes in SNA, SNB, and ANB are shown in Table 1. SNA improved in all cases after distraction and consolidation ($p < 0.001$, respectively). The mean preoperative SNB was 77.5°, and it decreased during distraction (74.9°). During postoperative orthodontic treatment, the mean SNB gradually increased, but was still smaller than the preoperative value.

Rotational changes to the maxilla and mandible (palatal plane and mandibular plane)

Changes in PP-FH and MP-FH are shown in Table 1. The mean preoperative PP-FH was 9.8°, and it had rotated clockwise at the completion of distraction. During maxillary fixation, we moved the maxilla upwards, resulting in counter-clockwise movement postoperatively at T2. The mean MP-FH increased with operation (T0-T1, $p < 0.001$), indicating mandibular backwards rotation (Fig. 1). Between T2 and T1, the mean MP-FH decreased by 2.9° ($p = 0.001$) (Fig. 2). The position of the teeth changed during postoperative orthodontic treatment, and the mandible was still rotated backwards.

Assessment of velopharyngeal function

One patient had mild to moderate hypernasality immediately after the second operation, but it had disappeared two months later. We did no pharyngeal flap operations, and no change in velopharyngeal function was detected in the other patients.

Fig. 1. Superimposition of T0 and T1. Clockwise rotations of the palatal plane and anteroinferior movement of point A were noted at the completion of distraction, which led to backwards rotation of the mandible. Maximum mouth opening decreased during this time.

Complications

Varying degrees of limited mouth opening were present in all patients at the completion of distraction. Six patients were unable to position one finger vertically between the upper

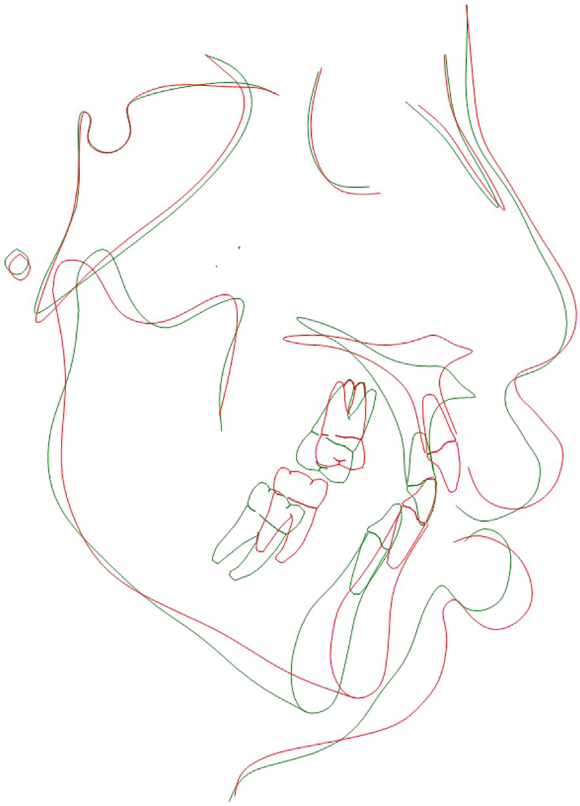

Fig. 2. Superimposition of T1 and T2 showing counter-clockwise rotation in the maxilla and mandible after maxillary fixation. Hypomobility in the mandible was relieved to a certain degree.

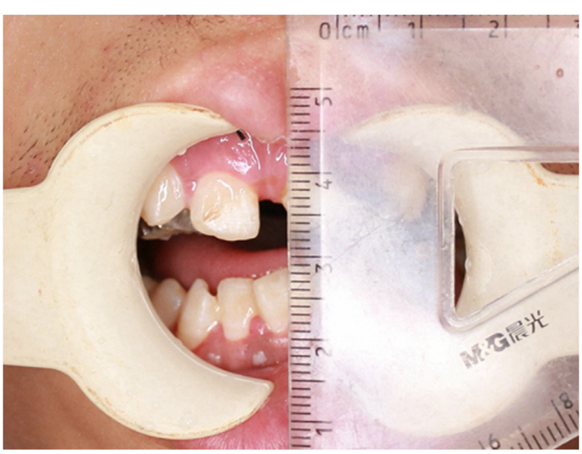

Fig. 3. Maximum mouth opening was significantly reduced at the completion of distraction. The patient was unable to position one finger, vertically aligned, between the upper and lower central incisors.

and lower central incisors up to the first distal interphalangeal folds (Fig. 3). This was relieved to a certain extent immediately after maxillary fixation (Fig. 4). Patients had completely recovered by one month after the operation.

Fig. 4. Reduced mouth opening was relieved to some degree after the second operation.

Discussion

External distraction osteogenesis is a well-established treatment for severe maxillary retrusion in cleft lip and palate,[7,8] and our cephalometric analysis showed that the mean horizontal movement was 12.0 mm immediately after distraction. The treatment gradually advances the midface, which allows for progressive osteogenesis and adaptation of the surrounding soft tissues. The gradual movement requires less counter-force to pull the maxilla into the appropriate position, which makes the maxilla more stable than after a traditional Le Fort I osteotomy.[9,10]

The distractors are usually left in place for a long consolidation period to prevent relapse and ensure formation of new bone, which imposes a heavy burden on patients and which may lead to physical and psychosocial problems. To overcome these, we removed the distractors after a short consolidation period and then fixed the maxilla. There was a 24.3% relapse one year postoperatively, which was similar to that found by Susami et al[11] who had a 25.2% relapse using a two-stage procedure that combined maxillary distraction with mandibular setback.

Overcorrection was necessary during distraction, and not only because of the relapse. During the second operation, it was easier to push the maxilla back than to draw it forwards. Overcorrection compared with the planned position was important for precise fixation using an occlusal guide plate, which was the main reason why the changes at point A decreased between T2 and T1.

The direction was important during distraction, as the location of the distraction force through the centre of the mass would produce linear advancement along its line of action. There would be movement downwards and forwards, or upwards and forwards, if the protractive force line of action superior or inferior was positioned to the centre of the mass. In general, the centre was at the apices of the maxillary premolars.[8,12] We used intranasal bone-borne traction hooks that extended from the base of the nostrils, and the force was applied above the centre of the mass. This caused clockwise rotation with a posterior open bite, with changes in PP-FH. We were unable to completely avoid this effect even by changing the angle of direction, but simultaneous use of a tooth-borne distractor may solve the problem.

Maximum mouth opening has been scored by many previous reports, and an initial decrease 1-6 months postoperatively has been described.[13] Reduced mouth opening is also a known complication of Le Fort III distraction and has been well described in craniofacial publications.[14] However, to our knowledge it has not been reported after Le Fort I distraction. We found varying degrees of limited mouth opening in all patients, six of whom had serious hypomobility of the mandible. From our cephalometric results we found clockwise rotation of the palatal plane and antero-inferior movement of point A at the completion of distraction. One reason for this was the location of the distraction force as previously described, and the other reason was that the distraction angle was 20° off the Frankfort horizontal plane. The two factors resulted in inferior movement of the maxilla, which led to backward rotation of the mandible. The mean increase of MP-FH was 9.9°, and the mean movement of point B was 5.0 mm backwards and 9.7 mm downwards, which could be the main reason for the decrease in maximum mouth opening in these cases, and which reminded us that the distraction angle should be reduced.

Another reason might be pain. The intranasal bone-borne traction hooks pass through the nasal mucous membrane, and excessive mouth opening could induce pain in this area. There are other possible reasons (as stated in previous studies) such as repositioning of the jaw, transient muscular adaptations, and residual areas of inflammation.[15] Limited mouth opening was also an important reason for doing the second operation. After maxillary fixation point B moved 2.6 mm anteriorly and 3.9 mm upwards, the limited mouth opening was relieved at a certain point. No patient had this side effect one year after the operation.

Velopharyngeal insufficiency after maxillary advancement is a well-known complication and is essentially reported in patients with borderline function preoperatively.[8] Le Fort I distraction advancement, which allows for progressive movement of the maxilla, favours adaptation of the velopharyngeal structures. Bevilacqua et al[16] found that no patients had increased hypernasal speech when midmaxillary internal distraction osteogenesis was used. Chanchareonsook et al[17] reviewed velopharyngeal insufficiency and found that operation had no impact, whereas we found no deterioration of velopharyngeal function in eight patients followed up for one year. However, one patient complained of hypernasality immediately after treatment. Careful examination of speech was still needed.

Conclusions

This method has advantages that include the short period for which patients had to wear distractors, which increased their acceptability. Our modified external device sufficiently advanced the midface. Though there were complications such as rotation and limited mouth opening with this technique, they were relieved eventually.

Financial disclosure

The authors have no financial interest declare in relation to the content of this article.

Conflict of interest

We have no conflicts of interest.

Ethics statement/confirmation of patients'permission

The Plastic Surgery Hospital, Chinese Academy of Medical Sciences, and Peking Union Medical College ethics committee approved the study. Patients' permission was obtained.

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