Limitations of Internal Distraction Devices in Mature Patients with Cleft Lip and Palate and Severe Midface Hypoplasia

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Abstract

Purpose We evaluated case-sensitive limitations of internal distraction devices (IDDs) to reveal further options for satisfactory outcomes.

Patients and Methods Two male cleft patients aged 22 years (P1) and 20 years (P2) with severe maxillary hypoplasia were treated over 18 months. Cephalometric analyses and analyses of A-point movements by superimposing lateral cephalograms were done before and after activation of IDDs and after bimaxillary surgery (BIMAX). **Results** After activating IDDs, the vectors deviated and clockwise rotation of the maxilla and vectors occurred. After 8 months, BIMAX was needed to carry out further maxillary advancement.

Conclusions IDDs could pave the way to achieve maximal advancement with BIMAX in a second step and justify the overall treatment when comparing the initial situation with outcome.

Keywords Maxillary advancement · Internal distraction devices · Cleft lip and palate · Vector deviation

Introduction

After repair of cleft lip and palate, patients frequently present a maxillary hypoplasia with a greater or lesser

extent of a class-III malocclusion and soft-tissue conditions compromised by rigid scar formation. Three-dimensional corrections of maxillary discrepancies in these patients remain a challenge for maxillofacial surgeons. Conventional orthodontic surgical methods frequently fail to reach expectations from the viewpoint of achieving normal occlusion and aesthetic proportions of the face after long-term follow-up [1–3]. Maxillary distraction osteogenesis has therefore become an important part of maxillofacial surgery because it is in particular a useful technique in the midface to overcome the perturbations and restrictions following growth by providing skeletal advancement and expansion of soft tissue at the same time [4–8].

On the basis of our experiences and despite using distraction devices, in some patients with matured cleft lip and palate, optimal orthodontic and surgical outcomes are not always possible due to several factors. These include: extremely rigid scars; excessive hypoplasia of the midface; missing teeth; failure of previously grafted alveolo-palatal cleft closure implicating the impossibility to form a harmonic teeth arch; or even patients' non-compliance with treatment. Nevertheless, it remains essential to achieve optimal results for each patient with regard to function and aesthetic parameters knowing that deficits compared with outcomes of routine cases will appear. At the endpoint of treatment of these patients, the résumé should show that at least treatment procedures lasting years were worthwhile and that results show an improvement and a fundamentally better status than before surgery.

The aim of this study was to evaluate the reasons and therapeutic options if midface distraction does not lead to optimal outcomes. Two cases of extreme skeletal dysgnathia after repair of cleft lip and palate and after velopharyngoplasty are described to reveal the understanding of individual satisfying solutions.

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Patients and Methods

Patients

Two male patients aged 22 years (patient 1) and 20 years (patient 2) with severe maxillary anteroposterior hypoplasia and massive palatal scarring were treated for skeletal dysgnathia (class-III malocclusion) over a period of 18 months. Both patients had bilateral cleft lip and palate. Numerous previous procedures focusing on the lip, alveolus and palate had been carried out (including velopharyngoplasty for speech improvement at the age of 6 years). At the time of first presentation for orthognathic surgery, patients had no symptoms of an obstructive sleep apnea-hypopnea syndrome (OSAHS) but presented with hypernasal speech. Both patients showed symptoms of a fetal alcohol syndrome since birth.

Distraction Planning

Patients underwent pre- and postoperative orthodontic treatment. The initial treatment plan consisted of Le Fort I internal distraction using a trans-sinusoidal maxillary distractor (TS-MD; Martin GmbH and Co Kg, Tuttlingen, Germany). Lateral cephalometric radiographs, cast-study models and stereolithographic (STL) models fabricated from CT scans were used to plan the distraction. Prediction tracing was initially done to determine maxillary advancement vectors. Therefore, lateral cephalograms were digitized and imported into a personal computer and transferred into the program Onxy Ceph Version 2.7.19 (Image Instruments GmbH, Chemnitz, Germany). Le Fort I osteotomies were completed on STL models. Internal distraction devices (IDDs) were then pre-bent and adapted to the STL models on each side of the maxilla. In patient 1, the distracting screws were oriented parallel to each other and to the sagittal plane, and then placed in the maxillary sinus in the predetermined vector. After sagittal splitting of the maxilla of the STL model, in patient 2, the vectors of the distracting screws were additionally set divergently at an angle of 5° to also expand the maxilla in transverse direction. A three-dimensional mock distraction was carried out on the STL models. The final position of the maxillary incisor was verified visually to be as predicted.

Surgery

Surgery consisted of Le Fort I osteotomy without advancing or down-fracturing the maxilla. Pre-bent IDD were then installed on both sides of the maxilla and the distraction screws placed in the maxillary sinus. After a five-day latency phase, distractors were activated by turning the activating rods at a rate of one complete turn daily

(half-turn two-times daily). This resulted in a predicted advancement of 0.5 mm per day along the predetermined vector. After 14 days of distraction, maxillary advancement arrested in both patients, and only a clockwise rotation of the maxillary complexes resulted in further distraction with closure of the open bite. Distraction was continued in both cases to provide a widening of the soft tissue and rigid scars. After reaching the full expanse of distraction devices after 30 days, the activating rods were cut and the devices left in situ for additional 8 months for bony consolidation.

In a second step, both patients underwent bimaxillary osteotomy (BIMAX) including Le Fort I osteotomy with complete mobilization of the maxilla for further maxillary advancement and bilateral sagittal split ramus osteotomy for mandible adjustment in combination with removal of the distraction screws and caudal distractor plates. The cephalometrically normal mandibles in both cases were not set back but instead lifted and rotated counter-clockwise in the bilateral sagittal split osteotomy lines. The planning of postoperative occlusion implicated an angle class-III relationship of the molars and an anterior cross bite. This was because it was not possible for orthodontic therapy to achieve an initial optimal occlusal scenario for a class I occlusion after surgery: after repair of cleft lip and palate, maxillary lateral incisors were lost in earlier years and anterior alveolus bone supply was poor. The maxillary central incisors stood on an imaginary direct axis between the canines and could not be moved into an anterior class-I occlusion with an over-bite and over-jet. In patient 1, 6 months after BIMAX, both mobile central incisors were extracted and a fixed prosthesis integrated for prosthetic rehabilitation and for stabilization of the occlusion. For aesthetic reasons, an anterior edge to edge bite was chosen because an over-bite would totally expose the incisors due to the short upper lip. In patient 2, a prosthetic rehabilitation could not be started up to the present day because of missing reimbursement of his medical insurance.

Cephalometric Analysis

All lateral cephalograms were obtained using a cephalostat and a defined object-to-focus distance of 1.5 m. Cephalograms were then scanned at 150 dpi for digitalization and transferred into the computer program and traced. Cephalometric analysis was done according to the description of Hasund and Jansen [9] using the following parameters: sagittal: SNA (prognathia angle of maxilla); SNB (prognathia angle of mandible); ANB (sagittal interbase angle); vertical: NL–NSL (inclination angle of maxilla); ML–NSL (inclination angle of mandible); NL–ML (vertical interbase angle) and ArGoMe (mandibular jaw angle). The maxillary and palatal related posterior airway space (PAS) was

determined using the planes PAS–uvula point (UT) and PAS–spina nasalis posterior (NL) according to the method of Hochban and Brandenburg ([10]). Cephalograms were taken 14 days before implantation of distractor devices (T0); after device implantation before activation in patient 2; and after eight days of distraction in patient 1 (T1) because patient 1 was not willing to have a lateral cephalogram before activation; at the end of distraction period (T2); and 3 months after BIMAX (T3). Additionally, in patient 1, cephalometric analysis was done after prosthetic rehabilitation (T4). To compare and evaluate the effort of distraction and BIMAX for maxillary advancement in millimeters, the traced cephalograms carried out at the time points T0 and T2, and T2 and T3, were superimposed using the NSL-plane as a reference. The amount of maxillary movement or its relapse in the sagittal direction was measured perpendicular from the A-point to the SNA of the superimposed cephalogram, and in the vertical direction as the distance between the A-point and a perpendicular line from the SNA in the amount of the A-point of the superimposed cephalogram. The total amount of movement was measured directly from A-point to A-point. The 112% magnificent ratio of cephalometric radiographs resulting from the object-to-focus distance of 1.5 m was corrected to 100%. Distraction vectors were represented by tracing along the long axis of the distraction screws. The angular difference of the distraction vectors was measured by superimposing the cephalograms T1 and T2.

Results

General Observations

All lateral cephalograms, the additional radiographs of the paranasal sinuses, and the orthopantomograms displayed no evidence of fracture or loosening of the distraction devices, screws or miniplates. No infections or wound-healing disturbances occurred.

Patient 1

A 2-mm anterior and a 4-mm posterior open-bite were presented preoperatively and were closed after the distraction period. Cephalometric measurements at T0, T2, and T3 are given in Table 1. In particular, NL–NSL increased and ML–NL decreased due to a clockwise rotation of the maxilla of about 20° at T2, whereas the deviation of the distraction vectors between T1 and T2 amounted to 15°. At T2, the maxilla (A-Point) was advanced by 9 mm (6-mm forward; 7-mm downward). After BIMAX at T3, the maxilla was de-rotated as indicated by the angular narrowing of NL–NSL and widening of ML–NL and by further advancement and cranial

Table 1 Development of cephalometric data of patient 1 and 2 during maxillary advancement with IDD and BIMAX (T0–T3)

	T0	T2	T3
Patient 1			
SNA	66	70	82
SNB	81	80	83
ANB	15	10	1
ML–NL	31	11	22
NL–NSL	5	27	18
ArGoMe	125	125	122
PAS–UT	9	13	14
PAS–NL	17	18	18
Patient 2			
SNA	75	79	82
SNB	82	83	84
ANB	7	4	2
ML–NL	36	25	24
NL–NSL	6	18	10
ArGoMe	136	136	134
PAS–UT	7	6	7
PAS–NL	11	11	12

Data are given in angular degrees (°), except distances PAS–UT and PAS–NL are given in millimeters (mm)

impaction of the maxilla. A-point advancement at T3 consisted of 15 mm in total (12 mm forward; 10 mm upward). The measurement of PAS–UT and PAS–NL at T0 covered the lower normal range. A widening of the PAS was observed for PAS–UT but not for PAS–NL at T2. In comparison, at T3 further significant extension of the PAS was not observed. At T4, a significant relapse greater than 2° of angular parameters or greater than 1 mm in the vertical or sagittal direction of the A-point could not be identified. Figure 1 displays clinical photographs at T0 and T4, Fig. 2 presents occlusal views from T2 to T4 and Fig. 3 shows cephalograms from T0 to T4.

Patient 2

An anterior open-bite at T0 of 10 mm was already closed at T2. The cephalometric measurements at T0, T2, and T3 are given in detail in Table 1. At T2, a rotation of the maxilla in a clockwise direction of about 12° was observed compared with T0. The aberrance of the distraction vectors between T1 and T2 amounted to 47°. At that time, cephalometric analysis presented a total movement of the maxilla (A-point) of 8 mm (4-mm forward; 6-mm downward). Direct measurement obtained a widening in transversal direction of 4 mm between the middle incisors. At T3, the A-point was moved 7-mm further forward and 3-mm upward, resulting in de-rotation of the maxilla but with presentation of recurrence of an open-bite.

Fig. 1 Lateral views of patient 1 (*above*) at T0 (*left*) and T4 (*right*), and patient 2 (*below*) at T0 (*left*) and T3 (*right*)

Fig. 2 Lateral and frontal occlusal views of patient 1 from T2 to T4

Measurement of the PAS–UT plane displayed pathological data at T0, T2, and T3, whereas measurement of PAS–NL was normal. Figure 1 displays clinical photographs at T0 and T3 and Fig. 3 shows the history in cephalograms from T0 to T3 for patient 2.

Discussion

Although recent studies suggest successful results for mid-face distraction in mature patients with cleft lip and palate [7, 8, 11] the outcome of using IDD for maxillary advancement in these patients was disappointing. A gain of maxillary advancement was achieved in both cases, but the aim to bridge the planned distance completely to reach a convexity of the face and an anterior over-bite failed. The problem was an unplanned clockwise maxillary rotation during distraction once a maxillary advancement started due to deviation between the distraction vectors at the beginning and at the end of distraction. This problem is well known [4], but it has not been described as severely as in this report, and has not been linked to the necessity of a bimaxillary osteotomy to overcome this misdirected distraction [12–14]. In a report of three patients with repaired cleft lip and palate, Gateno et al. [15] observed a clockwise rotation of the maxilla with a tendency to a posterior open-bite and a difference between the actual distraction vectors and the planned vectors. Vector deviations ranged from 9° to 26°, causing rotation of the occlusal plane between 2° and 9°. Interestingly, two of three cleft patients had pharyngeal flaps, and these patients presented the greatest rotations and the highest deviations between actual and planned vectors. In accordance with Gateno et al. [15], in the present study no correlation between deviation of the vectors and actual rotation of the maxilla could be found. One physical reason for the clockwise rotation of the maxilla during distraction even though these distractors were designed to be rigid is that the distraction force was applied above the maxillary center of resistance and so the rotation was introduced by a simultaneous forward movement. In contrast, some external devices are fixed with the teeth and so are below the center of resistance, causing counter-clockwise rotation of the maxilla and additionally offer the advantage to alter the vector during distraction [4, 16]. The extreme scarring in our patients after repair of cleft lip palate and after velopharyngoplasty should not be underestimated. These scars may not only be the reasons for insufficient maxillary growth and the underdevelopment of the midface [3], but also the reason of extreme maxillary rotation or even an arresting during distraction. In a retrospective view, it was obvious that the maxillary center of resistance in these cases was not only below the distraction force but additionally behind it in the region of the spina nasalis posterior, producing a

Fig. 3 History in cephalograms from T0 to T4 for patient 1 (*above*) and from T0 to T3 for patient 2 (*below*)

reinforcement of the rotation. After distraction in both patients, significant lengthening of the PAS–NL plane did not occur, providing evidence for a rigid scarring fixation of the maxilla and, in contrast to other studies finding a widening of the PAS [17, 18]. This scarring resistance was hard to overcome by the force of the distraction devices. As a result, by further activation of the distraction screws and a lengthening of the space between the cranial and caudal of the osteotomy line-inserted distraction plates, the rigidity of distraction screws may be overcome by the scarring maxilla. This may achieve an elastic bending of the distraction screws with a downward adjustment after rotation of the maxillary complex compared with the initial situation. Nadjmi et al. [13] reported a comparable phenomenon in which activation arms were apparently not strong enough and were bent, thereby stopping further distraction. Only after replacement with more rigid activation arms was distraction successful. Further studies evaluating the maximal possible load on internal devices should be done to clarify this point.

Additionally, some other factors may have played a more or less important part in distraction deviation. In particular in patient 2, it was critical to place the distractors parallel to each other on both sides of the maxilla, parallel to the sagittal plane, and additionally in a transverse angle of 5° to achieve a palatal widening as described in previous studies [8, 12]. Sufficient bony support was required to anchor the devices to the zygomatic buttress that could not be exactly evaluated with a SLT model. Some minor changes in bending of the distraction devices were

therefore necessary. Besides this experience and as opposed to external devices, the disadvantage of IDD that vectors can only be defined during the installation and once the devices are placed the vector of movement cannot be changed, is clearly shown in this report. The decision was initially made for internal devices for the comfort of the patients so as not to further lower their compliance [5, 19]. It may have been more practicable to apply external devices. As a promising alternative, an apparently feasible method has been described. In cases of severe scarring in cleft patients, maxillary advancement had been achieved by internal distraction only of the anterior part of the maxilla. With this method, the distraction of the scar formation and the pharyngeal flap could be avoided [14]. The disadvantages lie in the fact that a complicated preoperative orthodontic setup is necessary because the upper molars are not included in maxillary advancement, and that subsequent loading of dental implants is necessary for stabilization of the occlusion. Further studies are warranted to evaluate this method with respect to outcome and long-term stability.

Hence, the question may be asked: was it worth trying an internal distraction of the midface in these patients? It should be reconsidered that maxillary advancement greater than 6 mm is often difficult to achieve in patients with repaired cleft lip and palate with a primary bimaxillary osteotomy because of the firmness of maxillary scarring [5, 20]. A mean postoperative relapse of maxillary advancement up to 25% has been reported, and often it is necessary to concurrently

setback the mandible even if it is normal to cover an angle class III [21, 22]. Although maxillary advancement with IDD did not fulfill our expectations, a widening and loosening of scarring formations was achieved through the rotation. This effect allows the possibility to advance the maxilla in a second step with a bimaxillary osteotomy 15 mm in patient 1 and 9 mm in patient 2. These distances could not be overcome without internal distraction alone. As expected, deficits in aesthetics occurred and, with regard to occlusal parameters, an anterior cross-bite remains in both patients (which is covered with a fixed denture in patient 1). For this patient, a revision of the lip scar and a septorhinoplasty are planned.

In conclusion, satisfying results for these patients could be achieved. We feel that this justifies the overall treatment when comparing the initial clinical situation with the end result. For severe cases of midface hypoplasia due to matured cleft lip palate with velopharyngoplasty, we recommend to take into account a bimaxillary osteotomy after underachieved distraction and prompt definitive stabilization of the occlusion with prosthetic devices.

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