CLINICAL STUDY

Long-term results of maxillary distraction osteogenesis in nongrowing cleft: 5-years experience using internal device

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ABSTRACT

Patients with cleft lip and palate (CLP) related deformities frequently have maxillary hypoplasia in all dimensions. These patients usually present with class III malocclusions, retruded midfaces and narrow hard palates. The skeletal problems can be treated by means of Le Fort I maxillary procedures. Surgical and orthodontic correction of severe maxillary hypoplasia, as often seen in CLP patients, has however proved to be challenging. The magnitude of the advancement is often hampered and the post operative stability significantly affected by palatal soft tissue scarring. The slow distraction of bone and the histogenic abilities of distraction osteogenesis (DO) have made it an attractive alternative treatment option for the management of maxillary hypoplasia in these patients. This paper presents the treatment results of 15 nongrowing CLP patients with severe maxillary hypoplasia treated by means of intra oral distraction. The mean anterior distraction of the maxillas was 12.7 mm (9–15.0 mm). The long-term cephalometric and clinical evaluation after a minimum of 60 months (mean follow-up 71 months) proved to be stable. The treatment results revealed, that distraction osteogenesis in nongrowing CLP patients with severe maxillary hypoplasia proved to be a predictable and stable option (Tab. 2, Fig. 3, Ref. 26). Text in PDF www.elis.sk.

KEY WORDS: distraction osteogenesis (DO), cleft lip and palate (CLP), internal device, maxillary hypoplasia.

Introduction

Conventional Le Fort I osteotomy (one piece or segmental) with miniplate fixation is the treatment of choice for the correction of dentoskeletal deformities involving maxillary hypoplasia. This procedure however, has limitations particularly in cases requiring a greater than 8 mm advancement (2, 3, 4, 7, 16, 22, 23). To compensate for a large discrepancy between the maxilla and the mandible and to limit post operative relapse the clinician may have to consider to split the difference by means of a mandibular setback for the correction of the malocclusion. This approach may however result in a compromised esthetic treatment outcome.

Maxillary hypoplasia is a common dentofacial deformity found in patients with cleft lip and palate (CLP) and these patients usually present with Class III malocclusion, concave profile, retruded lower midface, and narrow hard palate. The Le Fort I osteotomy is currently the procedure of choice for the correction of maxillary deformities (8, 14, 15, 16, 19, 23). However for patients requiring large maxillary advancements (i.e. syndromic and CLP patients), advancement of the maxilla by means of distraction osteogenesis is a useful alternative treatment method. Distraction osteogenesis (DO) is currently an accepted treatment option for the slow and controlled repositioning of facial bones. Mc Carthy et al (12) reported the first clinical application of craniofacial distraction for the treatment of patients with hemifacial microsomia. In 1997 Polley and Figueroa (15) published an article on maxillary-midface advancement by means of distraction osteogenesis according to Ilizarov’s principles using an external distraction device. Patient’s discomfort and soft tissue scarring are certainly disadvantages of extra oral distraction and need to be considered. Improved design of intra oral distraction devices has largely circumvented the above disadvantages (3, 7, 16, 20).

Distraction procedure not only increases the bone volume, but also stimulates growth of the surrounding soft tissues. The soft tissue reaction is called „distraction histiogenesis“ and reduces soft tissues tension and its adverse effect on the repositioned bone (16, 19, 20, 24). Intra oral distraction devices are, however, also not without disadvantages. One of the most pertinent disadvantages of internal distraction devices is the fact that the distractor has only unidirectional vector of force leading to difficulty in achieving acceptable final occlusion (25, 26). In the authors view, intra oral distraction is indicated for the treatment of CLP patients requiring more than 8mm maxillary advancement. Furthermore most of these patients may require a staged approach involving an additional mandibular setback procedure (bilateral sagittal split osteotomy – BSSO). The decision regarding the choice of treat-
Tab. 1. Patients with one- and bilateral cleft lip and palate deformities.

<table>
<thead>
<tr>
<th>Patient No.</th>
<th>Gender</th>
<th>Age (years)</th>
<th>Diagnosis</th>
<th>Distraction</th>
<th>Follow-up months</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td>M</td>
<td>18</td>
<td>B</td>
<td>F</td>
<td>14</td>
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<tr>
<td>2.</td>
<td>F</td>
<td>17</td>
<td>U</td>
<td>B</td>
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<td>3.</td>
<td>M</td>
<td>20</td>
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<td>B</td>
<td>15</td>
</tr>
<tr>
<td>4.</td>
<td>M</td>
<td>22</td>
<td>B</td>
<td>F</td>
<td>12</td>
</tr>
<tr>
<td>5.</td>
<td>M</td>
<td>18</td>
<td>U</td>
<td>B</td>
<td>11</td>
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<tr>
<td>6.</td>
<td>M</td>
<td>17</td>
<td>U</td>
<td>F</td>
<td>10</td>
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<tr>
<td>7.</td>
<td>F</td>
<td>24</td>
<td>U</td>
<td>F</td>
<td>15</td>
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<td>8.</td>
<td>F</td>
<td>14</td>
<td>U</td>
<td>F</td>
<td>15</td>
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<tr>
<td>9.</td>
<td>M</td>
<td>22</td>
<td>U</td>
<td>F</td>
<td>12</td>
</tr>
<tr>
<td>10.</td>
<td>F</td>
<td>17</td>
<td>B</td>
<td>B</td>
<td>15</td>
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<tr>
<td>11.</td>
<td>M</td>
<td>21</td>
<td>U</td>
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<td>12.</td>
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<td>13.</td>
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<tr>
<td>15.</td>
<td>M</td>
<td>21</td>
<td>U</td>
<td>B</td>
<td>9</td>
</tr>
</tbody>
</table>

Results: 11 patient were male, 5 patients were female, average age 19.2 years, mean advancement 12.7 mm, mean follow-up 71 months
F = distraction as a final procedure, B = BSSO as an aditional procedure, Distraction obtained (mm), Follow-up period (months)

The distractors were activated after a latency period of 5 days and the maxilla advanced at the rate of 0.5 mm every 12 hours. A slight overcorrection was planned for all cases and the distraction continued until the planned position was obtained. After a consolidation period of 12 weeks the distractors were removed. No additional retention devices were used after removal of the distractors. In some patients additional elastics were placed to guide the maxilla into the final planned occlusion. Six patients required an additional bilateral sagittal split osteotomy (BSSO) and mandibular setback to accomplish the final planned occlusion (Tab. 1).

Standardized lateral cephalometric radiographs were obtained in natural head posture immediately before surgery (C1), 2 weeks after removal of distractors (C2) and finally a minimum of 60 months post-distraction(C3). Antero-posterior maxillary skeletal changes were evaluated by measuring the A-point-Nasion-Sella (ANS) angle at each interval. The pre-treatment, short term post-treatment, and long term post-treatment relationship between the mandible and the maxilla was evaluated by measuring the A– point-Nasion-B-point (ANB) angle on C1, C2 and C3.

(Tab. 1) All patients underwent pre- and postsurgical orthodontic dental alignment.

The surgical procedures were performed under general anesthesia using naso-endotracheal intubation. A circumvestibular incision was used to expose the maxilla. The osteotomy design was planned according to the aesthetic and functional requirements of each patient. To facilitate accurate placement of the internal distractors (Zurich, KLS Martin Group, Tuttlingen, Germany) the footplates were accurately bent and fixed with 2 screws on each side of the planned osteotomy line. The screws and appliances were then removed and the osteotomies completed. Following down fracture, the maxilla was mobilised and tested to confirm ease of advancement. The devices were then replaced using the previous screw holes as guide and finally fixed by placing all the other screws (25).

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To evaluate vertical maxillary changes a line was constructed through A-point perpendicular to Sella-Nasion (SN) (SN-A) and the distance measured in milimeters on C1, C2, and C3 (Fig. 1, Tab. 2). Vertical maxillary changes were measured by comparing the NS line -NL line angles at C1, C2 and C3 (Tab. 2). The incisor overjet was clinically measured at each interval, however this assessment was not considered relevant as in 6 patients mandibular setback procedures were performed and post operative orthodontic treatment would have influenced this assessment.

Results

All 15 patients demonstrated occlusal and esthetic improvement (Figs 2 and 3). Nine patients required only maxillary distraction to achieve the planned functional and esthetic result. However a large discrepancy between the maxilla and the mandible in 6 cases demanded an additional bilateral sagittal split mandibular setback procedure to limit maxillary distraction and improve post operative stability. The mean maxillary advancement achieved by distraction was 12.7 mm, (9 to 15 mm) measured on the distractor. The follow-up period after surgery ranged from 61 to 89 months (mean 71 months) (Tab. 1).

Cephalometric assessment (Tab. 2).

Cephalometric measurements (Fig. 1) revealed the following: the SNA angle increased by a mean of 12 degrees (C2–C1) (84–72 = 12 degrees) 2 weeks after removal of distractors. The maxilla relapsed slightly in the long term (mean 1 degree) (C2–C3)

Fig. 2. 17-years old female, unilateral CLP (patient 6, Tab. 1). a, b, c) cephalograms C1, C2, C3, d) patients’ profile before DO, e) patients’ profile 69 months after DO.
The anterior facial height increased slightly following the distraction (mean 2 degrees) (NS line-NL line angle increased from a mean of 8 degrees (C1) to a mean 10 degrees (C2)) and relapsed in the long term by 1 degree (mean 10 degrees (C2) to a mean of 9 degrees (C3)). Following the distraction the ANB angle increased from –7 degrees (C1) to +2 degrees (C2) with a slight relapse to +1 degree in the long term (C3) and can be considered clinically stable. The incisor relationship changed from a negative overjet to a positive overjet after distraction and remained positive in the long term however was not considered relevant as post operative orthodontics could have influenced this observation (Tab. 2). Some skeletal relapse occurred in all cases at point A in the horizontal and vertical direction however changes proved to be minimal (less than 2mm) and clinically insignificant (Tab. 2).

Discussion

Correcting the facial and occlusal deformities of CLP patients with severe maxillary hypoplasia remains challenging. The treatment planning, orthodontic treatment and orthognathic surgery for these patients are more complex than in non cleft patients for several reasons. One major factor is the presence of severe scarring of the lip and palate which limits maxillary development and renders surgical advancement of the maxilla difficult and often unstable. The choice between conventional orthognathic surgery and distraction osteogenesis (DO) depends mainly on the required amount of maxillary advancement. Several studies report that the amount of advancement and consequent post-operative relapses are the main problems (1, 2, 38, 16). Conventional Le Fort I osteotomy in the cleft patients seems to have a strong tendency to relapse.

![Fig. 3. 22-years old male, bilateral CLP (patient 4, Tab. 1). a, b, c) cephalograms C1, C2, C3, d) patients’ profile before DO, e) patients’ profile 79 months after DO.](image-url)
because of the necessity of large advancement, large discrepancies between bony segments, palatal scarring and tightness of the upper lip. Bone grafts in posterior defects between the maxillary tuberosity and the pterygoid plates to prevent relapse after maxillary advancement were recommended (8). Other authors used miniplate fixation along with bone grafting to improve stability (8, 17, 18). The disadvantages of autogenous bone grafting is the potential donor site morbidity, resorption or infection of the bone graft and tendency to relapse. There is a paucity of reports on the longterm stability of the maxilla following DO advancement in nongrowing patients (10). No randomized controlled trials have yet been conducted to compare DO and conventional surgery for CLP patients (3). Relapses following DO maxillary advancement in the long term are reported to be minimal, due to gradual advancement of the maxilla (1, 5, 6). A benefit of gradually advancement is histogenesis of the adjacent soft tissues as well as better perseverance of vascular supply. The term „large maxillary advancement“ means advancement more than 8 mm based on our criteria and also based on clinical evidence from several studies (9, 10). Cheung et al (3) reported that the magnitude of relapse tended to be correlated with magnitude of maxillary advancement. Precious DS (16) stated that maxillary advancement exceeding 10 mm is a limit to conventional orthognathic surgery for CLP patients, and distraction should then be considered as an option. Numerous reports have noted the instability of forward and inferior repositioning of the maxilla, mainly in CLP patients (17, 18, 19). Profit et al (18) reported that nearly two thirds of patients in their study had more than 2 mm skeletal relapse (20 % had more than 4 mm relapse). In a randomized clinical study Cheung et al (2) examined patients with CLP-related maxillary hypoplasia treated by means of conventional Le Fort I maxillary osteotomy or DO. Statistically better skeletal stability was noted in the distraction group. There are also several long-term clinical follow-up studies. Rachmiel et al (20, 21) obtained a mean advancement of 13.7 mm with relatively stable results (relapse rate 7.3 %) after a 2-year follow-up. Wiltfang et al (24) observed a mean relapse of only 6 %, 2 years after surgery. In a radiographic evaluation Kusnoto (11) and Rachmiell (21) found by means of a modified tomogram technique bone formation in the pterygoid region after maxillary distraction. The bone trabeculae could be seen six weeks after distraction. We recommend a prolonged period of consolidation (minimum 12 weeks) to improve stability. Rachmiel et al (20) published a study where advancement of the maxilla was combined with an increase in the vertical dimension. They reported an increase in vertical dimension of the face due to slight downward movement of the maxilla. This resulted in a clockwise and posterior rotation of the mandible. In their study the vertical changes were measured and compared. They compared the SN-mandibular plane angle and lower facial height before and after distraction. The oblique osteotomy and vector of distraction allows for advancement and slight vertical midface elongation (26). Results from our study support these conclusions.

During the consolidation period the distractor device serves as a retainer. It is covered by soft tissues and is well accepted by patients. A disadvantage of internal devices is the necessity of an additional surgery for removal. The results of our study showed that maxillary advancement in nongrowing CLP patients by the means of intraoral distraction devices is a viable alternative to conventional Le Fort I osteotomy.

References


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