Infant Orthopedics Has No Effect on Maxillary Arch Dimensions in the Deciduous Dentition of Children With Complete Unilateral Cleft Lip and Palate (Dutchcleft)


Objective: Evaluation of the effect of infant orthopedics on maxillary arch dimensions in the deciduous dentition in patients with unilateral cleft lip and palate.

Design: Prospective two-arm randomized controlled clinical trial with three participating cleft palate centers.

Setting: Cleft palate centers of the Radboud University Nijmegen Medical Center, Academic Center of Dentistry Amsterdam, and University Medical Center Rotterdam, the Netherlands.

Patients: Children with complete unilateral cleft lip and palate (n = 54) were included.

Interventions: Patients were randomly divided into two groups. Half of the patients (IO+) had a presurgical orthopedic plate until surgical closure of the soft palate at the age of 52 weeks; the other half (IO-) did not undergo presurgical orthopedics.

Mean outcome measures: Maxillary arch dimensions were assessed on dental casts at 4 and 6 years of age with measurements for arch width, arch depth, arch length, arch form, and the vertical position of the lesser segment. Contact and collapse were assessed also.

Results: There were no clinically significant differences found between IO+ and IO- for any of the variables.

Conclusions: Infant orthopedics had no observable effect on the maxillary arch dimensions or on the contact and collapse scores in the deciduous dentition at the ages of 4 and 6 years. Considering the Dutchcleft results to date, there is no need to perform infant orthopedics for unilateral cleft lip and palate patients.

KEY WORDS: arch dimension, cleft palate, collapse, deciduous dentition, infant orthopedics, multicenter, randomized clinical trial, treatment outcome

The effect of infant orthopedics (IO) on maxillary arch dimensions in unilateral cleft lip and palate (UCLP) has been studied for decades, but controversy regarding the effect of IO on the maxillary arch still exists. Advocates of IO claim that the presurgical orthopedic plate molds the alveolar segments into a better arch form and prevents the tongue from positioning in the cleft. In this way, the dentomaxillary development would improve (McNeil, 1954, 1956; Fish, 1972; Graf-Pinthus and Bettex, 1974; Hotz and Gnoinski, 1976, 1979; Weil, 1987; Gnoinski, 1990; Gruber, 1990; Kramer et al., 1994; Ball et al., 1995; Berkowitz, 1996; Mishima et al., 2000). Opponents of this therapy claim that lip surgery alone has the same effect and that the presurgical orthopedic plate is only an expensive appliance used to comfort the parents by starting treatment at the earliest moment possible (Pruzansky and Aduss, 1967; Huddart, 1974, 1979; Huddart and Huddart, 1985; Ross, 1987; Kramer et al., 1992; Mars et al., 1992; Shaw et al., 1992a, 1992b; Prahl et al., 2001).

Submitted July 2005; Accepted January 2006.

Address correspondence to: Dr. Kuijpers-Jagtman, Radboud University Nijmegen Medical Center, Department of Orthodontics and Oral Biology, 309 Tandheelkunde, PO Box 9101, 6500 HB Nijmegen, the Netherlands. E-mail a.kuijpers-jagtman@dent.umcn.nl.
Several studies describe the effect of IO on maxillary arch dimensions, but most are cohort studies in which UCLP patients treated with IO are compared with a control group of noncleft children, or case series that study changes in maxillary arch dimensions after IO treatment without comparison to a control group (Huddart et al., 1969; Robertson and Fish, 1975; Hotz and Gnoinski, 1979; Sarnäs et al., 1984; Kuijpers-Jagtman, 1985; Kramer et al., 1996; Friede and Katsaros, 1998; Kozelj, 2000). Kuijpers-Jagtman (1985) and Kozelj (2000) described that during IO the cleft narrowed in the anterior part, and the anterior arch depth increased less than in noncleft controls. Although the appliance was maintained after lip surgery, the cleft width decreased a considerable amount during the first 6 weeks after the operation. Later, a segmental displacement with the center of rotation at the tuberosities was found. At the age of 8 years the posterior arch width was not significantly different from the control group, but the anterior arch was narrower than in the control group.

As illustrated by the publications mentioned above, it cannot be concluded whether or not IO is an effective treatment approach. Therefore, a prospective randomized clinical trial was performed in three cleft palate centers in the Netherlands (i.e., the cleft palate centers of Nijmegen, Amsterdam, and Rotterdam) to investigate the effect of infant orthopedics with a passive plate in children with complete UCLP (Kuijpers-Jagtman and Prahl-Andersen, 2006). The first results, up to 1.5 years of age, showed that IO had a temporary effect on maxillary arch dimensions that did not last beyond surgical soft palate closure (Prahl et al., 2001). Also, IO could not prevent collapse of the maxillary arch (Prahl et al., 2003). No differences between IO+ and IO− could be shown in the occlusion at the ages of 4 and 6 years (Bongaarts et al., 2004). Data published by Konst et al. (2004) show the cost-effectiveness of the speech outcome at the age of 2.5 years. Speech therapists were asked to rate the speech quality of 10 IO+ children and 10 IO− children on a 10-point scale. The IO+ group had a significantly better rating for speech. The resulting cost-effectiveness ratio was 1041 euros for 1.34 points of speech improvement (Konst et al., 2003c, 2004). An evaluation of the speech data at the age of 6 has to be done to see whether the cost-effectiveness will change due to speech therapy from the ages of 3 to 6. More detailed findings on speech have been published elsewhere (Konst et al., 1999, 2000, 2003a, 2003b).

The purpose of the part of the trial presented here was to evaluate the effect of IO on maxillary arch dimensions in deciduous dentition in UCLP children, ages 4 and 6 years. The hypothesis tested was that the maxillary arch dimensions in the IO+ group were larger than in the IO− group and that less collapse occurred in the IO+ group compared with the IO− group.

**METHODS**

In a previous publication, a detailed description was given with respect to the experimental design, treatment assignment, treatment protocol, and operators (Prahl et al., 2001). A summary of the most important issues is given below.

The study was designed as a prospective two-arm randomized controlled clinical trial in the cleft palate centers in Nijmegen, Amsterdam, and Rotterdam, the Netherlands. The local ethical committees approved the study protocol. The inclusion criteria were complete UCLP infants born at term, both parents Caucasian and fluent in the Dutch language, and trial entrance within 2 weeks after birth. Exclusion criteria included soft tissue bands and other congenital malformations. Figure 1 shows the sample until the age of 6 with the reasons for exclusion from evaluation. When the parents agreed to participate in the study, they were asked to sign an informed consent. Between 3 and 6 months of age, all included children were assessed by the geneticist of their own cleft lip and palate team as being nonsyndromic.

**Treatment**

Half of the patients were treated with IO by means of passive plates until surgical soft palate closure (n = 27), and half did not receive a plate (n = 27). The plates were made on a plaster cast using compound soft and hard acrylic. The IO+
children had their plates adjusted every 3 weeks to guide the maxillary segments, by grinding at the cleft margins; maxillary growth and emergence of deciduous teeth indicated the necessity for a new plate. After surgical lip closure, the plate was replaced the same day. Checkups were planned every 4 to 6 weeks following lip surgery. The plate was maintained until soft palate closure. The IO group visited the clinic at 6 weeks, as well as before and after lip surgery and soft palate closure. In both groups, lip surgery was performed at the age of 18 weeks using the Millard technique. Soft palate surgery was performed at the age of about 52 weeks according to a modified von Langenbeck method. Hard palate repair was scheduled together with bone grafting. In the studied age period (until 6 years of age), other interventions were performed if indicated, including pharyngoplasty (n = 22), lip revision (n = 13), facial mask treatment (n = 1), plate to facilitate speech (n = 15), and closure of the anterior palate (n = 6). These interventions were equally distributed over the IO+ and the IO− group.

Data Acquisition

In order to evaluate arch dimensions, impressions were taken at ages 4 and 6. Plaster casts were fabricated. To eliminate bias, all models were duplicated and trimmed in the same way. In this way the examiners were not able to identify a patient or a cleft palate center.

The maxillary casts were analyzed three-dimensionally using the Reflex Microscope (Reflex Measurement, Somerset, UK; Speculand et al., 1988; Drage et al., 1991; Seckel et al., 1995). First, reference points were marked on the casts (Fig. 2). To calculate interexaminer error in marking casts, two observers marked the reference points. To be able to mark the casts blindly, both original models and duplicated casts were used. Points L′, P′, T′, I′, C′, and C′ correspond with the points used by Prahl et al. (2001), who evaluated the maxillary arch dimensions of the Dutch cleft children up to the age of 78 weeks. The other points were used as proposed by Movers (1976), Derijcke et al. (1994), and Heidbüchel and Kuijpers-Jagtman (1997). Second, the reference points were digitized by the two observers. With the digital coordinates, the distances and angles shown in Table 1 could be calculated.

In addition, the casts were examined for presence of contact or collapse of the alveolar segments, as was done by Prahl et al. (2003), a method comparable to Pruzansky and Aduss (1964). The method (Prahl et al., 2003) is an ordinal scoring system. Contact was scored as absent (score 0) or present (score 1), and collapse was scored as absent (score 0), slight (score 1), moderate (score 2), or severe (score 3). From these scores, four groups were formed: NC-NO = no contact and no overlap; NC-O = no contact and overlap; C-NO = contact and no overlap; C-O = contact and collapse. Four observers scored all models for contact and for collapse.

FIGURE 2 Explanation of the reference points used: I—The top of the interdental papilla between the (deciduous or permanent) central incisors; L′—Lesser segment margin, where the continuation of a line marking the crest of the ridge turns from the oral side to the nasal side at the anterior end of the segment; P′—The larger segment margin, where the continuation of a line marking the crest of the ridge turns from the oral side to the nasal side at the anterior end of the segment (P′ = L′ when the segments touch each other); I1/I1—Centroid of the central incisor; I2/I2—Centroid of the lateral incisor; Ce/Ce—Centroid of the canine; P1/P1—Centroid of the first deciduous molar or first premolar; P2/P2—Centroid of the second deciduous molar or second premolar; C(5)/C(5)—The most occlusal point of the cusp of the canine; P(5)/P(5)—The most occlusal point of the palatal cusp of the first deciduous molar or first premolar; P(5)/P(5)—The most occlusal point of the palatal cusp of the second deciduous molar or second premolar; C/C—The top of the interdental papilla between the (deciduous or permanent) canine and first premolar/first deciduous molar; T, T—Tuberosity points, at the junction of the crest of the ridge with the outline of the tuberosity; M—Midpoint between T and T′; Occl—Plane formed by the palatal cusp of the M1 on both sides and the cusp of the canine on the nonleft side. Centroid is the intersection of four points (X). The middle of (1) and (2) is A, the middle of (3) and (4) is B; the centroid is the midpoint of A and B. (1) distal midpoint: the point on the distal point of the tooth, midway between the buccal and lingual surfaces; (2) mesial midpoint: the point on the mesial point of the tooth, midway between the buccal and lingual surfaces; (3) most lingual point: at the lingual fissure location for the permanent molars and the second deciduous molars; for the premolars, first deciduous molars, canines and incisors, it is the most lingual point on the lingual surface; (4) most buccal point: at the buccal fissure location for the permanent molars and the second deciduous molars; for the premolars, first deciduous molars, canines, and incisors, it is the most buccal point on the buccal labial surface.
3. A kappa value between .81 and 1.00 indicates a very good agreement, whereas a kappa between .61 and .80 indicates good agreement. The measurements in the vertical direction have low reliability. Therefore, C(5)-occl, P1(5)-occl, P2(5)-occl were excluded from further analysis. Two other measurements with low reliability were maintained to make comparisons possible with the other measurements and research done by Prahl et al. (2001).

### Treatment Effect

Mean values and standard deviations for all variables for both ages are given in Table 4. Measurements at 0 to 2 weeks were performed.

### TABLE 2 Sample Characteristics; Some Variables Are Presented in Percentiles Because of Skewness (P10, P50, and P90)

<table>
<thead>
<tr>
<th>Variable</th>
<th>IO+ (n = 27)*</th>
<th>IO− (n = 27)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gender: girl/boy (n)</td>
<td>20/7</td>
<td>21/6</td>
</tr>
<tr>
<td>Side of cleft: left/right (n)</td>
<td>17/10</td>
<td>18/9</td>
</tr>
<tr>
<td>Patients per center: 1/2/3 (n)</td>
<td>7/11/9</td>
<td>7/10/10</td>
</tr>
<tr>
<td>Age, 4-year casts (y)</td>
<td>mean: 4.0</td>
<td>mean: 4.0</td>
</tr>
<tr>
<td>range: 3.7–4.3</td>
<td>range: 3.8–4.5</td>
<td></td>
</tr>
<tr>
<td>Age, 6-year casts (y)</td>
<td>mean: 6.0</td>
<td>mean: 6.0</td>
</tr>
<tr>
<td>range: 5.8–6.2</td>
<td>range: 5.9–6.4</td>
<td></td>
</tr>
<tr>
<td>Age at trial entrance (d)</td>
<td>0 3 7</td>
<td>1 6 13</td>
</tr>
<tr>
<td>Birth weight (g)</td>
<td>2660 3350 4020</td>
<td>2920 3600 4280</td>
</tr>
<tr>
<td>Cleft width at birth (mm)</td>
<td>9.5 12.5 14.4</td>
<td>8.6 12.4 16.4</td>
</tr>
<tr>
<td>Age lip repair (d)</td>
<td>117 127 142</td>
<td>117 125 138</td>
</tr>
<tr>
<td>Age soft palate closure (d)</td>
<td>355 375 438</td>
<td>301 367 389</td>
</tr>
</tbody>
</table>

* IO+ = patients with presurgical orthopedic plate; IO− = no orthopedic plate.

### RESULTS

At intake 54 patients participated in the study. An overview of the sample characteristics is given in Table 2. Two IO+ children hardly used the plate; in one case, the plate was mistakenly worn until 78 weeks. These children remained in the IO+ group according to the intention-to-treat principle. The mean duration of IO was 50 (± 16 weeks) SD. The flow diagram in Figure 1 shows the reasons for nonevaluation.

### Measurement Reliability

The interexaminer errors and the reliability coefficients for the interexaminer agreement for the maxillary measurements are shown in Table 3. The kappas for interexaminer agreement of the contact and the collapse scores also are shown in Table 3. A kappa value between .81 and 1.00 indicates a very good agreement, whereas a kappa between .61 and .80 indicates good agreement. The measurements in the vertical direction have low reliability. Therefore, C(5)-occl, P1(5)-occl, P2(5)-occl were excluded from further analysis. Two other measurements with low reliability were maintained to make comparisons possible with the other measurements and research done by Prahl et al. (2001).
and at 78 weeks of age from an earlier publication on the same sample (Prahl et al., 2001) also are shown in Table 4. No significant differences were found between the IO− and IO+ groups at age 4 and at age 6, except for I-TT*, and angle M-T-C(5) at the age of 4 years and angle M-T-C(5) at 0 to 2 weeks. The arch depth (I-TT*) at the age of 4 years was larger in the IO− group; at 0 to 2 weeks of age and at 78 weeks (Prahl et al., 2001). The mean severity score for collapse at the age of 4 years was 1.32 (±0.95 SD) for the IO− group and 1.05 (±0.50 SD) for the IO+ group. The severity scores for the age of 6 years were 1.72 (±0.98 SD) for IO− and 1.39 (±1.06 SD) for IO+. The mean score for contact at 4 years of age was 0.73 for IO− and 0.78 for IO+. At the age of 6 years IO+ scored a mean of 0.78 and IO−, 0.90.

There were no significant differences found for collapse and contact between IO+ and IO− at the ages of 4 and 6 years. Further, looking at IO+ and IO− for the combined groups of no overlap (NC-NO and C-NO) compared with the combined

### Table 4: Number (n†), Means, Standard Deviations, and Confidence Intervals (CI) of the Measured Variables (in mm or Degrees) for IO+ (Bold) and IO− (Normal) at the Age of 0 to 2 Weeks, 78 Weeks, and 4 and 6 Years;* Variables of the Ages 0 to 2 Weeks and 78 Weeks are Copied From Prahl et al. (2001); See Figure 2 for Definitions

<table>
<thead>
<tr>
<th>Variable</th>
<th>0–2 wk</th>
<th>78 wk</th>
<th>4 y</th>
<th>6 y</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>n Mean (SD)</td>
<td>n Mean (SD)</td>
<td>[95% CI]</td>
<td>n Mean (SD)</td>
</tr>
<tr>
<td>Ce-Ce'</td>
<td>22 33.4 (2.0)</td>
<td>18 36.0 (3.0)</td>
<td>21 39.91 (2.27)</td>
<td>18 42.63 (2.99)</td>
</tr>
<tr>
<td>C-C'</td>
<td>24 32.7 (2.3)</td>
<td>20 29.8 (3.0)</td>
<td>22 27.58 (2.28)</td>
<td>22 27.16 (2.70)</td>
</tr>
<tr>
<td>I-TT'</td>
<td>22 25.9 (2.8)</td>
<td>18 32.0 (2.4)</td>
<td>19 30.14 (2.62)**</td>
<td>18 33.36 (2.54)</td>
</tr>
<tr>
<td>L'-T'</td>
<td>22 37.38 (3.65)</td>
<td>22 39.31 (4.58)</td>
<td>22 38.37 (4.88)</td>
<td>22 39.65 (4.89)</td>
</tr>
<tr>
<td>P'-T</td>
<td>22 54.46 (4.87)</td>
<td>22 55.35 (3.89)</td>
<td>23 53.37 (3.33)</td>
<td>22 56.24 (4.97)</td>
</tr>
<tr>
<td>Total arch length</td>
<td>22 65.5 (5.6)</td>
<td>12 82.0 (4.4)</td>
<td>22 91.85 (7.74)</td>
<td>22 94.66 (7.49)</td>
</tr>
<tr>
<td>Angle M-T-C(5)</td>
<td>22 82.5 (3.9)**</td>
<td>18 77.4 (5.1)</td>
<td>19 40.55 (3.05)*</td>
<td>18 43.17 (3.98)</td>
</tr>
<tr>
<td>Angle M'-T-C(5)*</td>
<td>23 56.0 (3.8)</td>
<td>14 80.1 (5.7)</td>
<td>21 43.37 (3.85)</td>
<td>19 43.46 (6.98)</td>
</tr>
<tr>
<td>Angle P'-C(5)-T</td>
<td>22 95.0 (5.4)</td>
<td>18 86.2 (3.8)</td>
<td>19 72.58 (4.61)</td>
<td>18 69.86 (4.39)</td>
</tr>
</tbody>
</table>

† n may vary because of incidental missing values (e.g., maxillary tuberosities not visible on the model, shedding teeth).

* 0.5 ≥ p > 0.1; ** 0.1 ≥ p > .001. Differences between IO+ and IO− were tested with t tests. The level of significance is indicated with p values.
overlap groups (NC-O and C-O), no significant differences were found.

**DISCUSSION**

To compensate for shortcomings of earlier studies, the design of the present study was a prospective two-arm randomized controlled clinical trial (Prahl et al., 2001). The number of patients involved in the study decreased from 54 to 45 in the age groups of 4 and 6 years, but the number remained larger than in most published studies. Sarna¨s et al. (1984) and Mishima et al. (1996, 2000) both compared IO+ with IO− patients. Mishima et al. (2000) sampled 12 IO+ children and 8 IO− children. Sarna¨s et al. (1984) compared 24 IO+ patients with 18 IO− patients. However, neither study was designed as a randomized clinical trial.

The dental casts were digitized by means of a Reflex Microscope (Reflex Measurement). As is known from Drage et al. (1991), a Reflex Microscope (Reflex Measurement) is best used by trained observers. However, untrained observers can use the microscope well after some practice. As was also found in this study, errors were found to be greatest in the z-axis, along the axis of the eye. Errors were also rather high at the margins of the segments. The study of Speculand et al. (1988) shows that it is possible to generate reproducible results for redigitization with an intraexaminer error of less than .15 mm for linear measurements. This was not found for all measurements in this study. The measurement errors found in this study for landmark positioning (marking points) are comparable to those reported by Seckel et al. (1995). The errors for the measurements of contact and collapse done in this study are comparable to those reported by Prahl et al. (2001).

A few significant differences between IO+ and IO− are shown in Table 4, but they do not show a consistent pattern over the different periods. All significant differences faded away at the age of 6 years. Therefore, the few inconsistent significant influences at the age of 4 years may be either temporary or falsely significant, probably due to the large number of tests. The confidence intervals mentioned in Tables 4 and 5 are not extremely large. Only the angle P-C(5)-T and the contact-collapse variables show a large interval, which could point to a type II error. These variables should be interpreted with caution.

Many studies have been published about the effect of IO on maxillary arch dimensions or on collapse or contact of the alveolar segments, but most studies are nonrandomized. The two studies with the best research design were published by Mishima et al. (1996, 2000) and Sarna¨s et al. (1984). Mishima et al. (1996) reported that in a quasi-randomized trial at the age of 18 months, the Hotz plate seemed to stimulate growth of the segments, could prevent collapse of the segments after lip closure, and resulted in less steepness of the segments, combined with more forward migration of the lesser segment toward the larger segment. In 2000, Mishima et al. reported that at the age of 4 years, the width of the palate was larger in the group treated with Hotz plates than in the group treated without plates. Sarna¨s et al. (1984) followed a group of 24 IO+ children and 18 IO− children in a retrospective two-group cohort study and evaluated them at the ages of 3 and 19 months. All patients had lip surgery at the age of 3 months and palatal surgery at the age of 19 months. In the IO+ group the plate was worn until the moment of palatal surgery. The IO+ group had larger transverse dimensions and less rotation of the greater segment. Although the first results in Dutchcleft
showed that IO had a temporary effect on the maxillary arch dimensions, this did not last beyond surgical soft palate closure (Prahl et al., 2001). Also, no significant differences were found in Dutchcleft in width measurements at the ages of 4 and 6 years. In addition, the results of Dutchcleft are not really comparable to the Mishima and Sarnas studies because those studies did not use a randomized study design.

IO could not prevent collapse of the maxillary arch (Prahl et al., 2003). Table 5 shows that, over time, the number of children with contacting and overlapping segments increases for both groups, IO+ and IO−. At birth all children have no contact and no overlap between the alveolar segments, and at the age of 6 years the majority (97.8%) has contact, collapse, or both. This means that the impact of lip surgery and palatal surgery is much greater than the effect of IO.

The results of this part of the Dutchcleft study are in agreement with the other findings of this trial to date. Except for a small but significant improvement in speech development, no positive or negative influence of IO was found in the Dutchcleft study (Severens et al., 1998; Konst et al., 1999, 2000, 2003a, 2003b, 2004; Prahl et al., 2001, 2003, 2005; Bongaarts et al., 2004; Kuijpers-Jagtman and Prahl-Andersen, 2006).

**Conclusion**

IO did not influence the maxillary arch dimensions, the collapse, or the contact between the alveolar segments at the ages of 4 and 6 years. Therefore, from the orthodontic point of view, there is no need to perform IO in children with UCLP.

**References**


Huddart AG, Huddart AM. An investigation to relate the overall size of the maxillary arch and the area of palatal mucosa in the cleft lip and palate cases at birth to the overall size of the upper dental arch at five years of age. *J Craniofac Gen Dev Biol.* 1985;suppl 1:89–95.


