Chapter 5

Maxillary arch width in unoperated BLCA and BCLP subjects
Summary

In this chapter a study is described on maxillary dental arch width of unoperated adult subjects with a bilateral cleft lip and alveolus (BCLA) and a bilateral cleft lip, alveolus and palate (BCLP). The sample consisted of 18 unoperated BCLA subjects and 13 unoperated BCLP subjects. A non-cleft sample (n=24) from the surrounding population served as controls. Alginate impressions of the participating subjects were made and the dental casts were digitized three-dimensionally, using an industrial coordinate measuring machine. Maxillary transversal dimensions were calculated, using the cusps of the teeth as reference points.

The results show that the transversal maxillary arch dimensions in the BCLA group were comparable to the controls. Only at the canine level a significant difference was found. The intercanine distance, which is close to the alveolar cleft, was 4.3 mm (SE 1.4) smaller in the BCLA-group in comparison with the control group (p=0.002). This was also expressed by the upper arch ratio showing that in BCLA the mean intercanine width is 56.8% of the intermolar width, while this value is 64.2% (p=0.0009) in the control group. In BCLP a comparable pattern was found. At the canine level the mean transversal width was even 7.2 mm (SE 1.9) smaller in comparison with the control group (p=0.0003), while all other transversal dimensions were not significantly different from the controls. The upper arch ratio was 51.8% in the BCLP group and 64.2% in the control group and this difference was significant (p=0.0004).

We may conclude that the cleft as a congenital malformation has an intrinsic, although limited effect on the dento-alveolar development of the maxilla and only in the canine region. These results are important for our understanding of the iatrogenic effects of the surgical repair of the lip and/or palate, which might eventually lead to the development of more appropriate surgical techniques and better orthodontic management.
5.1 Introduction

The common orofacial clefts can be divided into three main categories: 1) isolated cleft lip and/or alveolus, 2) combined cleft lip, alveolus and palate, and 3) isolated cleft palate. Each category can be subdivided into complete and incomplete clefts, and unilateral or bilateral clefts. Besides these types of clefts there are rare types like median or oblique facial clefts (Tessier, 1976). The incidence varies considerably among races and depends also on the type of cleft (Vanderas, 1987; Calzolari et al, 2004; Shaw, 2004). In about 10 to 20% of the newborn children with a cleft the deformity is bilateral (Abyholm, 1978; Jensen et al, 1988; Kriens, 1989).

Bilateral cleft lip and alveolus (BCLA) is a rather mild form of cleft. This might be the reason that in most studies on clefts BCLA patients are mentioned only incidentally. In the cephalometric study of Ortiz-Monasterio et al (1974) a sample of 450 patients with different types of clefts was registered. Only 33 (7,3%) of them showed a BCLA, but the majority of these patients had undergone surgery prior to the registration. Mars et al (1990) studied 423 young and adult cleft patients with different types of clefts, but only 10 (2.4 %) showed a BCLA and all of them already had undergone surgery before registration. Lekkas et al (1997) analyzed a sample of 267 unoperated adult cleft patients. They found only 18 individuals with BCLA or 6,8% of the total sample. Except for the latter publication, to our knowledge there are no other publications concerning transversal measurements of the maxilla either on early operated or unoperated adult BCLA patients. On the other hand also in BCLA patients at least minor aberrations may be expected in the cleft region as was also found in UCLA (see chapter 5). Moreover the lateral incisor is more frequently absent in the BCLA group (Lekkas et al, 2001) and consequently there could be more lateral compression in the canine area to expect.

Bilateral complete cleft lip, alveolus and palate is the severest type of the common orofacial clefts. In spite of the relatively low incidence of BCLP a lot of studies have been devoted to BCLP because of the severity of the implications for the growth and final development of the facial skeleton and the dentition. After maturity almost all early operated patients with BCLP show a severe compression of the maxillary lateral segments and at least a moderate skeletal deficiency of the maxilla (Hagerty et al, 1964), for which extensive orthopaedic and/or orthodontic treatment is necessary. The extent of the skeletal deficiency, however, is difficult to predict (Kramer et al 1996,
Maxillary arch width in unoperated BCLA and BCLP subjects


Not much has been published concerning dental casts analysis in BCLP and the dental arch width is often rather estimated than really measured (Hayward, 1983). Nystrom and Ranta (1990) compared CP, UCLP, BCLP and non-cleft controls at the age of 3 years after palatal surgery at two different ages. They found that maxillary arch dimensions were smaller in the cleft groups as compared to the controls, but the timing of palatal closure did not affect arch dimensions in the cleft group. This is in contrast to a later study that found that timing might have an influence i.e. upper and lower intercanine width tended to be smaller after early hard palate closure, but only at 3 years of age (Melissaratou and Friede, 2002). In a longitudinal study on 22 BCLP patients from 3 up until 17 years of age Heidbüchel and Kuijpers-Jagtman (1997) found, in spite of the prolonged orthodontic treatment, a significantly smaller maxillary dental arch width in BCLP patients compared to the control group. Already from the age of three years on the arch width was smaller in the early operated BCLP children. This reduction of the arch width was most pronounced in the canine area. At the age of 17 years the reduction of the intercanine width was almost 10 mm in the BCLP group as compared to the non-cleft controls. At the level of the first molar the maxillary width was 8 mm smaller in BCLP subjects.

Studies on unoperated adult BCLP patients are rare. Besides the low incidence of BCLP the rudimentary medical and dental care, under-nourishment, endemic diseases like TBC, malaria tropica and respiratory tract diseases reduce the life expectation of patients with clefts in general, and more particular those with the severe types of clefts. Therefore there are only a few publications analyzing more than 10 adult unoperated BCLP patients. These patients are mostly assessed by means of lateral cephalograms only (Ortiz-Monasterio et al, 1959; Bishara et al, 1985; Da Silva Filho et al, 1997). Analysis of the dental arch width by means of dental casts is even more scanty. Bishara et al (1985) found that the maxillary intercanine width in BCLP patients was smaller compared to UCLA and UCLP patients in their sample. Also arch collapse was more pronounced in the BCLP group as
compared to the UCLP and UCLA group. Sidhu et al (1982) analyzed 10 patients with unoperated or late repaired BCLP and compared them with sufficient individuals from the surrounding population. They found that the arch width in the canine and first premolar region was not significantly different from the controls, but the intermolar width in the cleft group was significantly smaller compared to the control group. Da Silva Filho et al (1997) analyzing the maxillary dental arch morphology of 31 unoperated adult BCLP patients, found even more compression of the dental arch as in their sample arch width was not only smaller at the molars but also in the premolar and canine region compared to non-cleft individuals. This is in contrast to Motohashi et al (1994), who analyzed 24 unoperated BCLP subjects by means of AP cephalograms and they found no difference in non operated BCLP and non operated UCLP.

The study of operated BCLP patients has its limitations related to the collection as well as the evaluation of the sample. Many studies on BCLP concern only very young patients that have nearly always undergone extensive orthodontic treatment (Heidbüchel et al, 1994; Jain and Krogman, 1983). Therefore it is not possible to get insight into the final development of the maxilla of early operated BCLP patients without the influence of orthodontics. Moreover it is not really possible to predict the final development of a child’s face by studying facial growth at a very young age as due to the progressive and accumulative nature of the defect by the time the pubertal growth spurt is completed the facial deformity is often more severe. By analyzing facial growth and maxillary arch dimensions of unoperated adult BCLP subjects it is possible to get more insight into the real intrinsic growth potential of the maxillary structures separating this from the effect of surgical and orthodontic treatment. Therefore the purpose of the study described in this chapter is to investigate the maxillary dental arch width in a sample of unoperated adult BCLA and BCLP subjects and to compare the results with a non-cleft control group from the same population.

5.2 Materials and methods

Transversal maxillary arch dimensions of patients with unoperated BCLA and BCLP were measured on dental casts. The sample consisted of 18 unoperated adults with a bilateral cleft lip and alveolus (BCLA) and 13 unoperated adults with a complete bilateral cleft lip and palate (BCLP). The
control group consisted of 24 randomly selected non-cleft individuals from the surrounding population (see table 2.3 and 2.4). All patients were documented with dental casts, cephalograms and standard intra-oral and extra-oral photographs. After collection of the preoperative data all cleft patients were treated surgically in a single surgical procedure.

The dental casts were digitized three-dimensionally using an industrial coordinate measuring machine (Zeiss Numerex; Carl Zeiss®; Stuttgart, Germany). With this bridge-type system accurate single-point data acquisition is possible by using a touch probe. The linear accuracy is up to 0.002 mm.

For every molar, 2 points were recorded: the tip of the distobuccal cusp and the tip of the mesiobuccal cusp. In the case of abrasion of a cusp, the centre of the abraded cusp was used as the reference point. For the premolars and the canines, the tip of the buccal cusps was recorded. Between corresponding points at the right and left side the following distances in the maxilla were calculated:

- 171 - 271 distance between distal cusps of the right and left second molar.
- 172 - 272 distance between mesial cusps of the right and left second molar.
- 161 - 261 distance between distal cusp of the right and left first molar.
- 162 - 262 distance between mesial cusp of the right and left first molar.
- 151 - 251 distance between buccal cusps of the right and left second premolar.
- 141 - 241 distance between buccal cusps of the right and left first premolar.
- 131 - 231 distance between cusps of the right and left canine.

The upper arch ratio (%) was calculated as 131-231 distance / 161-261 distance * 100.

Means and standard deviations were calculated for all variables. Occasionally in the cleft groups models were missing or teeth were extracted. Therefore the numbers differ per variable. The t-test was used to determine whether the mean values of the cleft groups showed significant differences from each other and from the control group. The level of significance was set at p<0.05.
5.3 Results

Table 5.1 shows means and sd (in mm) for the transversal maxillary arch dimensions in the canine region, and the premolar / molar area in adult unoperated BCLA and BCLP subjects as compared to the control group. Box-whisker plots of the maxillary arch width are shown in figure 5.1.

From the table is obvious that the transversal maxillary arch dimensions in the BCLA group were comparable to the controls. Only at the canine level a significant difference was found. The intercanine distance, which is close to the alveolar cleft, was 4.3 mm (SE 1.4) smaller in the BCLA-group as compared to the control group (p=0.002). This was also expressed by the upper arch ratio showing that in BCLA the mean intercanine width is 56.8% of the intermolar width, while in the control group this value is 64.2% (p=0.0009).
Table 5.1  Mean and SD (in mm) of transversal maxillary dental arch dimensions for unoperated BCLA and BCLP and the non-cleft controls. For explanation of the variables see Materials and methods section.

**COMPARISON OF MAXILLARY TRANSVERSAL ARCH DIMENSIONS (IN MM) BETWEEN UNOPERATED ADULT BCLA, BCLP AND THE CONTROL GROUP**

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<th>N</th>
<th>MEAN</th>
<th>SD</th>
<th>GROUP</th>
<th>N</th>
<th>MEAN</th>
<th>SD</th>
<th>DIFF</th>
<th>S.E</th>
<th>t-VALUE</th>
<th>p-VALUE</th>
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**DIFF** = difference in maxillary arch width and upper arch ratio between BCLA, BCLP and control.

**S.E** = standard error of the difference

**UPPER ARCH RATIO** = 131-231 distance / 161-261 distance * 100 (%)
In BCLP a comparable pattern was found. At the canine level the mean transversal width was even 7.2 mm (SE 1.9) smaller as compared to the control group, while all other transversal dimensions were not significantly different from the controls. The upper arch ratio was 51.8% in the BCLP group and 64.2% in the control group and this difference was significant (p=0.0004).

**Figure 5.2** Box-whisker plot of maxillary arch width (in mm) in unoperated adult UCLA and BCLA subjects compared to the control group.

7-db = 171-271 distance; 7-mb = 172-272 distance; 6-db = 161-261 distance; 6-mb = 162-262 distance; 5-b = 151-251 distance; 4-b = 141-241 distance; 3-b = 131-231 distance.

In the box-whisker plots, shown in figure 5.2 and 5.3, the data for BCLA and BCLP are compared to the values for UCLA and UCLP as presented in chapter 4. No significant differences were found between unilateral and bilateral clefts.
Maxillary arch width in unoperated BCLA and BCLP subjects

**Figure 5.3** Box-whisker plot of maxillary arch width (in mm) in unoperated adult UCLP and BCLP subjects compared to the control group. 7-db = 171-271 distance; 7-mb = 172-272 distance; 6-db = 161-261 distance; 6-mb = 162-262 distance; 5-b = 151-251 distance; 4-b = 141-241 distance; 3-b = 131-231 distance.

5.4 Discussion and conclusions

Although some significant differences for maxillary arch width were found between the BCLA and BCLP group and the non-cleft controls, the results should be interpreted with caution as the sample size in our study was rather small. Of the total sample of 267 unoperated subjects only 18 (6.8%) had a BCLA, while 13 (4.9%) had a BCLP. Especially for BCLP these percentages are lower than could be expected based on the incidence for BCLP as reported in the literature (Kriens, 1989). Presumably the life expectation of patients with a complete BCLP in remote areas of Indonesia is lower compared to patients with milder types of clefts. A regional ENT survey on a sample of 25 UCLP patients by Hardjowasito (1989) revealed that 80% of the patients had hearing loss and radiographically sclerotic mastoid was found
indicating repeated middle ear infection. It has also been shown that in children with BCLP more peroperative respiratory complications have to be expected than in children with less severe clefts (Takemura et al, 2002). The fact that in our BCLP sample no individuals were found older than 40 year of age supports the assumption that patients with BCLP in less developed countries die earlier than patients with a mild type of cleft.

In the present study both in BCLA and BCLP arch widths were essentially normal except for the canine region where the arch width was significantly smaller (-4.3 and -7.2 mm, respectively) compared to the control group. This smaller intercanine width may also be partly explained by the frequent absence of the lateral incisors (Lekkas et al, 2001). Because no publications are available dealing with transversal measurements of dental arch width of early operated adult BCLA individuals, a comparison between early operated and unoperated adult BCLA is not possible. Maxillary transverse measurements on AP head films in a group of 24 children with BCLA, who all had there primary lip and palate surgery within the first year of life, showed that at the age of 5.8 years the width of the maxilla in BCLA was comparable to that of patients with a bilateral cleft palate only, but data on a non-cleft control sample was not reported (Liao et al, 2002). The results of the present study are in accordance with the studies of Sidhu et al (1982) and Bishara et al (1985). Especially the observation of Bishara et al (1985) that even in adult unoperated BCLP patients the developmental disturbances are rather limited to the vicinity of the cleft agrees with the findings in our study. In the BCLP sample (n=33) of Da Silva Filho et al (1997, 1998), however, the entire maxillary dental arch seemed to be affected by the presence of the cleft as the arch width in the lateral segments was also smaller than in their control group, while the intercanine width was even more restricted (-10 mm) than in our sample. However, in an earlier study from the same research group on AP cephalograms, in which at least partially the same BCLP sample (n=24) was analyzed, no significant difference in maxillary width at the skeletal level was found between clefts and controls (Motohashi et al, 1994), although the unoperated BCLP group demonstrated a significantly larger interorbital distance, nasal width, bicondylar width, and bigonial width than the non-cleft group. The reason why in our study the intrinsic deformity was much more limited than in the study of Da Silva Filho et al (1997) is difficult to explain. The differences could be partly attributed to the small sample size and a different racial background, but it also might be possible that due to the poor health care infrastructure or
undernourishment of our sample the more severe forms of BCLP have not reached adulthood. Additional research would be necessary in order to clarify these contradictions.

From the findings of the present and previous investigations on unoperated adult individuals, it could be summarized that there are differences in the dento-alveolar development of patients with different bilateral cleft types compared to non-cleft individuals. We may conclude that the cleft as a congenital malformation has an intrinsic, but limited effect on the dento-alveolar development of the maxilla only in the canine region. These results are important for our understanding of the iatrogenic effects of the surgical repair of the lip and/or palate, which eventually might lead to the design of more appropriate surgical techniques and better orthodontic management of these cases. The present findings in this relatively limited group of persons with bilateral clefts indicate that there is still a need to examine in detail a larger number of persons with untreated bilateral clefts. Ideally, these adult persons should be followed up after their operation to get more insight into the effect of surgery itself without interference of growth.

5.5 References


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