Chapter 7

General discussion and conclusions
Summary

In this chapter the results, as reported in this thesis, are discussed. The study focussed on maxillary and mandibular dental arch width, and on the width and elevation of the palatal shelves in patients with different types of unrepaired clefts in comparison to non-cleft controls from the same racial population. From the findings it can be concluded that there is an intrinsic or functional effect of the cleft itself on dentofacial development of the maxilla and to a lesser extend on the mandible. Each type of cleft has its own intrinsic characteristic dental arch form. However, compression of the maxilla is limited to the vicinity of the cleft in the anterior region. When the palate is involved, the palatal shelves seem also to be deficient and positioned more cranially. These findings support the hypothesis that the intrinsic deviation, that is already present, might become clinically manifest due to surgery.

There is a continued need to investigate the effects of different surgical procedures on maxillofacial growth and dento-alveolar development. Supplemental to that, the study of unrepaired clefts could teach us more about the natural history of clefts. Special attention should be given to bilateral cleft conditions as very few studies with a sufficient sample size have been published, neither about repaired nor about unrepaired clefts. Short-term and long-term follow-up of adult patients operated after facial growth had ceased, could provide more insight into the effect of surgery itself, excluding growth as a confounding factor. It is obvious that careful attention should be given to the anatomical deviating palatal shelves and the consequences for the surgical reconstruction of the palate in order to modify present surgical techniques for achieving better long-term dentofacial development in patients with clefts.
Chapter 7

7.1 Introduction

From clinical observation it is clear that patients with repaired complete clefts often have a facial and dento-alveolar morphology that differs from individuals who were not affected by orofacial clefting when they had reached maturity. The aetiology of this deviating dentofacial appearance is still partly unknown. Firstly, there may exist a different morphogenetic growth pattern, intrinsic to the cleft itself. Secondly, adaptive changes could be responsible due to the mechanical presence of the cleft or lack of continuity of the tissues involved. Thirdly, iatrogenic factors could also influence subsequent growth (Kuijpers-Jagtman, 1995). Besides animal research and the clinical study of growth in repaired clefts, it has long been recognized that the study of adult patients with unrepaired clefts, could also provide more insight into the ‘natural history’ of a cleft (Ortiz-Monasterio et al, 1959; Mestre et al, 1960; Bishara et al, 1986; Millard, 1980; Mars and Houston, 1990). The present study focussed on maxillary and mandibular dental arch width, and on the width and elevation of the palatal shelves in patients with different types of untreated clefts in comparison to non-cleft controls from the same racial population.

7.2 Methodology and design of the study

Between 1986 and 1997 we collected dental casts of 267 adult individuals with 4 types of unrepaired clefts: UCLA, UCLP, BCLA and BCLP. These data were collected during 9 cleft missions to Nusa Tenggara Timur and Timor Timur. The project was set-up with four main goals:

- To provide free surgery for children and adults with clefts who could not afford surgery,
- To provide training for Indonesian doctors to foster self-sufficiency,
- To collect scientific data for the study of the natural history of clefts,
- To investigate maternal dietary zinc deficiency in association with the occurrence of orofacial clefts in their offspring and to study the effect of supplemental oral zinc on the prevention of clefts.

The latter aim is beyond the scope of this thesis and the results have been published elsewhere (Hidayat et al, 1997).

By 1997 we had operated almost 2400 patients, giving them a normal appearance. It should be emphasised that we have always attempted to treat
all individuals that presented themselves with clefts. That means that also a lot of children were operated who did not take part in this study as they did not meet the age criteria for inclusion. Records were taken of all subjects with second permanent molars in occlusion (n=337), who appeared to have a UCLA, BCLA, UCLP or BCLP malformation on clinical inspection and who were operated after record-taking. In the final analysis 70 patients (21%) had to be excluded as they did not meet the inclusion criteria as described in chapter 2. The remaining 267 individuals with unrepaired clefts form the sample of this study. Looking at the reasons for exclusion (see paragraph 2.3.1) selection bias does not seem to play a role in the final outcome of this study.

The 6 most extensive projects on adult unoperated cleft patients in third world countries, that have been previously reported in the literature, were located in Mexico (Ortiz-Monasterio et al, 1974), in Puerto Rico (Law and Fulton, 1959; Mestre et al, 1960), in Mexico and India (Bishara et al, 1985, 1986), in Brazil (Da Silva Filho et al, 1993) and in Sri Lanka (Mars and Houston, 1990; Mars et al, 1990). Table 7.1 gives an overview of the samples in the studies. In 1985 and 1986 Bishara and co-workers and later on Mars and Houston (1990) summarized the limitations of previous studies on this topic. Most studies have a small sample size with a large age range. Quite often they are a mixture of different types of clefts and they describe different stages of treatment. They include really untreated patients, partially operated, and late operated individuals. Furthermore non-cleft controls and/or controls of treated patients from the same population are often not available. Postoperative follow-up of patients, that were treated in the course of the project, is scarce.

Most studies that were published earlier focussed on facial development and therefore were mainly conducted on lateral or antero-posterior head films. In the present study we have also collected cephalometric data, but these will be published elsewhere. We were particularly interested in maxillary and mandibular arch dimensions, as dental cast analysis had not often been employed in unrepaired clefts. When we started this study in 1986 only a few studies could be found in the literature that deal with dental cast analysis (Pruzansky, 1955; Law and Fulton, 1959; Innis, 1962; Foster, 1962; Hagerty et al, 1964; Mazaheri et al, 1971; Stöckli, 1971; Wada and Miyazaki, 1975; Bishara et al, 1976; Crabb and Foster, 1977; Sidhu et al, 1982; Bishara et al, 1985). In all these studies the main topic of interest was the sagittal relation of the molars and the canines.
Table 7.1  Overview of the sample size for complete UCLA, UCLP, BCLA and BCLP groups and for the control groups in the seven largest studies on unrepaired clefts, reported in the literature. Patients that had an incomplete cleft or an earlier repair are not represented in the table. The samples of patients older than 13 years of age are marked in gray.

<table>
<thead>
<tr>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Method</td>
<td>Returned Cephalostat</td>
<td>Returned Cephalostat</td>
<td>Returned Cephalostat</td>
<td>Returned Cephalostat</td>
<td>Returned Cephalostat</td>
<td>Returned Cephalostat</td>
<td>Returned Cephalostat</td>
</tr>
<tr>
<td>UCLA &lt;13y</td>
<td>-</td>
<td>-</td>
<td>13</td>
<td>32</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>UCLA &gt;13y</td>
<td>8</td>
<td>-</td>
<td>76*</td>
<td>-</td>
<td>26</td>
<td>50</td>
<td>168</td>
</tr>
<tr>
<td>UCLP &lt;13y</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>65</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>UCLP &gt;13y</td>
<td>42</td>
<td>8</td>
<td>222*</td>
<td>24**</td>
<td>44</td>
<td>78</td>
<td>68</td>
</tr>
<tr>
<td>BCLA &lt;13y</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>4</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>BCLA &gt;13y</td>
<td>-</td>
<td>-</td>
<td>21*</td>
<td>6</td>
<td>-</td>
<td>-</td>
<td>18</td>
</tr>
<tr>
<td>BCLP &lt;13y</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>16</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>BCLP &gt;13y</td>
<td>5</td>
<td>-</td>
<td>54*</td>
<td>7</td>
<td>40</td>
<td>13</td>
<td>-</td>
</tr>
<tr>
<td>N Total &lt;13y</td>
<td>-</td>
<td>-</td>
<td>330</td>
<td>-</td>
<td>236</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>N Total &gt;13y</td>
<td>55</td>
<td>8</td>
<td>373*</td>
<td>24**</td>
<td>83</td>
<td>168</td>
<td>267</td>
</tr>
<tr>
<td>Non-cleft controls</td>
<td>36</td>
<td>-</td>
<td>81</td>
<td>-</td>
<td>65</td>
<td>24</td>
<td>-</td>
</tr>
<tr>
<td>Repaired controls</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
</tbody>
</table>

* 6 yrs of age  
** 7 - 32 yrs of age

When dental arch measurements were included, the measuring methods were mainly two-dimensional (Crabb and Foster, 1977; Sidhu et al, 1982; Hardjowasito and Latief, 1988; Hardjowasito, 1989; Da Silva Filho et al, 1997). These two dimensional measuring methods fail to take into account the three-dimensional aspect of the deformity. Therefore it is difficult to draw conclusions about the effect of the presence of a cleft on the final dento-alveolar development of individuals with unrepaired clefts.

For this reason we aimed to perform a three-dimensional dental cast analysis, using an industrial coordinate measuring machine (Zeiss Numerex; Carl Zeiss®; Stuttgart, Germany) with a linear accuracy up to 0.002 mm. To our knowledge McCance et al (1990) are the only other researchers, besides a
preliminary report from our group (Derijcke et al, 1994), who did a three-dimensional cast analysis in unrepaired clefts, and in both studies the Reflex Microscope (Reflex Measurement Ltd., Somerset, UK) was used for this purpose. The Numerex system that was used in our study is using a touch probe to digitize the landmarks whereas the Reflex Microscope is a non-touch system. However, the reliability of the measurements seems to be comparable for both systems when comparing the magnitude of the measurement errors.

Already in 1971, Stöckli recognized the need for three-dimensional research into the spatial relationship of the maxillary segments in the frontal as well as the sagittal plane. However, performing three-dimensional surface area measurements of the palatal surface, is not really possible with the system we have used in our study. Nowadays, fast reliable 3D surface laser scanning systems are on the market that enable us to take any physical item and digitize it. From there three-dimensional computer models can be created and this will make it possible to scan the dental casts from the present study in order to get more insight into the size of the palatal shelves.

7.3 Discussion of the results

As part of the present study mandibular arch width was analyzed and it was found that in patients with clefts of the lip and alveolus only (UCLA and BCLA), the mandibular transversal dimensions were essentially normal. However, in complete UCLP, where the palate is involved, the posterior mandibular transversal dimensions were wider than in the controls. This corresponds with the wider upper arch dimensions in the same area. For BCLP individuals, neither in the upper nor in the lower jaw, differences in posterior dental arch width could be found. However, A-P cephalometric analysis on the transverse craniofacial morphology has shown that operated and unoperated BCLP patients have a significantly larger interorbital distance, nasal width, bicondylar width, and intergonial width (Ishiguro et al, 1976; Athanasiou et al, 1990; Motohashi et al, 1994). The sample of BCLP individuals in our study was possibly too small to draw reliable conclusions from this type of cleft.

The present study is the first one to investigate a really large sample of unrepaired UCLA over 13 years of age (see table 7.1). The BCLA group was more limited in number, but still one of the largest number reported so far.
The results show that in both groups the effects of the presence of a cleft remain restricted to the alveolar region. However, when the palate is also affected, a different pattern for maxillary arch width is found. At the level of the second permanent molar the maxilla was found to be wider in UCLP than in UCLA and the controls. The values point to a mesiopalatal rotation of the second permanent molar that has not been described previously in the literature. In our UCLP sample the intermaxillary width at the level of the first molar and second premolar was found to be normal. More anteriorly, in the first premolar and canine region, the arch width was significantly smaller in the UCLP group compared to both the UCLA group and the non-cleft control group. Changes of the general arch form of the maxilla in UCLP to a V-shape as McCance et al (1990) have already described was also observed with several individuals in our sample. As contrasted with McCance et al (1990) the changes of the shape of the dental arches in our sample may not only be attributed to the constriction of the entire maxilla, but is also due to a combination of enlargement of the maxillary width at the second molar area combined with constriction in the first premolar and canine region.

The study of maxillary arch width in BCLA and BCLP patients and the result 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, that even in BCLP adult unoperated patients, the developmental disturbances are rather limited to the vicinity of the cleft was in agreement with our findings.

In UCLP and BCLP patients the palatal shelf width was clearly smaller than in the non-cleft control group. This finding suggests an intrinsic deficiency in the amount of available tissue, not only at the cleft side but also at the non-cleft side that is assumed to be normally developed. Geometric three-dimensional analysis of serial palatal casts of operated patients has given important information about the palate’s physical changes that occur under the influence of growth and surgery (Berkowitz, 1990, 1999). Catch-up growth was found during the first 2 years of life and before palatal surgery: the lesser segment appeared to grow more rapidly than the non-cleft segment. A second period of accelerated growth was found 6 to 12 months after physiologic (non-scarring) surgery of the palatal cleft, and this extended to 6 years of age (Berkowitz, 1996). However, our study shows that even if this catch-up growth occurs, the palatal shelves still remain deficient at the adult stage.

The palatal shelves were also positioned cranially in comparison with the control group. Interposition of the tongue between the palatal shelves,
which are not connected to each other, might be responsible for this upward rotation of the shelves. The clinical implications of these findings are obvious. The smaller palatal shelf width and the larger elevation of the shelves result in a wider cleft. The surgeon who is trying to bridge the gap, especially when using the Langenbeck technique, creates a great empty space between the elevated palatal mucosa and the denuded bone of the palatal shelves. The scar tissue developing between these two structures, contracts, which possibly results in transversal compression of the maxilla, flattening of the palate and malposition of the teeth, as frequently seen in early operated cleft lip, alveolus and palate patients. Therefore surgical techniques should be developed that take into account the intrinsic deviations, which might result in a better long-term dentomaxillary development after surgical closure of the cleft.

7.4 General conclusions

From the findings of this study it can be concluded that there is an intrinsic or functional effect of the cleft itself on dentofacial development. Each type of cleft has its own intrinsic characteristic dental arch form. The more extensive the cleft, the more extensive the effect on the dental arch. However, compression of the maxilla is limited to the vicinity of the cleft in the anterior region. When the palate is involved the palatal shelves seem to be deficient and positioned more cranially as well. These findings support the hypothesis that surgery might influence subsequent maxillary growth, which makes the intrinsic deviation, that is already present, clinically manifest.

7.5 Recommendations for future research

Although the results of cleft lip and palate treatment have improved enormously over the last decades there are still patients that show a severely disturbed dentofacial growth pattern. There is an urgent and continued need to investigate the effects of different surgical procedures on maxillofacial growth and dento-alveolar development. Supplemental to that, the study of unrepaired clefts could tell us more about the natural history of clefts. Special attention should be given to bilateral cleft conditions since hardly any studies
with a sufficient sample size have been published, neither on repaired nor on unrepaired clefts.

Short-term and longer term follow-up of adult patients that were operated after facial growth had ceased, could provide more insight into the effects of surgery itself, excluding growth as a confounding factor. It is obvious that careful attention should be given to the anatomical deviating palatal shelves and the consequences for the surgical reconstruction of the palate. CT scanning before and after operation could provide more insight into the anatomical diversity among patients with orofacial clefts. In addition to these clinical studies, new techniques of palatal surgery, taking into account the anatomy of the defect and the size and position of the palatal shelves, should be tested in animal experiments in order to modify present surgical techniques for achieving better long-term dentofacial development in patients with clefts.

7.6 References


BERKOWITZ S. A multicenter retrospective 3D study of serial complete unilateral cleft lip and palate and complete bilateral cleft lip and palate casts to evaluate treatment: part 1: the participating institutions and research aims. Cleft Palate Craniofac J 1999;36:413-424.


HARDJOWASITO W. Studi sumbing bibir dan langit2 unilateral pada penderita penderita akil baliq dan dewasa yang belum dioperasi. Disertasi Universitas Airlangga. Surabaya (Indonesia); 1989.


