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Maxillary arch width in unoperated adult bilateral cleft lip and alveolus and complete bilateral cleft lip and palate

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Structured Abstract

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Objectives – To study maxillary arch width in adult patients with bilateral cleft lip and alveolus (BCLA) or with complete bilateral cleft lip and palate (BCLP), who have not had any surgery.

Setting and Sampling Population – Eighteen patients with BCLA, 13 patients with BCLP, and 24 controls from remote areas of Indonesia collected over 10 years.

Materials and Methods – Dental casts were digitized three-dimensionally using an industrial coordinate measuring machine (CCM) (Zeiss Numerex; Carl Zeiss®, Stuttgart, Germany). Transversal distance between molars was measured on the tip of the distobuccal cusp and the tip of the mesiobuccal cusp, and for premolars and canines, the tip of the buccal cusps was recorded. Means and standard deviations were calculated for all variables. *t*-Test was used to determine whether the mean values of the cleft groups showed significant differences from each other and from the controls. Level of significance was set at $p < 0.05$.

Results – Transversal arch dimensions in the BCLA group were comparable to the controls except at the canine level. Intercanine distance, which is close to the alveolar cleft, was 4.3 mm (SE 1.4) smaller in the BCLA group ($p = 0.002$). In the BCLP group, a comparable pattern was found. At the canine level, mean transversal width was 7.2 mm (SE 1.9) smaller compared to the control group, but no significant differences were found in the other transversal dimensions.

Conclusions – Small differences are found in transversal dimensions in patients with BCLA and BCLP compared to a control group. Differences are most outspoken in the area near the cleft.

Key words: cleft palate; maxillary growth; unoperated

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Introduction

There are several forms of orofacial clefts, yet the incidence varies considerably among races and the type of clefts (1–3). The majority of clefts is unilateral, but in 10–20% of all newborn children with a cleft the deformity is bilateral (4–6). For the bilateral clefts, there are different levels of severity of which probably the best documented are the bilateral cleft lip and palate (BCLP), but the bilateral cleft lip and alveolus (BCLA) is only mentioned incidentally. The BCLA is a rather mild form, and in known studies about epidemiology of clefts, these clefts are usually small in numbers, going from 2.4 to 7.3% (7, 8). Although the incidence of BCLP is also low, more studies have been dedicated to the BCLP group because of the implications for growth and final development of the facial skeleton and dentition.

The effect of a BCLP on growth and final development of the facial skeleton and dentition has mostly been studied in operated patients. Many of these studies dealing with the facial growth of patients with cleft palate have mainly used cephalograms to assess growth (9–17). When skeletal transversal measurements of the maxilla are included, AP head films are used (11, 18–20). These studies show that patients with a BCLP have at least a moderate skeletal deficiency of the maxilla and compression of the lateral maxillary segments. The extent of the skeletal deficiency, however, was difficult to predict (21, 22).

Measurements on dental casts of BCLA and BCLP have not been performed a lot (17, 23), and some studies show conflicting results about the effect of early palatal surgery and arch dimensions. Some found that timing of palatal closure had no additional effect on arch dimensions (24), whereas a later study 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 (25). In a longitudinal study on 22 patients with BCLP from 3 up until 17 years of age, Heidbüchel and Kuijpers–Jagtman (26) found, in spite of the prolonged orthodontic treatment, a significantly smaller maxillary dental arch width in patients with BCLP compared to the control group. Already from the age of 3 years on, the arch width was smaller in the early operated BCLP children. This reduction in the arch width was most pronounced in the canine area. At the age of 17 years, the reduction in the intercanine width was almost 10 mm in the BCLP group when compared to the non-cleft controls. At the level of the first molar, the maxillary width was 8 mm smaller in subjects with BCLP.

There is not much known about growth deficiencies in operated and unoperated patients with BCLA, but at least some minor aberrations and more lateral compression in the canine area might be expected (8).

The effect of the presence of a cleft on normal maxillary growth in BCLA and BCLP is not completely known mainly because all studies are carried out on treated patients. Studies with unoperated patients are carried out using cephalograms (17, 27–29) and/or analysis of the dental arch width by means of dental casts. These studies found that the maxillary width up to the first premolar was significantly smaller in patients with UCLP, BCLP, and CP compared to normal subjects (17) and that intercanine width in patients with BCLP was smaller than in the patients with UCLP and UCLA and that arch collapse was more pronounced in the BCLP group (28). Others found that the molar width was significantly smaller in the BCLP group compared to normal age-related population (29, 30), analyzing the maxillary dental arch morphology of 31 unoperated adult patients with BCLP, they found even more compression of the dental arch as in their sample arch width was smaller not only in the molars but also in the premolar and canine region compared to non-cleft individuals.

By analyzing facial growth and maxillary arch dimensions of unoperated adult subjects with BCLP, it is possible to get more insight into the real intrinsic growth potential of the maxillary structures and to separate this from the effect of surgical and orthodontic treatment. Therefore, the purpose of this study was to investigate the maxillary dental arch width in a sample of unoperated adult subjects with BCLA and BCLP and to compare the results with a non-cleft control group from the same population.

Materials and methods

Sample

The material for this study was collected through a cooperation between the University of Brawijaya, Faculty of Medicine (Malang, Indonesia), Universitas Indonesia, Faculty of Dentistry (Jakarta, Indonesia) and the University Medical Center Leiden, Department of Oral and Maxillofacial Surgery (Leiden, The Netherlands). This study is part of a bigger study on unoperated cleft lip and palate with a total population of 267 adult patients. Patients were collected during nine expeditions between 1986 and 1997 undertaken in the remote areas of Indonesia. Out of the 2400 patients seen during these expeditions, 267 totally unoperated adults were found. Out of these unoperated adults, a sample of 18 unoperated adults with a BCLA and 13 unoperated adults with a complete BCLP were found. The control group consisted of 24 randomly selected non-cleft individuals from the surrounding population. Patients were considered adult at the age of 13 or, when the exact age was not known to the family, patients were considered adult when the permanent dentition had erupted into full occlusion. Although facial growth at this age is still not fully complete in literature on unoperated clefts, this age is generally used at the start of the adulthood stage. A patient was considered unoperated if he had not undergone any kind of surgical or orthodontic treatment previously. Patients with submucosal cleft palate were not included in this study. All patients were documented with dental casts, cephalograms, and standard intra-oral and extra-oral photographs. After collection of the pre-operative data, all patients with cleft were treated surgically in a single surgical procedure.

The dental casts were digitized three-dimensionally using an industrial coordinate measuring machine (CCM) (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, two 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 center 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 the 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.

Statistics

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 $p < 0.05$.

To determine the measurement error, 40 dental casts were digitized twice by two independent observers (BL and a technician). Intra- and interobserver measurement errors were calculated according to Dahlberg's formula (31). The intra- and inter-observer errors for transversal distances were small ranging from 0.10 to 0.27 mm. The median measurement error was 0.15 mm.

Results

Table 1 shows means and standard deviations (in mm) for the transversal maxillary arch dimensions in the canine region, the premolar and the molar area in adult unoperated subjects with BCLA and BCLP when compared to the control group. Box-whisker plots of the maxillary arch width are shown in Fig. 1.

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 when 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% ($p = 0.0009$).

In the BCLP group a comparable pattern was found. At the canine level, the mean transversal width was even 7.2 mm (SE 1.9) smaller compared to the control group, but no significant differences were found in the other transversal dimensions. The upper arch ratio was 51.8% in the BCLP group and 64.2% in the

Table 1. **Comparison of maxillary transversal arch dimensions (in mm) between unoperated adult BCLA, BCLP, and the control group**

Group	N	Mean	SD	Group	N	Mean	SD	Diff	S.E	*t*-value	*p*-value
Intercanine width (131–231)											
BCLA	18	31.8	5.5	Control	24	36.1	1.9	−4.3	1.4	−3.17	0.002
BCLP	10	28.9	5.9	Control	24	36.1	1.9	−7.2	1.9	−3.81	0.003
BCLA	18	31.8	5.5	BCLP	10	28.9	5.9	2.9	2.3	1.30	0.19
First premolar width (141–241)											
BCLA	17	43.0	3.6	Control	24	44.0	2.1	−1.0	1.0	−1.01	0.31
BCLP	11	42.3	3.0	Control	24	44.0	2.1	−1.7	1.0	−1.67	0.09
BCLA	17	43.0	3.6	BCLP	11	42.3	3.0	0.7	1.3	0.57	0.56
Second premolar width (151–251)											
BCLA	17	48.4	3.9	Control	24	48.7	2.3	−0.3	1.1	−0.29	0.77
BCLP	13	47.9	3.8	Control	24	48.7	2.3	−0.8	1.2	−0.65	0.51
BCLA	17	48.4	3.9	BCLP	13	47.9	3.8	0.5	1.4	0.32	0.75
First molar width at distal cusps (161–261)											
BCLA	16	55.7	2.9	Control	24	56.3	2.6	−0.6	0.9	−0.65	0.51
BCLP	12	55.0	4.1	Control	24	56.3	2.6	−1.3	1.3	−1.00	0.32
BCLA	16	55.7	2.9	BCLP	12	55.0	4.1	0.7	1.4	0.50	0.62
First molar width at mesial cusps (162–262)											
BCLA	16	54.5	3.2	Control	24	54.9	2.6	−0.5	1.0	−0.50	0.61
BCLP	12	52.9	4.1	Control	24	54.9	2.6	−2.1	1.3	−1.58	0.11
BCLA	16	54.5	3.2	BCLP	12	52.9	4.1	1.6	1.4	1.10	0.27
Second molar width at distal cusps (171–172)											
BCLA	16	58.4	3.7	Control	24	58.8	2.7	−0.4	1.1	−0.37	0.71
BCLP	12	60.2	4.4	Control	24	58.8	2.7	1.4	1.4	1.03	0.30
BCLA	16	58.4	3.7	BCLP	12	60.2	4.4	−1.8	1.6	−1.17	0.24
Second molar width at mesial cusps (172–272)											
BCLA	16	58.7	3.8	Control	24	58.9	2.8	−0.2	1.1	−0.14	0.88
BCLP	12	58.6	4.7	Control	24	58.9	2.8	−0.3	1.5	−0.17	0.86
BCLA	16	58.7	3.8	BCLP	12	58.6	2.7	0.1	1.6	0.06	0.95
UPPER ARCH RATIO (%)											
BCLA	16	56.8	8.2	Control	24	64.2	3.2	−7.4	2.2	−3.43	0.0009
BCLP	10	51.8	10.4	Control	24	64.2	3.2	−12.4	3.3	−3.71	0.0004
BCLA	16	56.8	8.2	BCLP	10	51.8	10.4	5.0	3.9	1.30	0.19

DIFF, difference in maxillary arch width variables between BCLA vs control, BCLP vs control, and BCLA vs BCLP.
SE, standard error of the difference.
UPPER ARCH RATIO; 131–231 distance/161–261 distance * 100 (%).

control group, and this difference was significant ($p = 0.0004$).

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. These percentages are lower especially for BCLP than could be expected based on the incidence for BCLP as reported in the literature (6). Presumably, the life expectation of patients with a

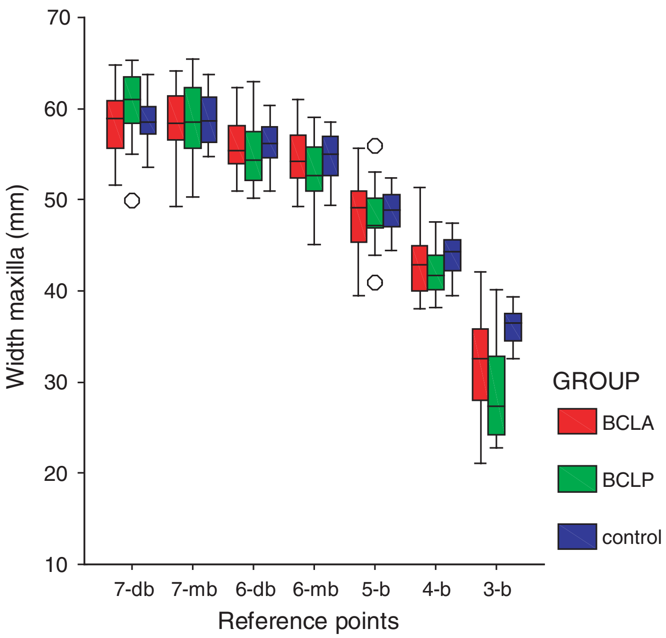

Fig. 1. Box-whisker plot of maxillary arch width (in mm) in unoperated adult subjects with BCLA and BCLP compared to the control group. 7-db = second molar width at distal cusps (171–271); 7-mb = second molar width at mesial cusps (172–272); 6-db = first molar width at distal cusps (161–261); 6-mb = first molar width at mesial cusps (162–262); 5-b = second premolar width at buccal cusps (151–251); 4-b = first premolar width at buccal cusps (141–241); 3-b = intercanine width (131–231).

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 patients with UCLP by Hardjowasito (32) 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 peri-operative respiratory complications have to be expected than in children with less severe clefts (33). The fact that in our BCLP sample no individuals were found older than 40 years 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 (34). 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 their 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 were not reported (20).

The results of the present study are in accordance with the studies of Sidhu et al. (30) and Bishara et al. (28). The observation especially of Bishara et al. (28) that even in adult unoperated patients with BCLP, 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. (29, 35), 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 (19). The reason why in our study the intrinsic deformity was much more limited than in the study of Da Silva Filho et al. (29) 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 because of 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 to clarify these contradictions.

From the findings of the present and previous investigations into unoperated adult individuals, it could be concluded that there are differences in the dentoalveolar development of persons 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 dentoalveolar 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.

Clinical relevance

Patients with cleft palate need extensive treatment from early age until adulthood. Early surgery though has an effect on growth of the maxillofacial complex. Yet the question is how far the cleft itself affects growth of the maxillofacial complex. By analyzing facial growth and maxillary arch dimensions of unoperated adults, it is possible to get more insight into the intrinsic growth potential of the maxillary structures and to understand more of the iatrogenic effects of 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.

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