

ORIGINAL ARTICLE

Maxillary arch dimensions in cleft infants in Northern Finland

VIRPI HARILA¹, LEENA P. YLIKONTIOLA², RIITTA PALOLA¹ & GEORGE K. SÁNDOR^{2,3}

¹Department of Oral Development and Orthodontics, ²Department of Oral and Maxillofacial Surgery, Institute of Dentistry, University of Oulu, Oulu Cleft Lip and Palate Center, Oulu University Hospital, Oulu, Finland, and ³Regea Institute for Regenerative Medicine, University of Tampere, Tampere, Finland

Abstract

Objective. The aim of this study was to examine the maxillary arch dimensions in cleft lip and/or palate infants in Northern Finland before surgery. **Materials and methods.** The subjects consisted of 70 Finnish cleft patients born between 1997–2004 in Northern Ostrobothnia Hospital District in Finland. The study casts were obtained before surgery at the mean age of 5.6 months (SD = 2.2). There were 42 children with cleft palate (CP) (26 girls/16 boys), 13 with unilateral cleft lip and palate (UCLP) (eight girls/five boys), eight children with cleft lip (CL) (two girls/six boys) and seven with bilateral cleft lip and palate (BCLP) (two girls/five boys). Conventionally-used landmarks were marked on study casts and cleft width, arch circumference, anterior and posterior arch width and arch length were measured with a digital sliding calliper. The statistical method was ANOVA. **Results.** The prevalence of CP in this study, 60% of all clefts, is higher than the average standards. There were statistically significant differences in cleft width, posterior and anterior arch width, arch length and arch circumference, when different cleft groups were compared. When differences between girls and boys were compared, boys had larger cleft size and arch dimensions generally, but the results were not statistically significant. **Conclusions.** The results show the large variation in the severity of cleft lip and/or palate deformity at birth and in maxillary arch dimensions between different cleft types. It also demonstrates the effect of phenotypic variability within the groups of cleft lip and/or palate.

Key Words: cleft width, dental casts, maxillary morphology

Introduction

Cleft lip and/or palate are the most common congenital craniofacial birth defects. Generally clefts are divided into two groups, isolated cleft palate (CP) and cleft lip with or without cleft palate (CL/P). Orofacial clefts arise in about one in 700 live births [1]. CP occurs in 0.4/1000 live births while CL/P has a higher prevalence; 1/700 [2]. In Finland, the prevalence of CP is 0.97/1000 [3]. There is variability in the prevalence depending on geography, ethnicity and socioeconomic position [4,5]. Rates of cleft lip with or without cleft palate are high in parts of Latin America and Asia (China, Japan) and low in Israel, South Africa and southern Europe. Rates of isolated cleft palate are high in Canada and parts of northern Europe and low in parts of Latin America and South Africa. Comparisons between ethnic groups within the US and the UK and studies of immigrants to the US from Japan and China indicate that migrant groups have rates of cleft lip with

or without cleft palate closer to those of the area from which they originated than those in the area into which they have moved [6].

Cleft lip with or without cleft palate is most frequent in males and isolated cleft palate in females across various ethnic groups. With the severity of cleft, the sex ratio varies [3,6]. In white populations, the sex ratio for CL/P is ~2:1 (male:female). In white populations the male excess in CL/P becomes more apparent with increasing severity of cleft [6,7]. CL/P and CP are often associated with other major congenital anomalies. General defects seem to be more frequent in CP patients than for those with CL/P [6]. In CP cases, 55% has been reported to be associated with syndromes, while 14% of CL/P cases are syndromic [8]. The etiology of CL/P and CP are thought to be multifactorial, but both genetic and environmental factors play a role. The role of inheritance in CL/P and CP was described in the 1940s and confirmed in the 1980s [9–11].

Correspondence: Virpi Harila, DDS, PhD, Department of Oral Development and Orthodontics, Institute of Dentistry, University of Oulu, PL 5281, 90014 Oulu, Finland. Tel: +358 407137073. Fax: +358 8 5375560. E-mail: virpi.harila@oulu.fi

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Table I.

Cleft type	<i>n</i>	Girls	Boys	Mean age (months)	SD
CL	8 (11.4%)	2 (25%)	6 (75%)	5.2	3.8
CP	42 (60.0%)	26 (61.9%)	16 (38.1%)	6.2	1.3
UCLP	13 (18.6%)	8 (61.5%)	5 (38.5%)	5.0	2.6
BCLP	7 (10%)	2 (28.6%)	5 (71.4%)	3.4	1.2
Total	70	38 (54.3%)	32 (45.7%)	5.6	2.2

CL, Cleft Lip; CP, Cleft Palate; UCLP, Unilateral Cleft Lip and Palate; BCLP, Bilateral Cleft Lip and Palate.

The size of the cleft in infancy is determined by the tissue deficiency and also by the degree of separation of the maxillary segments. The severity of the deformity at birth varies remarkably [12,13]. The initial severity of the deformity may have a potential effect on dental arch relationships and treatment outcome. Reduced maxillary arch dimensions in cleft children has been reported [14,15]. The most frequent malocclusion in cleft infants is cross-bite and deficient maxillary growth [14,15]. Studies of the associations between the initial cleft size and malocclusions in the primary dentition have varying results. Hellquist et al. [16] reported among CP children with large antero-posterior clefts of the palate the highest frequency of cross-bite and smallest inter-canine and inter-molar dimensions in the primary dentition. Suzuki et al. [17] also found significant correlation between the width of the palatal cleft at birth and the posterior arch width at 4 years of age in UCLP children. On the other hand, Johnson et al. [13] did not find any correlation between the severity of the initial cleft and dental arch relationships at 6 years of age in UCLP children.

The aim of this study was to examine the maxillary arch dimensions and the size of cleft in CP, CL and CL/P infants before primary surgery in Northern Finland.

Materials and methods

The subjects consisted of 70 Finnish cleft infants born between 1997–2004 in Northern Ostrobothnia Hospital District in Finland. The documents of the patients were taken according to the Eurocleft program. There were 42 children with cleft palate (CP), 13 with unilateral cleft lip and palate (UCLP), eight with cleft lip (CL) and seven with bilateral cleft lip and palate (BCLP). Alginate impressions were taken before the first surgical operation and casts were made at the mean age of 5.6 months (SD 2.2) (Table I).

The reference points and linear measurements were performed on the maxillary casts using an electronic measuring device. The method used in this study has been described earlier [12,18–20]. All the landmarks have a biologic meaning (signifying some anatomic

structure or special point on such a structure). Cleft dimensions are described in terms of cleft widths, anterior cleft width is the alveolar cleft along the crest of the ridge, middle cleft width is at the intersection of the inter-canine point line and posterior cleft width is at the intersection of the inter-tuberosity line [13,21]. Cleft width, arch circumference, anterior and posterior arch width and arch length were measured with a digital sliding calliper by one operator (Figure 1). Statistical analysis used was the analysis of variance (ANOVA). The intra-class correlation coefficient was used for the reliability of the measurements.

Results

In this study, the prevalence of CP was 60.0% of all clefts, which is higher than the average standards

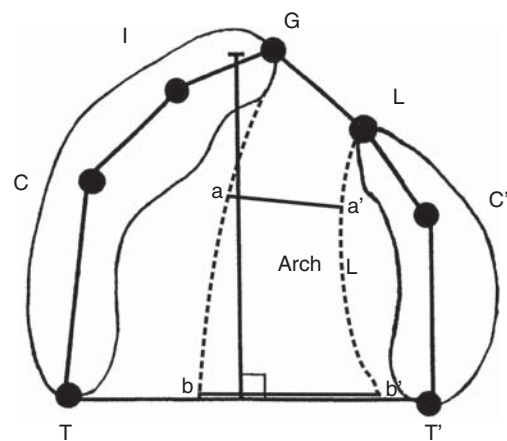


Figure 1. Reference points marked on the study casts. G = mid-point of the margin of the alveolar process medial to the cleft; L = mid-point of the margin of the alveolar process lateral to the cleft; I = point of intersection between the alveolar ridge and groove of the median labial frenum; C, C' = anterior arch width; point of intersection between the alveolar ridge and groove of the lateral labial frenum; T, T' = posterior arch width; tuberosity points, junction of the alveolar ridge with the outline of the tuberosity; arch circumference = T-C-I-G + L-C'-T'; arch length = G perpendicular to T-T'-line [13,21]. Cleft dimensions are described: G-L = Anterior cleft width – the alveolar cleft along the crest of the ridge; a-a' = Middle cleft width – soft tissue cleft margin is represented by the broken line; b-b' = Posterior cleft width – soft tissue cleft margin is represented by the broken line.

(Table I). There were statistically significant differences in cleft width, posterior and anterior arch width, arch length and arch circumference between cleft groups and between boys and girls. The results are shown in Table II. The arch circumference was largest in CL patients and smallest in CP patients ($p < 0.0001$). The anterior arch width was largest in UCLP patients and smallest in CP patients ($p < 0.0001$). The posterior arch width was largest in UCLP patients and smallest in CL patients ($p < 0.005$). The arch length was largest in BCLP patients and smallest in UCLP patients ($p < 0.0001$). When the width of cleft was compared between different cleft groups, the anterior cleft width was largest in UCLP infants ($p < 0.027$), the middle cleft width was largest in BCLP infants and the posterior cleft width was largest in UCLP infants ($p < 0.0001$).

When boys and girls were compared, boys generally had larger cleft size and arch dimensions (Table III). Significant differences were found in the measurement I-C distance between the point of intersection between the alveolar ridge and groove of the median labial frenum (I) and point of intersection between the alveolar ridge and groove of the lateral labial frenum (C) (Figure 1), boys having larger dimensions ($p < 0.009$). The posterior arch width was also larger in boys compared to girls ($p < 0.012$).

The intra-examiner reliability of the dental cast measurements were determined from duplicate recordings in 20 casts, 1 month after the first measurement. The correlation coefficients were calculated and the intra-examiner correlation coefficient generally varied from 0.82–0.99, showing good

correlation of repeated measurements, except for the measurement T-T' (0.57).

Discussion

The method of the measurements used in this study has been described earlier [12,18–20]. A well-constructed investigation of landmark precision suggests that errors can be substantial, up to 10%, and the experience of the operator is important [18]. It is also recommended that the measurements made on the dental casts are made by the same operator.

Measurements in the cleft region are subject to greater error than other measurements, especially the posterior cleft width as it is constructed [22]. Alveolar cleft was the second best of the analysed measurements in the study by Seckel et al. [18]. The quality of the study casts is also important, in addition to the experience of the examiner, especially in the T-T' region, a measurement which has been reported to have very poor reproducibility [18,22]. The intra-examiner correlation coefficient in this study generally varied from 0.82–0.99, showing good correlation of repeated measurements. The measurement T-T' had the poorest intra-examiner correlation, only 0.57, so the results concerning those areas are thus not highly reliable.

Previous data in other populations suggest the variability of the prevalence of cleft lip with or without cleft palate being 3.4–22.9 per 10 000 births and of isolated cleft palate being 1.3–25.3 per 10 000 births [6]. The epidemiology of cleft palate in various European countries has been investigated in the study

Table II. Comparison of the measurements (mm) between cleft types.

Measurement	CP ($n = 42$) M (SD)	UCLP ($n = 13$) M (SD)	BCLP ($n = 7$) M (SD)	CL ($n = 8$) M (SD)	p -value
Age (months, SD)	6.24 (1.32)	5.04 (2.64)	3.36 (1.2)	5.16 (3.84)	
Anterior cleft G-L	—	7.13 (4.83)	—	1.07 (0.60)	0.027*
G-I	—	8.88 (1.45)	—	9.32 (1.04)	0.534
G-C	—	8.60 (1.89)	10.54 (2.24)	—	0.180
I-C	14.42 (1.30)	14.99 (1.83)	—	15.14 (1.04)	0.265
C-T	20.11 (1.47)	18.33 (1.79)	19.61 (1.65)	21.05 (2.75)	0.003**
L-C'	—	11.51 (2.91)	9.63 (1.87)	10.63 (1.48)	0.289
C'-T'	20.01 (1.87)	17.34 (2.70)	18.67 (1.80)	20.31 (1.96)	0.001***
C-C'	25.74 (2.03)	31.08 (4.30)	29.27 (2.57)	28.23 (1.60)	0.0001***
T-T'	30.34 (2.37)	32.82 (2.52)	31.91 (2.96)	29.65 (1.91)	0.005**
Arch circumference	68.88 (3.72)	69.49 (5.93)	74.10 (4.50)	75.49 (4.78)	0.0001***
Arch length	25.75 (1.96)	25.19 (2.04)	30.21 (4.80)	28.99 (2.42)	0.0001***
Middle cleft width, a-a'	—	9.01 (6.22)	10.19 (2.91)	—	0.0001***
Posterior cleft width, b-b'	7.73 (4.71)	13.96 (5.63)	12.69 (4.06)	—	0.0001***

* $p < 0.05$, ** $p < 0.005$, *** $p < 0.001$

Table III. Comparison of the measurements between different cleft types and between boys and girls.

Measurement (mm)	CP girls ($n = 26$) M (SD)	CP boys ($n = 16$) M (SD)	UCLP girls ($n = 8$) M (SD)	UCLP boys ($n = 5$) M (SD)	BCLP girls ($n = 2$) M (SD)	BCLP boys ($n = 5$) M (SD)	CL girls ($n = 2$) M (SD)	CL boys ($n = 6$) M (SD)	p -value
Age (months)	6.36 (1.2)	6.12 (1.44)	5.16 (2.4)	4.8 (3.12)	4.68 (1.2)	2.88 (0.6)	5.28 (3.36)	5.16 (4.32)	
Anterior cleft	—	—	7.15 (4.90)	7.10 (5.28)	—	—	—	—	0.99
G-L									
G-I	—	—	8.61 (1.43)	9.41 (1.63)	—	—	9.96	9.19 (1.11)	0.99
G-C	—	—	9.25 (2.63)	7.95 (1.46)	11.18 (4.82)	10.29 (1.22)	—	—	0.50
I-C	14.38 (1.37)	14.49 (1.20)	14.20 (1.47)	16.57 (1.53)	—	—	14.02 (0.80)	15.51 (0.84)	0.009*
C-T	20.00 (1.41)	20.29 (1.59)	18.26 (2.02)	18.45 (1.56)	20.77 (0.83)	19.15 (1.72)	22.85 (0.71)	20.45 (2.97)	0.13
L-C'	—	—	11.02 (2.98)	12.47 (3.12)	8.50 (2.30)	10.08 (1.75)	—	—	0.32
C'-T'	19.97 (2.09)	20.19 (1.51)	17.30 (2.84)	17.42 (2.80)	19.85 (2.84)	18.20 (1.38)	20.16 (1.41)	20.36 (2.22)	0.70
C-C'	25.54 (2.01)	26.06 (2.08)	30.71 (4.68)	31.67 (4.06)	27.18 (3.75)	30.11 (1.82)	27.91 (0.02)	28.33 (1.87)	0.28
T-T'	30.46 (2.15)	30.14 (2.75)	32.21 (2.53)	33.80 (2.41)	29.70 (3.53)	32.80 (2.57)	27.68 (1.63)	30.32 (1.59)	0.012*
Arch circumference	68.77 (3.98)	69.06 (3.38)	69.05 (4.86)	70.19 (7.93)	72.30 (3.21)	74.82 (5.05)	76.09 (1.66)	75.29 (5.60)	0.62
Arch length	25.77 (2.02)	25.72 (1.93)	24.70 (2.28)	25.96 (1.47)	27.74 (9.21)	31.20 (3.02)	29.64 (1.15)	28.78 (2.77)	0.25
Middle cleft width, a-a'	—	—	8.24 (6.35)	10.78 (6.49)	8.21 (2.31)	10.98 (2.94)	—	—	0.36
Posterior cleft width, b-b'	7.80 (4.78)	7.21 (4.74)	13.34 (6.24)	15.20 (4.73)	11.57 (8.03)	13.15 (2.79)	—	—	0.63

* $p < 0.05$

by Calzolari et al. [23]. The prevalence of CP varied significantly in Europe and also within countries with an European mean value of $6.2 \times 10\,000$. The highest CP frequency was observed in Finland, essentially due to an excess of isolated CP. The prevalence rate of isolated cleft palate was 9.3 per 10 000 births (live births, still births and interrupted pregnancies) in Finland [23]. This finding was also confirmed in this study, the prevalence of CP being 60% of all clefts is higher than the average European level and reflects the speciality of cleft type distribution of northern Finland. The population in the Ostrobothnia Hospital District area is very homogenous compared to other study populations. The complex model of inheritance and the frequently conflicting results in different populations on the role of genes that constitute risk factors suggest the presence of real biological differences [23]. Several associations in craniofacial dimensions between parents and children with CLP emphasize the importance of genetic factors in the genesis of non-syndromic orofacial clefting [24]. CLP family members have been reported to have shortened palatal dimensions and their faces are disproportionately wide (width/height) [24]. The association of distinct craniofacial characteristics between parents and affected children emphasizes the importance of genetic factors in the development of clefting. The sidedness of the cleft in affected children has been found to be related to the asymmetry in the nasal cavity width in their parents [24,25].

Orofacial clefts have a major impact on craniofacial morphology. The severity of cleft is usually evaluated from its appearance at birth and there is great variability in tissue deficiency and the severity of the initial cleft. The width of the cleft may affect the surgical repair and thus also outcome. A wider cleft may generate more scar tissue and affect the growth of the maxilla.

In this study significant differences were found in cleft width, posterior and anterior arch width, arch length and arch circumference between different cleft types and between boys and girls. The anterior arch width was largest in UCLP patients and smallest in CP patients. The posterior arch width was also largest in UCLP patients, being smallest in CL patients. When the cleft width was concerned, the anterior cleft and posterior cleft width was largest in UCLP infants and the middle cleft width was largest in BCLP infants in this study. It has been reported earlier, that in UCLP the morphological features include a lateral displacement of the incisive papilla and a wider anterior arch [26] and the results of the present study confirm these findings. The arch length was largest in BCLP patients and smallest in UCLP patients, reflecting protrusion of the premaxilla in bilateral cleft lip and palate cases. The posterior arch width was significantly larger in boys compared to girls. With increasing severity of cleft, boys excess

becomes more apparent compared to girls [6,7]. Mid-face retrusion has always been a concern in cleft patients. There are reports of decreased size and retropositioned maxilla in operated cleft individuals of various ages [27–29]. Infants with clefting of the secondary palate have been reported to have short and retropositioned lateral parts of the maxilla, reduced height and increased width of the posterior maxilla [30,31]. Some of the disturbances are caused by the primary anomaly directly and other features are related to the surgical interventions needed.

The results concerning the effect of the initial cleft width on the clinical outcome has been variable. The study by Johnson et al. [13] demonstrated that the severity of the initial cleft does not correlate with the outcome in terms of dental arch relationship in UCLP patients and the initial presentation of UCLP is probably a poor predictor of outcome. Mazaheri et al [32] found wide variability in initial arch form and a variable pattern of arch development from birth to 5 years old in different cleft types. Kramer et al. [33,34] have reported differences in the palatal dimensions of complete and incomplete cleft lip and palate patients after cheiloplasty. They also suggested that palatal growth is more dependent on the type and severity of the cleft than the palatoplasty. Unoperated clefts of the primary palate are characterized by pre-maxillary protrusion, which diminishes after lip surgery [31]. The results of the study by Peltomäki et al. [12] indicate that less favourable maxillary growth is anticipated because of large cleft and short arch circumference. They reported that all patients with UCLP are not the same at birth and remain to be characterized by the severity of initial deformity, which has important clinical implications. The outcome of treatments and the growth of maxilla may be anticipated according to the initial severity of the cleft and less related to given treatment. They have suggested that the treatment protocol could vary according to the severity of the initial cleft deformity. According to their findings UCLP patients with small cleft and large arch circumference would probably have a different outcome with regards to maxillary growth from a child with a large cleft and small arch circumference. The data by Chiu et al. [20] suggest that, in patients with complete UCLP, there is a significant relationship between initial cleft severity and maxillary growth. Patients with a small cleft area have more protruded maxilla at the age of 9 years than those with a larger cleft area. Measurements and evaluation of the initial size of the cleft and arch dimensions of the maxilla are important for treatment planning.

The sex ratio varies between males and females with the severity of clefting. Cleft lip with or without cleft palate is more common in males, whereas isolated cleft palate is more frequent in women across various populations and the present study confirms

these findings. In Caucasoid populations, the sex ratio for cleft lip with or without cleft palate is ~2:1 (male:female) [6]. Studies about the male excess of cleft lip with or without palate were also confirmed in this study, suggesting the importance of genetic susceptibility. It has been found that significant differences of maxillary growth increments between males and females and the gender have been suggested to play a certain role in growth changes within the first 6 months of age [35]. There is individual variability in growth and improvements of arch form and the intrinsic potential seems to be analogous to that in non-cleft patients [36]. It has been reported that there are differences in maxillary arch dimensions of normal children of different ethnic origins at birth and in the extent of growth variation during the first 4 months of age [37]. The results of this study indicate that cleft children have a large variability in initial cleft severity and the results are in accordance with earlier reports [12,13,19]. The evaluation of the initial cleft severity is important for treatment planning and predicting the maxillary growth.

Conclusions

The results show the large variation in the severity of cleft lip and/or palate deformity at birth and in maxillary arch dimensions between different cleft types. They also demonstrate the effect of phenotypic variability within the groups of cleft lip and/or palate.

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