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Research Report

Craniofacial morphology of children with complete unilateral cleft lip and palate following labioplasty and palatoplasty

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ABSTRACT

Background: *A complete unilateral cleft lip and palate generally results in asymmetry of the midface. The lack of continuity in the perilabial musculature through the midline contributes to a malpositioning of the underlying osseus structures which are often underdeveloped.* ***Purpose:*** *The purpose of this study was to determine whether there are differences in the craniofacial morphology among children with complete unilateral cleft lip and palate following labioplasty and palatoplasty as compared with children without cleft lip and palate at the same pubertal age.* ***Methods:*** *A series of 14 consecutively treated subjects with complete unilateral cleft lip and palate following labioplasty and palatoplasty were compared with 14 pubertal stage-matched controls with normal craniofacial structure. Pubertal stage was determined with cervical vertebral maturation (CVM) method improved by Baccetti et al, 2002. Lateral cephalograms were used for comparison. An unpaired t-test was run for 14 subjects with complete unilateral cleft lip and palate and 14 normal subjects.* ***Results:*** *There were significant cephalometric differences in anterior cranial base length (p = .002), cranial base length (p = .001), maxillary length (p = .000), mandibular length (p = .000), mandibular ramus height (p = .000), mandibular body length (p = .002), and upper anterior face height (p = .004). There was no significant cephalometric difference in posterior cranial base length (p = .051), lower anterior face height (p = .206), posterior face height (p = .865), growth pattern/ facial type (p = .202).* ***Conclusion:*** *There were craniofacial morphology differences between children with complete unilateral cleft lip and palate post labioplasty and palatoplasty and children without cleft lip and palate at the age of pubertal. Children with complete unilateral cleft lip and palate post labioplasty and palatoplasty had shorter length of the anterior cranial base, cranial base, maxilla, mandible, mandibular ramus height, mandibular body, and upper anterior face height as compared with children without cleft lip and palate at the age of pubertal.*

Key words: *Cephalometrics, complete unilateral cleft lip and palate, craniofacial morphology*

ABSTRAK

Latar belakang: *Celah bibir dan langit-langit unilateral komplit umumnya menghasilkan asimetri wajah bagian tengah. Berkurangnya kontinuitas otot di sekitar bibir yang melewati garis tengah wajah mengakibatkan malposisi struktur tulang di bawahnya yang seringkali kurang berkembang.* ***Tujuan:*** *Tujuan penelitian ini adalah untuk menentukan apakah terdapat perbedaan morfologi kraniofasial anak dengan celah bibir dan langit-langit unilateral komplit pasca labioplasti dan palatoplasti dibandingkan anak tanpa celah bibir dan langit-langit pada usia pubertal.* ***Metode Penelitian:*** *Sejumlah subyek penelitian berupa 14 orang anak penderita celah bibir dan langit-langit pasca labioplasti dan palatoplasti dibandingkan dengan 14 orang anak yang normal pada masa pubertal yang sama. Masa pubertal ditentukan menggunakan metode cervical vertebral maturation (CVM) yang dikembangkan oleh Baccetti dkk, 2002. Dilakukan perbandingan hasil pengukuran sefalogram lateral dari kedua kelompok. Uji-t tidak berpasangan dilakukan untuk mengetahui perbedaan gambaran kraniofasial antara kelompok anak dengan celah bibir dan langit-langit unilateral komplit pasca labioplasti dan palatoplasti dan kelompok anak normal.* ***Hasil:*** *Terdapat perbedaan bermakna pada panjang basis kranium anterior (p = .002), panjang keseluruhan basis kranium (p = .001), panjang maksila (p = .000), panjang mandibula (p = .000), tinggi ramus mandibula (p = .000), panjang badan mandibula (p = .002), tinggi wajah anterior atas (p = .004). Tidak terdapat perbedaan bermakna pada panjang basis kranium posterior (p = .051), tinggi wajah anterior bawah (p = .206), tinggi wajah posterior (p = .865), pola pertumbuhan/tipe wajah (p = .202).* ***Kesimpulan:*** *Terdapat perbedaan morfologi kraniofasial antara anak dengan celah bibir dan langit-langit unilateral komplit pasca labioplasti dan palatoplasti dibandingkan anak tanpa celah bibir dan langit-langit*

pada masa pubertal. Anak dengan celah bibir dan langit-langit unilateral komplit pasca labioplasti dan palatoplasti memiliki panjang basis kranium anterior, panjang keseluruhan basis kranium, panjang maksila, panjang mandibula, tinggi ramus mandibula, panjang badan mandibula, dan tinggi wajah anterior atas yang lebih pendek dibandingkan anak tanpa celah bibir dan langit-langit pada masa pubertal.

Kata kunci: *Sefalometri, celah bibir dan langit-langit unilateral komplit, morfologi kraniofasial*

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INTRODUCTION

Cleft lip and/or palate is a common congenital malformation, with incidence between 1/500 to 1/1000 births worldwide and is associated with a high incidence of geographic origin, racial background/ethnicity, and social economic status.[1] These inherited disorders occur because of disruption to normal mechanisms during early development in the embryonic face. Cleft can vary from a small base on the edge of the lip vermilion to a complete separation of the alveolar ridge, to the bottom of the lip and nose.[2] Cleft is due to hypoplasia of the mesenchyme layer resulting in a failure of the unification of the medial nasal and maxillary processes, whereas cleft palate occurs due to failure of palatal shelves to fuse.[3] Etiology of congenital abnormalities is multifactorial with genetic and environmental factors play an important role through a complex mechanism at the molecular stage during embriogenesis.[4]

Patients with cleft lip and/or palate have many risk factors including inadequate nutrition, pulmonary aspiration, impaired masticatory function, aesthetic problems, hearing loss, speech impairment, and psychosocial, and growth disorders.[2] Shapira found an incidence of 74% for missing maxillary lateral incisors and 18% for missing second premolars in children with cleft lip, cleft palate, or both.[5] When not absent, the maxillary lateral incisor on the cleft site is nearly always abnormal in size and shape, and the second premolars show delay in development and eruption.[5,6] Root development was significantly delayed, especially on the side of the cleft.[7] Intrauterine growth and development of the nasomaxillary complex in patients with clefts often show anterior and posterior cross bite and midfacial deficiencies with class III malocclusion tendency.[8] Growth pattern in patients with cleft can be influenced by the type of cleft, post-surgical scar tissue, orthodontic/orthopedic, and alveolar bone graft.[9]

Lack of information about craniofacial morphology of children with complete unilateral cleft lip and palate (CULP) during pubertal encourage writers to do research on craniofacial morphology of children with complete unilateral cleft lip and palate during pubertal who had undergone labioplasty and palatoplasty using cephalometric analysis. Determination of pubertal age in this study was done by the method of cervical vertebral maturation (CVM) developed by Baccetti *et al*.[10] The purpose of this study was to determine whether there are differences in the craniofacial morphology among children with complete unilateral cleft lip and palate post labioplasty and palatoplasty as compared with children without cleft lip and palate at the same pubertal age.

MATERIAL AND METHODS

This study was a cross-sectional study using lateral cephalometric radiographs from children with complete unilateral cleft lip and palate and normal children. The radiographs were taken during routine orthodontic examination. The study was conducted in cleft lip and palate unit RSAB Harapan Kita-Jakarta and RSGM Faculty of Dentistry Universitas Indonesia-Jakarta during April-June 2012.

Patients were included if they fulfilled the following criteria: 1) patients who were diagnosed with non-syndromic complete unilateral cleft lip and palate corresponding visual inspection by oral surgeon listed on the medical record; 2) birth weight 2500-3500 g;[11] 3) has a lateral cephalometric radiographs with good clarity and contrast; 4) in the pubertal growth period (CVMS II and III according to the CVM index developed by Baccetti *et al*.;[10] 5) has had labioplasty and palatoplasty with the same surgical protocol (Cronin technique labioplasty and pushback partial split flap palatoplasty). The exclusion criteria of subjects: 1) being/ have been treated orthodontically; 2) has alveolar bone grafting performed previously.

Criteria for inclusion of control: 1) Class I jaw relationship, determined by cephalometric analysis (ANB

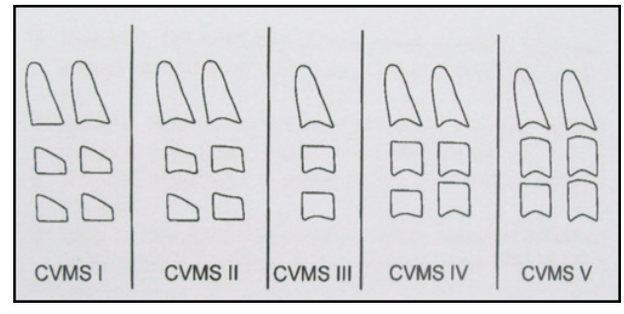

Figure 1. The newly improved cervical vertebral maturation (CVM) method.[10]

2°±2°); 2) class I incisor relationship (insisif classification of the British Standards Institute). Using unpaired numerical analytic formulas with a 5% error, as many as 14 of the samples are obtained in the group of children with complete unilateral cleft lip and palate and 14 samples in the control group.

A conventional cephalometric approach was used to examine the data. The parameters evaluated on the lateral cephalogram are listed in Table 1.

Lateral cephalometric radiographs of children with complete unilateral cleft lip and palate and normal children were observed in a darkened room, and a black surround was used on the light box to eliminate excess light and facilitate landmark identification. Each radiograph was then classified according to the cervical vertebral maturation (CVM) method improved by Baccetti *et al.*[10]

The outlines of cervical vertebrae and cephalometric landmarks were traced on cephalometric tracing paper (Ortho Organizers) with a 3H lead pencil. Digital caliper (Mitutoyo, Japan) was used to measure the distance between the landmark, cephalometric protractor (Ortho Organizers) was used to measure the angle.

To establish intraexaminer repeatability between the first and the second measurement 6 sefalogram were randomly selected (consisting of 3 subjects with cleft lip and palate and 3 normal subjects) performed with 1 week interval. Because cephalograms demonstrated different magnification, adjustment for enlargement factor was made. Data processing and statistical analysis performed on the data from each study variable. Statistical tests performed by univariat analysis to obtain the mean, the maximum and minimum, and standard deviation of each group. Data analysis was performed with unpaired t-test to examine craniofacial morphology differences between children with complete unilateral cleft lip and palate post labioplasty and palatoplasty and the normal children as controls.

Table 1. Operational definition of variables[12]

Variables (Cephalometric measurement)	Definition
Anterior cranial base length	Distance from point S to point N (mm)
Posterior cranial base length	Distance from point S to point Ba (mm)
Cranial base length	Distance from point N to point Ba (mm)
Maxillary length	Distance from ANS to PNS (mm)
Mandibular length	Distance from point Ar to point Pog (mm)
Mandibular ramus height	Distance from point Ar to point Go (mm)
Mandibular body length	Distance from point Go to point Pog (mm)
Upper anterior facial height	Distance from point N to point ANS (mm)
Lower anterior face height	Distance from point ANS to point Me (mm)
Posterior face height	Distance from point PNS to point Go(mm)
Y-axis	Angle from FHP and S-Gn line (degree)

Table 2. Cephalometric Landmark on hard tissues[13]

Cephalometric Landmark	Definition
S	Sella: mid point of the fossa hipophysealis/sella turcica
N	Nasion: anterior point at the frontonasal suture
Ba	Basion: most posteroinferior point of the clivus
ANS	Anterior nasal spine: most anterior point of anteroposterior profile of the upper jaw
PNS	Posterior nasal spine: most posterior point of the bony palate defined by the junction of the hard palate, the soft palate, and the extension of the pterygomaxillary fissure
Ar	Articulare: the intersection of a line along the posterior border of the mandible and the inferior border of the basilar occipital bone
Pog	Pogonion: most anterior point of the mandibular profile in the mental region
Go	Gonion: point of intersection between the line bisecting the posterior and inferior border of the mandible and the contour of the chin
Me	Menton: most inferior point of the mandibular symphysis
Frankfort Horizontal Plane	FHP: Po-Or
Gn	Gnathion: most anteroinferior point on mandibular symphysis
Po	Porion: most superior point of the external auditory meatus
Or	Orbitale: the deepest point on the infraorbital margin

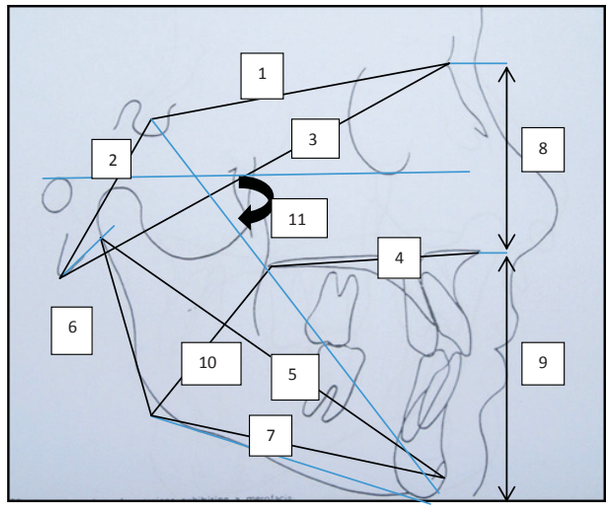

Figure 2. Measurement used in the cephalometric analysis[12] Linear measurements: 1) Anterior Cranial Base Length (S-N); 2) Posterior Cranial Base Length (S-Ba); 3) Cranial Base Length (N-Ba); 4) Maxillary Length (ANS-PNS); 5) Mandibular Length (Ar-Pog); 6) Ramus Height (Ar-Go); 7) Mandibular Body Length (Go-Pog); 8) Upper Anterior Facial Height (N-ANS); 9) Lower Anterior Facial Height (ANS-Me); 10) Posterior Facial Height (PNS-Go); Angular measurement: 11) Y-axis (FHP-S-Gn).[12]

RESULTS

Twenty eight subjects were finally selected. Characteristics of the groups are presented in table 3 with 14 subjects with complete unilateral cleft lip and palate which consist of 9 male subjects and 5 female subjects, whereas of the 14 subjects without cleft lip and palate which consist of 10 female subjects and 4 male subjects.

The measurement errors were calculated using Dahlberg formula. The error of measurement did not exceed 1.2 mm for linear measurement and 0.87 º for angular measurement. The normality test (Levene tests) was performed to asses differences in variance of each measurement. Normal distribution of data obtained thus followed by paired t-test to test the intergroup differences of measurement. The differences was considered significant for $p \leq 0.05$. The results of intra-observer suitability test shows no statistically significant difference between the first and second measurements for linear and angular measurements (p = .157 in linear measurements and p = .076 in angular measurement).

Table 3. Distribution of study subjects by sex and agethe cleft group and the normal group

	Cleft				Normal			
	N	%	Mean	SD	N	%	Mean	SD
Male	9	64.28	12.22	.83	4	28.57	12.75	1.71
Female	5	35.72	10.60	1.34	10	71.43	12.60	1.43
	14				14			

Shapiro-Wilk test results show that the overall measurement data has a normal distribution and Levene test results showed homogeneous data. With a normal distribution of data, unpaired t-test was performed to test the research hypothesis. The $p \leq 05$ value considered as statistically significant differences between the two groups with 95% confidence intervals were calculated for the difference of means for each variable.

There were statistically significant differences in the length of the anterior cranial base (SN, p = .02) and overall length of cranial base (N-Ba, p = 0.001) between patients with complete unilateral cleft lip and palate compared with control patients, but there was no statistically significant difference in the length of the posterior cranial base (S-Ba, p = .051).

There were statistically significant differences in the length of the maxilla (ANS-PNS) among patients with

Table 4. Intergroup differences in measured varibles detected by un*paired* t-test; 95% confidence interval (CI) calculated for the difference between means

Variable	Cleft group		Normal group		Difference	p
	Mean	SD	Mean	SD		
S-N	64.42	3.82	68.95	3.29	-4.53	.002*
S-Ba	45.70	4.74	48.65	2.59	-2.95	.051
N-Ba	99.12	5.88	106.53	5.12	-7.41	.001*
ANS-PNS	48.73	4.42	58.70	5.60	-9.98	.000*
Ar-Pog	96.42	6.14	105.78	5.71	-9.36	.000*
Ar-Go	38.96	3.84	44.11	2.86	-5.15	.000*
Go-Pog	71.07	5.53	78.73	5.99	-7.65	.002*
N-ANS	49.94	4.49	54.80	3.67	-4.86	.004*
ANS-Me	60.02	3.99	61.99	4.02	-1.97	.206
PNS-Go	40.70	6.70	40.34	4.26	0.36	.865
Y axis	63.50	3.06	61.71	4.08	1.79	.202

* $p \leq 05$ statistically significant difference

complete unilateral cleft lip and palate with a control group (p = .000).

There were statistically significant differences in the length of the mandibular ramus (Ar-Go, p = .000), length of mandibular body (Go-Pog, p = .002), and overall mandibular length (Ar-Pog, p = .000) between group of complete unilateral cleft lip and palate patients with a control group.

Anterior facial height was divided into upper anterior facial height (N-ANS) and the lower anterior facial height (ANS-Me). There is a statistically significant difference (p = .004) in the upper anterior facial height (N-ANS) between the groups but there was no statistically significant difference (p = .206) in the lower anterior facial height (ANS-Me).

Posterior facial height (PNS-Go) did not show a statistically significant difference (p = .189) between the groups of patients with cleft lip and palate in the control group. There was no difference in the pattern of growth and the facial type, measured as the acute angle formed by the FHP plane and S-Gn line (Y-axis) between the groups of patients with complete unilateral cleft lip and palate with a control group (p = .202).

DISCUSSION

This study was conducted with a cross-sectional design, the researchers conducted the measurement variable at a given moment. All subjects were observed only once and the subject variable measurement performed during the inspection. The research was conducted by utilizing lateral cephalometric radiograph made for the purposes of orthodontic treatment in patients with cleft lip and palate (secondary data) so as to prevent the patient from additional radiation. Cross-sectional design has the main advantage that is relatively easy, inexpensive, and the results are obtained faster. It can be used to investigate many variables at once, and can be used as a basis for further research that is more conclusive.[14]

Although the selection and determination of the subject of the research were conducted in a cleft center but there were constraints in choosing subjects that meet the criteria: 1) the limited number of pubertal age patients that owned lateral cephalographic radiograph because they were in the phase of evaluating the need to do alveolar bone grafting to prepare canine eruption in the area of the gap so most of the pubertal age patients only have orthopantomogram (OPG); 2) social-economic factor limiting orthodontist to refer patients to make a lateral cephalometric radiograph because it would add to the burden of the cost of care; 3) there were many variation of severity in cleft lip and palate so treatment needs may also differ between individual patients thus causing differences in treatment stages although patients were in the same age ranges.

Chronological age, dental development, sexual maturation, voice changes, and height are all ways that have long been used to assess skeletal maturation, but can not be used to determine the peak of the pubertal growth.[15,16] Hand-wrist radiographs can be used to assess the skeletal maturation.[15] However, the complexity in identification of landmarks can lead to less accurate predictions and the child will be exposed to additional radiation.[17] To overcome these shortcomings CVM used as a method of assessment of skeletal maturation stages by observing changes in the cervical spinal form and the CVM method is a reliable indicator for assesing skeletal maturity.[16] The peak of the pubertal growth occurred between CVMS II and III.[10] This method eliminates the need for additional radiographic exposure because it can be observed on routine lateral cephalometric radiographs.[16,18]

Lateral cephalometric radiographs used in this study to see the craniofacial morphology differences between children with of complete unilateral cleft lip and palate post labioplasty and palatoplasty with children without cleft lip and palate as it is a standardized method of measuring.[12] Cephalometric analysis is the most commonly used method to determine the dentocraniofacial morphology.[19] Cephalometric analysis is also a diagnostic support to determine the type and pattern of growth in the face so that the clinician can determine facial disharmony.[20] Lateral cephalometric shows craniofacial morphology and anatomical structures on the lateral direction.

The intraexaminer reliability were tested using paired t-test. Intraexaminer measurements test performed on the data showed that there was no significant difference between the results of the first measurement and the second measurement. This findings showed good intraoperator reliability. Levene tests showed homogeneous data (p >.05), this may be due to the inclusion criteria determining which sets the level of skeletal maturation as a guide in determining pubertal age in both groups and this result increased internal validity of the research result.

The results of this study showed a significant difference in the length of the anterior cranial base (SN) and overall cranial base length (N-Ba) between groups of children with cleft lip and palate compared with normal children. The length of the anterior cranial base (SN) and overall cranial base length (N-Ba) in the group of children with complete unilateral cleft lip and palate shorter than normal children.

Maxillary complex is the most affected part of the craniofacial structures because the existence of a gap, but growth disorders in children with complete unilateral cleft lip and palate are not limited only at the maxilla. It can be shown from the results of this research that there is a significant difference in both the length of the maxilla (ANS-PNS) and the dimensions of the mandible; mandibular length (Ar-Pog), mandibular ramus height (Ar-Go), and mandibular body length (Go- Pog) between groups of children with complete unilateral cleft lip and palate compared with the children without cleft lip and palate at pubertal age. The growth of the maxilla in the horizontal direction as measured by the distance between

the ANS-PNS in groups of children with cleft lip and palate that was shorter than the control group. It can not be determined for certain whether this was due to the surgical procedure but the growth of the maxilla in patients with cleft lip and palate who had surgery often restricted in the 3-dimensional direction.[21]

Mandibular length (Ar-Pog), mandibular ramus height (Ar-Go), and mandibular body length (Go-Pog) in children with complete unilateral cleft lip and palate significantly different compared with children without cleft lip and palate at the age of pubertal. The dimensions of the mandible in children with complete unilateral cleft lip and palate at pubertal age was relatively shorter than those of children without cleft. This is interesting because the mandible is not affected by either a gap or surgical procedures such as labioplasty and palatoplasty, however this is in accordance with the results of da Silva et al, who stated that patient with cleft lip and palate have a shorter mandibular length than individual without cleft lip and palate.[21] Mandibular length that is shorter in children with complete unilateral cleft lip and palate balanced with a cranial base length that is also shorter, which implies the existence of an equivalent growth in the mandible with anterior cranial base in the anteroposterior direction.

Although both the upper anterior facial height (N-ANS) and the lower anterior facial height (ANS-Me) in groups of children with complete unilateral cleft lip and palate shorter than normal children, but significant differences occur only in the upper anterior facial height (N -ANS) and there is no significant difference in the lower anterior facial height (ANS-Me) between the two groups. Significant differences in upper anterior facial height can be understood as a consequence of the existence of a gap in the maxilla. Maxillary growth affects not only in the anteroposterior and transverse direction, but also has an effect in the vertical direction.

Posterior facial height (PNS-Go) showed no significant difference between the two groups as well as the pattern of growth and the facial type (Y-axis). In a study conducted in adult patients with complete unilateral cleft lip and palate found a reduction in posterior facial height and the relationship between the rotation of the mandible in the cleft palate (downward and backward rotation) were also marked by the addition of gonial angle which also resulted in the addition of anterior facial height.[21] In this study there was no significant difference between the two groups in both the posterior facial height and Y-axis, but the Y-axis value in the groups of children with cleft lip and palate has a greater value than normal children (Table 4). This lateral cephalometric studies only examine the craniofacial structure in the anteroposterior and vertical direction, posteroanterior cephalometric or 3-D CT studies of the cleft are necessary to further examine the craniofacial morphology difference between the two groups in the transverse direction.

Based on this study it can be concluded that there were craniofacial morphology differences between children with complete unilateral cleft lip and palate post labioplasty and palatoplasty and children without cleft lip and palate at the age of pubertal. Children with complete unilateral cleft lip and palate post labioplasty and palatoplasty had shorter length of the anterior cranial base, cranial base, maxilla, mandible, mandibular ramus height, mandibular body, and upper anterior face height as compared with children without cleft lip and palate at the age of pubertal. The maxillary complex was most affected by cleft lip and palate but growth disturbance in chidren with complete unilateral cleft lip and palate were not restricted only at the maxilla.

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