Early Dental Arch Relationships as Treatment Outcome in Children with Complete Unilateral Cleft Lip and Palate in Northeastern Thailand

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The objective of the present study was to evaluate the dental arch relationships of children with complete unilateral cleft lip and palate (UCLP) as an early surgical repair outcome in the Northeastern part of Thailand. The study used a 5-year study model of 46 complete UCLP, non consecutive, patients whose primary repair had been performed at Khon Kaen University. The outcome of dental arch relationships was assessed using the 5-Year-Olds’ Index. Agreement of rating was assessed with weighted kappa statistics; both the intra- and inter-examiner agreements were high, indicating good reproducibility. The results indicated that the surgical outcome was graded as poor and very poor for 60% of patients and excellent, good, or fair for 40% of patients. This result was also poor compared with other overseas reports.

Keywords: Early treatment outcome, Dental arch relationships, Unilateral cleft lip and palate, 5-Year-Olds’ Index

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The malformations due to cleft lip and palate (CLP) present tremendous challenges for healthcare teams providing multi-disciplinary management1. There is general acceptance that treatment outcomes in cleft care need to be continually monitored; hence the development of new and better methods to assess their satisfactoriness. Previous studies suggest that both lip and palate repair may have a disproportionate influence on facial growth and dental arch development among young patients with CLP2,3. Dental arch relationships are likely therefore important indicators for the quality of cleft treatment outcome4.

There is a recommendation-as part of an internationally agreed protocol-that study models should be obtained at 5, 10, 15 and 20 years of age for individuals with cleft lip and palate5. The development of the 5-Year-Olds’ Index6,7 thus offers early assessment of one aspect of primary treatment outcomes prior to contamination by orthodontic treatment or alveolar bone grafting and this is likely to yield a truer assessment of the primary surgery. Furthermore, this index rates the dental arch relationships in the deciduous dentition of patients with complete unilateral cleft lip and palate (UCLP) into five categories: excellent, good, fair, poor and very poor6,7. The most important features of the classification include (a) anteroposterior arch relationships (b) followed by vertical relationships (deep bite, open bite) and (c) transverse relationships (extent of crossbite).

The Cleft Lip and Palate Center at Khon Kaen University, Thailand, was established in 1999. This Center is responsible for the management of patients born with CLP referred for treatment from districts of Northeast Thailand. This is the first time that the results of primary cleft surgery-using the relationships of the dental arch-have been assessed at this Center so it provides a level of information previously unavailable. The objective of the present study was to evaluate complete UCLP repair outcomes done at the Cleft Lip and Palate Unit, Faculty of Dentistry, Khon Kaen

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University, Northeast Thailand, by assessing the dental arch relationships on study models using the 5-Year-Olds’ Index(6,7).

**Material and Method**

**Sampling**
A retrospective study of non consecutive cases of patients with complete UCLP and available 5-year-old study models were identified from the database of the Cleft Unit. The criteria for inclusion of patients in the study were (a) having a repaired complete UCLP (b) being nonsyndromic and (c) having had the primary surgical repair undertaken at Khon Kaen University. A broad surgical protocol was employed, with clefts of the lip repaired at 3-6 months. Clefts of the secondary palate were closed at 10 and 12 months. In none of the cases was any primary bone grafting undertaken. A total of 46 subjects, who met the criteria, were identified. The mean age, when the present study models were obtained, was 5.25 years (SD, 0.42; range, 4 to 5.92).

**Measurement**
The present study models were graded by two examiners trained in the use of the 5-Year-Olds’ Index(6,7). The simplified assessment version of the 5-Year-Olds’ Index is shown in Table 1. The 10 reference models from Atack et al(6,7) were the study models used in the calibration course for the 5 Year Old Index. These study models represent the range of categories and have agreed upon scores. All study models were prepared in a standard fashion. Increasing the number of reference models into the present study models group helped to mask the present study models from the assessors. All 46 tested, study models and 10 reference models were independently assessed, twice per examiner, by two examiners. To minimize the possible influence of memory on the results, there was an interval of at least one week between each assessment.

The results of the assessments were used to illustrate the relationship of scoring for each examiner and also between examiners. The intra-examiner agreement was determined using the weighted kappa (k) statistic, which took into account the degree of disagreement within the results (>0.8 k value indicates very good agreement)(8). In addition, inter-examiner agreement was determined using components of variance, which indicate how much variation there was in the data (a) between observers (b) within observers and (c) between scorings. Statistical calculations were undertaken using the Statistical Package for the Social Sciences, Version 11 (SPSS Inc., Chicago, IL).

**Results**
The intra-examiner agreement was very good and the inter-examiner good (Table 2). The results of the grading of study casts from the KKU cleft database, using the Five Year Olds’ Index, are presented in Table 3.

**Discussion**
As part of an internationally agreed study protocol, the WHO(5) recommended that dental models should be obtained at 5, 10, 15 and 20 years of age for individuals with CLP. The rationale behind the development of the 5-Year-Olds’ Index(6,7) was that it

<table>
<thead>
<tr>
<th>Group</th>
<th>General Features Predicted Long-term</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>positive overjet with average inclined or retroclined incisors no crossbites or openbites good maxillary arch shape and palatal vault anatomy</td>
</tr>
<tr>
<td>2</td>
<td>positive overjet with average inclined or retroclined incisors unilateral crossbite/crossbite tendency ± open bite tendency around cleft site</td>
</tr>
<tr>
<td>3</td>
<td>edge-to-edge bite with average inclined or proclined incisors, or reverse overjet with retroclined incisors, unilateral crossbite ± open bite tendency around cleft site</td>
</tr>
<tr>
<td>4</td>
<td>reverse overjet with average inclined or proclined incisors unilateral crossbite ± bilateral crossbite tendency ± open bite tendency around cleft site</td>
</tr>
<tr>
<td>5</td>
<td>reverse overjet with proclined incisors bilateral crossbite poor maxillary arch form and palatal vault anatomy</td>
</tr>
</tbody>
</table>

±: with or without
Table 2. Intra-, and inter-examiner agreement

<table>
<thead>
<tr>
<th></th>
<th>Weighted Kappa</th>
<th>95% Confidential Interval</th>
</tr>
</thead>
<tbody>
<tr>
<td>Intra-examiner agreement</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Examiner 1</td>
<td>0.81</td>
<td>0.67-0.98</td>
</tr>
<tr>
<td>Examiner 2</td>
<td>0.86</td>
<td>0.69-1.00</td>
</tr>
<tr>
<td>Inter-examiner agreement</td>
<td></td>
<td></td>
</tr>
<tr>
<td>First assessment</td>
<td>0.72</td>
<td>0.58-0.80</td>
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<tr>
<td>Second assessment</td>
<td>0.77</td>
<td>0.60-0.89</td>
</tr>
</tbody>
</table>

Table 3. Comparison of the modified 5 Year Olds Index categories between other centers and Khon Kaen Cleft Database

<table>
<thead>
<tr>
<th>Index Grade</th>
<th>Bristol n = 46</th>
<th>Oslo n = 54</th>
<th>PMH* n = 54</th>
<th>KKU* n = 46</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ideal group (1 and 2)</td>
<td>35%</td>
<td>57%</td>
<td>28%</td>
<td>30%</td>
</tr>
<tr>
<td>Borderline group (3)</td>
<td>19%</td>
<td>28%</td>
<td>49%</td>
<td>10%</td>
</tr>
<tr>
<td>The worse group (4 and 5)</td>
<td>46%</td>
<td>15%</td>
<td>23%</td>
<td>60%</td>
</tr>
</tbody>
</table>

*PMH: Princess Margaret Hospital, Western Australia, 'KKU: Khon Kaen University Cleft Center

offers early assessment, but also long-term predictions, of the outcomes of primary surgery. This evaluation of the end-use quality of surgery enables a review and possibly modification of cleft management[9].

Comparisons of the outcomes of UCLP repair in the current study can be compared with reports form overseas centers at Bristol[10], Oslo[11], PMH (Perth)[12] and Khon Kaen (KKU) using the 5 Year Old Index (Table 3). Cases ascribed to either Group 4 or 5 represent poor surgical results which may require additional orthognathic surgery during late adolescence, whereas those classified as either Group 1 or 2 are considered as having good long-term growth outcomes. The KKU cleft database sample was assessed as having 30% of patients falling into either Group 1 or 2 while 60% had unsatisfactory results and fell into either Group 4 or 5. The “borderline group” (Group 3) comprised 10% of the sample. When these results are compared with other multicenter studies (Table 3), it can be seen that the results are indeed poorer than those achieved in the UK, Norway and Western Australia.

In Bristol, PMH and KKU, primary surgery was performed by a number of different operators; in Oslo, the records obtained were from a single surgeon. The better results produced by Oslo may indicate a benefit in high treatment volume and greater experience of the surgeon, as suggested in the Eurocleft study[9]. Timing of repair was also of interest; Bristol, PMH and KKU performed palatoplasty at 6 to 12 months, and in Oslo this procedure was performed at 18 months.

Other factors possibly affecting the differences in results of the center comparisons are ethnic and genetic backgrounds[13,14]. Studies of craniofacial morphology in Asian children reveal that they usually had a naturally occurring higher prevalence of underlying Class III skeletal relationships[13,14]; this might also be true among Thai children although no such study has been published. A naturally occurring higher incidence of underlying skeletal Class III relationship might lead to a higher proportion of the 5-Year-Olds’ Index categories of 4 and 5 that would not necessarily be related to treatment protocols. This would support the fact that the underlying skeletal relationships cannot be ignored when drawing conclusions regarding any effects from treatment on outcomes.

Dental caries and premature loss of primary teeth by extraction has an adverse influence on the occlusal relationship[15]. This was also implied in the results of the present study, as 90% of cleft groups had unfavorable oral health care and a high risk of early childhood caries (ECC) because of the habit of nighttime bottle feeding. In the present study, almost half of the patients had extensive caries in the upper anterior region used in making a decision for categorizing patients according to the 5-Year-Olds’ Index. This may
complicate assessing the results of surgical outcomes; so further studies about this effect are needed.

Conclusion

The current study supports dental arch relationship analysis as a valuable tool for early evaluation of treatment outcomes in UCLP patients, with the strong caveat that appraisal of other parameters (i.e., facial appearance, speech and hearing) is essential for completing the complex picture of cleft care outcomes.

Acknowledgement

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Potential conflicts of interest

None.

References

ผลลัพธ์การศัลยกรรมระยะแรกในผู้ป่วยเด็กปากแหว่งโหว่ชนิดข้างเดียวในภาคตะวันออกเฉียงเหนือของประเทศไทย

ทัศนีย์ วัตตรมังคละ, มนต์เย็น มโนสุดประสิทธิ์, สิทธิ์ กิตติพงษ์, วัลลภ จันทร์สว่าง, กมลวรรณ เจนวิถีสุชา

วัตถุประสงค์ของการศึกษานี้เพื่อศึกษาความสัมพันธ์ของส่วนโค้งแนวฟันในผู้ป่วยเด็กปากแหว่งโหว่ชนิดข้างเดียวในภาคตะวันออกเฉียงเหนือของประเทศไทยซึ่งใช้ประเมินผลลักษณะของการรักษาระยะแรกโดยใช้ความสัมพันธ์ของส่วนโค้งแนวฟันในผู้ป่วยปากแหว่งโหว่ชนิดข้างเดียวแบบสมบูรณ์ จำนวน 46 รายไม่เป็นผู้ป่วยรายต่อเนื่องกันที่มารับการรักษาโดยการผ่าตัดในระยะแรกที่มหาวิทยาลัยขอนแก่น และมีแบบฟันจำลองในการรักษาในขณะอายุ 5 ปีถูกรวบรวมเพื่อนำมาศึกษาหาความสัมพันธ์ของส่วนโค้งแนวฟัน โดยให้ค่าอายุ 5 ปีในการประเมินผลการศึกษาพบว่าผลการประเมินความเห็นสอดคล้องในผู้ประเมินและระหว่างผู้ประเมินอยู่ในเกณฑ์ระดับดี สำหรับการศึกษาของผลของการผ่าตัดระยะแรกของความสัมพันธ์ของส่วนโค้งแนวฟันและการรักษาพบว่ามันถูกต้องจากการผ่าตัดซึ่งพิจารณาจากความสัมพันธ์ของส่วนโค้งแนวฟันอยู่ในระดับดีและไม่ได้ที่สุดยอดละ 60 และอยู่ในระดับดีเยี่ยม ดีและปานกลาง ระดับละ 40 ดังนั้นผลลัพธ์จากการผ่าตัดจึงมีความคุ้มค่าในระดับที่มากกว่าการศึกษาในประเทศอังกฤษ, นอร์เวย์ และออสเตรเลียตะวันตก