Right Technique, Right Time: A Retrospective Analysis on Dental Arch Relationships for Patients with Total Unilateral Cleft Lip and Palate. A Comparison between Early and Late Closure of the Hard Palate.

Filippa Sibe
Tulika Ganoo
Right Technique, Right Time: A Retrospective Analysis on Dental Arch Relationships for Patients with Total Unilateral Cleft Lip and Palate. A Comparison between Early and Late Closure of the Hard Palate.

Filippa Sibe and Tulika Ganoo

Tutors: Mats Sjöström and Lena Björnström
ABSTRACT

Surgical closure of cleft palate improves function and esthetics but affects facial growth. The effect depends on age of the patient at the time for surgery. The study aimed to compare early versus late closure of the hard palate and its effect on dental arch relationship at the age of five for patients born with unilateral cleft lip and palate (UCLP).

The study included 40 non-syndromic, Caucasian children with UCLP. All patients had their surgeries performed by the regional cleft-team at University Hospital of Umeå, Sweden, according to the protocol for treatment of UCLP. The patients were divided into early closure (operation age: approximately 2 years, n = 20) and late closure (operation age 4 - 8 years, n = 20). Dental arch relationships were analyzed on dental casts (n = 36) or clinical photos (n = 4) taken at the age of five using the modified Huddart and Bodenham (mHB) scoring system.

The results showed that there was significant difference (P = 0.035) in mHB total score between early closure (median - 6.69) and late closure (median - 3.63). Children who had an early closure of the hard palate had a statistically significant lower mHB total score, and hence worse dental arch relationship compared to children with a late closure.
INTRODUCTION

Background information about cleft lip and palate (CLP)

CLP is a congenital combination of two conditions: cleft lip, where the medial nasal process and maxillary process fail to connect during the sixth embryonic week and cleft palate, wherein the maxillary process fails to fuse with the primary palate and the other maxillary process (Brand, 1994). The fusing of the palate occurs in an anterior to posterior direction, which implies that location of the cleft depends on when in the embryonic development, the failure occurred (Brand, 1994). The etiology is complex and believed to be a combination of genetic and environmental factors (Watson et al., 2001; Mossey et al., 2009; Lidral and Murray, 2004). Approximately 1.7 of 1000 children are born with CLP (Mossey et al, 2009).

Clefts vary in appearance and severity: from a tiny defect just affecting the lip or the uvula, to more severe involving many structures in the orofacial region at the same time (lip, nose, alveolar process, hard and soft palate). A total cleft can affect one side or both sides of the face (Hupp et al., 2014). 70 % of the total CLPs are unilateral, UCLP, which mean that the cleft affects the lip, nose, alveolar bone, hard and soft palate on only one side of the face (Mastroiacovo et al., 2011).

Treatment of UCLP is important as the condition affects patients psychosocially, biologically and physically. The muscular function of the mouth, speech and sometimes hearing as well as the development of jaw and teeth is affected (Hupp et al., 2014).

Presurgical treatment of UCLP

Orthopaedics is sometimes used to optimize the effects of surgery by implementing it presurgically and as a complement to primary surgeries. For example, naso-alveolar moulding encourages growth as it prevents collapse of the maxillary arch and reduces its severity by separating tongue from cleft area. Presurgical moulding makes the lip surgery easier by aligning the alveolar segments (Shetty et al., 2017). The effect of presurgical orthopaedics is debated, as certain studies have found that their effect on arch form is not significant and that lip repair (the first of the primary surgeries) has greater effect on arch form (Adali et al., 2012). Other studies show that the benefits can be found up to 6 years
of age and that it leads to correct maxillary arch form in patients with UCLP (Jorge et al., 2016; Shetty et al., 2017).

**Surgical treatment of UCLP**

Treatment procedures for UCLP differ around the world. Generally, the lip is operated as early as possible (at approximately 10 weeks). Then, the palatal cleft is closed either in one step (operating both the soft and hard palate during a single procedure) at approximately 8-18 months or two steps (operating the soft palate first and then the hard palate) (Hupp et al., 2014).

The cleft in the alveolar bone is treated with bone graft at the age of six to ten years to create conditions for the lateral incisor or canine to erupt to normal position in the dental arch. The bone graft creates a good bone support for the teeth in the cleft area (Hupp et al., 2014).

There are different techniques for hard palate closure as part of a one-stage process such as: the von Lagenback operation and the ‘push back’ operation developed by Veau. The soft palate can be operated by among others intravelar veloplasty, primary pharyngeal flap and Furlow’s double opposing Z-plasty (Watson et al., 2001).

In the two-stage procedure, the soft palate can be operated using among others, by the following methods: intravelar veloplasty, the Malek regime and the delaire regime. Good maxillary development may be found in intravelar veloplasty as the tension of the muscle slings brings together the two parts of the maxilla, narrowing the hard palate cleft and thus reducing the need to make any lateral incisions (Watson et al., 2001). Concerning the Malek regime and De Mey’s technique, “excellent maxillary growth is reported even after hard palate surgery has been completed by six months” (Watson et al., 2001). Closure of the palate in two stages shows good results on maxillary growth (Friede et al., 2012).

Several studies with the aim to investigate differences in impact on maxillary growth between various surgical techniques for palatal closure (for example the vomer flap technique and palatal flap technique) were considered to be of low quality. Therefore, there is no evidence that one specific method is better than the other to minimize the
negative effects on maxillary growth (Lee and Liao, 2013).

There seems to be no differences in maxillary growth between closure of the palate in one or two steps (Farranto et al., 2014).

**The relationship between surgical treatment of UCLP and dental arch relationships**

Patients with UCLP who have not been surgically treated for their defect seem to have a quite normal growth potential of their maxilla (Shi and Losee, 2014) and difference in growth compared to individuals without UCLP is insignificant (Bishara et al., 1976; Mars and Houston, 1990).

Surgical treatment of UCLP inhibits growth of the maxilla (Shi and Losee, 2014) by causing denervation, devascularization and disturbance of the periosteum and scarring (Watson et al., 2001). Of the primary surgeries the most negative effect on the maxillary growth is caused by the closure of the palate (Farranto et al., 2014). The bone graft treatment to close the cleft in the alveolar bone at age 9 - 11 shows minimal inhibitory effect on maxillary growth since most of the growth is completed at the time for the bone grafting. The closure of the cleft lip showed an inhibitory effect on maxillary growth in sagittal dimension (Shi and Losee, 2014).

Watson et al. (2001) refer specially to an article by Ross (in McCarthy) which states that despite defects in the cleft area, the teeth and maxillary bone can produce a satisfactory occlusion. According to it, the main impact on maxillary growth due to surgery is because of scar tissue produced. The scar tissue also disrupts the eruption of teeth and thus affects the form of the dental arch. Interestingly it also states that the “traditional techniques of surgery” do not have a great impact on maxillary growth and that instead, the greatest variable is the surgeon.

This view is explained by K. Salyer in his article “Unilateral cleft lip and cleft lip reconstruction” (Bardach and Morris, 1990), as he explains that the most important factor in achieving optimal results is having a proper diagnosis which requires an experienced, technically and theoretically skilled surgeon.
Early vs. Late closure of the hard palate

There are several advantages for early closure of the palate: better development of the palatal and pharyngeal muscles, improved ability to phonate sounds, better function of the auditory tube and better hygiene as the oral and nasal areas are separated. For the parents, early closure has advantages as they may have easier to feed the child. Further it has a positive effect psychologically on the baby and the parents (Hupp et al., 2014). Early closure also gives the child a better chance of develop normal speech (Watson et al., 2001).

However, there are certain disadvantages with early closure: surgery on younger children is more difficult because of smaller structures and early scar formation because of surgery can restrict maxillary growth (Hupp et al., 2014). Early surgical closure of the palate inhibits the maxillary growth in all dimensions (vertical, sagittal and transversal) (Shi and Losee, 2014).

To permit unrestricted maxillary growth the best solution is to wait with hard palate surgery as long as possible. It can be delayed at least until the deciduous teeth have erupted (Hupp et al., 2014). Long-term negative effects (reduced maxillary growth) of early surgery outweigh any short-term benefits (for example better aesthetics and easier feeding) but “for speech and hearing considerations, palatal closure at the age of 12 months is currently performed” (Kuokkanen et al., 2008).

A systematic review from 2014 concludes that late closure of the palate is better for the facial growth than early closure. But since there are more factors to have in mind when to decide the best timing for closure of the palate, for example speech development, there is no current evidence for the best timing (Farranto et al., 2014).

Evaluation of primary surgery on growth

There are several ways to evaluate the impact of primary surgery on maxillary growth, for example analyzing dental arch relationships or cephalometrics (American Cleft Palate – Craniofacial association, 2008).

From a clinical point of view, dental arch relationship is important, as it is essential for
oral function. Good dental arch relationship means a reduced need for orthodontic treatment and orthognathic surgery in the future.

**Analyzing the dental arch relationship**

For evaluation of the dental arch relationships in UCLP patients there are many indexes available:

*Goslon Yardstick:* analyses dental arch relationships in horizontal, vertical and transversal relation and categorizes in groups based on orthodontic treatment needs. As it categorizes based on treatment needs, it requires training and calibration (Haque *et al*., 2015).

*Five-year index:* developed by Atack to specifically analyze dental arch relationships at five years, so that surgeons can easily analyze treatment outcomes. It is also categorical and requires calibration (Haque *et al*., 2015).

*The Eurocran index:* developed by participants at Eurocran 2000-2004, is a modification of the GOSLON and the five-year index. It analyses both the dental arch relationship as well as the palatal form. This index requires previous knowledge about dental arch relationship and orthodontic treatment needs and thus is more difficult to learn (Haque *et al*., 2015).

*Modified Huddart and Bodenham index (mHB):* uses scores instead of categories. All primary teeth pair, except for the lateral incisors are scored based on their buccal-palatal relation (Appendix 1). The score for each tooth pair is then summarized to a total score for the dentition. The score can be used to see how much the dental arch relationship differs from the ideal arch (which would have a total score of 0) (Tothill and Mossey, 2007). It is not directly treatment linked as compared to the other indexes and does not include anterior-posterior or vertical scoring. However, it is simple to use and gives a simplified picture of the dental arch relationship (Haque *et al*., 2015).

There are several reported advantages of the mHB index. It is objective and reliable (Mossey *et al*., 2003; Haque *et al*., 2015). It is versatile: it can be used for patients at any age after eruption of primary teeth (Mossey *et al*., 2003; Tothill and Mossey, 2007; Haque *et al*., 2015). It is more sensitive to interarch discrepancies as it has a finer subdivision
(Hathorn and Mars, 1996; Manosudprasit et al., 2011; Haque et al., 2015) and it is easy to learn and does not require clinical experience (Tothill and Mossey, 2007).

**Treatment of UCLP at University Hospital of Umeå (NUS), Sweden**

The cleft team at NUS is responsible for treatment of cleft patients living in Norrbotten, Västerbotten, Västernorrland and Jämtland county. Over time, different surgical protocols have been used: since the eighties, plastic surgery of the lip is performed at the age of approximately three months, followed by two-stage closure of the palate. First, the closure of soft palate, performed at approximately six to eight months, followed by the hard palate a few years later. The team has changed the timing for hard palate closure from the age of four to eight years to performing it at the age of two years.

**Aim of the study**

The aim of the study is to retrospectively compare two different surgical treatment protocols (early versus late closure of the hard palate) and their effects on dental arch relationships at the age of five for patients with UCLP.

**MATERIALS AND METHODS**

**Materials**

Inclusion criteria were children with UCLP operated according to the surgical protocol for treatment of UCLP by the regional cleft team at NUS (Västerbottens läns landsting, 2014).

Children with UCLP combined with a syndrome diagnosis, children who had surgeries performed at other centers and children with non-Caucasian ethnic origin were excluded. The study was limited to children of Caucasian origin to limit the influence of genetic factors (the prevalence of prenormal occlusion differs among ethnic groups (Chang et al., 2006)).

The patients’ dental arch relationship was analyzed using study casts made when the child was five years old. In cases where study casts weren’t available, photos were used. Patients with no study casts or with photos of low quality were excluded from the study.
The full material consisted of 40 patients. 36 cases were evaluated with study casts and four with clinical photos. They were divided into two groups: Early closure of the hard palate and late closure of the hard palate. A summary of the patients is reported in Table 1.

By the age of five years, the “early closure” patients have undergone all the initial surgeries to treat their UCLP except the bone grafting procedure in contrast to the “late closure” group who only have had their two initial surgeries.

In both groups, the patients had still not received orthodontic treatment and thus, their occlusal relationships were a result of surgical intervention and genetic factors.

**Analysis of dental arch relationships**

The dental arch was analyzed using the modified Huddart and Bodenham (mHB) index. It was performed by four different examiners: an orthodontist, a maxillofacial surgeon both with a lot experience of working with cleft patients, and two dental students.

Before the analysis of the dental casts belonging to this study, training and calibration in using the scoring system was performed at three different occasions. The calibration was performed on dental casts of young children with CLP not involved in this study.

The dental casts were coded using a random number generator where in casts from both groups were assigned a number randomly. The operations history of the patients involved in the study (head surgeon, age at every operation, cleft location and gender) was noted.

The study was conducted two weeks after the coding. The scores were recorded by the four examiners independent of each other. The measurements were again repeated after two to four weeks to measure reliability.

Afterwards the key was broken, the models were divided into the correct groups, and the results were analyzed with Microsoft Excel and IBM SPSS using the Mann-Whitney U test. Mann-Whitney is a non-parametric test to identify differences between two independent groups and it can be used on data without normal distribution. The level of significance was set at 5%. Intra- and inter-examiner reliability was analyzed using Chronbach’s alfa reliability test which analyses reliability between measurements and a
score of 1 is achieved if the measurements are identical.

**Ethical reflection**

In this study, dental casts of five-year-old patients were used. Using dental study casts is recommended by the Americleft Study Project (American Cleft Palate – Craniofacial association, 2008) as these are routinely taken for any orthodontic intervention, the procedure is non-invasive /low risk for the patient and enables patient privacy protection.

By analyzing the treated patients in groups, no personal data is published and it is not possible to recognize an individual patient. As the study casts were previously made as part of the treatment of the patients, they experience no discomfort due to this study. Instead the aim is that through this study, future patients may have the possibility of a better treatment. The study is conducted for the quality assurance of the CLP team and data is collected as permitted by the law (SOSFS 2008:14, 2 kap. 20 §). The Ethics Forum at the Department of Odontology, Umeå University has approved the study.

**Obtaining relevant literature**

For background knowledge, books about facial growth and development, treatment of CLP and orthodontics available at the medical library in Umeå were consulted. A literature search was performed on PubMed with the following keywords: “Unilateral cleft lip and palate”, “UCLP”, “cleft lip and palate”, “prevalence”, “etiology”, “surgery”, “primary surgery”, “closure of palate”, “facial growth”, “maxillary growth”, “results”, “dental arch relationships” “Goslon yard stick”, “Huddart and Bodenham index”, “Eurocleft”. Relevant articles were also obtained from the tutors.

**RESULTS**

To analyze the differences between the groups, we chose to calculate the average mHB total score for each dental cast based on all examiners’ scores from both measurements. We found a statistical significant difference in the mHB total score between the early closure group and the late closure group (P = 0.035). The early closure group had a more negative score than the late closure group, thus they had a smaller upper dental arch in
relationship to their lower dental arch than the late closure group (Figure 1). The median of the early closure group was -6.69 while for the late closure group it was -3.63 (Table 2). There was no statistical significant difference in total score between females and males (Figure 2).

To evaluate where in the dentition the difference in score between the groups was located, we calculated a “front score”- total score for only the central incisors, and a “lateral score”- total score for all teeth pairs except incisors. There was no statistical significant difference in front score between early closure and late closure (p = 0.157). Instead the difference in score between early and late closure group was located in the lateral segment: the late closure group had a higher lateral score than the early closure group (Table 2). It was statistically significant (p = 0.030).

Intra-examiner reliability was as follows: Examiner 1 (0.97), Examiner 2 (0.98), Examiner 3 (0.82), Examiner 4 (0.80), considered good-excellent. Inter-examiner reliability based on the first measurements was 0.93; considered excellent.

**DISCUSSION**

The results of this study suggest that if closure of the hard palate is performed later in the child's life, the growth of the maxilla is less inhibited than if it is performed earlier. A delayed closure of the hard palate seems to give a more favorable inter-arch relationship at five years of age. This conclusion agrees with literature (Farranto et al., 2014). Other literature suggests that dental arch relationships are similar even if hard palate surgery is performed at different ages (but before 10 years of age) (Novarrez et al., 1993).

In Umeå, the cleft team has changed timing for closure of the hard palate, from late (at age 4 - 8 years) to now, early closure (at approximately 2 years). The results from our study indicate that there is a worse outcome on the inter-arch relationship at the age of five with the current treatment protocol for UCLP than with the protocol that the team previously had.

Whether this difference in the inter-arch relationship is relevant enough to revise the protocol and delay palatal closure can be discussed. As mentioned earlier, the timing for closure of the hard palate depends on combination of factors: maxillary growth, normal
development of speech, creating a better ability to eat and better comfort level for the patient (Hupp et al., 2014). Delayed closure of the hard palate has also been associated with higher numbers of speech errors compared to earlier closure (Willadsen et al., 2017). If the closure is performed later, this must also be weighed against the patient's experience of living with the cleft in the palate for a longer time. Of interest is also whether this difference in dental arch relationships at five years of age, impacts further need of treatment, for example orthodontic treatment and orthognathic surgeries and the final result.

An interesting follow-up to this study would therefore be to analyze the amount of orthodontic treatment and orthognathic surgery required based on jaw-relationship at five years of age and hence compare the following treatment plan for individuals based on which primary surgical treatment they received. Of interest would also be to compare the speech development and patient experience between the early and late closure group.

A broad conclusion from our study is problematic because of the small sample size, due to the limited number of patients at the center. This also meant that the study included patients that had slightly different care, for example: patients with delayed surgeries due to other illnesses or patients that developed slight complications and had additional surgery.

Patients in this study had varying cleft sizes which could have affected the result. However, we believe that the variation in cleft size is similar in both groups and hence the results are comparable.

Patients in the same group received different kinds of pre-surgical treatment to optimize the effects of surgery. However, the largest impact on maxillary growth is by surgery and hence the effect of pre-surgical treatment on jaw relation should be smaller. It is also difficult to determine the effect of the pre-surgical treatment as there is no record of compliance.

To minimize the effect of random measurement errors the scoring of dental casts was repeated and average mHB score of all the examiners was calculated. Therefore, random errors should not completely affect the results.
If this study was performed again, it would be desirable to have a larger sample size and find a way to exclude the impact of above mentioned variables, so that it could be certain that the score only depends on the timing of closure of the hard palate.

A strength with the study was the using of the mHB index that allowed us to have a high inter-examiner reliability, and which according to the literature, is an appropriate tool for evaluation of dental arch relationships in patients with UCLP (Hathorn and Mars, 1996; Mossey et al., 2003; Tothill and Mossey, 2007; Manosudprasit et al., 2011; Haque et al., 2015). The scoring system was easy to learn which was an important aspect for us, as the team who performed this study consisted of dental students with limited clinical experience.

In literature, the surgeon’s skills have been mentioned to be of great impact for the outcome of dental arch relationships and sometimes suggested it may be of greater impact than the timing of closure (Watson et al., 2001). Interestingly in this study, the surgeries in the late closure group were performed by different surgeons, therefore suggesting that the surgeons did this kind of surgery more rarely. The patients in the early closure group were treated by the same surgeon; hence the surgeon performed this kind of surgery more frequently. This implies that the early closure group had a worse dental arch relationship even though the surgeon was experienced in the field (has performed more UCLP surgeries).

This quality assurance work was conducted by the CLP team at NUS to analyze the effects of different surgical protocols on jaw relationships. We hope that this study opens up for future studies conducted by the CLP team and provides them with one of the puzzle pieces to continue improving their treatment plan for children with UCLP and optimizing the timing of their surgeries.

ACKNOWLEDGEMENTS

We want to thank our tutors of this project Mats Sjöström and Lena Björnström for sharing their knowledge and advices; and Anders Esberg for advice with the statistical analyzes.
REFERENCES


**Tables**

**Table 1.** Information about the patients included in the study for both the “Early closure group” and the “Late closure group”. The table shows number of patients, distribution of females and males, distribution of left and right sited UCLP and the patients’ age when the lip and nose surgery, soft palate surgery and hard palate surgery were performed.

<table>
<thead>
<tr>
<th></th>
<th>Total number of patients</th>
<th>Females</th>
<th>Males</th>
<th>Left sided UCPL</th>
<th>Right sided UCPL</th>
<th>Age at lip &amp; nose surgery</th>
<th>Age at soft palate surgery</th>
<th>Age at hard closure of cleft palate</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Early closure group</strong></td>
<td>20</td>
<td>5</td>
<td>15</td>
<td>10</td>
<td>10</td>
<td>3-11 months</td>
<td>4-11 months</td>
<td>1 year 4 months-2 year 9 months</td>
</tr>
<tr>
<td><strong>Late closure group</strong></td>
<td>20</td>
<td>7</td>
<td>13</td>
<td>10</td>
<td>10</td>
<td>1.5-10 months</td>
<td>6 months-1 year</td>
<td>4-8 years</td>
</tr>
</tbody>
</table>
Table 2. Modified Huddart and Bodenham total score, front score (score for central incisors) and lateral score (score for lateral teeth) for early and late closure groups.

<table>
<thead>
<tr>
<th>N</th>
<th>Mean ± Std. Deviation</th>
<th>Median</th>
<th>Min</th>
<th>Max</th>
<th>95% CI for mean</th>
<th>P-value</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Total score</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>20</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Early closure</td>
<td>-6.09 ± 3.59</td>
<td>-6.69</td>
<td>-14.00</td>
<td>2.00</td>
<td>-7.77 – -4.41</td>
</tr>
<tr>
<td></td>
<td>Late closure</td>
<td>-4.05 ± 3.73</td>
<td>-3.63</td>
<td>-15.00</td>
<td>2.00</td>
<td>-5.80 – -2.30</td>
</tr>
<tr>
<td></td>
<td>Front score</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>20</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Early closure</td>
<td>-1.40 ± 1.53</td>
<td>-1.37</td>
<td>-4.13</td>
<td>1.38</td>
<td>-2.11 – -0.68</td>
</tr>
<tr>
<td></td>
<td>Late closure</td>
<td>-0.75 ± 1.70</td>
<td>-0.50</td>
<td>-3.63</td>
<td>2.00</td>
<td>-1.55 – -0.05</td>
</tr>
<tr>
<td></td>
<td>Lateral score</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>20</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Early closure</td>
<td>-4.69 ± 2.62</td>
<td>-4.94</td>
<td>-11.75</td>
<td>0.13</td>
<td>-5.91 – -3.46</td>
</tr>
<tr>
<td></td>
<td>Late closure</td>
<td>-3.30 ± 2.61</td>
<td>-2.94</td>
<td>-11.63</td>
<td>0.38</td>
<td>-4.52 – -2.08</td>
</tr>
</tbody>
</table>
Figure 1. Distribution of modified Huddart and Bodenham total score for Early and Late closure group.
Figure 2. Distribution of modified Huddart and Bodenham total score for females and males.
APPENDIX:

**Image 1:** Scoring guideline for the modified Huddart and Bodenham index

Obtained from (Dobbyn et al. 2011)

For this index, if the maxillary tooth is located palatal to its mandibular equivalent, the pair gets a more negative score, while if the maxillary tooth is located buccal to the mandibular tooth, the gets a more numerically positive score.