# Successful Management of Cleft Lip and Palate Malformation without Pre-Surgical Infant Orthopedics

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***Objective:*** *To investigate the effect of lip closure on reduction of cleft palates when no pre-surgical infant orthopedics (PSIO) are used.* ***Study design****: Retrospective patient chart-review in our department for Cranio-Maxillofacial Surgery at the University Medical Centre Freiburg, Germany. 19 patients at the age of 5.9 ± 2.1 months with surgical treatment of uni- (UCLP), or bilateral cleft lip and palate (BCLP) without any use of PSIO were included.* ***Results:*** *Early soft tissue correction of the lip leads to an effective reduction of the maxillary arch without any use of PSIO. The presented conventional and digital measurements appeared to be reliable. A successful reduction of the cleft width (UCLP = 3.88 ± 2.42mm, BCLP = 7.33 ± 5.00mm), the width of the alveolar arch (1.91 ± 1.36mm) and the sagittal depth of the alveolar arch (3.07 ± 2.71 mm) could be achieved with the presented workflow.* ***Conclusions:*** *Cleft reduction was obtainable without PSIO when lip closure after Tennison-Randall was performed.*

*Keywords: Pre surgical infant orthopedics, cleft reduction, Tennison-Randall, infant orthopedics*

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## INTRODUCTION

Patients afflicted by CLP mostly experience a protracted treatment process from early infancy until adulthood.[1] Even though countless treatment concepts exist, there is still no generally accepted consent until today.[1] Despite the inhibitory effect on facial growth, the advantages of early reconstruction of orofacial structures have to be considered, such as an improved development of speech and better deglutition. Presurgical infant orthopedics (PSIO) are frequently used to decrease the extent of the cleft deformity before surgery and claim to facilitate subsequent operation procedures [2]. Contrary to this assumption treatment regimens without pre-surgical orthodontic treatment exist equally and lead to satisfying cleft reduction[2–4]. As epidemiological data underlines, the optimization of treatment concepts for patients with CLP is an international health issue, affecting a high number of patients worldwide. This study evaluates the change of the maxillary arch and cleft width in 19 patients with UCLP/BCLP, who did not receive PSIO during their treatment. In the first step two different measurement methods assessing maxillary arch dimensions were applied and analyzed regarding precision and reliability. In a second step the measurement results were analyzed considering the reduction of cleft width and maxillary arch dimensions to investigate the outcome of the presented treatment regimen.

## MATERIALS AND METHOD

19 patients with UCLP or BCLP treated at the medical center of the Albert-Ludwigs-University Freiburg, Germany between 2009 and 2019 were included in this study. The performing surgeon as well as the treatment method were the same for the entire study group. All patients received a two-step surgical treatment: In a first procedure at the age of 5.9 ± 2.1 months a lip closure was performed according to the technique of Tennison-Randall. At the age of 10.4 ± 1.8 months a closure of the palate was conducted according to the technique of Langenbeck-Veau-Ernst-Axhausen. Seven female and twelve male patients were included of which 11 patients had UCLP and 8 patients had BCLP. Ethical approval (No. 547/18) was obtained from the ethics committee of the Albert-Ludwigs-University of Freiburg, Germany. Exclusion criteria were differences in treatment (e.g. perioperative orthodontic treatment), syndromal forms of facial clefting and poor quality of the dental plaster casts. The present study evaluates two different measurement methods for the assessment of maxillary arch dimensions and cleft width. For every patient two plaster casts were produced at two different times– before and four months after lip closure. On each plaster model the distances S1-S1', S1-S2, S1'-S2', C2-C2', I-C2M (Table 1 and 2, Fig. 1 and 2) were determined by one rater manually and digitally at the beginning of the study and six weeks later. Dental cast impressions were taken during anesthesia at the beginning of each operation using Alginoplast ® (Kulzer Mitsui Chemicals Group, Hanau, Germany) for dental impression and special hard plaster pico-crema soft ® type 3 DIN EN ISO 6873 (Picodent, Wipperfürth, Germany) to create the plaster model. To evaluate changes in the maxillary arch and cleft region, the landmarks shown in Table 1 were set at the plaster models. Subsequently, the distances shown in Table 2 were determined with two different methods. Method 1 was carried out manually using a vernier caliper with a nonius of 0.05 mm (Scale type 16 FN, Mahr, Göttingen, Germany). The measurements were determined to the second decimal place. Method 2 included a digitalization step and subsequent computer-based measurement. First a 3D-surface scan of the plaster model was generated using a surface scanner (3shape, Kopenhagen, Denmark). Afterwards digital measurements were done using the software tool GOM Inspect (GOM, Braunschweig, Germany). Measurements between two landmarks were taken automatically and were specified to the third decimal place. The distance I-C2M could only be determined with the digital measuring technique, as virtual lines had to be constructed to get the required landmarks. Figure 1 and 2 show a digitized 3D-scan of a UCLP (Fig. 1) and a BCLP (Fig. 2) and the according measurements using GOM Inspect. The plaster models as well as the 3-D scans were measured by one observer with six weeks apart using both measurement methods two times. In collaboration with the Institute of Medical Biometry and Statistics of the University Freiburg, Germany the intra-rater reliability and accuracy of both methods were determined using a paired t-test. Subsequently, the different measurement methods were analyzed regarding a statistically significant difference using the one-sample t-test. A variation coefficient was calculated to evaluate the accuracy of both approaches. Eventually the significance of the reduction of cleft width and maxillary arch dimensions was investigated using a one sample t-test.

## RESULTS

To evaluate intra-observer reliability of the different methods, the measurement results of the digital and the manual measurement procedure were compared using a paired t-test (Fig. 3). No statistically significant differences of the two measurement procedures could be shown (t= 0.804, p= 0.424). Furthermore the differences of the first and second measurement round were compared concerning their differences and led equally to insignificant result for all measured distances (S1-S1': t=0.473, p=0.640, S1-S2: t=0.320, p=0.753, S1'-S2': t=(-)0.488, p=0.633, C2-C2': t=0.365, p=0.717).

As both techniques turned out to be reliable, the mean values of both techniques were considered for further investigations. The variation coefficient as quotient of the standard deviation for both methods appeared to be comparable. The coefficient of variation (COV) of the manual measurement method was 0.79 and for the digital measurement method 0.77. The clinical outcome was evaluated based on the reduction of cleft width and maxillary arch dimensions, described by the following distances S1-S1', S1-S2, S1'-S2', C2-C2', I-C2M (Fig. 1 and 2). A significant reduction of the mentioned distances could be accomplished for all five measurements. Table 3 shows pre- and postoperative distances as well as the resulting reduction of cleft width with standard deviation.

## DISCUSSION

The beneficial effects of PSIO in treatment of children with CLP are still discussed controversially. Separation of the nasal and oral cavity with orthodontic appliances can prevent abnormal tongue positioning. This seems to be helpful in guiding the direction of growth of the maxilla [5]. Additionally a positive reduction of the cleft width in the pre-surgical stage of treatment was observed when PSIO was used [3]. This may facilitate subsequent surgical closure of CLP [2]. Nevertheless recent meta-analyses demonstrate that the long-term effect of PSIO-treatment should be questioned critically [2–4]. Even though PSIO can help to reduce cleft width before surgery, equal results could be achieved in patients who did not receive any orthodontic treatment. Therefore, the benefits of PSIO should be evaluated carefully [2–4,6].

The non-significant paired t-test, which was performed to compare both measurement rounds for differences, indicates that both methods are reproducible. As the variation coefficient for both methods is comparable, both methods are equally precise and reliable. The manual method can be performed without much material costs and does not need any post-processing steps of the plaster model. One major advantage of the digital measurement technique is that complex 3D-analyses could be performed using the 3D-surface scan[7]. Furthermore, archiving of digital surface models is less costly and could easily be integrated in a pre-existing hospital computer environment. Digital measurements could be archived as well, while no damage is caused to the original model by digital measuring processes. Measurements of the dental casts by a second observer to evaluate inter-rater reliability were not performed in this study.

Besides technical questions, one major aspect of the study was to evaluate the success of treatment management in patients with CLP without PSIO. In UCLP the corresponding distance is S1-S1'. In BCLP S1-S2 is representative for the right and S1'-S2' for the left alveolar cleft (Fig. 1 and 2). With the proposed method UCLP

**Figure 1: Plaster model for UCLP with according distances.**

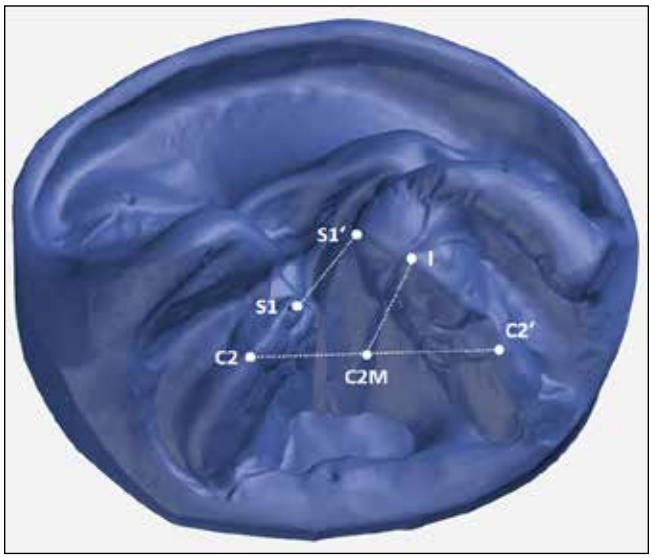


**Figure 2: Plaster model for BCLP with according distances.**

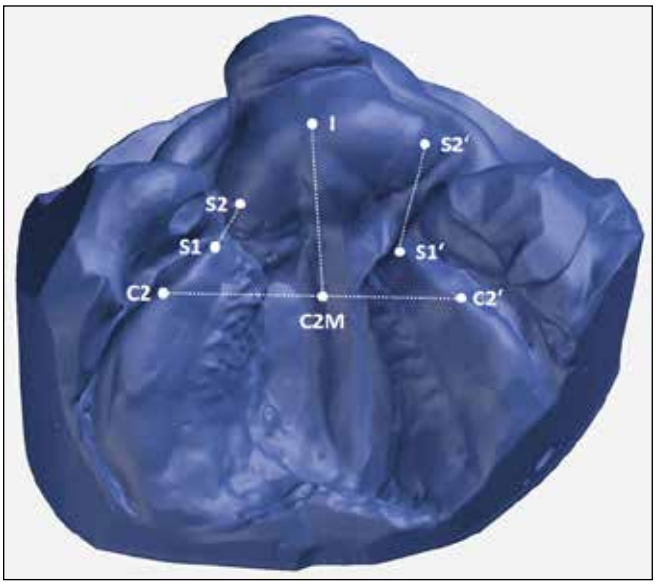


**Figure 3: Comparison of the manual and digital measurement methods. The mean value and confidence intervals for both measurement methods appeared to be comparable.**

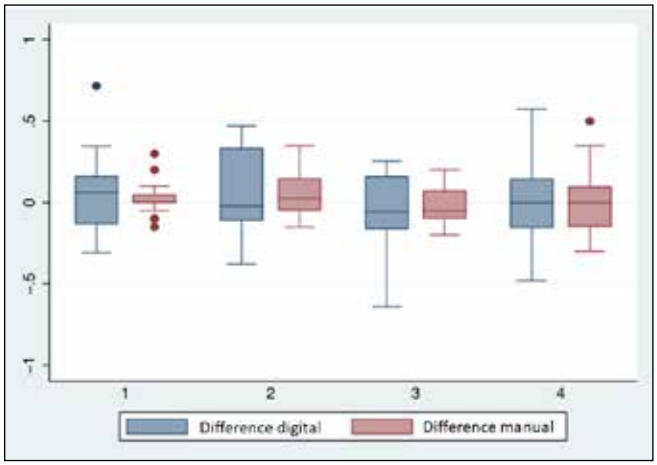


**Table 1**

| Landmark | Description |
|---|---|
| I | Intersection of the alveolar ridge with an imaginary line between the incisive papilla and the frenulum labii superioris.[9,10] |
| S1, S1' | UCLP: Distal and mesial cleft edge points. [9,10] BCLP: Lateral segment margin of cleft (S1 right, S1' left) [11] |
| S2, S2' | Only in BCLP: Premaxillary margin of cleft (S2 right, S2' left) [11] |
| C2, C2' | Canine points, where the lateral sulcus crosses the crest of the ridge (C2 right, C2' left) [11] |
| C2M | Middle point between C2 and C2' [11] |

**Table 2**

| Distance | Description |
|---|---|
| S1 – S1' | Connecting line of S1 and S1' |
| S1 – S2 resp. S1' – S2' | Connecting line of corresponding cleft edge points right (S1 and S2) resp. left (S1' and S2') |
| C2 – C2' | Connecting line of C2 and C2' |
| I – C2M | Connecting line of I and C2M |

**Table 3**

| Distance | Measurement method | Preoperative value in mm | Postoperative value in mm | Reduction in mm | p-value |
|---|---|---|---|---|---|
| S1 – S1' (UCLP) | digital | 8.90 ± 3.72 | 5.05 ± 2.33 | -3.86 ± 2.49 | 0.0004 |
| | manual | 8.87 ± 3.72 | 4.97 ± 2.36 | -3.9 ± 2.47 | 0.0004 |
| S1 – S2 (BCLP) | digital | 7.93 ± 4.19 | 3.88 ± 1.92 | -4.04 ± 3.14 | 0.0082 |
| | manual | 7.88 ± 4.12 | 3.81 ± 1.82 | -4.07 ± 3.12 | 0.0078 |
| S1' – S2' (BCLP) | digital | 7.57 ± 4.91 | 4.27 ± 2.44 | -3.3 ± 3.3 | 0.0253 |
| | manual | 7.56 ± 4.90 | 4.30 ± 2.49 | -3.26 ± 3.25 | 0.0251 |
| C2 – C2' (UCLP and BCLP) | digital | 31.00 ± 4.19 | 29.13 ± 4.11 | -1.88 ± 1.69 | 0.0001 |
| | manual | 31.05 ± 4.11 | 29.11 ± 4.10 | -1.94 ± 1.62 | 0.0001 |
| I – C2M (UCLP and BCLP) | digital | 12.47 ± 3.92 | 9.40 ± 2.29 | -3.07 ± 2.71 | 0.0001 |
| | manual | - | - | - | - |

were reduced by 3.88 ± 2.42mm and BCLP were reduced by 7.33 ± 5.00 mm. In addition, the width of the maxillary arch at the level of the canine points (C2-C2') was decreased by 1.91 ± 1.36 mm and the depth of the maxillary arch from the incisive papilla to the connection line C2-C2' (I-C2M) was decreased by 3.07 ± 2.71 mm. Thus, in our study group significant cleft width reduction was achieved without any use of PSIO. Furthermore the maxillary arch dimensions in the sagittal (C2-C2') and coronal plane (I-C2M) were reduced in size. Reduction of maxillary arch dimensions has shown to be one of the major aspects to improve the functional and aesthetic outcome in patients with CLP [8].

## CONCLUSION

The results of this study show that it is possible to achieve satisfying cleft width and maxillary arch reduction with surgical treatment only. As the use of PSIO is associated with additional costs and stress on the newborn patient, we recommend to critically evaluate its use in the management of patients with CLP.

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