Early One-Stage Repair of Complete Unilateral Cleft Lip and Palate

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Background: The purpose of this prospective study was to evaluate craniofacial morphology in children with complete unilateral cleft lip and palate treated at the Brussels cleft center after a 1-stage complete closure at 3 months and compare the results with a series of children operated on at 3 and 6 months of age according to the Malek surgical protocol.

Methods: A series of 72 consecutive patients who were operated on for nonsyndromic complete unilateral cleft lip and palate were included in this study at approximately the age of 10 years. Thirty-four were treated according to the Malek surgical treatment protocol: the soft palate was closed at a mean (SD) age of 3.04 (0.20) months, followed by simultaneous repair of the lip and hard palate at 6.15 (0.67) months. Thirty-eight underwent 1-stage all-in-one (AIO) closure of the lip and hard and soft palates at 2.98 (0.16) months. Craniofacial morphology was evaluated by means of a digital cephalometric analysis. Cephalometric data were compared with a noncleft control group (n = 40) matched according to age. The same 2 series of children were followed up until 15 years of age, and the results were again compared.

Results: Statistical analysis (analysis of variance with post hoc Tukey test) showed in both groups who were operated on a decreased anteroposterior growth compared with the children without cleft at 10 years but the AIO group only was not different from the group without cleft. The maxillary (MxP/ISN) plane was significantly (P = 0.002) increased in the Malek cleft group compared with the AIO group with cleft. At 15 years of age, a difference was not observed anymore between the 2 groups for the anteroposterior growth or for the maxillary plane inclination.

Conclusions: One-stage AIO closure based on the Malek surgical principles provided good anteroposterior midfacial morphology and resulted in less opening of the maxillary plane to the anterior cranial base.

Key Words: All-in-one, unilateral cleft lip palate, craniofacial morphology, cephalometry

Many different surgical treatment protocols and adjunctive orthopedic/orthodontic procedures have been described to treat children with unilateral cleft lip and palate (UCLP).1-4 In the cleft center of the Reine Fabiola Children’s Hospital, children with UCLP have been treated for more than 20 years by primary lip closure at 6 months followed by closure at the palate according to Veau-Wardill at 18 months. A retrospective study showed a bad anteroposterior growth in 66% of patients treated according to the later protocol.5 Therefore, since 1981, the Malek6,7 surgical technique and sequence, which consists of early soft palate closure at 3 months followed by simultaneous closure of the lip and hard palate at 6 months, has been adopted.

Based on the experience with the Malek protocol, 1-stage all-in-one (AIO) closure of the lip and hard and soft palates was introduced in 1988 in the Brussels cleft center to treat children with UCLP.8

One-stage AIO closure as a repair for patients with UCLP was first published in the literature by Farina9 in 1958. Other publications by Davies10 and Kaplan et al11-13 reported on long-term experience after AIO closure. The preliminary results of the Brussels cleft center with this technique were presented at the 7th14 and 9th15 International Congress on Cleft Palate and Related Craniofacial Anomalies. Other occasional reports on 1-stage repair in patients with cleft were from Davies and Fernandes,16 Standoli and Cordaro,17 Honigmann,18 and Honigmann and Scherrera.19 The first documented long follow-up series was published in 2008 by Fudaley et al.20

The purpose of this nonrandomized prospective study was to compare craniofacial morphology at the age of 10 years, after 1-stage AIO closure to a noncleft control group and to a group with cleft treated according to the Malek surgical treatment protocol, then observe the patients until the age of 15 years.

MATERIALS AND METHODS

The inclusion criteria for this nonrandomized prospective study were the following:

(1) Complete unilateral cleft lip, alveolus, and palate; patients with small Simonart bands were included
(2) Consecutive cases
(3) No associated syndrome or mental retardation
(4) White ethnicity
(5) Presurgical infant orthopedics with passive removable appliances
(6) Measurement of anterior and posterior cleft margins during initial primary cleft surgery
(7) All children must have received their primary and secondary surgeries at the Reine Fabiola Children’s University Hospital
(8) Lateral cephalometric radiographs at the age of 10 and 15 years.

Patients

Between November 1981 and August 1993, a total of 132 children with complete UCLP were operated on at the Reine Fabiola...
Children's University Hospital. A total of 72% fulfilled the inclusion criteria.

Thirty-four were operated on following the Malek protocol, and 38 underwent closure of their cleft after the AIO surgical procedure in the period from November 1987 to August 1993. The cleft size was peroperatively measured with a calliper anteriorly at the alveolar cleft margins and posteriorly at the level of the posterior nasal spine. One-stage AIO closure was only performed in case the posterior cleft distance was less than 10 mm; otherwise, the Malek protocol was adopted.

All patients were given prophylactic antibiotics such as cephalosporin for 5 days.

Malek Cleft Group

The soft palate was closed according to Malek and Psaume and Malek7 at a mean (SD) age of 3.04 (0.20) months. The malpositioned palatal muscles were detached from the palatal shelves and hamulus and carefully sutured in the midline.

The lip was repaired following Tennisson-Randall21 at a mean age of 6.15 (0.67) months, and the hard palate was closed in one layer using a vomer flap simultaneously with the lip closure. All primary surgical procedures were performed by 2 plastic surgeons, M.L. and A.D.M., who performed 14 (42%) and 20 (58%) of the primary operations, respectively. Children who were operated on starting from 1985 (n = 29) had primary nose correction according to McComb simultaneously with the lip closure. The anterior cleft distance ranged from 2 to 15 mm (mean, 10.96 [3.87] mm), whereas the posterior cleft distance ranged from 7 to 15 mm (mean, 10.96 [2.27] mm).

The mean operative times were 40 minutes (range, 35–60 min) for the first operation and 65 minutes (range, 55–85 min) for the second operation.

The mean hospital stays were 4 days for the first operation and 6 days for the second.

All-in-One Cleft Group

One-stage AIO closure of the lip and hard and soft palates was performed according to the Malek surgical principles at a mean age of 2.98 (0.16) months but in one operative session only. The lip was repaired following Tennisson-Randall; the hard palate was closed using a vomer flap without elevating the oral fibromucosa, whereas the soft palate was closed with great care to correctly reposition the palatal muscles. Most of the surgical procedures were performed by one surgeon (A.D.M.), who operated on 36 children (95%), whereas M.L. operated on 2 children (5%). Primary nose correction according to McComb was performed in all children simultaneously with the AIO closure. The anterior cleft distance ranged from 1 to 10 mm (mean, 6.11 [4.01] mm), whereas the posterior cleft distance ranged from 5 to 11 mm (mean, 8.17 [4.01] mm).

The mean operative time was 95 minutes (70–125 min).

The mean blood loss was 34 mL (range, 5–70 mL), and the mean hospital stay was 6 days.

Orthopedic/Orthodontic/Secondary Treatment

Both cleft groups underwent passive presurgical orthopedics using a removable acrylic appliance. Presurgical orthopedics was initiated immediately after birth except for the first 12 patients of the Malek group who received the plate approximately 6 weeks preoperatively as recommended by Malek. Active removable appliances were used during the deciduous and mixed dentitions to correct small anterior or lateral crossbites, if necessary.

Data on secondary surgical procedures of the Malek and AIO cleft groups are summarized in Figure 1.

Digital Cephalometry

Craniofacial morphology was evaluated by means of lateral cephalometric radiographs carried out on the patients at approximately the age of 10 years: AIO cleft group (age range, 8.9–11.5; mean age, 10.21 [0.73] y) and Malek cleft group (age range, 8.8–11.2; mean age, 10.48 [0.63] y). The children with clefts were compared with a sample of 40 untreated children (20 boys and 20 girls; age range, 9.2–11.0; mean age, 10.29 [0.42] y) with normal skeletal relationships randomly selected from the files of the Department of Orthodontics and matched according to age with the cleft samples. The Malek cleft and the noncleft control groups were already analyzed in previous conventional cephalometric studies.22,23 The same lateral cephalometric radiographs were taken at approximately the age of 15 years in both groups.

For this study, all lateral cephalometric radiographs were digitized using a calibrated scanner (Epson Expression 1600 Pro Epson Inc, Long Beach, CA; 1600 × 3200 dots per inch; color depth, 24 bits) at 800 dots per inch and stored into a Windows XP–based graphic Workstation (Pentium IV; 24 GHz; 512 megabytes; calibrated 17-inch color monitor; resolution, 1280 × 1024 pixels). Digital cephalometry was performed using the Onyx Ceph software (version 2.5.6; Image Instruments GmbH, Chemnitz, Germany). Digitizing of each landmark was done directly on the personal computer screen by 2 investigators (G.S. and N.C.) independently. Craniofacial morphology was evaluated using a modified lateral cephalometric cleft analysis that was validated in a previous study22 in which the hard tissue landmarks and cephalometric measurements were described. All linear variables were adjusted to the cranial base length (Ba-N) by the Onyx Ceph software.

Statistical Analysis

To determine the accuracy of tracing and digitizing, measurements were made twice of all patients independently. The measurement error was quantified by the SD of the differences between duplicate measurements on the same variable according to Bland.24

Because of the small AIO female cleft sample size and because our previous studies22,23 showed no significant sex differences in linear and angular measurements, male and female patients were pooled for the AIO cleft, Malek cleft, and noncleft control groups. Analysis of variance (with post hoc Tukey test) was used to compare the mean cephalometric values of the cleft and noncleft groups matched according to age. Differences were defined as statistically significant at the 0.05 level. All

FIGURE 1. Secondary surgery.
computations were performed using the SPSS statistical package (SPSS 11.5 for Windows).

## RESULTS

### Measurement Error

A total of 5544 hard tissue measurements were performed: 2856 linear and 2688 angular measurements. Analysis of duplicate measurements according to Bland\(^25\) revealed that 31 (94.0%) of 33 variables showed measurement errors of less than 1.40 degrees and 1.40 mm. This is in agreement with the literature.\(^26\)

### Cephalometric Craniofacial Morphology

The results of the cephalometric analysis at 10 years of age are summarized in Table 1.

The anterior cranial base (S-N) was less in both cleft groups compared with the noncleft group but reached only a significant level in the Malek cleft group (\(P = 0.003\); Table 1).

The AIO-Malek comparison data showed no significant differences regarding maxillary protrusion at the skeletal (Ba-N-ANS) or dentoalveolar (SNA) level. Compared with the noncleft control group, there was only a significant (\(P = 0.009\)) decrease in anterior maxillary height (R-PMP) and posterior maxillary height (R-PMD) was more open in both cleft groups compared with the control group and significantly (\(P = 0.002\)) increased in the Malek cleft group compared with the AIO cleft group (Table 1). Evaluation of facial depths and vertical relations showed no significant differences between both cleft groups. The AIO cleft group showed excellent anteroposterior development of the maxillary bone (Ba-ANS) and dentoalveolar process (Ba-A). The mandibular plane (MxP/SN) was more open in both cleft groups compared with the control group and significantly (\(P = 0.002\)) increased in the Malek cleft group compared with the AIO cleft group (Table 1). Evaluation of facial depths and vertical relations showed no significant differences between both cleft groups. The AIO cleft group showed excellent anteroposterior development of the maxillary bone (Ba-ANS) and dentoalveolar process (Ba-A). The mandibular plane (MxP/SN) was more open in both cleft groups but significantly more in the Malek (\(P = 0.000\)) cleft group as the AIO cleft group (\(P = 0.012\)) compared with the noncleft group (Table 1). Evaluation of mandibular sagittal position to the maxilla showed no significant differences at the skeletal (ANS-N-Pog), dentoalveolar (ANB), and dental (overjet) levels.

The results of the cephalometric analysis at 15 years of age are presented in Table 2. At 15 years of age, we could not compare our measurements to a control group of patients without cleft.

The analysis of the results showed no statistical difference between the 2 groups even for the inclination of the palatal plane.

## Secondary Surgery

A total of 18 patients (35%) in the Malek cleft group underwent lip correction at a mean age of 4.38 (2.13) years (Fig. 1). Three revisions (7%) were performed in the AIO cleft group. Ten patients (29%) in the Malek cleft group underwent a rhinoplasty consisting of correction of the nostril and ala associated with a septoplasty at 4.83 (2.71) years. In the AIO cleft group, a total of 11 patients (28%) underwent rhinoplasties at a mean age of 5.52 (2.69) years, consisting of correction of the nostril and ala in 8 (21%) and associated with a septoplasty in 3 patients (7%). A velopharyngoplasty was necessary in 4 Malek patients (11%) with cleft at a mean age of 5.07 (0.32) years and in 3 AIO patients (7%) with cleft at 5.5 years. In the Malek cleft group, a total of 21 fistulas (14 oronasal and 7 vestibulonasal) was observed, but closure was performed in 15 children (44%) at a mean age of 3.31 (1.57) years, whereas in the AIO cleft group, a total of 16 patients had fistulas (11 oronasal and 5 vestibulonasal). Twelve were closed for phonic reasons at a mean age of 4.80 (2.15) years. During the investigation time, secondary alveolar bone grafting was performed in 27 patients (72%) in the Malek cleft group at a mean age of 8.40 (1.05) years and in 23 patients (60%) in the AIO cleft group at a mean age of 8.61 (1.07) years.

## DISCUSSION

Evaluation of a new treatment protocol for patients with cleft is essential because it may have its specific influence on craniofacial morphology and growth. There is a common belief among cleft teams that early 1-stage repair of hard and soft palates with the lip affects maxillofacial development adversely. This has not been proven by long-term clinical studies.

One-stage AIO closure of patients with cleft lip and palate was introduced by Farina in 1958.\(^9\) However, since then, only few long-term clinical studies\(^10,11\) have been reported in the literature. Because none of the latter studies presented reliable evidence-based...
developed unique experimental animal models which includes a small.

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These experimental studies indicate that simultaneous lip and palate repair caused more severe craniofacial growth disturbances than lip or palate repair performed separately,27,28 or in sequence.29 These are however experimental studies and their results cannot be directly adapted to the clinical setting as started by Bardach et al.28 Savaci et al.6 compared the results obtained after 1- or 2-stage complete repair of UCLP between 6 and 12 months to a control group of unaffected children. The patients had their surgical orthopedics with a passive appliance. He concludes that 1- or 2-stage repair affects maxillolofacial growth in UCLP patients to the same degree. Therefore, they recommend the 1-stage closure for offers important advantages. Recently, a study of Fudaley et al.20 evaluated the long-term results of midfacial growth in preadolescent children after 1-stage simultaneous repair of cleft lip and palate with the same conclusion as Savaci et al. Our study evaluates and compares craniofacial morphologic after 1-stage AIO and 2-stage cleft closure in patients with UCLP at approximately the age of 10 years with a series of children without cleft. Then, the same series of children is evaluated by the same methods at approximately the age of 15 years.

data, Bardach et al.27 developed unique experimental animal models in rabbits and beagles28,29 to investigate the effect of simultaneous lip and palate repair on craniofacial growth. The results of these experimental studies indicated that simultaneous lip and palate repair caused more severe craniofacial growth disturbances than lip or palate repair performed separately,27,28 or in sequence.29 These are however experimental studies and their results cannot be directly adapted to the clinical setting as started by Bardach et al.28 Savaci et al.6 compared the results obtained after 1- or 2-stage complete repair of UCLP between 6 and 12 months to a control group of unaffected children. The patients had their surgical orthopedics with a passive appliance. He concludes that 1- or 2-stage repair affects maxillolofacial growth in UCLP patients to the same degree. Therefore, they recommend the 1-stage closure for it offers important advantages. Recently, a study of Fudaley et al.20 evaluated the long-term results of midfacial growth in preadolescent children after 1-stage simultaneous repair of cleft lip and palate with the same conclusion as Savaci et al. Our study evaluates and compares craniofacial morphologic after 1-stage AIO and 2-stage cleft closure in patients with UCLP at approximately the age of 10 years with a series of children without cleft. Then, the same series of children is evaluated by the same methods at approximately the age of 15 years.

A digital lateral cephalometric analysis that was validated for cleft research in a previous study24 was used to analyze skeletal craniofacial morphology. The AIO-Malek comparison data revealed only 1 significant difference (MxPL/SN at P = 0.002; Table 1). Although the maxillary plane (MxPL) was more open to the anterior cranial base (S-N) in both cleft groups compared with the noncleft group as in the study of Swennen et al.,27 Savaci et al.,27 and Fudaley et al.,29 it was significantly more open in the Malek cleft group compared with the AIO cleft group. This can be explained because posterior maxillary height (R-PMP) was significantly (P = 0.000) less in both cleft groups, whereas anterior maxillary (N-ANS) height was only significantly less in the AIO cleft group (P = 0.001) compared with the control group (Table 1).

The literature is confusing and conflicting regarding the use of vomer mucosal flaps for anterior hard palate repair. It has been reported that vomer flaps cause a reduction of maxillary vertical height, a more retruded maxilla, and an overclosure of the mandible owing to scar tissue adhesions that unite the vomer to the palatal shelves at the site of the repaired cleft.31,52 Evaluation of craniofacial outcome of the Oslo treatment protocol (primary lip closure using Millard’s procedure with simultaneous hard palate closure with a vomer flap at 3 months)31 followed by soft palate closure with a von Langenbeck palateplasty at 15 to 18 months revealed a reduction of posterior face height, a retrusive maxilla, but also a retrusive mandible.34 On the contrary, the 6-center study of treatment outcomes of the European Cleft Lip and Palate Research Group30,35 clearly identified the use of a vomer flap to close the anterior palate as a therapeutic factor associated with good outcome. Our previous studies22,23 confirmed the reduction in maxillary vertical height but revealed excellent anteroposterior maxillary morphology and a more open mandible in the Hannover and Malek cleft groups, which were both treated with vomer flaps.

Cephalometric assessment of Rene Malek cases by Ross36 also showed excellent midfacial development in 10-year-old patients with UCLP. The literature therefore seems to agree on the inhibiting effect of vomerine mucosal flaps on the vertical dimension of the maxilla but remains conflicting regarding the horizontal dimension of the midface. We suggest that the vomer flap in the AIO cleft group is responsible for the decrease in both posterior and anterior vertical heights. As far as the Malek cleft group is concerned, we suggest that posterior vertical maxillary height decrease is related to the veloplasty according to Malek and Psaume,6 which includes a small vomer flap.37–39 Moreover, according to Enlow and Hans,37 the direction of maxillary growth is primarily vertical during the initial stage of growth. Hence, the hard palate closure with a vomer flap in the Malek cleft group, which was carried out 3 months later compared with the AIO cleft group, might have caused less early anterior vertical maxillary growth disturbance.

This would explain the more open maxillary plane in the Malek cleft group. The mandible was more open to the anterior cranial base (MdPL/SN) in both cleft groups and more in the Malek cleft group (P = 0.000). As we already reported in previous studies22,23 and by Fudaley,20 we suggest that the increased inclination of the mandibular plane in the Malek cleft group is a consequence of mouth breathing and maxillary morphology. The increased inclination of the maxillary plane in the Malek cleft group and its concomitant more open mandibular plane illustrates this. However, the hypothesis of Molsted38 that presurgical orthopedics alters tongue position and causes the mandible to be inclined in a more backward direction is not supported by the results of this study. The AIO cleft group underwent presurgical orthopedics with passive appliances from birth and showed less backward mandibular inclination compared with the Malek cleft group, in which presurgical orthopedics was initiated 6 weeks before soft palate closure.

The AIO-Malek comparison data revealed no significant differences in the anteroposterior dimension of the midface. Moreover, the angles of maxillary protrusion (Ba-N-ANS, Ba-N-A) that reflect the forward translation of the maxilla and give a good indication of anteroposterior growth were excellent in the AIO cleft group and showed no significant differences compared with the Malek cleft group. In addition, the effective length of the maxilla (MxPL), which includes both basal maxillary length and the position of the alveolus on it, and pharyngeal depth (Ba-PMP), which indicates the anterior of the maxilla, were likewise excellent in the AIO cleft group and showed no significant differences to the Malek cleft group. Cephalometric measurement of the maxillary prominence to the anterior cranial base (SNA) is known as an insensitive measure of outcome35,39 and should therefore be interpreted with caution.

At 15 years of age, no difference was noted between the 2 groups for any of the studied measurement. This means that

| TABLE 3. Midfacial Morphology at 10 and 15 Years of Age: AIO |
|---------------|-------|-------|---------|
| Yr            |       |       | Tukey   |
| 10 y          | 15 y  |       |         |
| SNA           | 78.64 | 3.52  | 76.41   |
| SNB           | 75.14 | 3.33  | 74.69   |
| ANB           | 3.50  | 3.2   | 1.56    |
| MxPL          | 10.01 | 3.72  | 8.39    |
| MdPL          | 35.01 | 4.2   | 35.62   |

| TABLE 4. Midfacial Morphology at 10 and 15 Years of Age: Malek |
|---------------|-------|-------|---------|
| Yr            |       |       | Tukey   |
| 10 y          | 15 y  |       |         |
| SNA           | 76.49 | 4.79  | 76.44   |
| SNB           | 73.89 | 4.2   | 74.69   |
| ANB           | 2.60  | 3.51  | 1.54    |
| MxPL          | 12.85 | 4.12  | 10.15   |
| MdPL          | 38.18 | 5.03  | 36.36   |
the advantage of a better anteroposterior growth observed in the AIO series at 10 years of age is no more observed at 15 years of age. The comparison of the measurements at 10 or 15 years of age in the 2 series (Tables 3 and 4) confirms this observation.

Secondarily, operation was needed in both groups. A different number of corrections were needed for the lip, but the Malek group presented with larger clefts. Nose correction and pharyngoplasty were needed in a similar number in the 2 series. Fistulas were observed in both groups.

In the Malek group, the fistulas occurred most frequently at the junction of the hard and soft palates. At this level, during the second operation, consisting in the closure of the hard palate with the lip, we had to deal with some scar tissue at the level of the posterior vomer flap. This could be an explanation of the problem. In AIO closure, the fistulas observed mostly more anteriorly were smaller and gave seldom speech problems.

Despite our effort to perform a well-designed prospective comparative study, following the recommendations of Shaw et al., this study still has several shortcomings. First, although we attempted to avoid methodological biases, such as detection, exclusion, analysis, reporting, and proficiency bias, this study nevertheless clearly shows susceptibility and sampling biases. The anterior and posterior cleft dimensions were less in the AIO cleft group (susceptibility bias), and 2 different treatment protocols were used concurrently in the same cleft center (sampling bias). The patients were not randomized (selection bias) but selected by the surgeon for the AIO procedure, if the preoperative measured posterior cleft distance was less than 10 mm. Second, because of the prospective study design and the strict inclusion criteria, the size of the AIO cleft group at 15 years was still too small to discern statistically minor surgical effects using multiple comparison tests. Furthermore, the variable sample sizes might have influenced the statistical power of the pairwise post hoc Tukey tests.

However, as started by Shaw, as the number of operation and working surgeon decreases, the success of therapeutic results in increases.

CONCLUSIONS

Because both of the surgical protocols give rise to similar maxillofacial development outcomes regardless of the type of surgery, the 1-stage procedure offers several important advantages for the treatment of patients with UCLP.

1. only 1 anesthesia and hospitalization for primary surgery with minimalization of potential risks and less psychologic stress for the child and parents.
2. potential of less growth disturbance due to less scar tissue.
3. no more secondary operations, and
4. ideal for countries where the health system cannot afford multistage multidisciplinary treatment.

Although this paper presents long-term quantitative data on craniofacial outcome after 1-stage AIO closure at 10 and 15 years of age, evaluation of craniofacial morphology at the end of skeletal growth combined with dental cast analyses, speech, and hearing evaluations are mandatory in the future.

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