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B. Offert
P. Pazera
J. Janiszewska-Olszowska
K. Hozyasz
C. Katsaros
P. S. Fudalej

Dental arch relationship in 5-year-olds with complete unilateral cleft lip and palate after early alveolar bone grafting

Authors' affiliations:
Barbara Offert, Piotr S. Fudalej, Department of Pediatric Surgery, Institute of Mother and Child, Warsaw, Poland
Pawel Pazera, Christos Katsaros, Department of Orthodontics and Dentofacial Orthopedics, University of Bern, Bern, Switzerland
Joanna Janiszewska-Olszowska, Department of Orthodontics, Pomeranian Medical University, Szczecin, Poland
Kamil Hozyasz, Department of Pediatrics, Institute of Mother and Child, Warsaw, Poland
Piotr S. Fudalej, Department of Orthodontics, Palacky University, Olomouc, Czech Republic; Department of Orthodontics and Craniofacial Biology, Radboud University Nijmegen Medical Centre, Nijmegen, The Netherlands; Cleft Palate Craniofacial Unit, Radboud University Nijmegen Medical Centre, Nijmegen, The Netherlands

Correspondence to:
Piotr S. Fudalej
Department of Orthodontics and Craniofacial Biology
Radboud University Nijmegen Medical Centre
PO Box 9101
6500 HB Nijmegen
The Netherlands
E-mail: orthodontics@dent.umcn.nl

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Structured Abstract

Objective – To evaluate dental arch relationship in preschoolers with unilateral cleft lip and palate after early alveolar bone grafting (ABG).

Materials and methods – Three raters blindly assessed the dental arch relationship with the GOSLON Yardstick (using a 5-point scale, from 1 – very good to 5 – very poor outcome) in *Early-grafted* group (27 boys and 15 girls; mean age = 5.2 years, SD 0.5) and *Non-grafted* group (17 boys and 12 girls; mean age = 5.8 years, SD 0.8). The groups differed regarding the age when ABG was performed: between 2 and 4 years (mean = 2.4, SD 0.6) in the *Early-grafted* group and after 9 years in the *Non-grafted* group. The strength of agreement of rating was evaluated with kappa statistics.

Results – The intra- and inter-rater agreement was high ($\kappa > 0.800$). The mean GOSLON score in the *Early-grafted* group was 2.72 and in the *Non-grafted* group –2.64. The distribution of the GOSLON grades in the *Early-grafted* group was: 54.8% had a score 1 or 2, 23.8% – 3, and 21.4% – 4 or 5; in the *Non-grafted* group, 38.0% subjects scored 1 or 2, 41.4% – 3, and 20.6% – 4 or 5 ($p = 0.023$).

Conclusions – *Early* alveolar bone grafting carried out between the ages of 2 and 4 years was not found to negatively affect dental arch relationship by the age of 5 years. However, it is possible that such a negative effect could be found if a longer observation period (e.g. at age 10 years or age 15 years) was allowed.

Key words: alveolar bone graft; cleft palate; dental arch relationship; early bone grafting; maxilla; surgery; treatment outcome; unilateral cleft lip and palate

Introduction

Alveolar bone grafting (ABG) is an integral element of the treatment of cleft lip and palate. Its aim is to provide support for

cleft-adjacent teeth, to stabilize the maxillary segments, to eliminate the notched alveolar ridge, to support the alar bases, and to enable expansion of the interpremaxillary suture (1).

Currently, ABG carried out prior to eruption of the canine adjacent to the cleft – usually between 9 and 11 years of age – is a widely used method (2). Because bone grafting is associated with formation of growth-inhibiting scarring, the timing of ABG is based on the assumption that growth of the anterior maxilla is largely complete by 8 years of age (3), and a repair of the cleft alveolus with bone graft at 9–11 years should not compromise future maxillary development. Little deficiency of maxillofacial growth after this approach was confirmed later. For example, Semb (4) found that the anteroposterior and vertical maxillary growth in bone-grafted and non-grafted children with a unilateral cleft lip and palate was comparable. Similar conclusions were made by Daskalogiannakis and Ross (5) and Levitt et al. (6).

Alveolar bone grafting also offers the opportunity to close oronasal fistulas whose prevalence in children with cleft lip and palate may range up to 40% (7). Because oronasal fistulas have a negative effect on speech and speech development starts in the first year of life, early ABG might demonstrate beneficial influence on language skills. Furthermore, it could provide a bony support for the alar bases, hence, could improve nasolabial esthetics. Moreover, early ABG could create a bony environment for eruption and maintenance of cleft-adjacent lateral incisors, when present. Provided that facial growth and dental arch relationship are comparable to that following ABG performed during a mixed-dentition period, early ABG could offer advantages of mixed-dentition ABG without disadvantages of bone grafting performed around the time of lip repair (1). Unfortunately, there are no published data regarding growth effect of early ABG, performed before the timing recommended by Boyne and Sands (2).

Our previous study (8) demonstrated that an ABG carried out between 2 and 4 years of age resulted in a slight constriction of maxillary dental arch. We did not evaluate, however, an anteroposterior maxillary deficiency, which is more difficult to manage than a transversal deficiency. Therefore, the objective of this investigation is to assess anteroposterior maxillary growth impairment by evaluation of the dental arch relationship. The null hypothesis (H_0) tested in this study is that an ABG performed between 2 and 4 years of age does not affect the dental arch relationship.

Materials and methods

Subjects

Dental arch relationship was assessed in two groups of children with a non-syndromic complete unilateral cleft lip and palate (CUCLP) repaired with a one-stage closure of the cleft in the 1st year of life. Case eligibility was ascertained by clinicians using detailed diagnostic information from medical records, and patients with any other associated anomalies were excluded from the study.

The *Early-grafted* group comprised 42 children (27 boys and 15 girls) at the age of 5.2 years (range 4.1–6.1 years; le 1) taken from a series of 85 consecutive non-syndromic patients operated on from July 1999 to June 2006. The only inclusion criteria were as follows: 1) ABG carried out between 2 and 4 years of age and 2) availability of good-quality dental casts taken at the age of approximately 5 years. In this series, 67 of 85 children (78.8%) received ABG between 2 and

Table 1. **Characteristics of the *Early-grafted* and *Non-grafted* groups**

	Early-grafted	*Non-grafted*	*p*
Proportion of boys and girls (%)	64.4/35.6	58.6/41.4	0.629
Age in months at one-stage repair (SD)	6.0 (1.6)	8.9 (2.2)	<0.001
Age in years at alveolar bone grafting (SD)	2.4 (0.6)	N/A	N/A
Age in years at collection of dental casts (SD)	5.2 (0.5)	5.5 (0.8)	0.080

SD, standard deviation; N/A, not applicable; *p*, *p*-value.

4 years of age, whereas in the remaining 18 children, ABG was not performed. A maxillary arch constriction (cross-bite) was the reason for postponement of ABG in 8 of 18 children; in 10 children other reasons caused postponement of ABG. Of the 67 children who had received ABGs, only 42 subjects had dental casts taken at 5 years available (dropout rate – 37.3%).

The *Non-grafted* group comprised 29 children (17 boys and 12 girls) at the age of 5.5 years (range 4–6.7 years; Table 1) taken from a series of 61 consecutive non-syndromic patients operated from May 1993 to August 1996. The outcome in this group has been evaluated previously (9). The only inclusion criterion for the *Non-grafted* group was availability of good-quality dental study casts taken at 5 years. The dropout rate was 52.5%.

Surgical protocol

Infant orthopedic (IO) treatment was not carried out in any of the subjects. CUCLP was repaired according to the following protocol: lip was closed using a Tennison-Randal triangular flap technique; an extended vomer flap with a tight closure of the anterior palate was used for hard palate closure; a modified von Langenbeck technique with a dissection of all abnormal muscle insertions from the posterior edge of the hard palate up to the hamuli, which were always fractured, was used for the soft palate repair. No primary nose surgery was performed at the time of operation. However, in the *Early-grafted* group, secondary nose surgery (open rhinoplasty) was carried out at the time of ABG.

The mean age at one-stage repair of CUCLP was 6 months (SD 1.6; range 4.0–13.2 months) and 8.9 months (SD 2.3; range 4.8–15.8 months) for the *Early-grafted* and *Non-grafted* groups, respectively (Table 1). One experienced surgeon operated on all children of the *Non-grafted* group, and three experienced surgeons (including the one, who carried out surgical repairs in the *Non-grafted* group) performed one-stage repairs and ABG in the *Early-grafted* group.

The groups differed regarding the timing of ABG. In the *Early-grafted* group, iliac-crest solid bone grafting was performed at 2.4 years (SD = 0.6; range: 1.4–4.1 years). In the *Non-grafted* group, ABG was performed between 9 and 12 years. The surgical technique described by Boyne and Sands (5) was used in all subjects.

Methods

The GOSLON Yardstick (10) was used to rate dental arch relationship. According to the Yardstick, the dental arch relationship is graded from 1 to 5, where a grade 1 means very good, 2 – good, 3 – fair, 4 – poor, and 5 – very poor dental arch relationship. In a patient graded 1 or 2, a straightforward orthodontic treatment is required; in a patient graded 3, complex orthodontic treatment is required; whereas a patient graded 4 or 5 requires orthognathic surgery.

The 71 models were given random numbers to blind their origin and placed in random order. After calibration exercises, three raters (PP, CK, PF) scored the dental casts with the anchor models present throughout the rating sessions as a reference. The mean scores of all raters were subsequently used for statistical analysis. Categorization of the groups according to the score was as follows: grade 1 when mean score was ≤ 1.50; grade 2 when mean score was > 1.50 and ≤ 2.50; grade 3 when mean score was > 2.50 and ≤ 3.50; grade 4 when mean score was > 3.50 and ≤ 4.50; grade 5 when mean score was > 4.50. To evaluate intra-rater agreement, 20 randomly selected models were reassessed.

Statistical analysis

The dropout analysis in the *Non-grafted* group included χ^2 test to compare the drop-outs with the remaining subjects regarding dental arch relationship at the age of 11.2 years (SD = 1.7) (9).

Reliability of the scorings was evaluated by calculating the intra-and inter-rater agreement with proportionally weighted kappa statistics (11). Strength of agreement was defined according to Landis and Koch (12): poor ($\kappa < 0.20$), fair (0.21–0.40), moderate (0.41–0.60), good (0.61–0.80), and very good (0.81–1.00).

χ^2 test was run to compare a distribution of the GOSLON grades between the *Early-grafted* and *Non-grafted* groups.

A regression analysis was performed to investigate an association between the GOSLON score (dependent variable) and age at one-stage repair of CUCLP and presence/absence of alveolar bone graft (independent variables). The differences were considered significant for $p < 0.05$. The principles outlined in the Helsinki Declaration have been followed during the current investigation.

Results

Gender distribution was comparable in both groups (Table 1). Repair of CUCLP was performed 3 months earlier in the *Early-grafted* group than in the *Non-grafted* group (at 6 and 9 months, respectively), and the difference was statistically significant. Dental casts were made at comparable age in both groups.

The dropout analysis in the *Non-grafted* group showed that the dental arch relationship in the dropouts and the remaining patients was comparable ($p > 0.1$). The assessment of reliability of the method demonstrated that both intra- and inter-rater agreement were very good according to Landis and Koch (12) (Table 2).

Treatment outcome

The mean GOSLON scores were 2.72 (SD = 0.86) and 2.64 (SD = 1.05) for the *Early-grafted* and *Non-grafted* groups, respectively. Distribution of the GOSLON grades in both samples is presented in Figure. 1. There was a statistically significant difference between samples ($p = 0.023$). In the *Early-grafted* group, 54.8% of the sample had a score 1 or 2, 23.8% had a score 3, and 21.4% had a score 4 or 5. In the *Non-grafted* group, 38.0% subjects scored 1 or 2, 41.4% subjects scored 3, and 20.6% scored 4 or 5.

The regression analysis (Table 3) demonstrated that neither age at one-stage repair of CUCLP nor presence/absence of alveolar bone graft was associated with the GOSLON score.

Table 2. **Kappa values for the intra- and inter-rater agreement. Lower limit of 95% confidence interval (95% CI) is presented in the brackets**

		Inter-rater		
	Intra-rater	Rater 1	Rater 2	Rater 3
Rater 1	0.85 [0.68…]	N/A	0.84 [0.75…]	0.84 [0.74…]
Rater 2	1.00 [1.00]	0.84 [0.75…]	N/A	0.85 [0.76…]
Rater 3	0.89 [0.74…]	0.84 [0.74…]	0.85 [0.76…]	N/A

N/A, not applicable.

Discussion

The objective of this study was to evaluate the effect of *early* ABG performed between 2 and 4 years of age on dental arch relationship in unilateral cleft lip and palate. On the basis of the results of our previous study (8) suggesting that *early* alveolar bone grafts result in unfavorable dimensions of the maxillary dental arch, we

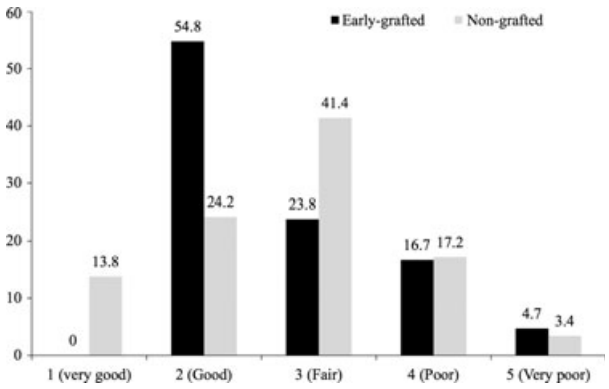

Fig. 1. The distribution of the GOSLON grades in the *Early-grafted* and *Non-grafted* groups. The distribution (%) is presented on the *Y*-axis. The GOSLON grades (1–5) are shown on the *X*-axis.

Table 3. **Results of the regression analysis with the GOSLON Yardstick score as dependent variable and age at repair of the cleft and presence or absence of alveolar bone graft as independent variables**

	Regression coefficient B	*p*	95% CI
(Constant)	3.200	0.000	2.317 to 4.083
Age at repair	−0.836	0.248	−2.268 to 0.595
Presence/absence of ABG	0.133	0.605	−0.377 to 0.642

$R^2 = 0.020$.
ABG, alveolar bone graft; CI, confidence interval; *p*, *p*-value.

hypothesized that ABG performed at this age might also negatively affect the dental arch relationship.

The detrimental facial growth effects of *primary* ABG carried out around the time of lip repair have been confirmed in numerous reports (13, 14). A common finding in the patients who had received *primary* ABG was inhibition of the anterior and vertical maxillary growth causing negative overjet and reduction of midfacial height. Consequentially, up to 50% of the patients eventually needed maxillary advancement surgery to achieve satisfactory outcome (14). In contrast, a postponement of ABG for 2–4 years seems to produce more favorable outcome because only 21% of patients from the *Early-grafted* group would likely need orthognathic surgery (were rated as having GOSLON 4 or 5 category). Furthermore, the proportion of patients with very good and good outcome (GOSLON 1 and 2 category, respectively) was more advantageous in the *Early-grafted* group (55%) than in the *Non-grafted* group (38%), whereas the proportion of patients with poor and very poor outcome (GOSLON 4 and 5, respectively) was comparable.

The dental arch relationship in the *Non-grafted* group had already been evaluated in the Warsaw–Oslo comparative study (9). The patients treated by the Oslo cleft team were assessed in numerous investigations (15, 16), and the outcome of the Oslo cleft center has been widely considered unofficial *gold standard* in treatment of cleft lip and palate. In the Warsaw–Oslo comparison, the dental arch relationship of 61 patients at the mean age of 10 years consecutively treated at the Warsaw cleft center was blindly compared with age- and sex-matched patients treated in Oslo. The outcome in both samples was similar. Because the *Non-grafted* group is derived from the 61-subject sample evaluated in the Warsaw–Oslo study and the dental arch relationship in the *Early-grafted* group is comparable with that in the *Non-grafted* group, it can be cautiously concluded that *early* ABG does not affect adversely the dental arch relationship. Longer observation time, however, is needed to substantiate it.

To the best of our knowledge, there are no published studies evaluating occlusion after ABG performed between 2 and 5 years of age; thus, we are unable to directly compare our results with data from other studies. However, alveolar repair according to the Milan protocol, an early secondary gingivo-alveoloplasty (EsGAP) combined with a hard palate closure (17), and the Warsaw protocol, *early* ABG, are performed at similar age. EsGAP differs from ABG, because instead of grafting bone, formation of new bone is induced by a creation of a tunnel of mucoperiosteal flaps sealing off the alveolar defect from both oral and nasal cavities. Nevertheless, the timing, design of the flaps, and a potential of ABG and EsGAP to induce scarring of the palate seem comparable. The outcome for the dental arch relationship of the Milan cleft team was assessed with the 5-year-olds Index by Flinn et al. (16). The results were very favorable – 63% 5-year-olds were graded 1 or 2 corresponding with very good or good outcome, respectively, and 7% graded 4 or 5 (poor or very poor outcome, respectively). Although our findings demonstrate a higher proportion of poor and very poor outcome than after the Milan protocol (21% vs. 7%), it should be emphasized that the rating of dental arch relationship at primary dentition stage with the GOSLON Yardstick (the current group) and the 5-year-olds Index (the Milan group) produces somewhat different scores – a higher proportion of very good and good outcomes when the latter index is applied (18). Consequentially, after adjustment for the difference between these two indices, the effects of *early* ABG and EsGAP on dental arch relationship seem to be comparable. This should be confirmed, however, in a study in which both samples are rated with the same index.

A regression model showed that timing of one-stage surgery did not influence the dental arch relationship (Table 3). It was demonstrated that facial growth impairment after cleft surgery is associated with scar formation (1). In consequence, if the post-surgical scarring develops early, when more residual growth is present, the more growth deficiency should be expected. Our previous results (8) were in agreement with this observation because they showed an increased mesiopalatal inclination of the lesser segment if the repair of the cleft was performed at earlier age.

In this investigation, the one-stage repair of the cleft was performed 3 months earlier in the *Early-grafted* group than in the *Non-grafted* group (6 vs. 9 months, respectively) and the expectation was that earlier repair had negatively influenced the dental arch relationship. The current findings cannot confirm a relationship between the timing of cleft repair and occlusal status at 5 years. A possible explanation is that palatal scarring predominantly affects the width of the maxillary dental arch and, to a lesser degree, anteroposterior position of the maxillary dentoalvealoar region, which affects the GOSLON rating the most. Also, it might have been too soon to identify a difference because this comparative study was carried out at a young age.

Usually, the dental arch relationship in 5-year-old children with CUCLP has been evaluated with the 5-year-olds Index (19). This index uses analogous to the GOSLON Yardstick, a 5-point scale for grading. However, a recent study by Mars et al. (18) questioned the validity of the 5-year-olds Index, especially as a predictive measure in 5-year-olds, because the results of the longitudinal assessment at 5 years with the 5-year-olds Index disagreed with the results of the evaluation at 10 years with the GOSLON Yardstick. For example, the 5-year-olds Index scores at 5 years compared with the GOSLON scores at 10 years showed a weighted kappa score of only 0.090, which corresponds with poor concordance. The authors stated that the GOSLON Yardstick used at 5 years is more reliable than the 5-year-olds Index as a predictive tool. This was also suggested by Noverraz et al. (20). Therefore, the use of the GOSLON Yardstick in the present study is justified.

Assessment of the results of cleft surgery many years before completion of growth and/or therapy is always burdened with the risk that it does not truly reflect the final effect of treatment. Moreover, detrimental or beneficial effects of a given treatment may not be detectable at an early age. On the other hand, early prediction of outcome is particularly desirable in the cleft lip and palate field because treatment is prolonged and identification of any harmful treatment modalities shortly after their implementation would allow early modification of treatment protocol. Also, if pure effects of surgery are to be assessed, an evaluation before orthodontic or speech therapy is indispensible. Weighing the risks and the benefits of early assessment, it should be stated that potential advantages such as identification of harmful or unnecessary therapeutical modalities usually outweigh the costs of evaluation. From this perspective, the evaluation of the effects of *early* ABG was highly desired.

Conclusions

On the basis of the current findings, it can be concluded that *Early* alveolar bone grafting carried out between the ages of 2 and 4 years was not found to negatively affect dental arch relationship by the age of 5 years. However, it is possible that such a negative effect could be found if a longer observation period (e.g. at age 10 years or age 15 years) was allowed.

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