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Correlations between initial cleft size and dental anomalies in unilateral cleft lip and palate patients after alveolar bone grafting

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ABSTRACT

Objective To determine in individuals with unilateral cleft lip and palate the correlation between initial cleft size and dental anomalies, and the outcome of alveolar bone grafting.

Methods A total of 67 consecutive patients with non-syndromic unilateral complete cleft lip and palate (UCLP) were included from the cleft lip and palate-craniofacial center, Uppsala University Hospital, Sweden. All patients were operated by the same surgeon and treated according to the Uppsala protocol entailing: lip plasty at 3 months, soft palate closure at 6 months, closure of the residual cleft in the hard palate at 2 years of age, and secondary alveolar bone grafting (SABG) prior to the eruption of the permanent canine. Cleft size was measured on dental casts obtained at the time of primary lip plasty. Dental anomalies were registered on radiographs and dental casts obtained before bone grafting. Alveolar bone height was evaluated with the Modified Bergland Index (mBI) at 1 and 10-year follow-up.

Results Anterior cleft width correlated positively with enamel hypoplasia and rotation of the central incisor adjacent to the cleft. There was, however, no correlation between initial cleft width and alveolar bone height at either 1 or 10 years follow-up.

Conclusions Wider clefts did not seem to have an impact on the success of secondary alveolar bone grafting but appeared to be associated with a higher degree of some dental anomalies. This finding may have implications for patient counseling and treatment planning.

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KEYWORDS

Dental status; initial cleft size; unilateral cleft lip and palate

Introduction

Cleft width at birth is highly variable in the cleft lip and palate deformity (1–3). The severity of a cleft is usually assessed by its appearance at birth. There is a tissue deficiency and/or displacement of the palatal segments and the alveolar segments (4). The impact of the initial cleft width on various treatment outcomes in these patients has been studied to some extent (1,2,5). Initially wider clefts were correlated with increased transverse dental arch dimensions and less cross-bite occlusion in the primary dentition (6), with inhibited maxillary growth (7) as well as with higher prevalence of velopharyngeal incompetence (8). Initial cleft size as measured prior to palate closure was shown to have no impact on dental arch relationship in mixed dentition as evaluated by the GOSLON Yardstick (9). Further, individuals with smaller initial cleft width and with more remaining palatal tissue showed better maxillofacial growth, as did patients with less facial deformities and more tissue volume of the upper lips at the time of lip repair (7,10).

It has also been recognized that wider clefts are generally associated with increased difficulty of primary surgical repair as well as with an increased occurrence of postoperative palatal fistulae (11). However, it has also been demonstrated that maxillary development mainly depends on the actual

treatment performed rather than the severity of the initial cleft width (12).

Secondary alveolar bone grafting (SABG) to the cleft area is performed in order to provide bony support for teeth adjacent to the cleft, to stabilize and restore the structure of the maxillary arch and to close any remaining oronasal fistulae (13–16). Successful SABG is vital for orthodontic tooth movement into the grafted bone and for achieving a complete dental reconstruction of the alveolar ridge. The surgical procedure is performed in conjunction with orthodontic treatment at the time of permanent canine eruption (14,17,18). The importance of the orthodontic preoperative planning and treatment, entailing expansion of the maxillary arch and restoration of arch width prior to bone grafting as well as close follow-up around the SABG procedure, is widely recognized. Dental anomalies such as hypodontia, peg-shaped teeth, supernumerary teeth, and crown and root malformations occur more frequently in cleft patients (19) and constitute additional complicating factors for the treatment planning.

The previously reported long-term outcome analysis after SABG in the same cohort of patients showed that the preoperative degrees of dental anomalies such as enamel hypoplasia, canine inclination, and incisor inclination and rotation were positively correlated to the reduction in alveolar bone height observed at 10 years after SABG (20).

The aim of the present investigation was to examine whether initial cleft size is correlated to this reduction in alveolar bone height seen in some patients at long-term follow-up and to the severity of dental anomalies in the cleft area in unilateral cleft lip and palate (UCLP) patients.

The hypothesis is that initially wide clefts are associated with inferior outcomes after the SABG procedure and with a higher frequency of dental anomalies.

Material and methods

Subjects

The patient cohort of the present investigation has previously been investigated with regard to the correlation between dental anomalies and long-term SABG outcomes (20). The data set was originally composed of 67 Caucasian subjects with non-syndromic UCLP, who underwent SABG between 1987 and 1997 by the same surgeon (Valdemar Skoog). One patient was excluded due to the lack of dental cast at the primary lip plasty. Approval from the Uppsala Ethics Committee was obtained (reference number 2012/401), and patient consent was obtained.

Surgical procedure

The surgical protocol included lip closure according to Skoog (1969) at 3–4 months, two-stage palatal closure with soft palate at 6 months, and residual cleft in the hard palate at 24 months. The SABG procedure has been described previously, and one experienced plastic surgeon was responsible for all operations (20). Briefly, the cleft was exposed through both vestibular and palatal approaches in all patients. Deciduous teeth projecting into the cleft space were extracted. Cancellous bone was harvested from the iliac crest and grafted to the entire cleft space. The mean age of the subjects at the time of SABG was 10.0 years (range 8.5–12.0).

Cleft width

Cleft size was measured on dental study casts obtained in connection to the primary lip plasty at 3–4 months of age. The reference points and linear measurements used in this study have been described in previous studies (Figure 1) (6,21,22). After an initial inter-operator calibration of the landmarks between the two authors F.J. and E.R., F.J. performed all measurements and scorings. Distances were measured to the nearest 0.01 mm using a digital caliper.

Alveolar bone height according to the Bergland Index, canine inclination, central incisor inclination and rotation. These variables were recorded as reported in previous studies (17,20,23).

Tooth status. Intra-oral radiographs obtained prior to SABG were assessed for tooth status of the central incisor in the cleft area according to a previously described tooth status index (24).

Lateral incisor status. Lateral incisor status was dichotomized into presence (group 1) or absence (group 2) of lateral incisor (hypodontia).

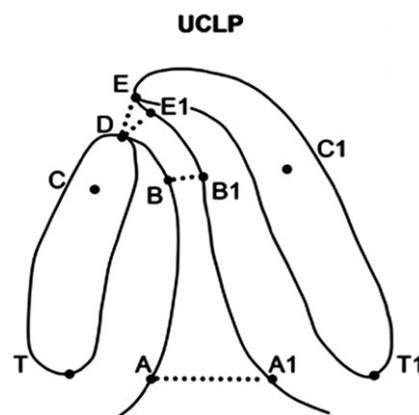


Figure 1. Schematic drawing of an infant maxillary dental cast with unilateral cleft lip and palate (UCLP). Landmarks and distances used in the linear measurements are demonstrated. D–E = cleft width at the level of the alveolar processes anteriorly; D–E1 = smallest cleft width at the level of the alveolar processes anteriorly; T–T1 = posterior width of the alveolar arch in the tuber area; A–A1 = width of the cleft at the level of T–T1; B–B1 = width of the cleft at the level of C–C1; B–B1/C–C1 = ratio of the cleft width related to the total alveolar arch width anteriorly at the level of the canine points; A–A1/T–T1 = ratio of the cleft width related to the total alveolar arch width posteriorly at the level of the tuberosity points.

Reliability of recordings

The reliability and intra-rater reproducibility of the measurements/scorings of cleft widths were determined from randomized duplicate recordings in 20 UCLP patients. Intra-class correlations coefficients (ICC) were calculated for intra-rater reliability and agreement of repeated measurements. The intra-rater reliability was considered excellent (0.96–0.97). Weighted kappa coefficients calculated for the other measurements/scoring were reported previously (20). The kappa coefficients varied from 0.58 (moderate) to 0.90 (almost perfect) agreement of repeated measurements.

Statistical analysis

Median, mean, and range values were calculated for all variables. Relations between cleft widths at different levels as well as ratios were analyzed with Spearman rank correlation. Multivariate analysis was not suitable due to the non-parametric data.

Results

Descriptive measurements

Transverse cleft widths showed a large inter-individual range (Table 1). In infancy the median separation between the alveolar processes anteriorly (D–E) was 6.2 mm. Median anterior cleft width (B–B1) was 6.3 mm. Median posterior cleft width (A–A1) was 7.5 mm.

Correlations between initial cleft size, dental status, and the success of alveolar bone grafting

Alveolar bone height. There was no significant correlation between initial cleft width and alveolar bone height at either 1 or 10-year follow-up (Table 2).

Table 1. Descriptive statistics with mean, standard deviation, minimum, median, and maximum for: cleft size measurements in millimeters (mm), cleft size ratios, degrees of dental anomalies, and modified Bergland Index.

Variable	n	Mean	SD	Minimum	Median	Maximum
Incisor inclination	67	37.09	13.00	0.00	40.00	70.00
Incisor rotation	67	60.82	29.27	0.00	45.00	90.00
Modified Bergland grade 1-4	67	1.03	0.17	1.00	1.00	2.00
Enamel hypoplasia	67	1.06	0.83	0.00	1.00	2.00
Canine inclination	67	1.12	0.64	0.00	1.00	2.00
Modified Bergland Index at 10-year follow-up	67	1.58	0.53	1.00	2.00	3.00
D-E	66	6.40	2.68	1.20	6.22	11.83
D-E1	66	3.10	2.58	0.00	2.41	10.31
C-C1	66	25.18	2.87	18.99	24.62	33.83
B-B1	66	6.49	2.65	1.37	6.26	15.06
BB1-CC1	66	0.25	0.08	0.06	0.26	0.47
T-T1	66	30.39	2.82	24.56	30.10	36.94
A-A1	66	7.61	2.45	3.11	7.56	13.20
AA1-TT1	66	0.25	0.06	0.12	0.25	0.37

Table 2. Spearman correlation between initial cleft width and alveolar bone height.

	Modified Bergland at 10-year follow-up	Modified Bergland grade 1-4
AA1-TT1	0.05509	-0.19858
	0.6604	0.1099
	66	66
A-A1	0.11919	-0.20509
	0.3405	0.0985
	66	66
BB1-CC1	0.21202	-0.12045
	0.0874	0.3354
	66	66
B-B1	0.18771	-0.15301
	0.1312	0.2200
	66	66
C-C1	0.09007	-0.17906
	0.4720	0.1503
	66	66
D-E	0.18749	0.04558
	0.1317	0.7163
	66	66
D-E1	0.09605	-0.15301
	0.4430	0.2200
	66	66
T-T1	0.15272	-0.10093
	0.2209	0.4201
	66	66

Tooth status. As reported in our previous study (20) we found that the degree of enamel hypoplasia was positively correlated with the relative anterior cleft width (B-B1/C-C1) ($r = 0.24$, $P = 0.0498$) (Figure 2). There was no correlation between enamel hypoplasia and posterior cleft width.

Canine inclination. Prior to bone grafting 10 patients had a grade 0 canine inclination, 39 patients had grade 1, and 18 patients had grade 2 (data from Jabbari et al. (20)). There was no correlation between cleft width and canine inclination.

Incisor rotation and inclination. In our previous publication (20) on these patients we found that 10 patients had grade 0 incisor rotation, 25 patients grade 1, and 32 patients grade 2 when examined before operation. Further, eight patients had grade 0 incisor inclination, 45 patients grade 1, and 14 patients grade 2. Central incisor rotation was positively correlated with the relative anterior cleft width (B-B1/C-C1) ($r = 0.25$; $P = 0.042$) (Figure 3), with cleft width at the level of the alveolar processes anteriorly (D-E) ($r = 0.32$; $P = 0.0074$)

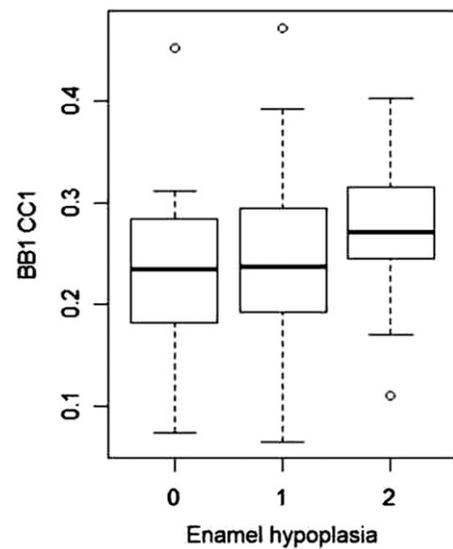


Figure 2. Enamel hypoplasia of the central incisor in the cleft area versus relative anterior cleft width (B-B ratio = B-B1/C-C1) as measured on dental casts obtained in connection to the primary lip plasty.

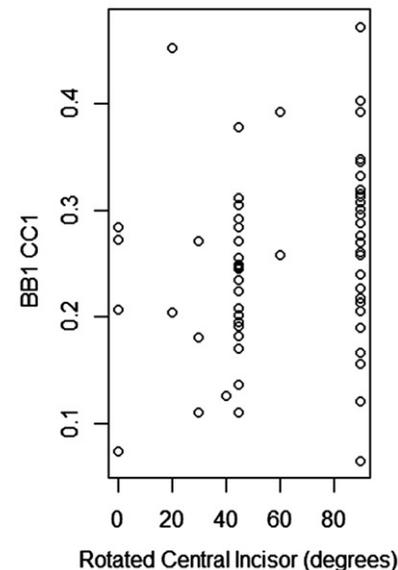


Figure 3. Rotation of the central incisor (degrees) in the cleft area versus relative anterior cleft width (B-B ratio = B-B1/C-C1) as measured on dental casts obtained in connection to the primary lip plasty.

(Figure 4), and with the smallest cleft width at the level of the alveolar processes (D-E1) ($r = 0.29$; $P = 0.0168$) (Figure 5). There was no correlation between cleft width and central incisor inclination.

Lateral incisor. There was hypodontia of the laterals in 20 patients, peg-shaped laterals in 39, and only 7 patients had normal dental shape in the cleft area. Both the posterior cleft width (A-A1) and the relative posterior cleft width (A-A1/T-T1) at the level of the tuberosity were positively correlated to presence of laterals ($P = 0.0082$ and $P = 0.0063$ respectively) (group 1). Thus, posteriorly wider clefts were associated with presence of laterals.

Discussion

The present study shows that the severity of initial cleft width does not correlate with the outcome after secondary alveolar

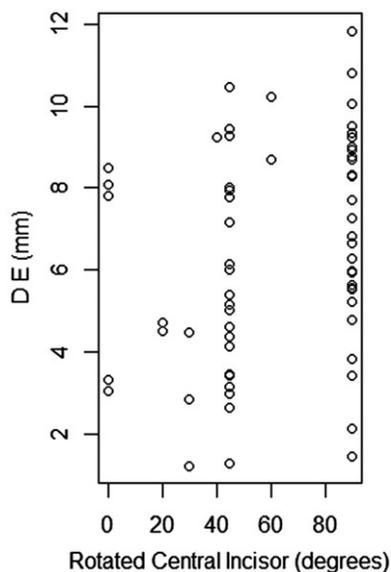


Figure 4. Rotation of the central incisor (degrees) in the cleft area versus cleft width at the level of the alveolar processes anteriorly D-E (mm).

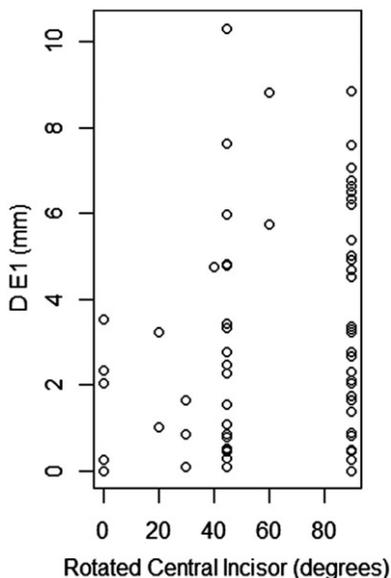


Figure 5. Rotation of the central incisor (degrees) in the cleft area versus smallest cleft width at the level of the alveolar processes anteriorly D-E1 (mm).

bone grafting. However, the initial cleft width correlated with the severity of some dental malformations. This is to the best of our knowledge the first study describing how initial cleft dimensions are related to the severity of dental anomalies.

Secondary alveolar bone grafting (SABG) has since its introduction by Boyne and Sands (15) become a well-accepted method for reconstruction and stabilization of alveolar defects in patients with cleft lip and palate. The outcome after SABG has mainly been assessed by measuring the height of the grafted bone on occlusal radiographs and has usually been graded according to the Bergland Index. Two-dimensional occlusal radiographs are readily available in all clefts units, and measuring the alveolar bone height is easily performed. Limitations of the method are that it does not provide a complete visualization of the alveolar cleft space and does not allow for assessments of the nasal bone height and grafted bone volume.

The impact of cleft size on various treatment outcomes in UCLP patients has been assessed to some degree. Initial cleft size was found to have an impact on early outcomes with regard to dental arch dimensions, such as cross-bite and inhibited maxillary growth (2,6).

The only report to date analyzing the correlation between cleft width and SABG outcomes showed that cleft width at the time of secondary alveolar bone grafting did not influence bone graft survival (25). Similarly, measuring cleft size on dental casts obtained at the time of primary lip plasty, we could not find any correlation between initial cleft size and SABG outcomes. Evidently, there is a strong impact of the skill of the surgeons and the treatment protocol on outcomes in cleft surgery. In our study one surgeon performed all procedures and then used one and the same surgical technique and protocol. This is most probably the main factor behind the excellent results obtained.

This study does, however, demonstrate several links between initial cleft dimensions and the severity of dental anomalies. The relative anterior cleft width correlated with the degree of enamel hypoplasia, i.e. a wider cleft at the level of the canine points was associated with more enamel hypoplasia of the central incisors. It has been shown that enamel hypoplasia, due to a deficiency in enamel formation and absence of the enamel surface (26), mainly occurs in teeth close to the cleft (27). This occurrence may be either attributed to the cleft deformity itself or to the early surgical correction of the defects (28). The positive link between cleft width and central incisor enamel hypoplasia points at the deformity itself as the main factor behind the disruption in enamel formation. All measurements reflecting anterior cleft width correlated positively with the degree of central incisor rotation. Impacted permanent canines occur with higher frequency in the cleft lip and palate population due to canine inclination (29). Moreover, we have demonstrated that high degrees of canine inclination have a negative impact on SABG (20). However, in the present study we did not find any correlation between initial cleft width and the position of the permanent canine at the time of bone grafting. A statistical correlation was also found between the presence of the lateral incisor in the cleft area and posterior cleft width; however, the implications of this finding are still unsure.

A correlation between variables does not imply that one has a causal effect on the other. However, the aim of the study was not to prove causal effects of cleft size on later dental status but to find possible associations in order to, at an early age, find children with future need for more extensive orthodontic treatment. Although we found no correlation between cleft width and the outcome of SABG, we did find a correlation with enamel hypoplasia and central incisor rotation, factors that were previously demonstrated to be associated with a reduction of bone height in the alveolar cleft with time (20). Thus, anteriorly wide clefts at infant age could signal a need for future more extensive orthodontic treatment in connection to the SABG procedure. Initial cleft width varies considerably, implying that a more individualized approach based on cleft morphology rather than strict adherence to a pre-established treatment protocol is desirable. Therefore, an early identification of patients at increased risk enables the

possibility to establish an appropriate interceptive treatment plan, entailing early identification of enamel defects and other related dental abnormalities.

Conclusions

The width of the cleft prior to lip plasty does not seem to have an impact on the success of secondary alveolar bone grafting, but is positively correlated to the degree of central incisor rotation and enamel hypoplasia. Consequently initial cleft dimensions could be used as an early indicator of future need for more extensive orthodontic treatment in connection with secondary alveolar bone-grafting.

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Disclosure information

The authors report no conflicts of interest.

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