Long-term stability of alveolar bone grafts in cleft palate patients

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Introduction: Many authors have examined the surgical bone treatment of cleft palate patients, but no study has emphasized the role of orthodontic therapy. The aims of this study were to evaluate the long-term stability of bone grafts when using an orthodontic-surgical protocol, to determine the success of bone grafts in minor vs severe clefts, and to develop a qualitative method for assessing the success of bone grafting. Methods: Forty-nine patients were included in this study. Occlusal x-rays were taken before (T0), immediately after (T1), and at least 1 year after bone grafting (T2). Two radiographic parameters were analyzed adjacent to the cleft side: the vertical bone level (Bergland scale) and the horizontal bone level (Witherow-derived scale). Results: The bone graft success at T2 was 91.84% (95% confidence interval, 84.55-96.41). The severity of the cleft before grafting was not statistically correlated with success at T2 ($P < 0.05$). The concordance rate between Bergland and Witherow values was 87.07% (95% confidence interval, 82.69-90.69). The variables analyzed (sex, age, type of cleft, lateral incisor agenesis) were not statistically correlated ($P < 0.05$) with the stability of bone graft. Based on the results, the only factor involved in the stability of the graft seems to be dental age at the time of bone grafting and the orthodontic therapy before and after grafting. Conclusions: It seems appropriate to recommend early application of a surgical-orthodontic protocol to treat cleft lip and palate patients, prevent postoperative bone resorption, and guarantee correct positioning of the teeth. (Am J Orthod Dentofacial Orthop 2012;142:289-99)
protocol, to determine the success rate of bone grafts in minor vs severe clefts, and to develop a qualitative method for assessing the success of bone grafting.

MATERIAL AND METHODS

In this retrospective clinical trial, we analyzed the records of 446 complete cleft lip and palate patients, recruited from the Regional Hospital of Vicenza (Italy), Cleft Palate Patients National Reference Centre, between 1994 and 2011. There was only 1 data manager (D.T.), and all records were digitized. The study population consisted of white patients matched for sex and age.

Patients were included in the study based on the following criteria: (1) congenital complete cleft lip and palate; (2) treatment with the protocol of the Regional Hospital of Vicenza and the University of Ferrara; (3) digital occlusal radiographs before (T0), immediately after (T1), and at least 1 year after bone grafting (T2); (4) performance of the alveolar bone graft by the same surgeon; (5) digital occlusal x-rays exposed with a standardized radiologic technique (long-cone technique and root direction perpendicular to the palatine midline, compared with 2 other projections, to prevent radiologic superimpositions, image artefacts, and interpretation errors) by the same radiologist; (6) standardized radiographic evaluations; and (7) informed consent from each patient. Patients were excluded if they had syndromic cleft lip and palate. A total of 117 patients met all inclusion criteria. The study was double-blinded, and the statistical analysis was performed by an external statistician. Of the 117 patients who met the inclusion criteria, 68 were excluded for various reasons, leaving 49 patients in the study cohort (Table I). A flow chart of the study participants is shown in the Figure. Table II gives the sex distribution. There were 45 unilateral and 4 bilateral clefts. Among the 45 patients with unilateral clefts, 15 were right sided, and 30 were left sided. Thirty-two patients demonstrated lateral incisor agenesis, and 8 patients had agenesis of other teeth, contralateral to their clefts.

All patients had a surgical-orthodontic protocol in accordance with the Regional Hospital of Vicenza and the Postgraduate School of Orthodontics at the University of Ferrara in Italy. The chronologic order of the treatment, according to the protocol, is outlined below.

1. At birth: placement of a passive palate plate, constructed of soft resin.
2. At 3 months of age: soft-palate surgery and lip and nostril surgery with definitive lip repair.
3. At 6 months of age: unilateral and bilateral rhinoplasty.
4. At 8 to 12 months of age: initiation of speech therapy.
5. At 18 to 20 months of age: hard palate surgery and continuation of speech therapy.
6. At 5 years of age: placement of a hyrax-type rapid palatal expander bonded onto the deciduous teeth, with palatal and buccal arms reaching to the canines, activated at a quarter turn per day. The average activation period depended on the degree of maxillary constriction. A 2- to 3-mm overcorrection at the molars was recommended to counteract relapse. Immediately after the expansion, a maxillary traction device (Delaire mask) was applied with 450 g of force per side for 14 hours per day for 7 months. The rapid palatal expander was passively maintained in the mouth for 1 year after the expansion.
7. At 6 to 9 years of age: placement of a passive transpalatal bar (0.036-in wire) bonded onto the first permanent molars (to prevent transverse relapse) and continuation of speech therapy.
8. At 9 to 11 years of age: placement of an alveolar graft in the cleft side, using bone from the iliac crest, once canine root development was between one fourth and two thirds of its final root length as identified on the occlusal x-ray.
9. At 9 to 15 years of age: orthodontic fixed therapy with preadjusted 0.022 × 0.028-in brackets to obtain alignment, leveling, malocclusion correction, and canine repositioning in the arch. Orthodontic therapy began when canine root development was between one fourth and two thirds of its final root length. Orthodontic appliances were kept in place through the transitional phase of the dentition until all teeth had erupted and definitive orthodontic treatment was completed, when all occlusal and functional goals of therapy were achieved.
10. At 18 years of age: plastic (lip revision, nose revision, rhinoplasty, nasal septum surgery, adenoid surgery, sinus mucous membrane surgery, scar surgery) or maxillofacial surgery and implantation if necessary. In this study, no patient received jaw surgery, plastic surgery, or implantation.

Table I. Patients excluded from the study

<table>
<thead>
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<tbody>
<tr>
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<tr>
<td>Bone graft unnecessary</td>
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<tr>
<td>Bone graft not currently performed</td>
</tr>
<tr>
<td>Digital radiographs not exposed</td>
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<td>Patients excluded from study</td>
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</tbody>
</table>

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Each patient had digital occlusal x-rays taken before the alveolar bone graft surgery (T0). The radiographs were taken using an AC Cefla dental machine with a CEI OXC 070G x-ray tube (Anthos, Imola, Italy). The machine was set at 65 kV and 7.5 mA. The focus-to-skin distance was 20 cm. Exposure time varied depending on the size of patient but was consistent for preoperative and postoperative x-rays and varied from 0.7, to 0.86, to 1.0 seconds. The film was oriented parallel to the occlusal plane, and the central beam was directed perpendicular to the cleft side for the first projection and at 70° to the film in the mesial and distal directions for the second and third projections. Then x-rays were processed with a dental vista scan machine (model 2130-51; Dürr Dental, Bietigheim-Bissingen, Germany) with DBSWIN software (Dürr Dental).

A numeric code was assigned to each patient before the evaluations to ensure blinding of the reviewers.
At T0, qualitative categorizations of cleft severity were made by using occlusal x-rays and based on the amount of bone support on each root adjacent to the cleft site. A standard for evaluation was adopted according to 2 scales: the Bergland vertical bone evaluation and the Witherow-derived horizontal bone evaluation. These scales are shown in Table III (types I, II, III, and IV and scores of 1, 0.5, and 0 were qualitative values used to indicate gradations in the presence or absence of bone). We compared each radiograph with its corresponding 2 other projections (the tube was directed at 70° to the film in the mesial and distal directions) to prevent image artefacts and interpretation errors. Two values were obtained for each cleft. The first value was related to the distal surface of the tooth on the mesial side of the cleft (site 1), and the second represented the mesial side of the tooth on the distal side of the cleft (site 2). In patients with bilateral clefts, both clefts were evaluated; however, only 1 value was recorded in the table because of identical results.

All evaluations were performed by a blinded examiner (D.T.) and repeated 3 times on 3 different days, with a 30-day interval between evaluation sessions, to assess intraexaminer variability. Twelve randomly selected patients were assessed by 3 blinded and experienced examiners (U.B., A.G., G.S.) to evaluate interexaminer variability.

Based on the results, the patients were divided into the following groups (Table IV): (1) minor cleft: type I, score 1 or 0.5, on both teeth adjacent to the cleft; (2) moderate cleft: types II and III, score 0.5, on both teeth adjacent to the cleft; and (3) severe cleft: type IV, score 0, on both teeth adjacent to the cleft. Patients were also included in the gravest group if there was a different value on the roots adjacent to the cleft.

According to the protocol, an alveolar bone graft on the cleft side and an immediate postsurgical occlusal x-ray were performed on each patient. The patients were then examined monthly to monitor the orthodontic therapy. At T2, a long-term occlusal x-ray was obtained, and a qualitative evaluation of bone level was performed on the teeth adjacent to the cleft at T1 and T2. On 12 randomly selected patients, these evaluations were made by the first examiner and the 3 other examiners to evaluate interexaminer variability.

The first examiner obtained 294 values and repeated each measurement 3 times. The other 3 examiners obtained 216 values, resulting in a total of 1098 evaluations (Tables V and VI).

By using this method, both of the following criteria had to be met for a successful outcome at T2: (1) vertical and horizontal bone values equal to or better than the bone values at T1; and (2) type I and score 1 (vertical and horizontal bone levels approximately normal), with bone support greater at T2 than at T0.

The patients’ records were reviewed for additional biometric data. Particular attention was paid to type of cleft (unilateral or bilateral); age at the 3 evaluation times; sex; any supernumerary teeth, diminutive lateral incisors, and peg-shaped lateral incisors in the cleft area; any supernumerary or diminutive teeth not in the cleft area; absence of any teeth adjacent to the cleft and the probable cause thereof; and the incidence of regrafting.

The average age at the time of bone grafting was 10.25 years, with a range of 8 to 14.7 years (Table VII). The average age at follow-up was 12.16 years, with a range of 10.1 to 15.6 years. The average time of follow-up after grafting was 1.87 years, with a range of 1 to 4.8 years (Table VIII).

At T2, 38 patients (77.55%; 95% confidence interval [CI], 63.38–88.23) had not experienced canine eruption, and 11 patients (22.44%; 95% CI, 11.77–36.62) had canine impaction requiring a surgical flap and orthodontic recovery.

### Statistical analysis

The percentage of bone graft success was calculated at T1 and T2. To evaluate whether the severity of the
In addition, we evaluated several variables to determine whether the Bergland scale was comparable with the Witherow-derived scale (0 < Kendall W < 1). The Kendall W expresses the level of agreement of multiple assessments from the same or different operators. This coefficient is a real number between 0 (no agreement) and 1 (full agreement). A Kendall W less than 0.7 indicates little agreement and that the measurement method can lead to inaccurate results, whereas a Kendall W greater than 0.9 indicates strong agreement and that the results are likely to be accurate.

We calculated the Kendall coefficient of concordance (0 < Kendall W < 1) to control for intraexaminer and interexaminer repeatability and reproducibility.

In addition, we evaluated several variables to determine whether they had statistically significant effects on the long-term stability of the bone grafts at T2. (1) The potential effects of sex and age (at bone grafting) were analyzed by using a linear regression analysis, which is an approach to modeling the relationship between a scalar variable Y and at least 1 variable called X. Proceeding to the statistical analysis, the patients were further divided into those who were 10 years or younger and those older than 10 years, in accordance with sample medians equal to 10 years old (P <0.05).

(2) The effects of type of cleft (unilateral or bilateral) were assessed by using a binomial test (P <0.05). This determines the statistical significance of the deviations from the expected and the observed distributions into 2 categories. (3) Lateral incisor agenesis was also assessed by using a binomial test (P <0.05). (4) Finally, we used the Wilcoxon signed rank test to determine the surgical success rates (P <0.05). This is a nonparametric statistical hypothesis test used to assess whether population means differ when comparing 2 related samples or repeated measurements of 1 sample. It is useful in evaluations of ordinal-scale variables.

The chi-square test (P <0.05) was used to verify the correlation between the severity of the cleft palate at T0 and any lost teeth adjacent to the cleft at T2, and whether site 1 had a different Bergland value than site 2 at any of the 3 measurement times. A binomial test (P <0.05) was used to evaluate whether lateral incisor agenesis at T0 was correlated with the spontaneous eruption of the canines at T2, and whether there was an association between the type of cleft at T0 and any lost teeth adjacent to the cleft at T2.

The statistical analysis was performed with the R 2.12 × 64 software (copyright 1998-2012 by Kurt Hornik), and the statistical significance level was set at 95% (P <0.05).

RESULTS

Success rate and grading systems

The success rate at T1 was 70.41% (95% CI, 60.34-79.21). The success rate at T2 was 91.84% (95% CI, 84.55-96.41). The binomial intervals of confidence did not intersect; therefore, the T2 success rate was significantly greater than that at T1.

The correlation between cleft palate severity and success rate at T2 was not statistically significant (P = 0.64). The Kendall coefficient of concordance was 0.99 for both the Bergland and the Witherow-derived scales (0 < Kendall W < 1), indicating that these scales were comparable.

It was acceptable to use the Kendall W because the concordance rate between the Bergland and Witherow-derived values was 87.07% (95% CI, 82.69-90.69), and the error method was not relevant.

Table IX shows the cross-tabulation of the Bergland and Witherow-derived evaluations by first operator and first evaluation. It highlights the error point in which 37 type I Bergland values were classified as a Witherow-derived score of 0.5.

After the alveolar bone graft, there were no oronasal filling was radiographically evident on occlusal x-rays.

Table V. Sample distribution for Bergland grading system (first operator’s first evaluation)

<table>
<thead>
<tr>
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<th>I</th>
<th>II</th>
<th>III</th>
<th>IV</th>
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<tr>
<td>T0</td>
<td>19</td>
<td>48</td>
<td>18</td>
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<td>T1</td>
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<td>T2</td>
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Table VI. Sample distribution for Witherow-derived grading system (first operator’s first evaluation)

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<tr>
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<tr>
<td>T2</td>
<td>0</td>
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<td>90</td>
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Table VII. Sample age at time of bone grafting

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<th>Minimum</th>
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<td>10.25</td>
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<td>49</td>
<td>8.00</td>
<td>14.70</td>
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Table VIII. Follow-up time

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<tr>
<td>1.87</td>
<td>1.10</td>
<td>49</td>
<td>1.00</td>
<td>4.80</td>
<td>1.30</td>
</tr>
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</table>

The statistical analysis was performed with the R 2.12 × 64 software (copyright 1998-2012 by Kurt Hornik), and the statistical significance level was set at 95% (P <0.05).
In our 3 groups (minor, moderate, and severe clefts), no regrafting was necessary.

**Variables affecting the long-term stability**

As previously mentioned, we determined the inter-examiner variability by calculating the Kendall coefficient of concordance for the 3 series of values. The coefficient was 0.98 for both Bergland and Witherow-derived values (0 < Kendall W < 1), signifying a high degree of repeatability. The interexaminer variability was also determined by using the Kendall coefficient of concordance for the 3 series of values. It was 0.99 for both Bergland and Witherow-derived values (0 < Kendall W < 1), signifying a high degree of reproducibility.

There was no statistically significant correlation between the patient’s sex or age and the success of the alveolar bone graft at T2 (P = 0.08 and P = 0.94, respectively).

There was no statistically significant influence of the type of cleft (unilateral or bilateral) on the long-term stability of the bone graft (P = 0.84).

There was no statistically significant influence of lateral incisor agenesis on the long-term stability of the bone graft at T2 (P = 0.32).

The Bergland vertical bone evaluation resulted in P = 0 for T1 vs T0, and P = 1 for T2 vs T1. The Witherow-derived horizontal bone evaluation resulted in P = 0 for T1 vs T0 and P = 0.13 for T0 vs T2. Based on the P values, there was a statistically significant Bergland and Witherow-derived value improvement only from T0 to T1.

**Clinical features and bone graft**

At T0, there was a statistically significant difference between the Bergland bone levels observed at sites 1 and 2 (P = 0), with site 2 having a greater bone level than site 1, as shown in **Table X**.

At T1 and T2, there was no statistically significant difference between Bergland bone levels at sites 1 and 2 (Bergland grading system with P = 0.47).

Spontaneous canine eruption at T2 was not associated with lateral incisor agenesis (P = 0.34) or cleft palate severity at T0 (P = 0.77).

Teeth lost adjacent to the cleft at T2 did not correlate with cleft palate severity (P = 0.64) or type of cleft at T0 (P = 1).

**DISCUSSION**

The main purpose of this study was to evaluate the long-term stability of alveolar bone grafts with an orthodontic-surgical protocol. Our success rates were 70.41% (95% CI, 60.34-79.21) at T1 and 91.84% (95% CI, 84.55-96.41) at T2; these correspond well with results reported in the literature (90%, Bergland et al in 378 patients; 83%, Amanat and Langdon in 34 patients; 91%, Long et al in 43 patients; 73%, Kindelan et al in 38 patients; 72%, Da Silva Filho et al in 50 patients; 91%, Lilja et al in 70 patients; and 95%, Jia and Mars in 55 patients).

We used the Kendall coefficient of concordance to evaluate method error, intraobserver repeatability, and interobserver reproducibility. The statistical analysis indicated that the Kendall W value was close to 1, and the method error was insignificant for the study. Furthermore, there was 87.07% agreement between the Bergland and the Witherow-derived values; therefore, we can assume that this is a simple and accurate qualitative method for assessing the success of bone grafting.

Bergland et al evaluated the height of the interdental bone ridge adjacent to the erupted canine on the cleft side, using a qualitative scale and radiographs taken at least 1 year after surgery. Long et al measured contours of the grafted bone with a qualitative method from the occlusal or periapical radiographs of 43 patients with 56 total cleft sites, taken at least 6 months after surgery. The mean follow-up time was 3.1 years (range, 0.6-8.1 years). We focused on the bone adjacent to the cleft side and measured its contours with 2 qualitative scales to assess the bone in the vertical and horizontal directions. On the occlusal radiographs of 49 patients at least 1 year after bone graft surgery, 98 cleft sites were analyzed (2 per subject). The mean follow-up time was 1.87 years (range, 0.6-8.1 years). Our follow-up was at least 1 year rather than 6 months after surgery, as reported by Long et al. This 1-year interval after the bone graft was thought to allow for adequate bone maturation to occur.

Various studies have confirmed the reliability of qualitative measurements in occlusal and periapical x-rays.

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**Table IX.** Cross-tabulation of the first operator’s first evaluation

<table>
<thead>
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<th>Type</th>
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<tbody>
<tr>
<td>I</td>
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<td>37</td>
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<td>II</td>
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<tr>
<td>III</td>
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<tr>
<td>IV</td>
<td>13</td>
<td>0</td>
<td>0</td>
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</tbody>
</table>

**Table X.** Sample distribution for sites 1 and 2 for the Bergland bone level types

<table>
<thead>
<tr>
<th>Site 1 (n)</th>
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<th>II</th>
<th>III</th>
<th>IV</th>
</tr>
</thead>
<tbody>
<tr>
<td>8</td>
<td>24</td>
<td>5</td>
<td>12</td>
<td></td>
</tr>
<tr>
<td>Site 2 (n)</td>
<td>11</td>
<td>24</td>
<td>13</td>
<td>1</td>
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</tbody>
</table>

There were 12 patients (24.48%; 95% CI, 13.34-38.87) with diminutive teeth outside the cleft area. None had supernumerary teeth far from the cleft side.
for assessing the stability of bone grafts.\textsuperscript{17,25,34,40} However, no authors have used 2 scales at the same cleft site and at the same time. Because of its high repeatability and reproducibility, the latter method is statistically more accurate. It also allows for a 2-dimensional (vertical and horizontal) assessment of the long-term success of bone grafts, exposing the patients to a low dose of cerebral radiation, in accordance with the radiation protection rules and the ethical, regulatory, and scientific principles governing clinical research in the Declaration of Helsinki (2004). Furthermore, this qualitative method eliminates the measurement errors that result from radiographic image distortions. The only error was on the minor cleft side, where some Bergland type 1 values were classified as Witherow-derived scores of 0.5.

In this study, the average age at bone grafting was 10.25 years, with a range of 8 to 14.7 years. This is similar to the data reported by Paulin et al,\textsuperscript{41} Brattström and McWilliam,\textsuperscript{42} and Long et al,\textsuperscript{17} thereby highlighting the consensus about when to perform secondary bone grafts.

The most important clinical finding in our study was that cleft severity does not statistically correlate with the rate of surgical success at T2. This finding is consistent with the report of Long et al.\textsuperscript{17} Furthermore, regression analysis showed a low correlation between the presurgical cleft width and the success of alveolar grafts. Therefore, minor, moderate, and severe clefts appear to have the same long-term stability.

Our study did not identify any clinically apparent oronasal or palatal fistulae after surgery; this agrees with the studies of Russell and McLeod\textsuperscript{43} and Long et al,\textsuperscript{17} who reported only 1 patient with a residual oronasal fistula.

In our 3 groups (minor, moderate, and severe clefts), there was no regrafting; this is lower than any rate of regrafting reported in the literature (less than 2\% in the studies of Hall and Posnick,\textsuperscript{22} Bergland et al,\textsuperscript{25} and Long et al\textsuperscript{17}). Helms et al\textsuperscript{26} reported a higher rate of regrafting (19\%) in both the secondary early and delayed graft groups, probably due to the dental age of their patients at the time of bone grafting.

Another interesting observation was that linear regression analysis demonstrated no statistically significant correlation between the patient’s sex and age (at bone grafting) and graft success at T2. Therefore, sex and age did not influence the long-term stability of bone grafts in our patients. Furthermore, there was no significant correlation between type of cleft (unilateral or bilateral) at T0 and the long-term stability of the bone graft at T2. This indicates that the type of cleft did not influence the success of bone grafts. We also did not find a significant influence of lateral incisor agenesis on the long-term stability of bone grafts at T2. This is consistent with data from Russell and McLeod.\textsuperscript{43}

Others factors that possibly contribute to the variability in graft success were excluded in this study because of these factors: the same surgeon and the same surgical technique were used, the same bone was grafted (in contrast, Long et al\textsuperscript{17} used cancellous bone from the iliac crest in 95\% of patients and cranial bone in 5\% of patients), and the eruption stage of the teeth in the distal cleft segment (canine) was identical for all patients. We could also exclude variations due to surgeon or surgical technique because we found a statistically significant improvement of Bergland and Witherow-derived values from T0 to T1.

Another important clinical finding of our study is that, at T0, there was a statistically significant difference in the Bergland bone levels between site 1 (mesial side of the cleft) and site 2 (distal side of the cleft). We found that site 2 had a higher bone level than did site 1; this is consistent with the findings of Teja et al\textsuperscript{44} and Long et al.\textsuperscript{17} Both studies attributed this difference to the eruption of the mesial tooth into an unfavorable position before the alveolar bone grafting. In addition, both authors suggested that, unlike the apex of the distal tooth, the apex of the mesial tooth was not always covered by bone. In this situation, the clinician should not use orthodontic force to prevent root resorption. At T1 and T2, there were no statistically significant differences between the Bergland bone levels at sites 1 and 2. These data correlated with good stability of the bone grafts and indicated the periodontal health of both mesial and distal teeth adjacent to the cleft side.

Another interesting finding of this study was that, at T2, 38 of the 49 patients (77.55\%; 95\% CI, 63.38-88.23) had spontaneous canine eruption through the grafted bone. The success rate in this sample is comparable with the average percentage reported in the literature (75\%). Previously reported success rates of bone graft are 97\% (Turvey et al\textsuperscript{23}), 95\% (Troxell et al\textsuperscript{45}), 95\% (Long et al\textsuperscript{17}), 92\% (Enemark et al\textsuperscript{24}), 85\% (Bergland et al\textsuperscript{25}), 80\% (Boyne and Sands\textsuperscript{15}), 73\% (Nicholson and Plint\textsuperscript{46}), and 72\% (Da Silva Filho et al\textsuperscript{27}). Bone graft integration was radiographically and histologically verified.\textsuperscript{47} However, El Deeb et al\textsuperscript{21,48} reported spontaneous eruptions in only 27\% and 41\% of the canines. They attributed their findings to the bone graft material and the surgical technique used. Bergland et al\textsuperscript{25} clarified that wide flaps of alveolar mucosa impede canine eruption and increase further surgical exposure.

We believe that the following factors are involved in spontaneous canine eruption in cleft patients: (1) arch development with previous orthopedic expansion, (2)
creation of space for canine eruption with orthodontic therapy, and (3) periodontal health of the canine with respect to the root during the graft surgery. In addition, the grafted bone itself plays a basic role by triggering a cytokine cascade through interactions between the bone and the canine follicle. The molecular basis of this phenomenon is not well characterized yet. Occasionally, canines require an orthodontic stimulus to erupt in the correct position. In our study, 11 patients (22.44%; 95% CI, 11.77–36.62) had canine impactions and required a surgical flap and orthodontic recovery. The exposure percentage of the canines varies in the literature, ranging from 9% to 17%, and the need for orthodontic traction is 5% to 56%. Patients with alveolar clefts have a 20-fold increased risk for canine impaction (18.9% compared with the general population (1%-2%).

Further research is needed to determine the correlation between canine impaction and bone height, because canine impactions might affect the bone height vs having a canine erupt into the graft. In this study, we found only 1 patient with canine impaction and incomplete bone height at T2. Therefore, canine impaction seemed not to affect the bone height after the bone graft.

In this study, 65.31% (95% CI, 50.36–78.33) of the patients had lateral incisor agenesis; this is lower than reported by Akcam et al (70.8%–97.1%) but higher than reported by Dewinter et al (50%). The high rate of dental anomalies on the cleft side might be due to a deficient blood supply or ectodermal and mesodermal tissues during embryogenesis. We found that spontaneous canine eruption at T2 was not significantly associated with lateral incisor agenesis. This is in accordance with the study of Gereltzul et al, who reported no effect of the lateral incisor on canine eruption. However, Russell and McLeod studied 101 cleft sides in patients with complete unilateral or bilateral cleft lips and cleft palates and found that 61% of those with abnormally positioned canines also had abnormal lateral incisors. In addition, there were more canines with an abnormal vertical position before alveolar bone grafting when the lateral incisor was missing. We concluded that the absence or anomalies of the lateral incisors might influence canine eruption and increase the risk for canine impaction. However, thus far, no data support this relationship.

Spontaneous canine eruption at T2 was not significantly associated with cleft severity at T0. This finding was never previously investigated, and it might result from complete healing of the cleft side after bone grafting, allowing a physiologic canine eruption through the remodeled bone.

We observed a difference between the type of cleft and spontaneous canine eruption on the cleft side. In patients with bilateral clefts, there was only 1 with a normal canine pattern of eruption at T2. But with only 4 patients (8.16%, 95% CI, 2.27–19.6) in the bilateral cleft group, we cannot use statistics to draw inferences about these 2 groups. However, alterations in eruption were probably due to a more serious impairment of the maxillary structure. The presence of deciduous teeth, lateral incisor guidance, and interactions between cytokine and bone are all well-characterized factors that influence canine eruption, which is usually absent or more compromised in bilateral than in unilateral cleft patients.

In this study, only 3 patients (6.12%; CI, 1.28–16.87) had lost teeth adjacent to the cleft side at T2. Two patients had peg-shaped lateral incisors, and 1 patient had microdontic supernumerary teeth instead of a lateral incisor; these were extracted during the graft surgery. This finding concurs with the findings of Long et al, who reported the loss of 3 teeth, accounting for a failure rate of 5%. In contrast, Helms et al reported failure rates of 22% and 59% in their study. All teeth lost were lateral incisors with anomalies in size, root and crown shapes, and altered periodontal support. Tooth loss adjacent to the cleft at T2 did not correlate with cleft severity or type of cleft at T0. This finding has not been investigated previously.

We also assessed any supernumery and diminutive teeth outside the cleft area. There were 12 (24.48%; 95% CI, 13.34–38.87) patients with diminutive teeth outside the cleft area. None had supernumerary teeth far from the cleft side. Previously, Dewinter et al also reported agenesis outside the cleft area in 27.2% of patients, and Brattström and McWilliam reported a 27.8% rate of agenesis in unilateral cleft lip and cleft palate patients. In contrast, Akcam et al showed rates from 12.5% to 52.8% for noncleft side agenesis in the anterior, premolar, and molar regions; this was explained by the severity of the cleft phenotype.

This study confirms the benefit of secondary bone grafts. By eliminating the gap, rehabilitating the alveolar morphology, and facilitating the eruption of teeth, bone grafting allows alveolar bone to develop and supports the normal functions of the teeth.

Previous studies have confirmed the importance of several variables involved in the long-term stability of alveolar bone grafts. These include the initial severity of the cleft, the surgical technique and the surgeon’s technical skills, the handling of tissues during the graft procedure, the type of graft material, the periodontal status of teeth in the proximal segment at the bone grafting, and the eruption stage of the teeth in the distal segment.
We analyzed the variables of age (at bone grafting), sex, cleft severity and type of cleft, lateral agenesis, and grading system by keeping other variables constant, such as the surgeon and the surgical bone-graft technique, the radiologist and the radiographic technique, the type of graft material, the eruption stage of the teeth in the distal segment, and the treatment protocol. According to our data, there was no significant correlation between age or sex and the long-term stability of the bone graft. There was also no significant association between cleft severity (minor, moderate, severe) at T0 and the success of the alveolar graft at T2, or between the type of cleft (unilateral or bilateral) and the stability of the alveolar graft. Finally, we found no significant correlation between lateral incisor agenesis on the cleft side and the long-term success of bone grafts. These findings implicate other factors that might cause the variability in the long-term stability of bone grafts.

Based on the evidence from this clinical trial, it is reasonable to assume that factors involved in the success of the bone graft are dental age at the time of bone grafting and the surgical and orthodontic protocol. Previous studies have emphasized the importance of exploiting the eruptive force of the incomplete canine root on the cleft side and performing appropriate surgical procedures to aid in tissue healing and bone-graft stability. However, no prior studies have addressed the importance of orthodontic therapy on the long-term stability of bone grafts. Only 1 study mentioned orthopedic treatment as a probable variable in the success of the primary bone graft; however, there was no orthodontic protocol, and the orthodontic role in the study population was unclear. Da Silva Filho et al underline the role of the bone graft in canine eruption and emphasized that pregraft orthodontic treatments yield better access for the surgeon and that postgraft orthodontic procedures can correct the positions of the permanent teeth in the grafted area. However, they did not consider that canines that are protruding through the bone graft have a functional role in the stability of the bone graft. Feichtinger et al reported that the average loss of bone was 52% in the third year after surgery and clarified the importance of canine and lateral incisor eruption through the graft. They did not emphasize the role of orthodontic therapy.

We agree with the study by Long et al and believe that it is important to achieve orthopedic maxillary protraction and expansion before graft surgery. If expansion is carried out after bone grafting, fistulae can appear, leading to an additional surgical procedure. The palatal expansion increases the transverse maxillary dimension and the sagittal projection of the nasal spine and improves the growth prognosis and the percentage of spontaneous canine eruptions. Even though Russell and McLeod asserted that this conclusion is not supported by the literature, many studies have confirmed the benefits of rapid maxillary expansion in noncleft patients and cleft patients. In noncleft patients, separation occurs in the midpalatal suture. However, in cleft patients, there is a separation between the maxilla and the premaxilla without osseous gain, as well as asymmetric bone movement in unilateral cleft patients because of their maxillofacial structure. Therefore, many authors agree on the necessity and efficiency of early transverse expansion in cleft patients.

After surgery, it is necessary to prevent a relapse by preserving the transverse maxillary dimension with a transpalatal bar. It is also necessary to encourage transverse alveolar development through orthodontic multi-bracket appliances. The application of biologic force facilitated tooth eruption on the grafted bone and contributed to its long-term stability. Biology supports the resorption of bone in the absence of functional stresses. In this case, the eruption of teeth (lateral incisor and canine) and their proper placement through bone grafting are the functional elements of the long-term stability of bone grafts. In the case of diminutive teeth and residual spaces after orthodontic therapy, the grafted side must be restored with functioning prosthetic teeth (implants) as soon as possible to prevent further bone loss. The long-term stability of dental implants in the cleft side ranges from 82.2% to 98.6%.

A limitation of this study was the absence of a control group. However, not treating patients with cleft lips and cleft palates is unethical because of a consensus on the therapeutic benefits.

It is reasonable to assume that our protocol is an adequate form of treatment for cleft lip and palate patients. It reduces the need for maxillofacial surgery at the completion of growth, and it positively affects the long-term stability of bone grafts with a greater survival rate of the teeth adjacent to the cleft.

CONCLUSIONS

Based on our data, it seems appropriate to recommend the early application of a surgical-orthodontic protocol to treat cleft lip and palate patients, prevent postoperative bone resorption, and guarantee correct positioning of the teeth.

Our findings indicate the following.

1. The success rate of bone grafts at T2 was 91.84% (95% CI, 84.55–96.41), suggesting a high percentage of success with this therapy protocol.
2. Cleft severity was not statistically correlated with success at T2, demonstrating the protocol’s efficacy and its ability to repair clefts of varying severity.

3. The concordance rate between the Bergland and the Witherow-derived scales was 87.07% (95% CI, 82.69-90.69), indicating that this is a simple and reliable 2-dimensional method for assessing the success of bone grafts as a useful clinical and experimental tool.

Additional studies involving this surgical-orthodontic protocol need to be conducted with a larger sample size and a longer follow-up period.

REFERENCES