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Retrospective analysis of clinical outcomes in bilateral cleft lip and palate patients after secondary alveolar bone grafting and premaxilla osteotomy, using a new dento-maxillary scoring system

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ABSTRACT

Evaluation of relevant clinical outcomes in patients with bilateral cleft lip and palate (BCLP) after secondary aveolar bone grafting (SABG) and premaxilla osteotomy (PMO), through the use of a new scoring system.

Data were collected retrospectively from all patients with BCLP who were operated on between 2004 and 2014, at the end of follow-up. The treatment protocol consisted of SABG + PMO in patients aged between 9 and 13 years. At the end of follow-up, the following parameters were scored: (un)interrupted dental arch, skeletal sagittal relationship, bone height using the Bergland/Abyholm criteria, and the presence of postoperative fistula. These parameters were combined to produce a dento-maxillary scoring system, giving a final score between 1 and 10. For statistical analysis, the independent *t*-test was used.

Of 55 children, 45 were suitable for analysis. The mean age at time of surgery was 12.0 years (8.9–16.4 yrs), and the mean follow-up time was 11.7 years (5.8–15.8 yrs). The average number of surgeries executed under general anesthesia was 6 (range: 3–11). The average dento-maxillary score in this patient cohort was 7.6 (1–10; median: 8). Among these patients, 31 had an uninterrupted dental arch; the average Bergland/Abyholm score was 2.07; 30 patients exhibited an Angle class I incisor relationship; and, in 38 cases, the oronasal communication was closed after SABG + PMO treatment. A significant effect of fistulas was seen on dento-maxillary score ($p = 0.001$). Specifically, a significant effect of fistulas was seen on interrupted dental arch ($p = 0.002$) and on Bergland/Abyholm score ($p = 0.037$).

The proposed dento-maxillary scoring system is a straightforward tool that can be used to describe and analyze the amount of dento-maxillary rehabilitation at the end of the treatment. Persistence of oronasal fistulas in patients with BCLP has a significant impact on interruption of the dental arch, and can influence dental results at the end of the second decade.

1. Introduction

The treatment of patients with cleft lip and palate is planned and executed to achieve an acceptable end result, in which stigmata are corrected to produce a balanced, symmetrical face with harmonic proportions. One of the end points in cleft care is an orthognathic relationship with a complete dental arch, which contributes to both aesthetics and function (Bittermann et al., 2016). To achieve this for patients with bilateral cleft lip and palate (BCLP), correct timing of the secondary alveolar bone grafting (SABG) procedure, which can be combined with a premaxillary osteotomy (PMO), is important. Successful bone grafting facilitates dental rehabilitation with the patient's dentition or with a fixed prosthesis (Vellone et al., 2017). The timing of the procedure is chosen to support successful eruption of the canine or lateral incisor into the bone graft, and therefore reduce the risk of the development of complications at the end of growth (Stoelinga et al.,

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1990; Bittermann et al., 2020). The reason for performing an SABG + PMO procedure is not only the support of teeth or dental implants. The treatment is also executed to stabilize the alveolar ridge, to provide bony support and favourable periodontal health to the teeth adjacent to the alveolar cleft, to facilitate eruption of the impacted canine, to close residual oronasal fistulas, and to support the alar base of the nose. All these factors are addressed through application of the SABG + PMO procedure (Rawashdeh and Telfah, 2008). Residual bone height after the execution of SABG + PMO procedures is important to achieve a complete dental arch (Abyholm et al., 1981; Jia, 2006; Janssen et al., 2017). Early SABG + PMO, performed on patients before the age of 10, yields the best results in terms of residual bone height and the ability to guide the canine into the bone graft (Bergland et al., 1986; Andlin-Sobocki et al., 1995; Bittermann et al., 2020). If a tooth adjacent to the cleft is absent or hypoplastic, closure of the diastema can be achieved by moving adjacent teeth orthodontically into the dental gap, by means of a segmental osteotomy of the small fragment, by auto-transplantation of redundant teeth in the grafted cleft area (Tanimoto et al., 2010), or with prosthodontics, for example via an adhesive bridge or a dental implant (Zachrisson, 2004).

The sagittal dental relationship at the end of the treatment period makes an important contribution to the stigmata of patients with cleft lip and palate. About 30–50% of these patients have retruded maxillae, which can be an indication of the need for maxillary advancement osteotomy or distraction (Good et al., 2007; Voshol et al., 2012). In an earlier study, a maxilla osteotomy was indicated in 46% of patients with BCLP (Bittermann et al., 2018).

The literature reports that a favourable end result in BCLP treatment requires approximately five to eight surgeries with general anesthesia (Cohen et al., 1995; Pai et al., 2019). In the Department of Oral and Maxillofacial Surgery of the Wilhelmina Children's Hospital at the University Medical Center Utrecht in The Netherlands (WCH cleft team Utrecht), patients have been treated by the cleft team through lip closure, soft-palate closure, hard-palate closure, removal of deciduous teeth in the cleft area, SABG with PMO, and, if necessary, a pharyngoplasty or (bi)maxillary osteotomy. Finally, an optional secondary rhinoplasty can be added if desirable.

In the literature, various scoring systems have been described that assess outcomes after cleft lip and palate surgery with respect to overall facial morphology (Mosmuller et al., 2013; Mulder et al., 2018) or specific parts of facial morphology, for example the Bauru yardstick, by which the development of the maxilla and its relation to the mandible are scored (Ozawa et al., 2011). The diseased, missing, or filled teeth (DMFT) scoring method is one system by which the dental situation is classified (Anaise, 1984). There are various questionnaires that cover mastication and oral health, through which the patient perspective can be measured. These scoring systems cover small and separate items that relate to the outcome of cleft lip and palate care. More recently, 3D imaging and analysis has also become an important method for analysis of postoperative results. This study aimed to present a method that covers all aspects of dento-maxillary rehabilitation and can be followed using available clinical and radiological data, giving a simple overall score.

In this retrospective study by the WCH cleft team at Utrecht, the end results of treatment of patients with BCLP were analyzed. Of special interest were cases where an orthognathic maxillary relationship with an uninterrupted dental arch was established. The study was performed on patients with BCLP who had undergone SABG + PMO and orthodontic treatment, and eventually prosthodontic rehabilitation.

A practical dento-maxillary scoring system was proposed to evaluate the clinical outcomes at the end of BCLP treatment.

2. Materials and methods

This study was a retrospective consecutive cohort study of all children with complete BCLPs who underwent SABG + PMO at WCH Utrecht between 2004 and 2014. The study was performed at the end of follow-up at our institution. Secondary rhinoplasties were performed after the maxillofacial rehabilitation, and therefore this procedure was part of this analysis. Treatment consisted of SABG + PMO and was timed at two-thirds through the developmental stage of the root of the maxillary canine or the lateral incisor if present. Patients were aged between 9 and 13 years.

2.1. Primary closure

Patients had been treated according the surgical BCLP protocol, which involved closure of the lip at approximately 6 months of age, according to a modified Millard or Tennison technique (Millard, 1958). Closure of the soft palate had been accomplished according to the procedure described by Sommerlad (2003) at 7–9 months of age. Closure of the hard palate had been performed as described by von Langenbeck (1861) at 3–6 years of age, with the modification that the palatal flaps were dissected epiperiosteally. The treatment protocol is shown in Table 1.

2.2. Orthodontic protocol before and after SABG + PMO

As there were no defects in the mandible, orthodontic treatment began with the creation of a mandibular dental arch, by means of fixed appliances, between the ages of 9 and 11 years, dependent on the timing of the grafting operation. Prior to the SABG + PMO, a short interceptive orthodontic maxillary arch expansion procedure was completed with a quad-helix devices or a removable appliance. Quad-helix devices were chosen for a bilateral transversal crossbite, and removable appliances were chosen in cases of unilateral or frontal crossbite. This expansion was necessary not only to enlarge the operating area and to facilitate access to it, but also to determine the future intermaxillary transverse relationship. Additionally, the aim of this procedure was to position the premaxilla in a postitive sagittal overbite and overjet to the mandibular arch, if possible. About 2–3 months after SABG + PMO, a final, long-term, active orthodontic treatment took place to create correct dental intra- and interarch relationships. Two orthodontists carried out all orthodontic treatments using fully fixed appliances. The bone in the cleft was functionally loaded by regulating adjacent teeth into the bone graft, to ensure bone continuity in the newly created alveolar ridge. After completion of the orthodontic treatment, the maxillary and mandibular front teeth were retained permanently from cuspid to cuspid with bonded retainers. In addition, transverse expansion of the maxillary arch was retained with a removable appliance, to be worn at night for the patients' lifetimes.

2.3. Surgical protocol: SABG + PMO

Planning involved mock surgery, using a custom metal splint that was pre-bent and soldered over a dental cast model to stabilize the premaxilla. Operations were performed with the use of general anesthesia. Prophylactic intravenous clindamycin (13 mg/kg) was administered at the start of surgery and continued three times daily for 3 days postoperatively. A PMO was carried out to correct the position of the premaxilla and to improve access to the nasal floor for watertight closure of the nasal mucosa. The premaxilla was replaced in a positive sagittal overjet and overbite. The premaxilla was fixated apically to the vomerine bone with a 0.4 mm stainless steel wire. After fixation of the premaxilla, the nasal layer was closed, and the premaxilla fixated with the metal splint. Both sides

Table 1
Treatment protocol.

Age	Procedure
6 months	Closure of lip (Millard, Tennison)
7–9 months	Closure of soft palate (Sommerlad)
3–6 years	Closure of hard palate (Langenbeck)
6–9 years	*Pharyngoplasty if necessary*
9–13 years	Secondary alveolar bone grafting with premaxilla osteotomy
18 years	*Orthognathic surgery if necessary*
20 years or above	*Secondary rhinoplasty if necessary*

were grafted in one procedure. For grafting, a mandibular symphyseal bone graft was preferentially used; if this was not possible, because of insufficient bone or anatomical variations, an iliac crest graft was used. After bone grafting, the oral mucosa was closed with slowly resorbable Vicryl 4-0 sutures. During the first postoperative week, the wound was protected with iodoform-vaseline gauze covered with a zinc oxide-eugenol paste. The metal splint was removed after 6 weeks.

2.4. Prosthodontic protocol

In patients with BCLP, lateral incisors are frequently absent or hypoplastic. In these cases, they may be removed during the SABG + PMO procedure. In cases where teeth are missing, an interrupted maxillary arch in good relationship with the mandible is a prerequisite for prosthetic replacement of teeth. If sufficient bone is present, it is preferable to move canines orthodontically and even (pre)premolars mesially. The diastema of a missing tooth is thus placed more distally in the dental arch to a position where aesthetics play a less important role.

In the studied cases, if there was insufficient bone for implantation, the interruption in the dental arch was either bridged by a fixed adhesive bridge or, if extra teeth were missing, it was replaced with a removable (cast cobalt-chromium) prosthesis.

2.5. Data collection

The following baseline data were collected: sex, age at surgery, follow-up time, age at final X-OPT, type of bone graft, postoperative fistula with reoperation, pharyngoplasty, type of osteotomy (Le Fort I or bimaxillary osteotomy), number of surgeries, number of surgeries exceeding six. Data on the placement of tympanotomy tubes and rhinoplasties were not recorded for this study.

2.6. Scoring system

A dento-maxillary scoring system was proposed to measure parameters that influence clinical outcome, specified at the level of the maxillary arch, hard palate, and dentition. These parameters were considered critical in evaluation of maxillary and dental treatment outcomes (Table 2). The parameters used in the scoring system were: the (un)interrupted dental arch, sagittal relationship (lateral cephalometric radiograph), Bergland/Abyholm criteria (Bergland et al., 1986), and the presence of fistulas. Sagittal relationship was scored as a negative overbite and overjet, an end-to-end relationship, or a positive sagittal overbite and overjet. The ultimate goal was to create a practical and quick scoring system for patients with BCLP, which evaluated the end result of dento-maxillary treatment. As such, a score between 1 and ten 10 was applied, comparable to a visual analogue scoring system, with 1 being the worst outcome and 10 the best.

Analysis of the dental arch, the sagittal relationship, and the Bergland/Abyholm score (Bergland et al., 1986) was performed by two raters, KB and RE, and scores were discussed until consensus was reached. The Bergland/Abyholm score is divided into four grades, with grade 1 being the best result and 4 the worst (the dento-maxillary scoring system is shown in Table 2, with reference images in Fig. 1). The Bergland/Abyholm score was measured by use of panoramic X-rays, as recommended by Schultze-Mosgau et al. (2003). The Bergland/Abyholm score, which was to be incorporated into the dento-maxillary scoring system, was measured for each side. These scores ranged from 0 to 1.5. As BCLP has two cleft sides, the maximum Bergland/Abyholm score was 3.

2.7. Statistical analysis

The baseline characteristics of all patients were reported as categorical variables. The Statistical Package for the Social Sciences (SPSS for Mac, release 24.0.0.0, 2016, SPSS Inc.) was used for all statistical analyses. The independent T-test was used to calculate the effect of the preoperative parameters, an osteotomy, a pharyngoplasty, the presence of fistulas and number of surgeries on the outcome in the dento-maxillary scoring system. The level of significance was set at $p < 0.05$.

3. Results

3.1. Baseline data

Of 55 children with BCLP who were treated, the records of 45 were suitable for analysis. Ten patients were excluded because the data needed for analysis were not available. All pertinent clinical baseline data for the 45 patients included in this study are presented in Table 3. The mean age at time of surgery was 12.0 years (8.9–16.4 yrs), and the mean follow-up time was 11.7 years (5.8–15.8 yrs). Panoramic and cephalometric radiographs were taken at a mean age of 19.5 years (15.04–28.9 yrs). Seven of the 45 patients underwent revisional surgery because of postoperative oronasal communications. Fourteen patients received a pharyngoplasty because of velo-pharyngeal insufficiency. Twenty-three patients were treated with a (bi)maxillary osteotomy because of either a retruded maxillary position or a malocclusion that could not be corrected by orthodontics alone.

3.2. Number of surgeries

The average number of surgeries conducted under general anesthesia was 6 (range: 3–11). Additional surgeries are listed in Table 4.

3.3. Scoring system

The average dento-maxillary score in this patient cohort was 7.6 (range: 1–10, median: 8). Table 5 summarizes all parameters separately. In the dental arch analysis, 31 patients had an uninterrupted dental arch. The average Bergland score was 2.07. In 30 of

Table 2
Scoring of dento-maxillary treatment results.

Parameter	Description	Score	Modality
1. Dental arch between both maxillary canines	Both sides with interruption with or without removable prosthetics	0	Panoramic radiographs
	One side interrupted	1	
	Two sides without interruption by use of implants or fixed prosthetics	2	
	Two sides uninterrupted without prosthetics	3	
2. Incisor relationship	Negative overbite and overjet	0	Lateral ceph
	End-to-end	1	
	Positive overbite and overjet	2	
3. Oronasal fistulas	Persisting fistula	0	Patient files
	Fistula closed after revision surgery	1	
	Fistula closed by SABG + PMO	2	
4. Bergland/Abyholm criteria (per side[a])	Grade 4: no bone	0	Panoramic radiographs
	Grade 3: Bone level less than ¾ of normal bone level	0.5	
	Grade 2: At least ¾ of normal bone level	1	
	Grade 1: Normal bone level	1.5	
Maximal total score	Dento-maxillary BCLP score	10	

[a] Bergland/Abyholm score per patient is measured on each cleft side separately.

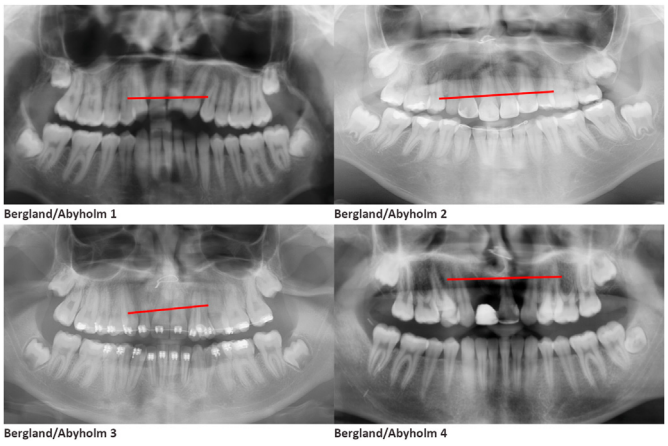

Fig. 1. Bergland/Abyholm criteria reference images.

the patients, a final positive overbite and overjet incisor relationship was achieved. In 38 patients, the oronasal communication was closed after SABG + PMO. In seven patients, the oronasal communication was closed through application of an additional procedure.

Table 6 shows the individual effects of separate parameters on the dento-maxillary score. Only the parameter relating to fistulas showed a statistically significant effect on the total score ($p = 0.001$).

Table 7 shows the relationship between parameters of the dento-maxillary scoring system. Both the presence of fistulas and the Bergland/Abyholm score proved to be of significant relevance to the presence of an uninterrupted dental arch ($p = 0.002$ and $p = 0.035$, respectively).

4. Discussion

Our study analyzed the end results of alveolar cleft closure, orthodontics, and prosthodontic rehabilitation of 45 patients with BCLP who were treated in a dedicated cleft-care centre. The results were presented through application of a newly proposed dento-maxillary scoring system. A previous study addressed dental arch morphology and skeletal relationships (Bittermann et al., 2018). This new scoring system is intended to cover the complete dento-maxillary result of BCLP treatment. It applies four typical factors for measuring dental maxillary outcome. To analyze the results at bone level, the criteria developed by Bergland/Abyholm (Bergland et al., 1986) are used and extended through consideration of the presence or absence of postoperative fistulas, as applied by others (Schultze-Mosgau et al., 2003). Dental factors play an important role in BCLP stigmata (Posnick and Kinard, 2020). Therefore, the complete or incomplete nature of the dental arch and the sagittal relationship of the incisors are included as factors in this scoring system.

4.1. Surgical protocol

The protocol used in our study was secondary aveolar bone grafting (SABG) combined with a premaxilla osteotomy. The eruption of the canine was used as guidance for planning the surgery. Different protocols have been planned in the literature, including secondary alveolar bonegrafting without osteotomy of the premaxilla, or early secondary alveolar bone grafting, with use of the incisors as guidance for planning (Fahradyan et al., 2019). For optimal timing of alveolar bone grafting, not only is the residual bone important, but also the residual growth and eventually growth retardation if surgery is done at an early age (Padwa et al., 1999). More recent research shows promising results for an early alveolar bone grafting procedure, but this needs more investigation of skeletal growth (Siegenthaler et al., 2018).

In our study, a premaxilla osteotomy was performed in all cases to gain access to the nasal floor and ensure watertight closure of the nasal mucosa. In the presented patient group, access to the nasal floor was difficult due to the nearly complete closure of the hard palate in earlier surgeries. A review addressing different protocols for closure of the alveolar cleft and the effect on outcome was carried out by Bittermann et al. (2015). 51.1% of the patients in the present cohort were treated with a (bi)maxillary osteotomy to correct their skeletal profile. Bartzela et al. presented a study comparing different surgery protocols between cleft centers, including those performing a premaxilla osteotomy. They did not find significant differences in growth between centers when using the Bauru yardstick (Bartzela et al., 2010). However, it remains possible that midfacial growth is affected by osteotomy of the premaxilla. The effect of other and earlier surgical procedures should also not be ruled out.

Recently, standardization of the evaluation of cleft lip and palate care using patient-reported outcome measures (PROMs) has been advocated. Also, the International Consortium for Health Outcomes Measurement (ICHOM) has started to develop a set of questionnaires and guidelines for cleft lip and palate care. The parameters that are recommended for scoring are: mastication, oral health, dental health, and occlusion (Allori et al., 2017). Mastication and

Table 3
Baseline clinical data.

	Number ($n = 45$)	
Male	26	58%
Female	19	42%
Mean age at time of surgery	12 yrs	range: 8.9–16.4 yrs
Mean follow-up	11.7 yrs	range: 5.8–15.8 yrs
Mean age when final X-panoramic taken	19.5 yrs	range: 15.0–28.9 yrs
Bone graft mandibular symphysis	33	82.2%
Bone graft crista illiaca	8	17.8%
Postoperative oronasal communications with reoperation (fistulas)	7	16%
Pharyngoplasty	14	31%
(Bi)maxillary osteotomy	23	51%
Average no. of surgical procedures	6	range: 3–11
Number of patients with > six surgeries	15	33%

Table 4
Overview of patients who underwent extra surgical procedures under general anesthesia ($n = 45$).

Patient number	Extra procedures beyond six	Type of procedure
1	1	Redo closure soft palate
2	1	Redo closure lip
3	4	Redo closure lip, redo closure hard palate, redo SABG + PMO procedure, surgical removal of maxillary incisor
4	1	Redo closure lip
5	1	Redo SABG + PMO procedure
6	4	Redo closure soft palate, Redo closure lip, redo SABG + PMO procedure, removal of osteosynthesis material
7	2	Redo pharyngoplasty, ligation of canine
8	2	Nose correction at early age, necrotectomy after nose correction
9	1	Tonsillectomy
10	2	Nose correction at early age, extraction of deciduous teeth
11	1	Redo SABG + PMO procedure
12	1	Gingiva correction
13	1	Pharyngeal fat graft
14	1	Additional fistula closure
15	2	Redo lip closure, additional lip correction

Table 5
Scores for factors that comprise the dento-maxillary scoring system ($n = 45$).

$N = 45$	Score	
Mean dento-maxillary score	7.6	SD 2.2
Dental arch analysis:		
Both sides with interruption with or without removable denture	6	13%
One side interrupted	2	4%
Two sides without interruption (with implants or fixed prosthodontics)	6	13%
Two sides without interruption	31	69%
Bergland/Abyholm gradation (number of sides = 90):		
Mean score (1.00–4.00)	2.07	SD 1.08
Score 1 or 2	63	76%
Score 3 or 4	27	24%
Incisor relationship:		
Negative overbite and overjet	9	20%
End-to-end	6	13%
Positive overbite and overjet	30	67%
Oronasal fistulas around premaxilla:		
Persisting fistula	0	0%
Fistula closed after revisional surgery	7	15.6%
Fistula closed after SABG + PMO	38	84.4%

oral health are scored by questionnaires. A decay-missing-filled (DMF) index (Anaise, 1984) is applied for dental health, and the Goslon yardstick (Bartzela et al., 2010) for occlusion. These scoring systems are designed to enable intercenter evaluation of all aspects of BCLP treatment. The patient-reported outcomes of this BCLP cohort had already been evaluated in an earlier study by Kappen et al. (2019). However, the proposed dento-maxillary scoring system is useful to provide a quick, overall clinical evaluation of dental, orthodontic, and prosthetic end results after BCLP treatment. It focuses on clinical outcome, not patient-reported outcome.

Table 6
Effects of additional surgeries (pharyngoplasty, Le Fort I osteotomy) and fistulas after SABG + PMO, and number of surgeries beyond 6, on the dento-maxillary scoring system end result ($n = 45$).

	n	Mean score	Range of scores	SD	*p*-value
All patients	45	7.6	(1–10)	2.20	
Pharyngoplasty	14	8.178	3–10	1.97	0.256
Le Fort I	23	7.304	1–10	2.61	0.324
Number of surgeries > 6	15	7.933	2–10	2.25	0.448
Fistulas	7	5.286	1–9	2.86	0.001

Calculated using the independent sample *t*-test.

Table 7
Effects of fistulas on different parameters of the dento-maxillary scoring system ($n = 45$).

	Mean	SD	*p*-value
Fistulas vs Bergland/Abyholm	1.5714	1.304	0.275
Fistulas vs dental arch	1.2857	1.380	0.002
Fistulas vs incisor relationship	1.4286	0.975	0.895
Bergland/Abyholm vs dental arch	1.500	1.109	0.037

Calculated using the independent sample *t*-test; $p < 0.05$.

Several other scoring systems concerning the outcomes of cleft lip and palate treatments are described in the literature. These scoring systems are designed to score maxillary growth and do not include the different dental aspects of cleft lip and palate treatment, i.e. surgery, orthodontics, and prosthetics. For instance, the Bauru yardstick (Ozawa et al., 2011) and the Huddart and Bodenham index (Huddart and Bodenham, 1972) were developed to analyze growth at the level of occlusion (Pai et al., 2019).

4.2. Validation and alveolar bone height

In our proposed scoring system, the validated Bergland/Abyholm criteria were used to analyze bone height. In 34 patients, the Bergland/Abyholm score was 1 or 2, which seemed to be a sufficient result that was comparable with those reported in the literature (Schultze-Mosgau et al., 2003). The three other factors that were added (dental arch, sagittal relation, and fistula) were objective parameters that did not require validation. This new method scored the complete treatment period throughout childhood until the age of at least 18 years on a scale from 0 to 10. This BCLP cohort scored an average of 7.62 in this new scoring system. It would be of interest to compare this score with those for other cleft care units.

4.3. Fistulas

It was observed that the presence of residual fistulas that had to be closed through application of additional surgery was associated with a lower score in the dento-maxillary scoring system. Our study showed a significant correlation between the presence of residual fistulas after closure of the alveolar cleft and the result for the dental arch at the end of follow-up ($p = 0.002$). Clinically relevant residual fistulas occur immediately around the premaxillary bone, and impair alveolar ridge integrity. This explains the significant relationship between the interrupted dental arch and the occurrence of fistulas. To the best of our knowledge, this correlation between the occurrence of fistulas and that of an interrupted dental arch has not been described previously.

In our study, fistulas were found to occur after SABG + PMO in seven patients. The presence of fistulas after the SABG + PMO procedure may be related to difficulties with closure of the several layers during surgery. It has been reported that nasal closure can be performed more accurately if SABG is combined with a PMO (Scott et al., 2017). Scott et al. described the consecutive, completed treatment of 44 patients with BCLP through SABG + PMO. They found residual fistulas in 11% of the patients (Scott et al., 2017), with a slightly lower percentage of fistulas after surgery.

Pepper et al. studied the presence of fistulas in uni- and bilateral cleft cases (Pepper et al., 2014). They reported an overall fistula rate of 10% post SABG, and an 8% rate in the bilateral cases. They also performed PMOs in all cases, as in our cohort. However, information on the pre- and postoperative positions of the premaxillae was missing in their report. The mean age at which surgery was performed in our study was 12.0 years, compared with 11.4 years in the Pepper et al. study. As timing is important in relation to outcome due to the eruption of the cuspid in the cleft, the higher percentage of fistulas in our study might be explained by the average age of 12.0 years. It should be mentioned that in this study, as well as that of Pepper et al. the age range of included patients was wide, which could make the groups less comparable.

4.4. Orthodontics

The orthodontic treatment in our study achieved uninterrupted dental arches on both sides in 69% of cases. In a study on timing of alveolar bone-grafting surgery in unilateral cleft patients, Enemark et al. reported successful outcomes without prosthodontics in only 39% of the patients (Enemark et al., 1985). Over the years, protocols have been modernized, and this has resulted in a better overall outcome for the patient.

In the literature it is shown that the orthodontics protocol is an important factor in achieving a good end result. Presurgical as well as postsurgical orthodontics are important in achieving the best end result. In their study, Liao and Huang did find better results with presurgical repositioning of the adjacent teeth and postsurgical movement of the teeth into the grafted area (Liao and Huang, 2015).

4.5. Prosthodontics

In cases of missing lateral incisors, our treatment protocol advocated that the distal teeth should be mesialized into the grafted area. If indicated, prosthodontic rehabilitation with an implant was then executed in the premolar region. This procedure has been found to be more reliable than implantation in the alveolar cleft area (Cune et al., 2004). Härtel et al. suggested that the use of implants in the grafted area can be reliable (Härtel et al., 1999). However, implants should be placed shortly after secondary alveolar bone grafting; the Utrecht cleft team does not consider this to be an option in children before the end of adulthood. By applying mesialization of the posterior teeth, as advocated by Semb and Ramstad (1999), natural teeth are retained in the aesthetic zone, and stable prosthodontics are positioned in the lateral part of the maxilla, outside the cleft area. In the dento-maxillary scoring system, this outcome would be scored as an uninterrupted arch. A second option would be to preserve the diastema and place an implant in the grafted area (Cassolato et al., 2009; Alberga et al., 2020). However, if fistulas occurred after SABG and PMO treatment, the amount of bone present in the cleft might be reduced, which would lead to difficulty in placement of implants in the grafted area or orthodontic mesialization of the distal teeth. As a result, these patients would be more likely to be fitted with removable prosthodontics, and this result would give lower overall scores in the dento-maxillary scoring system. This outcome is demonstrated in Table 7 by the association between a lower Bergland/Abyholm score and an interrupted dental arch.

4.6. Multiple surgeries

Patients with BCLP need to undergo multiple surgeries to reach an acceptable end result. It is generally seen in patients with clefts that they develop an aversion to surgery over time. Therefore, from a patient point of view it is important to reach an acceptable end result with as few surgeries as possible. The average number of surgeries under general anesthesia at the end of follow-up was 6 (range 3–11). This seems reasonable, since patients with BCLP often need six operations to achieve a satisfactory end result, with a positive vertical and sagittal overbite, an uninterrupted arch, and no fistulas found: four separate primary closures of lip, soft palate, hard palate, and alveolar cleft, with an additional pharyngoplasty and a maxillary osteotomy may be necessary. Late, secondary nose and/or lip corrections were not taken into consideration in this study, as these procedures were performed after final orthodontic treatment and orthognathic surgery, usually in the late teenage years or early adulthood. Cohen et al. reported that the average number of surgeries in patients with BCLP was 8. However, in 62% of their patients, a primary lip adhesion was performed, and they included secondary nose corrections in their data (Cohen et al., 1995). If these figures were excluded, their result would be comparable with that found for the Utrecht cohort. In the Utrecht centre, lip adhesion is rarely part of the BCLP treatment protocol; instead, performance of a lip adhesion results in at least one additional surgical treatment.

In our study, a pharyngeoplasty was performed in 14 patients (31%). The protocol used for closure of the hard palate was comparable to that used at the same centre for patients with a unilateral cleft lip and palate. A study was carried to investigate this group of patients, and a pharyngoplasty rate of 40% was found (Kappen et al., 2017). This group was compared with a patient group in a study by Lohmander et al. reporting a pharyngoplasty rate of 11%, which is considerably lower (Lohmander et al., 2012). It might be possible that these patients benefit from an early closure of the hard palate. However, the study by Kappen et al. demonstrated a very heterogeneous outcome between the different protocols (Kappen et al., 2017).

Pai et al. reduced the number of surgical procedures for patients with unilateral cleft lip and palate by combining speech-enhancing surgery with the SABG procedure. This reduced the total number of procedures to an average of 4.8; the researchers did not mention the range in their report (Pai et al., 2019). Alternatively, pharyngoplasty and closure of the alveolar cleft may be performed at different times.

5. Strengths and limitations

There were some limitations in our study, which may have some implications for the outcome of the research. For example, sample size and power were not calculated. All patients at the Utrecht centre with BCLP who were available in the selected time period were selected and included. The data for these patients were retrieved retrospectively by analyzing patient files. In this patient group, there was a wide range in age at surgery, follow-up time, and age at which the X-rays were taken. This was due to the fact that data were collected during regular consultations, which may influence the results of this study.

It is known that if secondary alveolar bonegrafting is performed at an older age, the results of the procedure may be worse. The wide age range for final follow-up radiographs may have had a limited effect, as all radiographs were taken at the end of the treatment protocol.

This study was able to demonstrate the end results for a large cohort of BCLP patients with a long follow-up period.

6. Conclusion

The proposed dento-maxillary scoring system is a straightforward and easy-to-use tool for describing and analyzing overall dento-maxillary reconstruction at the end of treatment for patients with BCLP.

An average of 7.6 on a scale from 1 to 10 was scored in the BCLP patient group.

This study shows that the persistence of oronasal fistulas in patients with BCLP has a relevant impact on interruption of the dental arch, and influences the dental result at the end of the second decade.

Declaration of Competing Interest

The authors declare that they have no conflicts of interest.

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List of abbreviations

BCLP	bilateral cleft lip and palate
SABG	secondary alveolar bone grafting
PMO	premaxilla osteotomy
SABG + PMO	secondary alveolar bone grafting and premaxilla osteotomy
Yrs	years
WCH cleft team Utrecht	Wilhelmina Children's Hospital cleft team at the University Medical Center Utrecht, The Netherlands

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Ethical approval

The Medical Ethics Committee of the Utrecht University Medical Center approved the protocol (14/417). All procedures that were performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committees, and with the 1964 Helsinki declaration and its later amendments, or comparable ethical standards. For this type of retrospective study, formal consent was not required.

References

Abyholm, F.E., Bergland, O., Semb, G., 1981. Secondary bone grafting of alveolar clefts. A surgical/orthodontic treatment enabling a non-prosthodontic rehabilitation in cleft lip and palate patients. Scand. J. Plast. Reconstr. Surg. Suppl. 15, 127–140.

Alberga, J.M., Stellingsma, K., Meijer, H.J.A., Oostenbrink, H.A., Vissink, A., Raghoebar, G.M., 2020. Dental implant placement in alveolar cleft patients: a retrospective comparative study on clinical and aesthetic outcomes. Int J Oral Maxillofac Surg 49, 952–959.

Allori, A.C., Kelley, T., Meara, J.G., Albert, A., Bonanthaya, K., Chapman, K., Cunningham, M., Daskalogiannakis, J., De Gier, H., Heggie, A.A., Hernandez, C., Jackson, O., Jones, Y., Kangesu, L., Koudstaal, M.J., Kuchhal, R., Lohmander, A., Long, R.E., Magee, L., Monson, L., Rose, E., Sitzman, T.J., Taylor, J.A., Thorburn, G., Van Eeden, M., Williams, C., Wirthlin, D.D.S.D., Wong, K.W., 2017. A standard set of outcome measures for the comprehensive appraisal of cleft care. Cleft Palate-Craniofacial J 54, 540–554.

Anaise, J.Z., 1984. Measurement of dental caries experience — modification of the DMFT index. Community Dent Oral Epidemiol 12 (43–46).

Andlin-Sobocki, A., Å, L., Eliasson, Paulin, G., 1995. Periodontal evaluation of teeth in bone grafted regions in patients with unilateral cleft lip and cleft palate. Am. J. Orthod. Dentofacial Orthop 107, 144–152.
Bartzela, T., Katsaros, C., Shaw, W.C., Rønning, E., Rizell, S., Bronkhorst, E., Okada, T.O., Pinheiro, F.H. de S.L., Dominguez-Gonzalez, S., Hagberg, C., Semb, G., Kuijpers-Jagtman, A.M., 2010. A longitudinal three-center study of dental arch relationship in patients with bilateral cleft lip and palate. Cleft Palate Craniofac J 47, 167–174.
Bergland, O., Semb, G., Abyholm, F.E., 1986. Elimination of the residual alveolar cleft by secondary bone grafting and subsequent orthodontic treatment. Cleft Palate J 23, 175–205.
Bittermann, G.K.P., van Es, R.J.J., de Ruiter, A.P., Frank, M.H., Bittermann, A.J.N., van der Molen, A.B.M., Koole, R., Rosenberg, A.J.W.P., 2020. Incidence of complications in secondary alveolar bone grafting of bilateral clefts with premaxillary osteotomy: a retrospective cohort study. Clin Oral Investig 24, 915–925.
Bittermann, G.K.P., de Ruiter, A.P., Bittermann, A.J., Mink van de Molen, A.B., van Es, R.J., Koole, R., Rosenberg, A.J., 2018. Midfacial growth and dental arch relationships in bilateral cleft palate following secondary alveolar bone grafting and orthodontic intervention: factors predicting a Le Fort I osteotomy at age 18. J Cranio-Maxillofacial Surg 46, 1764–1771.
Bittermann, G.K.P., de Ruiter, A.P., Janssen, N.G., Bittermann, A.J.N., van der Molen, A.M., van Es, R.J.J., Rosenberg, A.J.W.P., Koole, R., 2016. Management of the premaxilla in the treatment of bilateral cleft of lip and palate: what can the literature tell us? Clin Oral Investig 20, 207–217.
Bittermann, G.K.P., de Ruiter, A.P., Janssen, N.G., Bittermann, A.J.N., van der Molen, A.M., van Es, R.J.J., Rosenberg, A.J.W.P., Koole, R., 2015. Management of the premaxilla in the treatment of bilateral cleft of lip and palate: what can the literature tell us? Clinical Oral Investigations.
Cassolato, S.F., Ross, B., Daskalogiannakis, J., Noble, J., Tompson, B., Paedo, D., Ortho, D., 2009. Treatment of dental anomalies in children with complete unilateral cleft lip and palate at SickKids hospital, Toronto. Cleft Palate-Craniofacial J 46, 166–172.
Cohen, S.R., Corrigan, M., Wilmot, J., Trotman, C.A., 1995. Cumulative operative procedures in patients aged 14 years and older with unilateral or bilateral cleft lip and palate. Plast. Reconstr. Surg 96, 267–271.
Cune, M.S., Meijer, G.J., Koole, R., 2004. Anterior tooth replacement with implants in grafted alveolar cleft sites: a case series. Clin Oral Implant. Res 15, 616–624.
Enemark, H., Krantz-Simonsen, E., Schramm, J.E., 1985. Secondary bone grafting in unilateral cleft lip palate patients: indications and treatment procedure. Int J Oral Surg 14, 2–10.
Fahradyan, A., Tsuha, M., Wolfswinkel, E.M., Mitchell, K.A.S., Hammoudeh, J.A., Magee, W., 2019. Optimal timing of secondary alveolar bone grafting: a literature review. J Oral Maxillofac Surg 77, 843–849.
Good, P.M., Mulliken, J.B., Padwa, B.L., 2007. Frequency of Le Fort I osteotomy after repaired cleft lip and palate or cleft palate. Cleft Palate Craniofac J 44, 396–401.
Härtel, J., Pögl, C., Henkel, K.O., Gundlach, K.K.H., 1999. Dental implants in alveolar cleft patients: a retrospective study. J Cranio-Maxillofacial Surg 27, 354–357.
Huddart, A.G., Bodenham, R.S., 1972. The evaluation of arch form and occlusion in unilateral cleft palate subjects. Cleft Palate J 9, 194–209.
Janssen, N.G., Schreurs, R., Bittermann, G.K.P., Borstlap, W.A., Koole, R., Meijer, G.J., Maal, T.J.J., 2017. A novel semi-automatic segmentation protocol for volumetric assessment of alveolar cleft grafting procedures. J Cranio-Maxillofacial Surg i 1–5.
Jia, Y.L., 2006. Long-term outcome of secondary alveolar bone grafting in patients with various types of cleft. Br J Oral Maxillofac Surg 44, 308–312.
Kappen, I.F.P.M., Bittermann, D., Janssen, L., Pieter Bittermann, G.K., Boonacker, C., Haverkamp, S., de Wilde, H., van der Heul, M., Specken, T.F., Koole, R., Kon, M., Breugem, C.C., van der Molen, A.B.M., 2017. Long-term follow-up study of young adults treated for unilateral complete cleft lip, alveolus, and palate by a treatment protocol including two-stage palatoplasty: speech outcomes. Arch Plast Surg 44, 194–209.
Kappen, I.F.P.M., Bittermann, G.K.P., Stock, N.M., Mink van der Molen, A.B., Breugem, C.C., Swanenburg de Veye, H.F.N., 2019. Quality of life and patient satisfaction in adults treated for a cleft lip and palate: a qualitative analysis. Cleft Palate-Craniofacial J 56, 1171–1180.
von Langenbeck, B., 1861. Operation der angeboren totalen Spaltung des harten Gauments nach einer neuen Methode. Drsch Klin 8, 231.
Liao, Y.F., Huang, C.S., 2015. Presurgical and postsurgical orthodontics are associated with superior secondary alveolar bone grafting outcomes. J Cranio-Maxillofacial Surg 43, 717–723.
Lohmander, A., Friede, H., Lilja, J., 2012. Long-term, longitudinal follow-up of individuals with unilateral cleft lip and palate after the Gothenburg primary early veloplasty and delayed hard palate closure protocol: speech outcome. Cleft Palate-Craniofacial J 49, 657–671.
Millard, D.R., 1958. Columella lengthening by a forked flap. Plast Reconstr Surg Transplant Bull 22, 454–457.
Mosmuller, D.G.M., Don Griot, J.P.W., Bijnen, C.L., Niessen, F.B., 2013. Scoring systems of cleft-related facial deformities: a review of literature. Cleft Palate-Craniofacial J 50, 286–296.
Mulder, F.J., Mosmuller, D.G.M., De Vet, H.C.W., Mouës, C.M., Breugem, C.C., Van Der Molen, A.B.M., Don Griot, J.P.W., 2018. The cleft aesthetic rating scale for 18-year-old unilateral cleft lip and palate patients: a tool for nasolabial aesthetics assessment. Cleft Palate-Craniofacial J 55, 1006–1012.
Ozawa, T.O., Shaw, W.C., Katsaros, C., Kuijpers-Jagtman, A.M., Hagberg, C., Rønning, E., Semb, G., 2011. A new yardstick for rating dental arch relationship in patients with complete bilateral cleft lip and palate. Cleft Palate Craniofac J 48, 167–172.
Padwa, B.L., Sonis, A., Bagheri, S., Mulliken, J.B., 1999. Children with repaired bilateral cleft lip/palate: effect of age at premaxillary osteotomy on facial growth. Plast Reconstr Surg 104, 1261–1269.
Pai, B.C.J., Hung, Y.T., Wang, R.S.H., Lo, L.J., 2019. Outcome of patients with complete unilateral cleft lip and palate: 20-year follow-up of a treatment protocol. Plast Reconstr Surg 143, 359e–367e.
Pepper, H.B., Revington, P.J., Deacon, S., Thomas, S., 2014. Impact of secondary alveolar bone grafting on the symptoms of reflux in oronasal fistulas: a patient-related outcome study. Cleft Palate-Craniofacial J 51, 270–273.
Posnick, J.C., Kinard, B.E., 2020. Challenges in the successful reconstruction of cleft lip and palate: managing the nasomaxillary deformity in adolescence. Page Plast Reconstr Surg 591e–603e.
Rawashdeh, M.A., Telfah, H., 2008. Secondary alveolar bone grafting: the dilemma of donor site selection and morbidity. Br J Oral Maxillofac Surg 46, 665–670.
Schultze-Mosgau, S., Nkenke, E., Schlegel, A.K., Hirschfelder, U., Wiltfang, J., 2003. Analysis of bone resorption after secondary alveolar cleft bone grafts before and after canine eruption in connection with orthodontic gap closure or prosthodontic treatment. J Oral Maxillofac Surg 61, 1245–1248.
Scott, R., Scott, J., Stagnell, S., Robinson, S., Flood, T., 2017. Outcomes of 44 consecutive complete bilateral cleft lip and palate patients treated with secondary alveolar bone grafting and premaxillary osteotomy. Cleft Palate-Craniofacial J 54, 249–255.
Semb, G., Ramstad, T., 1999. The influence of alveolar bone grafting on the orthodontic and prosthodontic treatment of patients with cleft lip and palate. Dent Update 26, 60–64.
Siegenthaler, M., Bettelini, L., Brudnicki, A., Rachwalski, M., Fudalej, P.S., 2018. Early versus late alveolar bone grafting in unilateral cleft lip and palate: dental arch relationships in pre-adolescent patients. J Cranio-Maxillofacial Surg 46, 2052–2057.
Sommerlad, B.C., 2003. A technique for cleft palate repair. Plast Reconstr Surg 112, 1542–1548.
Stoelinga, P.J., Haers, P.E., Leenen, R.J., Soubry, R.J., Blijdorp, P.A., Schoenaers, J.H., 1990. Late management of secondarily grafted clefts. Int. J. Oral. Maxillofac. Surg 19, 97–102.
Tanimoto, K., Yanagida, T., Tanne, K., 2010. Orthodontic treatment with tooth transplantation for patients with cleft lip and palate. Cleft Palate-Craniofacial J 47, 499–506.
Vellone, V., Cirignaco, G., Cavarretta, B., Cascone, P., 2017. Canine eruption after secondary alveolar bone graft in unilateral cleft lip and palate patients. J Craniofac Surg 28.
Voshol, I.E., Van Der Wal, K.G.H., Van Adrichem, L.N.A., Ongkosuwito, E.M., Koudstaal, M.J., 2012. The frequency of Le Fort I osteotomy in cleft patients. Cleft Palate-Craniofacial J 49, 160–166.
Zachrisson, B.U., 2004. Letters to the editor. Am J Orthod Dentofac Orthop 126, A13–A15.