

Factors influencing the decision for orthognathic surgery in cleft palate patients

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Abstract:

In the complex rehabilitation of patients with cleft lip and palate, a secondary surgical correction is often necessary at the end of the growth period in order to optimize the skeletal jaw position.

The aim of this study was to retrospectively identify those factors in the initial diagnosis, treatment approaches and findings at the time of decision-making, which statistically describe the assignment of patients to a group receiving only orthodontic treatment and to a group receiving surgical correction of the jaw position.

The study assessed the findings and analyses of 55 adult patients with non syndromic cleft (38 who received orthognathic surgery at the end and 17 who only received orthodontic treatment). The 25 parameters, collected for the initial cleft formation, the treatment concept and the timing of the bone graft, also included additional parameters from cephalometrics and dental cast analysis.

A total of 1,210 items of the 55 patients were evaluated using a classification algorithm (CART analysis). The target was to identify the parameters that were responsible for assigning the patients to the specified groups.

As expected, both groups differed significantly in various parameters. However, an overbite depth indicator (ODI) of 73.7° can be interpreted as the threshold influencing assignment to the respective group. The probability that the patients who received orthognathic surgery were correctly assigned was 90% in contrast to all other parameters.

As a result, the ODI is a simple value which could be used to estimate the likelihood that a secondary surgical correction in cleft patients is needed.

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Introduction

The midface, maxilla and mandible exhibit characteristic growth changes in patients with operated cleft lip and palate. However, these are individually very distinctive. The growth potential of the maxilla is probably not primarily limited (1, 2).

After surgical correction, frequently described characteristic changes occur during subsequent growth (3, 4, 5, 6, 7, 8). Treatment measures play an additional role.

The maxilla is described as shortened and retro positioned in relation to the base of the skull, and the mandible is more posteriorly oriented, shortened and the mandibular plane is steep.

The impact which special treatment measures have on craniofacial growth and the development of the dentition has been the subject of several studies, but the results are inconsistent. It isn't possible to differentiate between intrinsic growth changes and the effects of various forms of treatment. In multi-center studies, different treatment concepts produced varying results; however the identification of causal factors was not possible (9).

Even the prognostic significance of the initial expression of the cleft formation is assessed differently. **Johnson et al.** (10) found no connection between the original extent of the cleft and the treatment outcome at the age of six. **Chiu et al.** (11) and **Wiggmann et al.** (12), however, described how the initial cleft expression had a significant effect on the growth of the maxilla. **Suzuki et al.** (13) reported a correlation between the cleft width and the lateral displacement of the nasomaxillary complex in unilateral cleft patients. The presence of a Simonart's band is attributed to having a positive influence on the future skeletal configuration in adulthood (14).

The positive effect of pre-surgical orthodontics (15) on the growth of the cleft segments and an earlier normalization of tongue function was demonstrated (16), but even here there are some critics (17).

The number of previous surgical interventions does not seem to be a valid predictor of the need for a secondary surgical correction (18).

However, it can be assumed that a combined orthodontic-maxillofacial surgical correction of the jaw position after growth has stopped seems to be a reasonable option for at least one-fifth of all cleft patients (19, 20). For this reason orthognathic surgery can be considered to be an integral part of the treatment concept (21) since the conditions for lip and nose correction can be significantly improved.

In addition to these individual aspects, the basic design of the treatment seems to play a crucial role in the long-term outcome (12).

When treating patients with cleft lip and palate, the team is confronted in almost every patient with the question of whether a secondary surgical correction in the form of an osteotomy is recommended or not. This is a serious decision for the patient and is influenced by many parameters.

Question: Is it possible to identify retrospectively patterns or specific parameters that influence the decision to carry out secondary surgical correction?

Patients and methods

Patients

Medical files of 55 patients at University Hospital Leipzig were made available for this retrospective study. These included 21 women and 34 men. Thirty-eight underwent surgical correction after growth stopped (surgery group) and 17 received purely orthodontic treatment (orthodontics group). Patients with syndromic cleft were not considered. An exchange of patients between the groups did not take place.

Parameters

Patient-related aspects, types of therapy and dental and skeletal factors were taken into account (Table 1). The selection was limited to quantifiable (ordinal / metric) features which were found in all patients. The selection cannot claim to be complete since, in view of the limited number of patients, there should be a significantly fewer number of parameters than this to obtain statistically meaningful results. Thus it was necessary to limit the number of parameters.

Patient- and treatment-related parameters were determined retrospectively using treatment documentation. The age refers to the age at which the final diagnosis (retention plan) of a purely orthodontic correction (orthodontics group) was made and, in the case of the surgery group, the age at which the need for a combined orthodontic-maxillofacial surgical therapy was initially diagnosed.

Nonexistent teeth were categorized into agenesis in the cleft region (missing laterals, Table 1) and teeth extracted due to pathological conditions (tooth loss).

The subtopics of concept, timing of the bone graft and veloplasty are substantially influenced by patients who were treated according to the concept of late palate closure (22). Patients with early closure of the cleft palate were treated according to the concept of functional rehabilitation (23). The majority of the patients were treated with plates according to Hotz / Gnoinski as a part of pre-surgical orthodontics (15, 16). Some patients in the group with late cleft palate closure received active treatment with plates according to McNeil.

The only distinction made between the type of orthodontic appliances used was whether they were removable or fixed.

The dental casts were analyzed using a digital caliper (Mannesmann, Mannesmann Brothers Tools GmbH, Remscheid, Germany). The survey was based on the measurement points to the transverse arch width (24, 25). A difference was calculated independently of standard values as a difference between the upper and lower dental arch width. This was done to better reflect the specific situation found in cleft patients. Negative values indicate a change in the direction of the crossbite.

Cephalometric analysis was performed using the OnyxCeph 3™ (image instruments GmbH, Chemnitz, Germany). All radiographs were evaluated by the same examiner.

A selection of values (SNA, SNB, ANB, NSBa, ML–NSL, NL–NSL and ML–NL) according to the analysis by Segner and Hasund (26) was supplemented by two complex parameters of the Denture Frame Analysis (27, 28). These were, firstly the ODI (overbite depth indicator), described as the arithmetic sum of the angle between AB line and mandibular plane and the angle between the upper jaw and the Frankfort horizontal base plane. This is regarded as the degree of the vertical relationship between the upper and lower jaw at a maximum correlation with the overbite. The second value was the APDI (Antero Posterior Dysplasia Indicator) which is the sum of three angles. These are the angle between the Frankfort horizontal plane and facial plane, the AB Line to the facial plane, and the angle of the Frankfort plane itself. This is regarded as the degree of the sagittal relationship between the upper and the lower jaw. The transferability to a Central European population was demonstrated (29).

Statistics

The differences between groups were tested using the t-test for independent samples and the Kruskal–Wallis test (30).

The CART analysis used here (Classification and Regression Trees) is a way to solve the statistical problem of classification (31), i.e. the assignment of objects to classes based on the expression of certain variables.

CART analysis was developed by **Breimann et al.** (32) and is a tool for classification and regression decision trees. It is based on a binary recursive partition. A parent node is exactly divided into two child nodes and this process is recursively traced further to the respective child nodes (33, 34).

All of the possible splits of the variables involved are calculated and the results are sorted according to their quality. The ones whose class is as pure as possible are selected. This splitting is recursively continued until no further division is possible or until a predefined termination criterion is reached. The generated decision tree is then shortened to improve the quality of decision (35, 36, 37).

The main advantage of this non-parametric method lies in its great robustness against outliers and in the fact that both categorical and continuous variables can be processed. In addition, there is invariance with regard to the monotonic transformation of the variables.

Medical applications have demonstrated that this methodology is well-suited (38, 34, 39).

All statistical calculations were performed with R 2.15.2 (33, 40).

Table 1: Overview of the selected parameters		
Parameter	Description	Values / Scale
Patient-related parameters		
Age	Age at begin / end of treatment	[years]
Gender	Gender of the patient	[male, female]
Initial cleft	Initial diagnosis	[Bilateral cleft lip and palate (BCLP), Cleft lip and palate (CLP), Cleft palate (CP)]
Missing laterals	At least one missing upper lateral incisor (cleft region)	[yes, no]
Tooth loss	Loss of teeth (caries, trauma, attachment loss...) other than cleft region	[yes, no]
Treatment related parameters		
Treatment concept	General treatment concept	[early, late, other]
Bone graft	Timing and/or presence of bone graft	[no, early, late]
Velopharyngeal flap		[yes, no]
Pre-surgical orthodontics	Use of plates before surgical closure of palate	[no, Hotz-Gnoinski, McNeil]
Lower bicuspid extraction	Extraction of lower bicuspid for orthodontic reasons - class III compensation attempt	[yes, no]
Orthodontic appliances	Orthodontic appliances predominantly used in this case	[removable, fixed]
Dental casts		
Anterior arch width difference	Difference between upper and lower anterior arch width according to Korkhaus	[mm]
Posterior arch width difference	Difference between upper and lower posterior arch width according to Korkhaus	[mm]
Overbite	Overbite and overjet measured on dental cast in occlusion	[mm]
Overjet		[mm]
Cephalogram		
SNA	Basic skeletal parameters according to Segner and Hasund	[°]
SNB		[°]
ANB		[°]
NSBa		[°]
ML-NL		[°]
NL-NSL		[°]
ML-NSL		[°]
ODI	Overbite depth indicator (Kim)	[°]
APDI	Anterior posterior dysplasia indicator (Kim)	[°]
Wits appraisal [19]	Distance of perpendicular lines to occlusal plane from points A and B	[mm]

Results

Comparison of the groups

The groups studied differ significantly in their age structure. The group of patients with secondary surgical correction is older by an average of eight years. The groups did not differ with regard to the distribution of gender and the severity of the cleft. The group with an isolated cleft palate was significantly unequally distributed. The groups did not differ with regard to the frequency of aplasia of the lateral incisor in the cleft region.

The different treatment approach was reflected in the points concept, bone graft, veloplasty and pre-surgical orthodontics.

The features that depict aspects of the therapy differ significantly from one another in most of their characteristics (Table 2). They do not differ in patients who had mandibular bicuspid extraction as a supposedly compensative measure.

Table 2: Treatment and patient-related parameters.

	Surgery [n=38]		Orthodontics [n=17]		p
	Mean	SD	Mean	SD	
Age [years]	24.3	10.39	16.5	5.10	0.0004 #
Gender	male 26 female 12		male 8 female 9		0.1350
Initial cleft	BCLP 9 CLP 26 CP 3		BCLP 7 CLP 10 CP 0		0.1183
Missing laterals	yes 30 no 8		yes 14 no 3		0.7725
Concept	early 2 late 34 other 2		early 9 late 7 other 1		0.0000 *
Bone graft	no 1 early 29 late 8		no 1 early 7 late 9		0.0044 *
Veloplasty	yes 21 no 17		yes 4 no 13		0.0163 *
Pre-surgical ortho- dontics	no 30 McNeil 6 Hotz 2		no 7 McNeil 2 Hotz 8		0.0015 *
Lower bicuspid extraction	yes 11 no 27		yes 5 no 12		0.6121
Orthodontic appli- ances	fixed 14 removable 24		fixed 16 removable 1		0.0002 *

t-test for independent samples
all other Kruskal-Wallis one-way analyses of variance

In the dental cast analysis there was no significant difference between the groups in terms of the overbite. The surgery group had a pronounced difference between the posterior and anterior arch widths of more than 4mm on average. The overjet was significantly smaller in the surgery group.

In the evaluation of the groups' cephalometric parameters, significant differences were observed in the SNB and ANB angles. SNB was significantly larger (77 °) in the group with secondary surgical correction than in the orthodontics group (73.2 °), but still below the standard value (79.8 °). SNA was reduced in both groups in terms of maxillary retrusion. The angle of the cranial base and the vertical parameters ML-NSL, NL-NSL and ML-NL did not differ (Table 3).

Table 3: Cast analysis and cephalometric parameters.

	Surgery		Orthodontics		p#
	Mean	SD	Mean	SD	
HZB_diff [mm]	-4,5	3,85	-1,3	2,49	0,0157 *
VZB_diff [mm]	-4,9	3,98	-0,1	1,86	0,0001 **
Overbite [mm]	2,5	2,82	1,9	1,64	0,4118
Overjet [mm]	-3,1	3,75	3,7	1,03	0,0000 **
SNA [°]	74,0	5,37	76,2	5,11	0,1201
SNB [°]	77,0	5,00	73,2	3,95	0,0025 *
ANB [°]	-3,0	4,54	3,0	3,40	0,0000 **
NSBa [°]	131,7	5,12	133,4	4,48	0,1997
ML-NSL [°]	36,9	8,00	35,4	6,44	0,435
NL-NSL [°]	8,1	4,49	8,6	3,92	0,7011
ML-NL [°]	28,5	7,33	26,8	6,50	0,3647
ODI [°]	60,9	8,81	75,4	6,39	0,0000 **
APDI [°]	89,4	9,18	77,4	6,87	0,0000 **
Wits [mm]	-4.2	4.27	0.4	2.47	0.0000 **

t-test for independent samples

The composite values of the ODI and APDI differed significantly. ODI was significantly smaller in the surgery group at 60.9° (population mean = 74.5°) while the APDI was larger at 89.4° (population mean = 81.4°). Both values in the orthodontics group approximated the mean values much more closely (ODI 75.4° and APDI 77.4°) and there was also a significantly lower standard deviation.

CART Analysis

CART Analysis was carried out using a classification algorithm, as categorical variables were included in the data matrix. A total of 1,210 patient items were included in the calculation.

Of the 22 parameters, the variables ANB, ODI and overjet were selected by the algorithm to construct the classification tree. A root node error of 0.3091 produced a resubstitution error of 3% (error rate in the existing data set) and a prediction error of 29% was calculated after cross-validation.

The plot of the number of splits compared to the relative error (Fig. 1) revealed that the first split showed the greatest information gain. Thus no more than two branches should form. Even the information gain produced by the second split was minimal (1-SE rule). The tree contains four endpoints (three splits), the smallest cross-validation error (xError) was found in nsplit = 1, i.e. only one branching would be sufficient in principle (Table 4).

Table 4: Summary of the model arranged from smallest tree (no split) to the largest tree (3 splits).

Complexity parameter (CP)	Number of splits	Relative error	Error	Standard Deviation
0.6471	0	1.0000	1.0000	0.2016
0.1177	1	0.3529	0.82353	0.1903
0.0100	3	0.1176	0.9411	0.1981

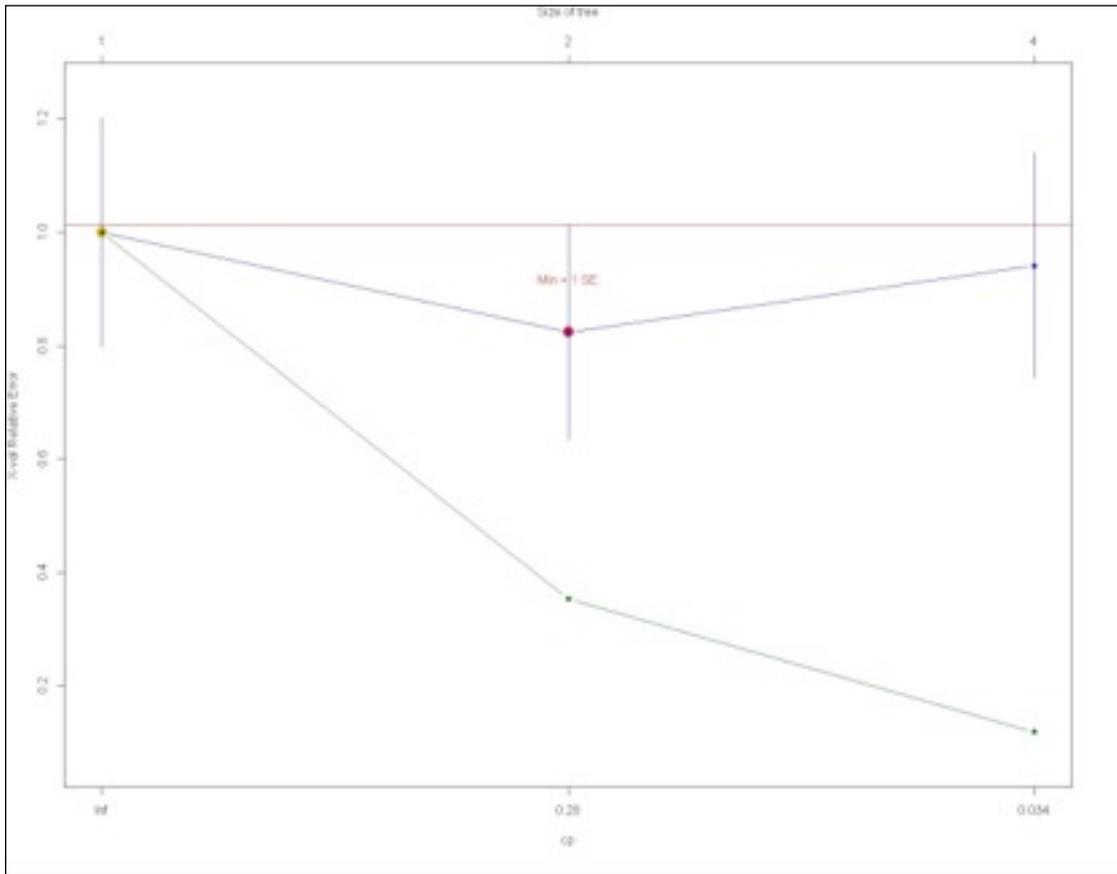


Figure 1: Comparison of the number of splits and relative error

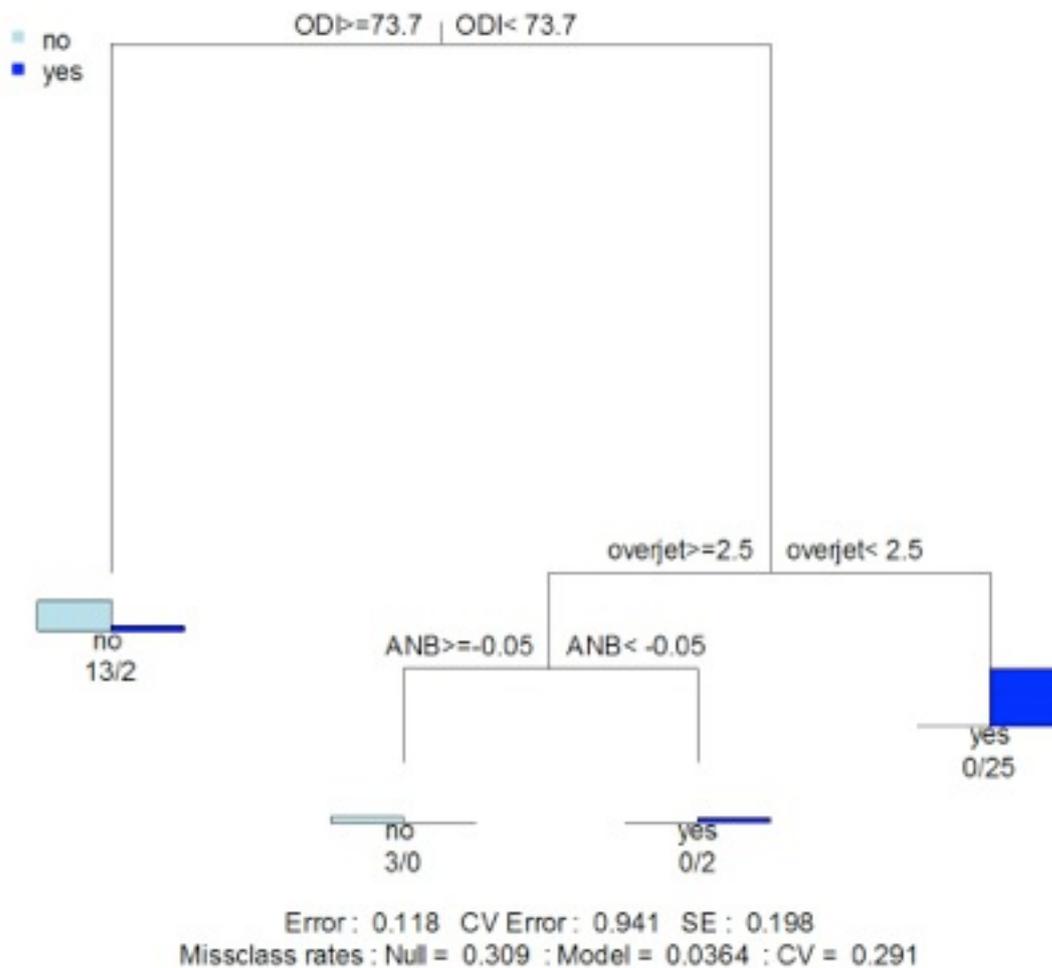


Figure 2: Classification tree

The first division (Fig. 2) was determined using an ODI with a value of 73.7° . Fifteen patients (orthodontic treatment - orthodontics group) were assigned to the left branch of the tree (Fig. 2) and 40 patients (orthognathic surgery group) to the right branch. Two patients (surgery = yes) were assigned to the left branch and 4 patients (surgery = no) were assigned to the right branch by classification error. This corresponds to an 86% correct assignment for the orthodontics group and a 90% correct assignment for the surgery group. An overjet of less than 2.5 mm in 25 patients led to a correct group assignment. In patients with a larger overjet, an ANB which was less than -0.05° (on average) was decisive.

From the small number of patients grouped according to ANB and overjet, ODI was determined to be a decisive parameter with a threshold of 73.7° (population mean = 74.5°).

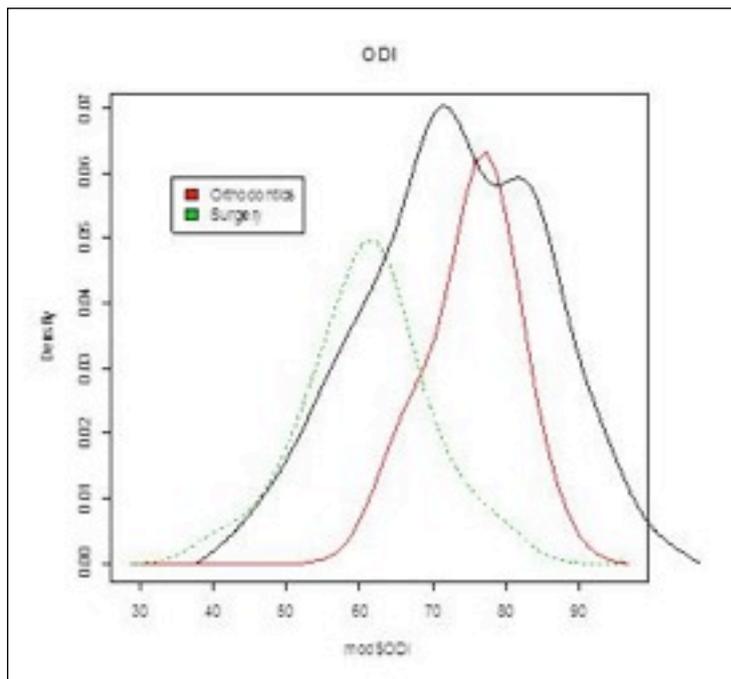


Figure 3: Density plot (55) of both groups compared with a scaled sample distribution (black) of ODI in the population.

Discussion

Several attempts to estimate the need for a combined orthodontic–maxillofacial surgical procedure on the basis of cephalometric parameters [12, 41] have been described in the literature. **Rabie et al.** (41) used a discriminant analysis and was able to show an ANB $< -5^\circ$ and a Holdaway angle (12°) as differentiation criteria. Other authors used multivariate statistics (42, 43), ROC analysis (44) or regression algorithms (42). Wit's value, overjet, ANB, SNA and maxillary basal plane angles are consistently cited (43) as crucial parameters. Due to the characteristic change in growth pattern, however, these considerations are only partly transferable to patients with cleft lip and palate.

In a follow-up of 107 six-year-old children, **Haliövaara et al.** (45) showed that none of these patients with cleft lip and palate required later surgical jaw correction if they had an ANB $> 4^\circ$. If the ANB was less than -1° all patients required an osteotomy of the maxilla.

The observed differences between the groups are the result of different therapeutic approaches (significant difference between the groups in the point Concept, Table 2), frequency of bone grafting and different orthodontic treatment strategies. This is reflected, among other things, in the findings of the dental arch width, overjet and ANB angle (Table 3). Furthermore, an individual dentoalveolar optimum is achieved in the group which received orthodontic treatment (8).

Since the groups showed no significant differences in the distribution of cleft formation characteristics, no correlation between initial severity and the need for future orthognathic surgery can be drawn (10).

In contrast, **Voshol et al.** (46) found an increase in the frequency of Le Fort I osteotomies depending on the occurrence and severity of the cleft (25% for cleft lip and palate). Other authors suggest a frequency of up to 66% (47, 48].

Regardless of all the other factors examined, an overjet of < 2.5 mm, an ANB angle of $< 0^\circ$ and an SNA angle of $< 74.5^\circ$ were statistically the most influential factors. This is largely consistent with the results of the above tests for Class III patients.

The effects expected from the type of the treatment concept, the presence of a velopharyngeal flap, agenesis of incisors in the cleft region and the type of orthodontic treatment could not be substantiated.

It is highly probable that an ODI of $< 74^\circ$ is the differentiating factor between the two groups ex post. This requires an explanation, since the population mean for ODI is 74.5° (29) (Fig. 3).

ODI is the numeric sum of the angle between the AB plane and the mandibular plane angle plus the maxillary base plane and Frankfort horizontal plane angle and represents the correlation with the overbite as given by Kim (27) with $r=0.588$. This means that by including the AB line, there is an extension of the vertical angle by a value which denotes the dentoalveolar relationships. **Wardlaw et al.** (49) already showed in the cephalometric analysis that compound parameters, such as the ODI, have a higher significance than singular values.

The cause of the observed effect can probably be postulated by overlaying the existing growth pattern with characteristic changes after surgical correction of the cleft. Patients with a more horizontal growth pattern may compensate for the vertical growth rotation typical in cleft patients (7) which would enable successful orthodontic treatment (Fig. 3).

At this point it should be noted that the skill of the surgeon is most likely very decisive, being an essential factor in long-term prognosis (50, 51, 18). It is known that the extent of surgical mucoperiosteal mobilization in the primary operation plays a major role in subsequent midfacial growth (52).

The patient's satisfaction with the treatment outcome relies on many factors (53) and is not only based on the measurable occlusal findings (54). Many patients seek surgical jaw correction in adulthood for aesthetic reasons (47). Improvements in rhinoplasty for cleft patients are often decisive in the decision to opt for a treatment strategy where the position of the maxilla is surgically corrected.

It should also be noted that aspects like aesthetics, facial symmetry, profile shape, and prosthetic rehabilitation were not considered in the present investigation.

Conclusions

The results indicate that the ODI could be a simple indicator for assessing the need for surgical correction in cleft patients in adulthood. All other investigated parameters did not significantly contribute in a statistical way in differentiating between patient groups. The different mean ages of the groups may cause a switch of some patients only treated orthodontically to the orthognathic surgery group. However, other authors are invited to test this method with their own data.

This prospective value has to be proven in other prospective studies and with a larger sample size.

The authors hereby declare no conflict of interest.

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