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Prediction of orthognathic surgery need in children with unilateral cleft lip palate: Dental arch relationships and 5-year-olds' index

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Objective: To evaluate whether dental arch relationships at 6 years of age can categorize treatment outcome and predict later need for orthognathic surgery in children with unilateral cleft lip and palate (UCLP).

Setting and sample population: A retrospective longitudinal single-center study. The study sample comprised 70 consecutive nonsyndromic children (47 boys) with complete UCLP operated on by pushback techniques during 1981-1989 and followed until early adulthood in the same cleft center.

Materials and Methods: Dental casts and maxillomandibular relationships were assessed before orthodontic treatment and secondary alveolar bone grafting at mean age 6.1 years (range 5.6-6.8) using the 5-year-olds' index and lateral cephalograms. The need for orthognathic surgery was retrieved from patient files. Student´s t test, Pearson's correlation, and Kappa statistics were used in statistical analyses.

Results: Orthognathic surgery frequency was 41% (29/70). Those needing orthognathic surgery comprised all 3 patients with an index score of 5 (very poor), 14 of 17 (82%) scoring 4 (poor), 10 of 26 (38%) scoring 3 (fair), and 2 of 19 (11%) scoring 2 (good). Of the five patients with index score 1 (excellent), none needed osteotomies. The mean index score was 2.9. The score was significantly better in those without orthognathic surgery (2.4 versus 3.6). A significant negative correlation existed between the 5-year-olds' index and cephalometric angles ANB and anb.

Conclusion: The use of 5-year-olds' index may help to predict treatment outcome and the clinical need for orthognathic surgery especially in patients with the lowest and highest index scores.

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Introduction

Children with unilateral cleft lip and palate (UCLP) need treatment and follow-up by multidisciplinary teams from birth to adolescence. The treatment goal is to achieve good speech, hearing, maxillary growth, facial esthetics, and psychosocial well-being with a reasonable burden of care. However, even with modern surgical techniques and multidisciplinary teams, patients with UCLP present an ongoing treatment challenge. Maxillary retrusion with anterior and lateral crossbites are typical findings regardless of the treatment approach.

Facial growth in repaired UCLP is characterized by a progressive retrusion of the profile relative to the cranial base involving the nasal bone, the mandible, but especially the maxilla.[1] Because of the growth discrepancy, a subgroup of UCLP patients need orthognathic surgery. Recently reported frequencies of orthognathic surgery in patients with UCLP range from 20.1% to 48.3%.[2-6]

For evaluation and comparison of the severity of malocclusion and crossbite in growing children with UCLP, several indices have been developed. These indices can also be used for categorization and prediction of treatment outcome. The ones most common for children with UCLP include the 5-year-olds' index[7], the GOSLON Yardstick[8], the Huddart/Bodenham (HB) index[9], and the Modified Huddart Bodenham (MBH) index.[10] The 5-year-olds' and GOSLON indices categorize occlusal outcome into five categories from excellent to very poor. The anteroposterior relationship (anterior crossbite) is the most important aspect, but assessment also includes vertical (deep bite, open bite) and transversal (extent lateral crossbite) relationships. The 5-year-olds' index is meant for the deciduous dentition, whereas the GOSLON Yardstick grades dental arch relationships in the late mixed and early permanent dentition. The HB and MHB use a numerical cumulative scoring of the crossbite. The more negative the score, the more severe the crossbite. These indices have proven reliable and capable of discriminating the quality of dental arch relationships in large inter-center studies[11-15] but there is lack of long-term follow-up studies for categorizing and predicting treatment outcome.

The aim of this study was to evaluate whether dental arch relationships grading with the 5-year-olds' index can categorize treatment outcome and predict long-term need for orthognathic surgery in patients with UCLP. The hypothesis was that children with later osteotomies would have poorer index scores than those who did not.

Materials and Methods

Patients

The patients comprised of consecutive 70 Finnish children (47 boys and 23 girls, mean age 6.1 years, range 5.6-6.8) with complete UCLP without syndromes who had been operated on and followed until early adulthood and the end of growth at the Cleft Palate and Craniofacial Center, Department of Plastic Surgery, Helsinki University Hospital. The patients attend regular follow-up clinics where both dental casts and cephalograms are taken. In this study, the dental casts and the cephalograms of the same 6-year-old follow-up visit were used. Eight patients were excluded from the study because of poor-quality or missing records. The girls were followed at least until 16 years and boys until 18. The primary operations were done between 1981 and 1989. Since then, our cleft center has entered the Scandcleft randomised controlled trial of primary surgery in UCLP, and the number of operating plastic surgeons as well as the methods and timing of primary palatal surgery have all changed.

Surgery and orthodontic treatment

Surgical information was retrieved from patient files. The methods of lip repair at the age of 3 to 6 months were modifications of Millard I and II. Palatal closure was at age 0.6 to 1.9 years by use of the Veau-Wardill-Kilner V-Y pushback operation (n=42) or the Cronin modification (n=28) of the Veau-Wardill-Kilner V-Y pushback operation. In the Cronin technique, performance of the V-Y pushback is exactly as in the Veau-Wardill-Killner repair with two mucoperiosteal palatal flaps, but it entails additional mucosal flaps from the floor of the nose to the nasal side of the soft palate. These operations were performed by 9 surgeons, most of them high-volume plastic surgeons but some of them residents in plastic surgery undergoing training.

Of the 70 children, 19 had undergone secondary operations before the 6-year-old models and cephalograms were taken. These included closure of fistula for 12 (6 children with and 6 without later orthognathic surgery), surgery for speech for 8 (4 children with and 4 without later orthognathic surgery), and 3 who had lip revision. None of the children had had bone grafting of the alveolar cleft or orthodontic treatment before the 6-year-old follow-up visit. In our cleft center, secondary alveolar bone grafting (with cancellous bone from iliac crest) is performed most preferably between 9 and 11 years of age before the eruption of upper canines. Orthodontic treatment is initiated before bone grafting. Maxillary protraction facemasks with skeletal anchorage are not used during treatment in patients with UCLP.

The main indication for orthognathic surgery is crossbite and maxillary hypoplasia with poor facial harmony and unbalanced profile. Surgery is also recommended for patients with vertical maxillary deficiency with edentulous appearance and an inadequate smile line. Bimaxillary osteotomy is usually necessary for correction of maxillary hypoplasia with facial asymmetries, canting of the occlusal plane, and severe anteroposterior discrepancy.

The patients were classified as needing a maxillary osteotomy based on the recommendation of the cleft team even if the patient refused the surgery. The cleft team (oral and maxillofacial surgeon or plastic surgeon and orthodontist) analyses the need for orthognathic surgery by clinical assessment, facial photographs, dental models and orthopantomograms (OPT), lateral cephalograms, and posteroanterior X-rays. In addition, CBCT data can be added.

Study models and cephalometrics

The 5-year-olds' index was used in assessment of dental-arch relationships into five categories from very poor to excellent. The ratings were done blindly by one senior orthodontist. Twenty randomly chosen models were rated twice in order to calculate intra-rater reliability. The definitions of the index with examples of the models are given in Figure 1.

Standard lateral cephalometric radiographs were taken at the same follow-up visit as the dental models. The lateral cephalometric radiographs were taken with the head positioned in the Frankfort horizontal plane with molar teeth occluded and lips in repose. The enlargement factor was corrected, and the cephalograms were traced blindly by a computer-connected digitizer by one senior orthodontist. Twenty randomly chosen cephalograms were digitized twice in order to calculate intra-rater reliability. The reference points and landmarks represented the relation of the maxilla and mandible to the cranial base (SNA, SNB), to each other (ANB), and the corresponding soft tissue relations (Sna, Snb, anb).

Statistical methods

Kappa statistics were calculated to assess reliability. Student’s t test was used to compare 5-year-olds’ index and the cephalometric variables in the groups with and without orthognathic surgery. The Pearson correlation was calculated to assess the relationship of the 5-year-olds’ index and the cephalometric angles. Test statistics with p-values equal to or less than 0.05 were considered significant.

Ethical issues

The study protocol was approved by Helsinki University Hospital and adhered to the principles outlined in the Declaration of Helsinki.

Results

Surgery and orthodontic treatment

Those with an overall later need for orthognathic surgery among the 70 numbered 29 (16 boys, 13 girls) (41%), although 4 patients refused the operation. Five patients had bimaxillary osteotomies because of maxillary hypoplasia with severe anteroposterior discrepancy, facial asymmetry, and canting of the occlusal plane.

All patients had later orthodontic treatment with fixed appliances but none of the patients used protraction facemasks or class III intermaxillary elastics with skeletal anchorage. All patients had cancellous alveolar bone grafts from iliac crest between 9-13 years of age.

Study models and cephalometrics

Those needing orthognathic surgery comprised all 3 patients with an index score of 5 (very poor), 14 of 17 (82%) scoring 4 (poor), 10 of 26 (38%) scoring 3 (fair), and 2 of 19 (11%) scoring 2 (good). Of the five patients with index score 1 (excellent), none needed osteotomies (Figure 2.).

The mean 5-year-olds' index score for the entire sample was 2.9 (SD 1.0). The proportion with fair, good, and excellent occlusion (index scores 3-1) was 71%. The mean index score was significantly better for those needing no orthognathic surgery (2.4; SD 0.8) than in those needing surgery (3.6; SD 0.8), ($p <0.001$.) No significant differences were detectable between boys or girls or patients undergoing the Veau-Wardill-Kilner V-Y pushback operation or the Cronin modification.

Patients who needed orthognathic surgery had significantly smaller mean values angles ANB and anb at the age of 6 years than those who needed no osteotomies (1.3 versus 3.7, $p<0.001$, and 4.7 versus 6.9, $p<0.001$) (Table 1). No cephalometric differences emerged between boys and girls or patients who had been operated on with the Veau-Wardill-Kilner V-Y pushback operation versus the Cronin modification.

Significant negative correlations ($p<0.001$) existed between with the 5-year-olds' index and the angles ANB and anb ($r = -0.674$ and -0.593).

Reliability

Very good levels of agreement were obtained in the 5-year-olds' index scores and cephalometric landmarks according to Kappa statistics. The intra-rater reliability of the 5-year-olds' index scores was 0.903. The intra-rater reliabilities of the cephalometric landmarks ranged from 0.891 (landmark n) to 0.924 (landmark A).

Discussion

Predictability of the 5-year-olds' index

In order to evaluate, categorize, predict, and compare the results of various surgical techniques in children with UCLP, what is essential is a reliable method of assessing long-term occlusal outcome and severity of crossbite. Based on our findings, the 5-year-olds' index could predict of treatment outcome and need for orthognathic surgery in early adulthood in patients with index scores 4 and 5 (poor and very poor). The predictive value was uncertain in index group 3 (fair), whereas in groups 2 and 1 (good and excellent), orthognathic surgery was not likely to be necessary. These findings agree with those of Miteff et al[6], who used the GOSLON Yardstick in 66 children aged 9 with UCLP but disagree with those of Suzuki et al[16], who found no correlation between GOSLON Yardstick scores and maxillofacial growth in 85 children with UCLP between age 5 and 15. On the other hand, the latter evaluated dental arch relationships and cephalometrics in patients who had undergone orthodontic treatment, whereas the patients of Miteff et al[6] and our study underwent their assessment before receiving orthodontic treatment or bone grafting. Orthodontic treatment and possible correction of crossbite may influence the GOSLON index positively.[17]

Recently the reliability and predictive validity of the 5-year-olds' index and GOSLON Yardstick was evaluated in patients with UCLP at 5, 7/8, 10, 15/16, and 19 years.[18] The predictive value of "good" dental arch relationship scores (1 and 2) over time was good in all age groups (n=106) whereas the prediction of cases in group 3 was very poor at all ages. Of the 5-year-olds starting in groups 3, 4, or 5, 60% had a good or fair dental arch relationship at 19 years.

An advantage of early assessment of outcome is the later planning of orthodontic treatment. In good and excellent cases, orthodontic treatment can usually be done conventionally, whereas in the very poor and poor cases, avoidance of orthodontic dental compensation of the crossbite and skeletal deficiency in patients who will likely need later orthognathic surgery is essential. A finding well documented in longitudinal cephalometric studies is that the maxillary growth deficiency in UCLP becomes progressively more apparent during the pubertal growth spurt.[19-21] A limitation in using the dento-occlusal relations before orthodontic treatment is that several factors may influence later facial growth and development. These include congenital dysmorphology of the midface, other variations intrinsically associated with the cleft and initial surgery, later functional adaptations, and additional surgical treatment.[1]

Controversy exists about the importance of primary surgery on the outcome of the cleft patient. The patients of our retrospective study underwent VY-pushback palatoplasty. In patients with UCLP, this method has resulted in increased scar tissue and increased prevalence of crossbites[22,23], as well as in increased need for orthognathic surgery.[24] Nowadays, the methods of primary operations as well as the number and volume of operating surgeons in our center have changed.

A shortcoming of this paper is the number and varying experience of number of surgeons, which introduces uncontrolled variables. Unfortunately, the material was small for statistical analysis. Shaw and Semb[25] concluded in the Scandcleft study that the familiarity with procedures and operator skill outweigh protocol in importance. Other limitations include single rater for dental models and cephalometrics, and the lack of objective cephalometric measurements for the decision on the need for orthognathic surgery. The strength of this paper is the long follow-up time of the same surgical protocol in the same center. The time lapse between primary surgery and measurement of outcome in early adulthood is a challenge in research of cleft lip and palate.

Measuring outcome with the 5-year-olds' index

The 5-year-olds' index was able to take into account the severity of the malocclusion as a whole and estimate discrepancies between anterior and posterior occlusion with good reliability, although no retrospective study can establish a true cause-and-effect relationship. The mean score was 2.9, comparable to scores of other studies involving Wardill-Killner pushback techniques[4,16,23,24], but is worse than the scores nowadays of 2.5 to 2.8.[14] One advantage of the 5-year-olds' index is that later in mixed and permanent dentition, the GOSLON Yardstick, with a similar 5-point scale can be used. The GOSLON Yardstick is the most popular index.[26]

An important aspect in the grading are borderline cases with an index score of 3. Mars et al[27] compared the 5-year-olds'index and the GOSLON Yardstick in the same 10-year-old UCLP patients. They recommend patients with edge-to-edge bite to be assigned to group 2 rather than group 3. Moreover, Peterson et al[24] found that GOSLON index groups 2 and 3 receive almost identical ranges of MHB scores in the 5-year age group. Merging groups 2 and 3 when assessing the primary dentition of 5-year-olds was thus their recommendation.

Another important aspect in the evaluation is, when judging whether anterior crossbite malocclusion is of dental or skeletal origin, to emphasize the apical base relationship. Whereas a total anterior and bilateral posterior crossbite is likely to be related to deficient maxillary growth (index scores 4 and 5), a simple anterior crossbite, especially on the cleft side, may be due to dental malposition. The prevalence of anterior crossbite and the dental arch dimensions did not differ between our 6-year-old children with UCLP who later needed orthognathic surgery and those who did not.[28] The incidence of crossbite in children with UCLP increases from 40% to 78% in early mixed dentition irrespective of the arch configuration of the deciduous dentition.[29] An additional consideration is the registration of accurate occlusal relationship with young children who may be in either the deciduous dentition stage or in the early transitional dentition stage.

The conventional methods of scoring arch relationships and the predictive validity of the GOSLON Yardstick has been questioned.[30] Altalibi et al[26] recommend considering the MBH index to be the standard for measuring outcome in patients with clefts, whereas Jones et al[31] support the use of the 5-year-olds' index and MBH index at 5 years of age and the GOSLON index at 10 years of age. The predictive validity was similar for MBH, GOSLON index and 5-year-olds' index with a 50-65% prediction of final outcome from 5 and 10 years. In the clinical use, the 5-year-olds' index is easy and fast, although it provides less information about the sites of occlusal discrepancy than does the MBH index. Furthermore, correlations exist between these dental indices.[32,33]

The need for orthognathic surgery

The comparison of studies on the incidence of orthognathic surgery in UCLP are hampered by several factors such as primary surgery, type and extent of the original cleft, small number of patients, and the clinical criteria of the cleft team for orthognathic surgery. Daskalogiannakis et al[34] used ANB angle, Harvold unit difference, and Wits appraisal to assess the need for orthognathic surgery. On the other hand, only 21% of the patients with orthognathic surgery fulfilled these cephalometric criteria in the study by Miteff et al.[6] Nowadays, the patient's subjective response and esthetic concerns receive more and more emphasis. Nevertheless, to create optimal facial esthetics and balance, a correct skeletal maxillomandibular relationship and stable occlusion are essential.

In this study, the skeletal and soft tissue and maxillomandibular cephalometric relationships (angles ANB and anb) were significantly smaller in the patients with orthognathic surgery. The fact that significant correlations existed between the 5-year-olds' index and the angle ANB and the corresponding soft tissue angle adds validity to the comparison. Cephalometric correlations between GOSLON Yardstick and angle ANB have also appeared earlier.[34] This may question the need for cephalometric x-rays and radiation at an early age, especially in young children showing poor cooperation. On the other hand, cephalometric variables, as well, (especially angle ANB) have proven to be useful in predicting need for later orthognathic surgery.[4,35-37] A problem, however, with the 5-year-olds' index is that this scoring method cannot predict facial asymmetry, canting of the occlusal plane, bimaxillary retrusion, or facial esthetics. This is especially important for patients with bimaxillary surgery. It is notable that none of the patients of the present study had used protraction facemasks with skeletal anchors during orthodontic treatment. Orthopedic treatment could have influenced maxillary growth but long-term reports in patients with UCLP are few, and relapse can be expected.[38] In addition, secondary alveolar bone grafting was performed between 9-13 years of age. Secondary alveolar bone graft before 8 years of age can have limited negative effect on craniofacial morphology [39], although a recent systematic review concluded that further studies are needed.[40]

Good et al[2] reported that the higher frequency of Le Fort I (47.4% in CLP patients) in their unit may reflect their preference for operative correction for the patients with poor midfacial aesthetics. We also recommend surgery for patients with poor esthetics and bimaxillary retrusion. The cost of orthognathic may also vary considerably between cleft centers.[35] In Canada, the high rates of orthognathic surgery in cleft patients (48.3% in UCLP) have been in part explained by the fact that Canada's national health care covers the cost of that surgical procedure in its entirety.[34] In Australia, where treatment cost is also covered, 36% of the patients with UCLP require orthognathic treatment.[6] In Finland, the costs are almost totally covered by State, but not all who would need orthognathic surgery want surgery.

Predicting the treatment requirements within a cleft unit is not only of value for audit purposes but also as an aid to calculating the resources necessary to provide ongoing treatment.[6] The higher the GOSLON score, the more complex and difficult is the treatment anticipated to correct a UCLP-related malocclusion.[6] On the other hand, the orthodontic treatment time in childhood with poor index scores may be shortened. Early information provided to the patient and the parents as to the possible orthognathic surgical treatment is also important in order to reduce their anxiety and give time for both patient and family to prepare for the later treatment. In cases with severe skeletal discrepancy (index score 5), two-stage treatment may be considered with an early osteotomy during growth and a finishing final surgical procedure at growth completion. This may lead to an increased burden of care for the patient but ensure successful correction of the severe maxillomandibular discrepancy and undesirable facial esthetics and thus improve the patient’s quality of life during growth.

Conclusions

The 5-year-olds index may help to predict treatment outcome and clinical need for orthognathic surgery in early adulthood especially in patients with index scores 4 and 5 (poor and very poor). The predictive value was uncertain in index group 3 (fair), whereas in groups 2 and 1 (good and excellent), orthognathic surgery was not likely to be necessary.

A significant negative correlation emerged between the 5-year-olds’ index and the angles ANB and anb. Because of this, the cooperation-level of the child and the need for cephalograms in 5- and 6-year-old children should be carefully considered.

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Declaration of Conflicting Interest

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Data availability

Research data are not shared because the permission of the study does not allow data sharing.

Author contribution

A.H. contributed to the design and implementation of the research, to the analysis of the results, and to the writing of the manuscript. J.L. contributed significantly to the writing of the manuscript, especially to the surgical text.

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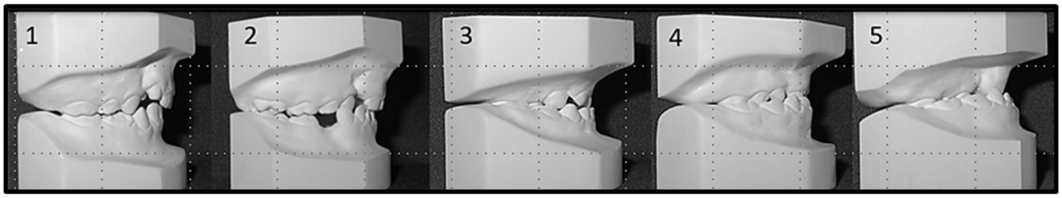
Legends to Figure 1.

The definitions of the 5-year-olds' index scores from excellent (1) to very poor (5) with examples of dental models.

Legends to Figure 2.

Distribution of 5-year-olds' index scores from excellent (1) to very poor (5) and need for orthognathic surgery in 70 children with UCLP.



Group	General Features	Outcome
1	Positive overjet with average inclined or retroinclined incisors No crossbites/open bites Good maxillary arch shape and palatal vault anatomy	Excellent
2	Positive overjet with average inclined or proclined incisors Unilateral crossbite/crossbite tendency ± Open bite tendency around cleft site	Good
3	Edge-to-edge bite with average inclined or proclined incisors; or reverse overjet with retroinclined incisors Unilateral crossbite ± Open bite tendency around cleft site	Fair
4	Reverse overjet with average inclined or proclined incisors Unilateral crossbite, ± bilateral crossbite tendency ± Open bite tendency around cleft site	Poor
5	Reverse overjet with proclined incisors Bilateral crossbite Poor maxillary arch form and palatal vault anatomy	Very Poor

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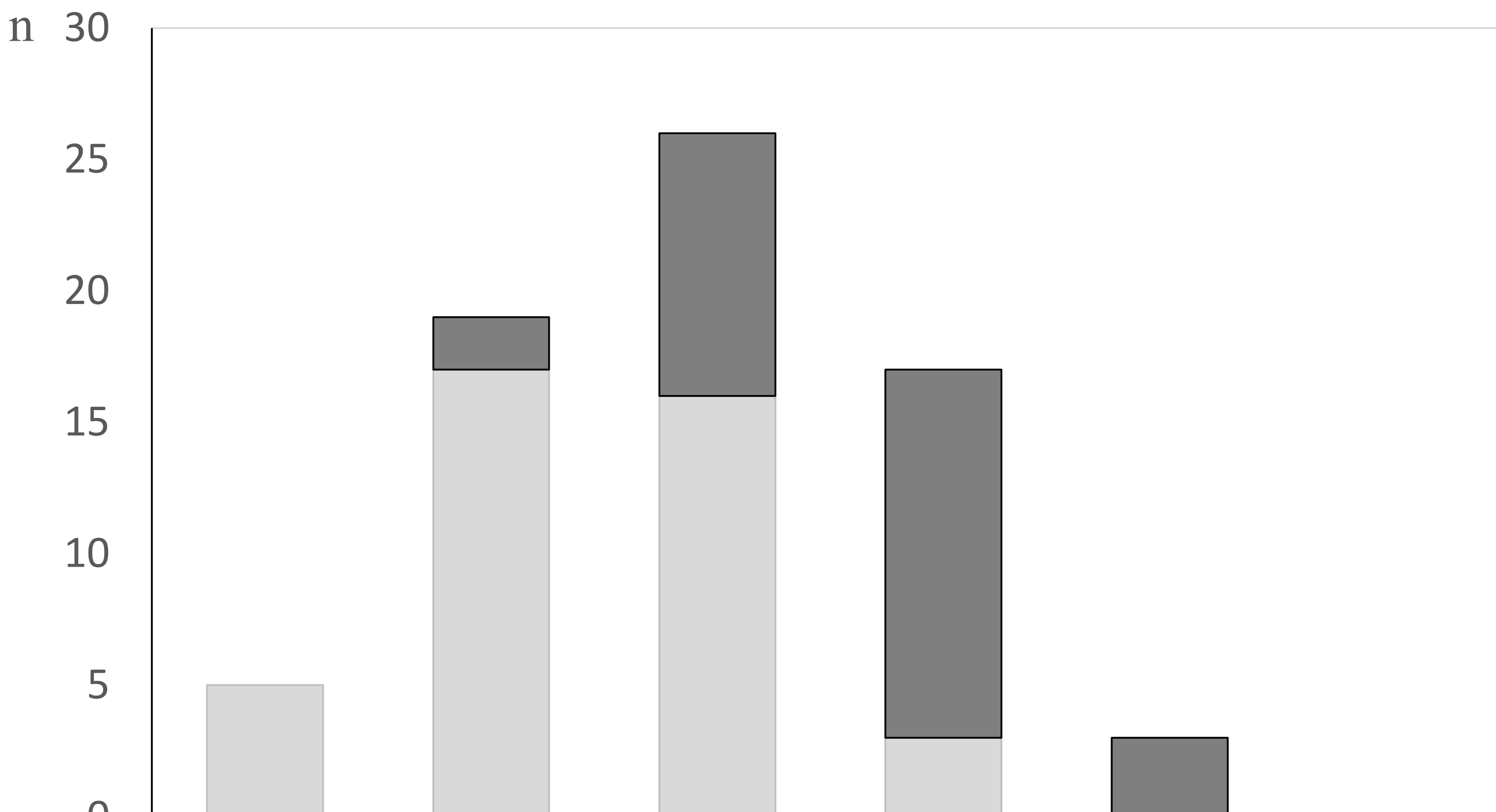

	Index score 1 (Excellent)	Index score 2 (Good)	Index score 3 (Fair)	Index score 4 (Poor)	Index score 5 (Very poor)	Total (n=70)
Orthognathic surgery		2	10	14	3	19
No orthognathic surgery	5	17	16	3		41