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Original Article

Maxillary arch dimensions, occlusion assessment and space conditions in patients with cleft palate in the period of deciduous dentition—A retrospective study

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ABSTRACT

Purpose: The aim of this study was to evaluate the maxillary arch dimensions, dentoalveolar relationships and spacing conditions in patients with cleft palate in comparison with the control group.
Methods: The subjects consisted of 31 children with cleft palate only (CP) aged 5.5 ± 0.51 (20 with a cleft of hard and soft palate (SHPC group) and 11 with a cleft of soft palate only (SPC group)). In the control group 30 subjects had a normal occlusion at age 5. Maxillary arch dimensions, dentoalveolar relationship according to the Huddard Bodenham index (HBI) and space conditions were compared with the control group.
Results: In all variables of maxillary arch dimensions except for total arch height the control group exhibited greater values. Most of the significant differences were on account of differences between the control and the SHPC group, with only three comparisons yielding significant results when comparing the two groups of children with clefts (SHPC vs SPC, IV-IV central: $p_{Dunnett\ T3}$= 0.0002, 95%$CI_{Difference}$=-9.9-(-3.18); V-V distopalatal cusps: $p_{Dunnett\ T3}$= 0.0002, 95%$CI_{Difference}$=-9.97-(-3.17); Total arch length: $p_{Dunnett\ T3}$= 0.0014, 95%$CI_{Difference}$=1.74–7.85). The three groups differed significantly in anterior HBI only ($H_{Kruskal-Wallis}$=15.56, p = 0.0067). The spacing conditions in both jaws were also shown to be significantly dependent on the group of subjects studied (Upper jaw: $\chi^2_{omnibus}$= 16.79, p = 0.0018; lower jaw: $\chi^2_{omnibus}$= 13.75, p = 0.0102).
Conclusions: The growth of the upper dental arch at the age of five is impeded in participants with CP in comparison to a control group. It is important to assess the effect of cleft subtypes on growth and development to get a better understanding.

1. Introduction

Orofacial clefts (OFC) are the most common congenital anomalies of the craniofacial complex. They can be unilateral, bilateral, complete or incomplete, and may involve the lip only, the palate only (CP) or both [1]. In Slovenia, the incidence of clefts in the period 1994 –2013 was 1.57 per 1000 live births. CPs were the most frequent, representing 45.8% of all cases [2]. The establishment of optimal maxillary growth and good dentoalveolar relationships are amongst the main goals of interdisciplinary treatment of patients with clefts.

Maxillofacial growth and development can be influenced by the type of cleft and by other factors, such as the degree of congenital tissue defect and the intrinsic growth potential of the craniofacial tissue [3,4].

There is very little knowledge about the growth and development of the upper dental arch and occlusion in children with cleft palate only. It has been shown that the dimensions of the dental arches were smaller in the CP group when compared to a control group at the ages of three and six [5]. At the age of four, the maxillary arch widths and the arch lengths were larger in the CP group than in unilateral and bilateral cleft lip and palate subjects who showed discontinuity of both buccal segments and had received two operations [3]. Concoran et al. also showed that clefts affecting the palate resulted in smaller maxillary arch dimensions than clefts affecting only the lip [6].

Two methods are normally used to assess occlusion in patients with a cleft: the GOSLON yardstick and the Huddart and Bodenham

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index. The GOSLON yardstick is a standardised ranking system designed by Mars et al. to assess treatment outcomes in 10-year-old children born with UCLP. The system ranks study models into five discrete categories. These categories reflect the severity of the malocclusion and the complexity of possible orthodontic treatment [7]. It has been demonstrated that the GOSLON yardstick has good measurement characteristics when used with children aged five [8]. The calibration of examiners is required in the use of the GOSLON yardstick [9]. The other measure to assess the dentoalveoar relationship was described by Huddart and Bodenham in 1972. Each maxillary tooth or midpoint of the arch is scored according to its relationship with the corresponding tooth in the lower jaw. The lateral incisors are not included because they may be absent or mispositioned. The scores of each tooth are added together; the more negative the cumulative score, the more severe the upper arch constriction. However, the score could also be a consequence of abnormal development of the lower dental arch. The original Huddart and Bodenham index was developed to assess occlusion in children with UCLP in deciduous dentition [10] and was later modified for use in mixed dentition [9]. It has been shown that it provides a useful tool for assessing occlusion in patients with all types of clefts, being objective, sensitive, and versatile. Moreover, the Huddart/Bodenham scoring system can be applied to scanned digital images without issue. The main advantage of the Huddart-Bodenham index is that it does not require calibration [11].

The spacing in the dental arches at the beginning of the eruption of the permanent teeth appears to be fundamental for their proper alignment [12]. An analysis of the spacing conditions at the end of the period of primary dentition could be one of the parameters for the evaluation of the growth and development of the dental arches [13].

The primary aim of this study was to evaluate the maxillary arch dimensions, dentoalveolar relationships and spacing conditions in patients with CP, and compare them to children with normal occlusion in deciduous dentition. The secondary aim was to explore whether the type of cleft, namely clefts of both hard and soft palate and of soft palate clefts only, is associated with maxillary arch dimensions, dentoalveolar relationships and tooth spacing condition outcomes.

2. Methods

The subjects consisted of 31 children (16 girls, 15 boys) with cleft palate only, born in Slovenia in the period 2013−2017. Of these, 20 presented with a cleft of both hard and soft palate (SHPC group) and 11 with a cleft of soft palate only (SHP group). No orthodontic treatment had been performed before the age of five. The medical documentation of the hospital was used to check the medical history of the participants. Patients with concurrent syndromes, a submucous cleft or missing documentation were excluded from this study. The control group consisted of 30 subjects in the deciduous dentition with normal occlusion (14 girls, 16 boys) at the age of 5.5 ± 0.28 (Fig. 1A). The study was designed as a retrospective study, meaning the data of both cleft palate groups and controls was acquired from hospital records. The data obtained included digital study models of the upper and lower dental arch, obtained from the subjects at age 5. These 3-D digital models are acquired during routine orthodontic examinations of children with clefts and are thus a part of hospital records. Alongside the study models, demographic and other diagnostic data was acquired from the records. The data obtained was then screened for inclusion and exclusion criteria and utilized in the study accordingly. All available records in the period were examined. No data additional to what was available in the hospital records was used or obtained for the purposes of this study. The study was approved by the Commission of the Republic of Slovenia for Medical Ethics − Reference No.: 0120−190/2022/3.

2.1. Surgical procedures

All subjects were treated at the Department of Maxillofacial and Oral Surgery of the University Medical Centre Ljubljana by two surgeons, according to the protocol for patients with cleft palate. The “straight-line” cleft palate repair technique with routine use of radical intravelar veloplasty, described by Sommerlad and Cutting, was used [14,15]. This technique involves extensive dissection of the levator muscles from the nasal and oral mucosa, the separation of the m. tensor and m. levator veli palatini, retropositioning of the levator musculature, and tensionless suturing with resorbable sutures. In complete cleft palate cases the elevation of the mucoperiosteal palatal flap from the oral side and subperiosteal elevation of nasal layer, with two layer closure in the hard palate, with radical intravelar veloplasty is performed in one operation at the age of 30 months [16].

2.2. Maxillary arch dimensions

Measurements of the transverse distances between the contralateral cusps of canines; the contralateral central pits of the first and second deciduous molars; the lowest midpoints of the palatal gingival contours of the contralateral canines; the contralateral first and second deciduous molars; and the distopalatal cusps of the second deciduous molar were taken on digital study cast models. In the sagittal plane the anterior and total arch lengths were measured (Fig. 1B-D). The mean value of two measurements performed by the same researcher was used for further analysis.

2.3. Dentoalveolar relationships

The anterior and posterior occlusion of each subject was analysed on digital dental casts according to the Huddard and Bodenham index (HBI) [10]. The anterior score (deciduous upper central incisors), the posterior left (left deciduous canine and molars), the posterior right (right deciduous canine and molars) and the total score were determined [17]. All the measurements were repeated twice by the same researcher.

2.4. Space conditions

Children without complete primary dentition were excluded (n = 2). The presence or absence of spaces was determined on the digital dental casts, based on the existence of proximal contact of the teeth. If proximal contact was visible or the teeth overlapped, the site was classified as “no spacing”. If there was no proximal contact, the site was classified as “spacing”. If the upper dental arch had “spacing” in 1 or 2 sites, the arch was considered as “crowding”. In the lower dental arch “crowding” was defined by the presence of 3 or fewer “spacing” sites [13]. Each cast was examined twice by the same researcher.

2.5. Statistical analysis

SPSS version 25 (IBM, Armonk, NY, USA) was used for all the data analyses and Microsoft Excel 2013 (Microsoft Corporation, Redmond, WA, USA) was used for visualisation. The data on the maxillary arch dimensions of the teeth and the data on the HBI values were analysed by a one-way ANOVA (ordinary and Welch’s ANOVA), with three groups. The data on the spacing and crowding of the teeth was analysed via χ^2 tests of independence. Dunnett’s T3 post-hoc tests were used on teeth placement data. A Kruskal-Wallis H test was used for the HBI data, with post-hoc Mann-Whitney U tests. The spacing and crowding data were post-hoc tested by χ^2 tests of independence. The Bonferroni correction was used on all a-priori and post-hoc analysis results. All the statistical tests performed were two-sided and a post-correction α of 0.05 was adopted. Measures of effect size [18,19] were also calculated.

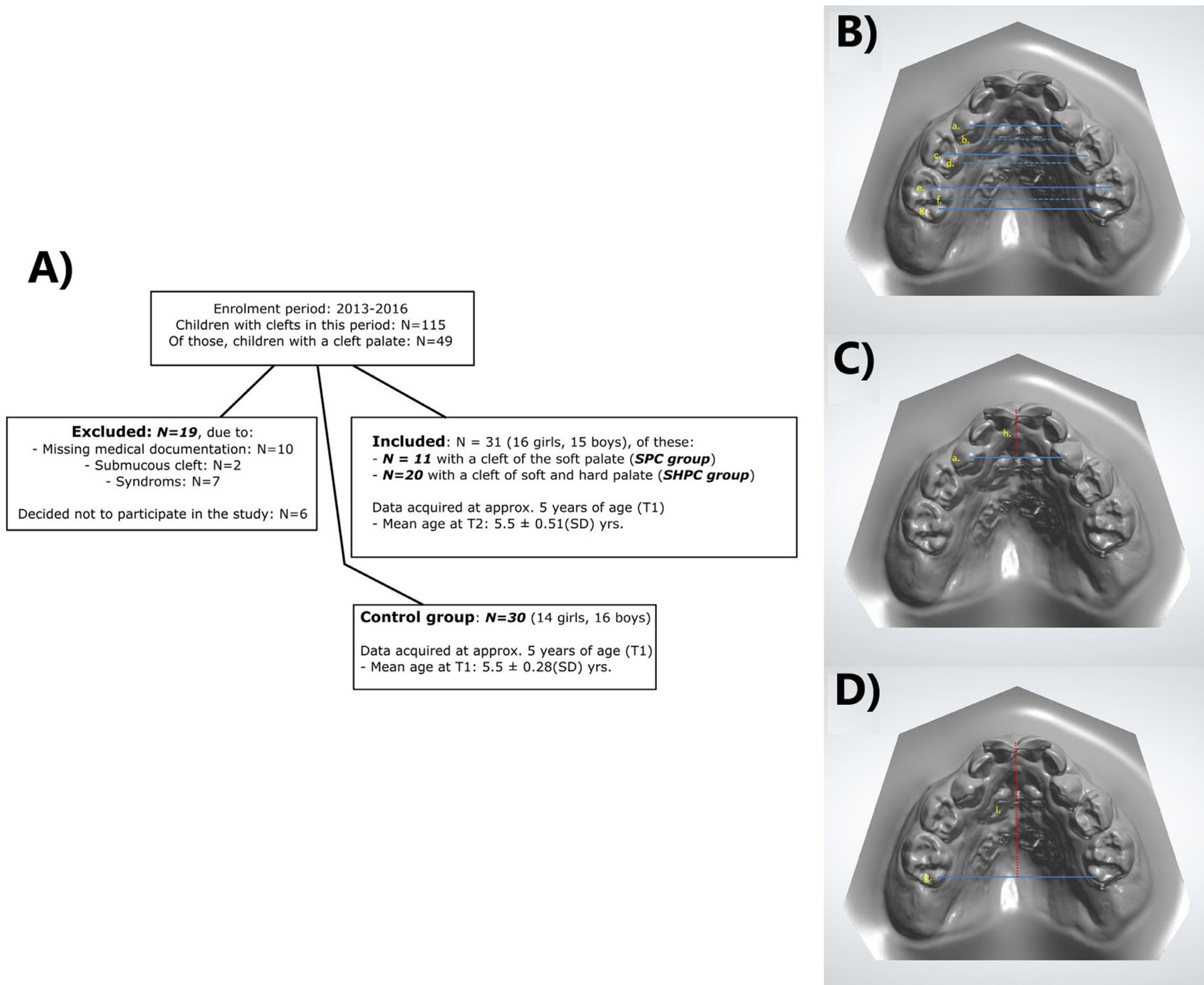

Fig. 1. The flowchart of recruitment and the measured maxillary arch dimensions. A) The flowchart of recruitment of the participants. B): (a) Intercanine width measured between the cusps (III-III cusp); (b) Intercanine width measured between the lowest midpoints of the palatal gingival contours (III-III gingival); (c) and (e) The distance between the central pits of the first and second deciduous molars (IV-IV central; V-V central); (d) and (f) Distances between the lowest midpoints of the palatal gingival contours of the contralateral canines and the contralateral first and second deciduous molars (IV-IV gingival, V-V gingival); (g) distance between the distopalatal cusps of the second deciduous molar (V-V dpc); C): (h) Anterior arch length (AAL); D): (i) Total arch length (TAL).

Three of the variables were non-normally distributed: TAL, IV-IV central and V-V central, as determined by the Shapiro-Wilk test of the normality of distribution. As the ANOVA [20] and the Dunnett's T3 test [21] are robust to normality violations, the ordinary ANOVA was used where no other assumptions were violated.

Two variables, the TAL and V-V central, also exhibited non-homogenous variance between the groups, so Welch's ANOVA was used to analyse the differences in these two variables. A Kruskal-Wallis ANOVA was performed to verify the results and showed the same significant differences.

The HBI data expectedly violated both the normality of the distribution and the homogeneity of the variance assumptions. Thus the Kruskal-Wallis H test with post-hoc Mann-Whitney U-tests was used.

3. Results

3.1. Maxillary arch dimensions

The results of the omnibus ordinary and Welch ANOVAs showed that there was a significant difference between the three groups in the following variables: III-III cusp, $F = 14.82$, $p = 0.0001$; III-III gingival, $F = 12.16$, $p = 0.0007$; IV-IV central, $F = 19.79$, $p<0.000$; V-V central, $F = 11.71$, $p = 0.0010$; V-V dpc, $F_{Welch}=33.95$, $p<0.0001$; and TAL, $F_{Welch}=7.79$, $p = 0.0483$. The results of post-hoc testing are depicted in Fig. 2.

Nine pairwise comparisons yielded statistically significant results. Most of these were on account of differences between the control group and the SHPC group, with only three comparisons yielding significant results when comparing the two groups of children with clefts (the SHCP and SPC groups). None of the comparisons showed a significant difference between the control group and the SPC group. The SHPC group and the control group significantly differed in the following variables: III-III cusp, III-III gingival, IV-IV central, V-V central, V-V dpc, and TAL. In all the variables except TAL the control group exhibited greater values. In the case of TAL, the SHPC group exhibited higher values than the control.

In the four variables where significant differences between the two groups of children with clefts were observed, the SHPC group showed smaller values for the IV-IV central distance. The TAL was greater for the SHPC group, while the V-V dpc was smaller.

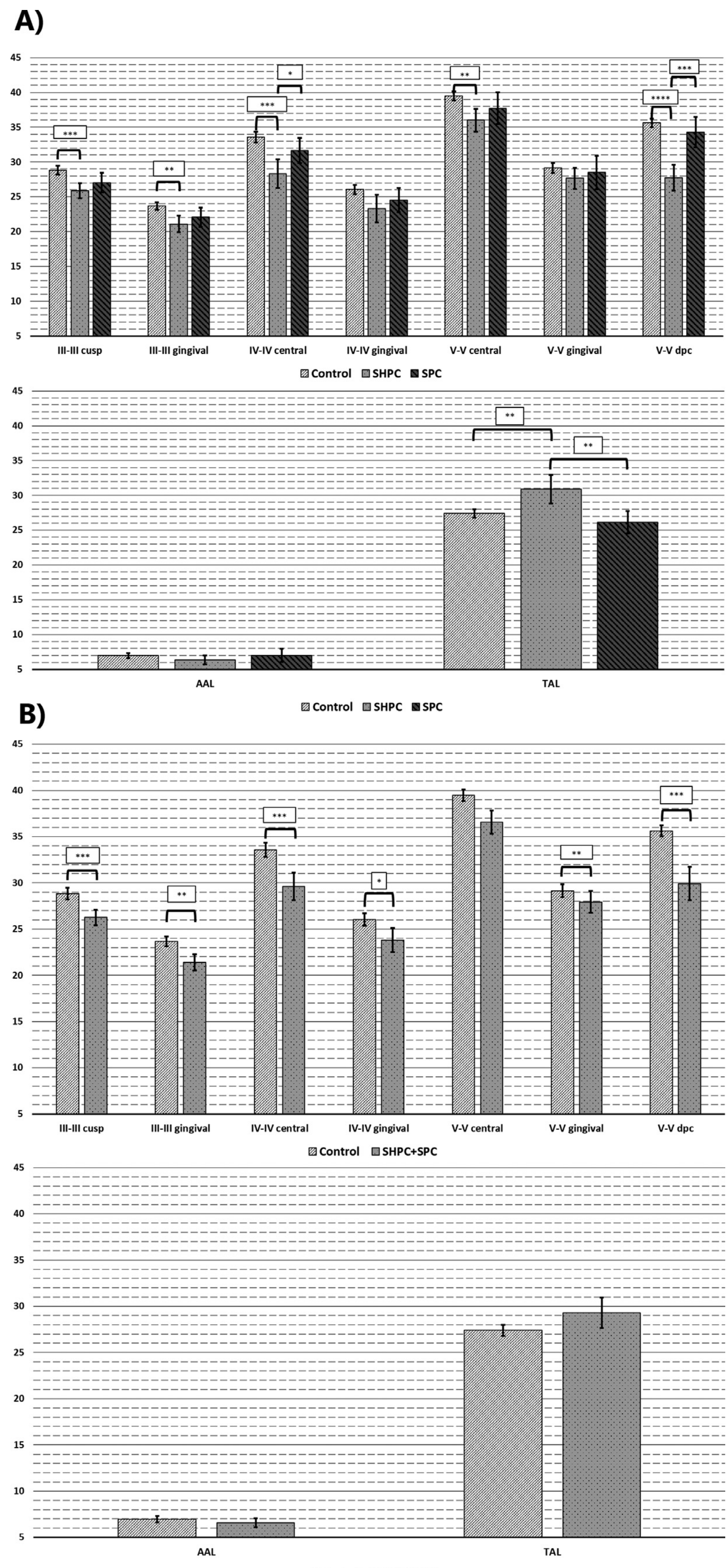

Fig. 2. A): The results of pairwise comparisons between the control, SHPC and SPC groups in transversal parameters (upper panel) and upper dental arch heights (lower panel). The significance bars point to the groups in which the differences were detected. B): The results of pairwise comparisons between the control, SHPC and SPC groups in transversal and anteroposterior dimensions. The error bars represent 95% CI of the means; the y-axis origin of the graph is at 5. Units are millimetres. The significance markers denote the following significance levels: *=0.05–0.01, **=0.01–0.001, ***=0.001–0.0001, ****<0.0001.

Table 1
Observed effect sizes and the difference in the% of explained variance due to grouping – transversal length and height data.

	η^2 – three groups	η^2 – two groups	Difference in% of explained variance
III-III cisp	0.363	0.305	5.813
III-III gingival	0.319	0.243	7.610
V-V central	0.311	0.251	5.984
V-V gingival	0.076	0.079	−0.323
IV-IV central	**0.462**	**0.344**	**11.806**
IV-IV gingival	0.219	0.191	2.779
V-V dpc	**0.624**	**0.369**	**25.487**
AAL	0.068	0.006	6.282
TAL	**0.301**	**0.015**	**28.616**

*Numbers in bold represent variables whose amounts of explained variance differed by more than 10% when the subjects were grouped into 2 or 3 groups. Units of variance are in%.

3.1.1. Comparison between children with clefts and the control group

To illustrate the importance of differentiating between the groups of children with different types of clefts, and to further explore the data of our study, we performed some additional post-hoc analyses of our data, where the data of the two groups of children with clefts was pooled into a single experimental group. This data was then compared to the values of the control subjects by Kruskal-Wallis H tests. The results are shown in Fig. 2B.

The effect sizes for all the variables that produced significant results on the ANOVA (Fig. 2A) were in excess of 0.14, which is considered to be a large effect size [22]. Furthermore, when the effect sizes of the three group division and two group division of participants based on cleft characteristics are compared, the three group division explains more variance (from 2–28% more) in all but the V-V gingival variable (Table 1).

3.2. The Huddart-Bodenham index

The descriptive statistics for the HBI can be found in Table A.1. The results of the Kruskal-Wallis H test showed that the three groups differed significantly in only one of the five tested variables, namely the anterior HBI, $H = 15.56$, $p = 0.0067$.

The post-hoc pairwise Mann-Whitney tests revealed one significant difference between the control and the soft palate cleft only group on the anterior HBI, where the soft palate cleft group exhibited significantly lower scores than the control group ($U = 80$, $W = 125$, $p = 0.0187$). The results of the post-hoc tests are shown in Fig. 3A.

3.2.1. Comparison between children with clefts and the control group

Additional analysis with both cleft groups pooled into one was carried out. A Kruskal-Wallis H test was used to analyse the data and the results are depicted in Fig. 3B.

A comparison of observed effect sizes, between the three group and two group mode of analysis is presented in Table 2.

Table 2
Observed effect sizes and the difference in the% of explained variance due to grouping – HBI data.

	η^2-three groups	η^2-two groups	Difference in% of explained variance
Total score	0.128	0.137	−0.924
HBI posterior left	0.035	0.032	0.288
HBI posterior right	0.130	0.145	−1.555
HBI posterior total	0.106	0.109	−0.263
HBI anterior score	**0.230**	**0.120**	**11.007**

*Numbers in bold represent variables whose amounts of explained variance differed by more than 10% when the subjects are grouped into 2 or 3 groups. Units of variance are in%.

3.3. Space conditions

When the incidences of crowding and spacing in the three groups are compared, the differences between the expected and observed incidences are unlikely to be independent of the group of subjects studied in both the upper ($\chi^2_{omnibus}$= 16.79, $p = 0.0018$) and lower jaw ($\chi^2_{omnibus}$= 13.75, $p = 0.0102$). The results of the post-hoc χ^2 tests of independence are shown in Fig. 4A.

3.3.1. Comparison between all the children with clefts and the control group

Once again, an analysis on the pooled cleft group and control group differences was performed. The results are illustrated in Fig. 4B. Again, the three group mode of analysis shows an increase in effect size. For the maxilla, when the subjects are assigned to three groups, the Cramer's $V = 0.755$, while when the subjects are assigned only to the control and cleft group, $V = 0.532$, which presents a difference of effect size of 0.222. A similar difference (0.211) can be seen in the mandible, with the three group division having an effect size of 0.72 and the two group division an effect size of 0.509.

4. Discussion

In this study we evaluated the transverse and sagittal maxillary arch dimensions, dentoalveolar relationships, and space conditions in children with cleft palate only (CP). The morphological data of CP was compared to a control group of children with no congenital anomaly and with normal occlusion, at five years old. We also explored the effects that the type of cleft exhibits on the morphological parameters.

Most patients (84.6%) with cleft palate only at the mean age of 6 years have a good prognosis according to the GOSLON Yardstick [23]; however, the growth and development of the craniofacial complex is not only affected by a cleft lip and palate; it is also affected in children with a cleft palate only [17].

All the measured transversal dimensions in the upper arch, except the distance between the midpoints of the palatal gingival contours of the second molar (V-V gingival), were significantly smaller in children with a cleft palate when compared to healthy controls at the age of five. In the group of participants with clefts of the soft palate only (the SPC group) the distances in the transversal plane were generally greater than in the SHPC group. The significant differences were found in IV-IV central and V-V dpc. The upper arch lengths did not differ significantly between the cleft and control groups, but when we analysed the participants with a cleft of soft palate only and clefts of hard and soft palate separately, the total arch length was, surprisingly, significantly smaller in the soft palate cleft only group.

This shows that it is important to differentiate between different types of clefts, as the different presentation in children with different types of clefts can skew research results, either in detecting greater differences due to similar but not equally expressed differences between healthy controls and children with various types of cleft (e.g. the V-V gingival, Fig. 2A, B), or by masking important differences between children with a specific type of cleft and healthy controls on variables that are particularly affected by the type of cleft present (e.g. the distance between the distopalatal cusp of the second molar, Fig. 2A, B). Further illuminating this point is the data in Table 1, where the effect sizes are compared. The differences in explained variance (η^2) ranged from 0.3% to 28.6% between the two modes of analysis. The variance of all the variables, where the difference of explained variance exceeded 10%, was better explained by the division of subjects into 3 groups (Table 1). This furthers the argument that the type of cleft is of great importance to morphological outcomes, and should be considered in any analyses of cleft-related data that includes measurements of the maxillary dental arch.

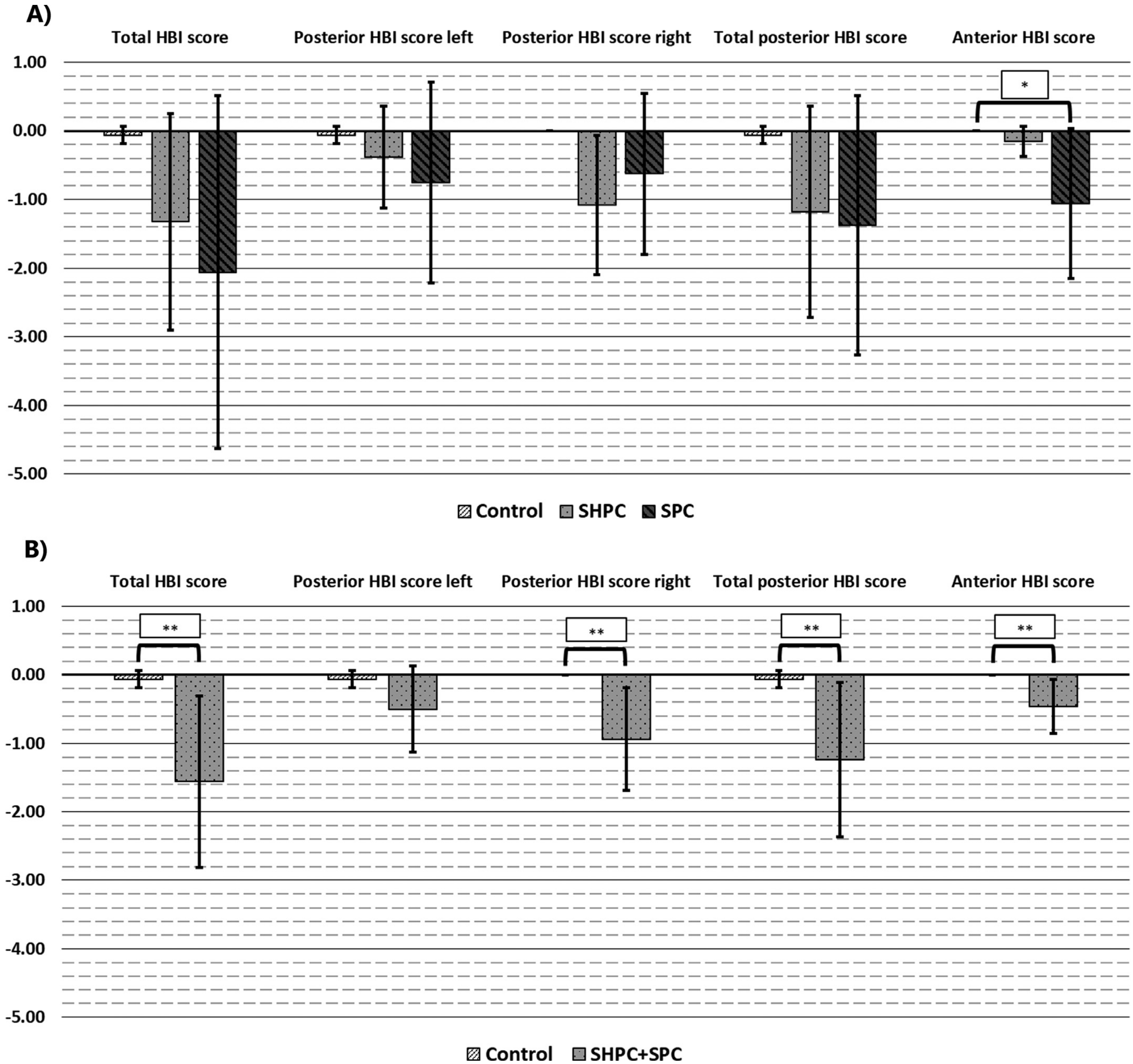

Fig. 3. A): The results of pairwise comparisons of the Huddart-Bodenham index scores of the control, SHPC and SPC groups. B): The results of pairwise comparisons of the Huddart-Bodenham index scores of the control and pooled cleft palate groups. The error bars depict 95% CI of the mean. The * denotes a difference significant at the 0.05−0.01 level. The ** denotes a difference significant at the 0.01−0.001 level.

There is a lack of studies comparing the dental arch morphology of different types of cleft palate to control groups. Most of what is available compared the maxillary arch dimensions between different types of clefts at different times [3,6], but Rando et al. showed that the upper intercanine distances in the control and unilateral cleft lip groups were significantly greater than those of the groups of participants with cleft palate only and with unilateral cleft lip and palate. However, the differences between cleft palate subtypes were not studied [24]. Smaller upper arch dimensions with cleft palate only in comparison to a control group at six years of age were also found by Nyström et al. Moreover, these authors also showed that in the period from three to six years of age, growth in the transversal plane was the most diminished in the group with total cleft palates, in comparison to the submucous cleft, soft palate and partial cleft palate groups [5].

Additionally, we were interested in the spacing of the teeth at the end of deciduous dentition in the upper and lower dental arches. A relatively simple measurement scheme was adopted, focusing on a binary classification of either spacing or crowding, as it was found that a simple space count in the primary dentition is of equal value to detailed space measurements [25]. Spacing in primary dentition is informative of space conditions in the permanent arch, where more spaces at the end of primary dentition mean less crowding in the permanent arch [12]. It is not possible to use this method as an accurate predictor of space conditions at an individual level but it provides a general overview.

In our study the post-hoc tests revealed that only for the control and SHPC groups the incidences of crowding and spacing, in both jaws, are unlikely to be independent of the group. In the control group, the upper jaw was much more likely to exhibit spacing of the

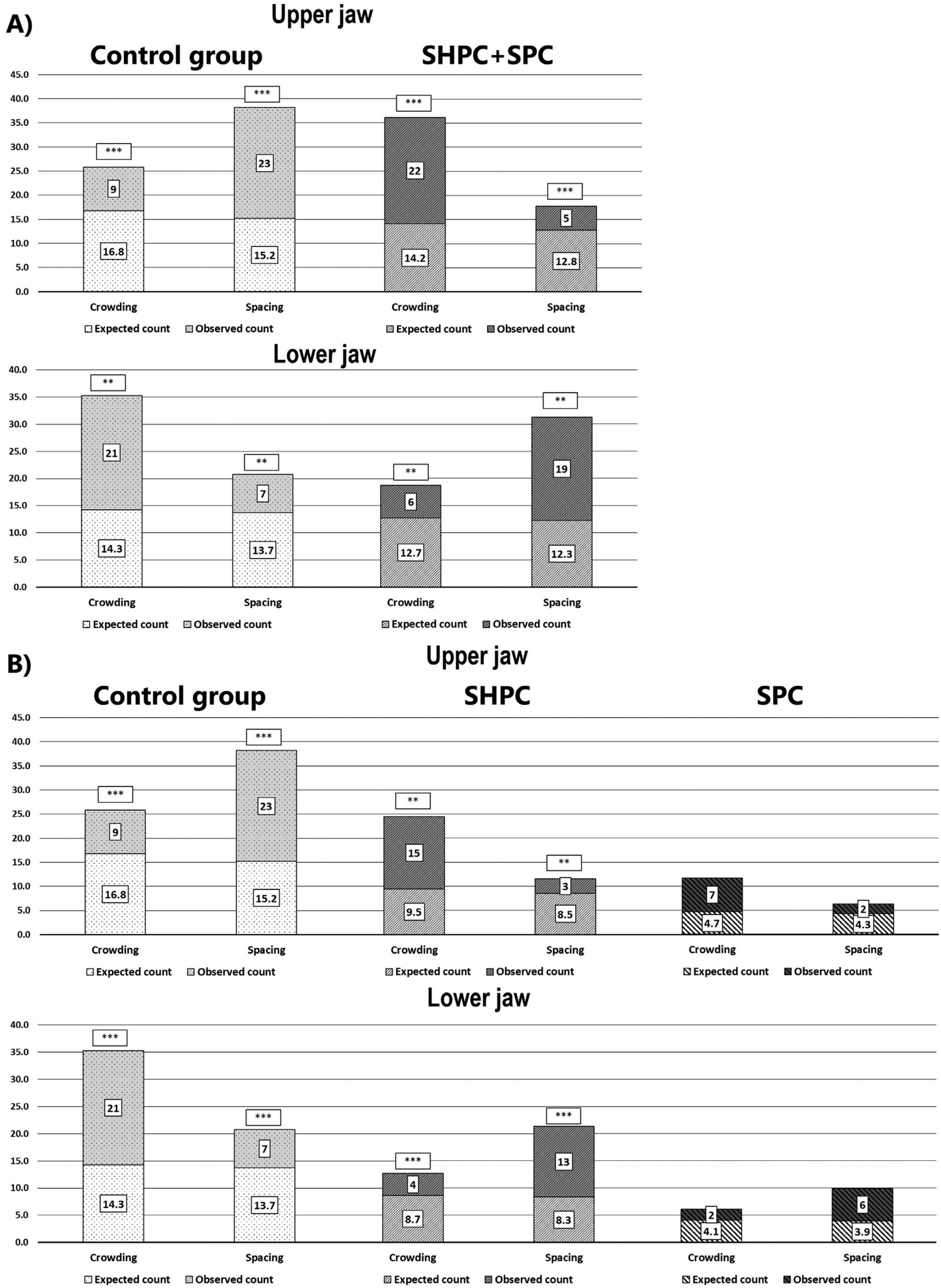

Fig. 4. A): The results of the post-hoc χ^2 tests of independence of the incidence of crowding and spacing in the upper and lower jaw for the control, SHPC and SPC groups. The y axis shows the counts of crowding and spacing. The expected (lower portion of graph) and observed (upper portion of graph) counts are depicted in a cumulative bar graph. Boxes with numbers in the columns show the respective counts. B): The results of the post-hoc χ^2 tests of independence of the incidence of crowding and spacing in the upper and lower jaw for the control group and all children with clefts. The ** symbol denotes statistical significance of the post-hoc χ^2 test result at the 0.01−0.001 level, and the *** denotes statistical significance at the 0.001−0.0001 level.

Table A.1
Descriptive statistics for HBI measurements.

	Mean	Maximum	Minimum	Standard deviation	95.0% Lower CL for Mean	95.0% Upper CL for Mean
Control	−0.06	0.00	−2.00	0.35	−0.19	0.06
SHPC	−1.32	0.00	−12.00	3.07	−2.90	0.25
SPC	−2.06	0.00	−7.00	3.08	−4.63	0.51
Control	−0.06	0.00	−2.00	0.35	−0.19	0.06
SHPC+SPC	−1.56	0.00	−12.00	3.03	−2.81	−0.31

* CL = Confidence level.

teeth, while the opposite was observed for the lower jaw. The SHPC group exhibited the opposite pattern of results, with more frequent crowding in the upper jaw and spacing in the lower. For the SPC group, no significant dependence of crowding or spacing on the group of subjects being studied was observed in either jaw. These results suggest that a prevalent crowding in the upper jaw, but more often spacing in the lower jaw, is characteristic of children with cleft palates. However, as can be seen in Fig. 4, this is not true for the SPC group. These results once again show the importance of differentiation between the different types of clefts.

Large effect sizes were found for both modes of analysis, leading to the conclusion that the effect of group, regardless of the mode of grouping, profoundly affects the incidence of spacing and crowding of teeth. Nevertheless, the difference in effect size measurements between the three groups division and two groups division study design was more than 0.2 for both variables, which would be considered a medium-sized effect in-and-of itself. In all three streams of data, the importance of the careful selection of experimental groups, based on the type of cleft, has been illustrated.

In this study we showed that the values of the total HBI and of the anterior HBI were significantly more negative in children with clefts in comparison to the control group. When we compared the HBI values of two subtypes of cleft palate, there were no significant differences between the groups, except for the value of the anterior HBI, where surprisingly the soft palate cleft group exhibited the most negative values. Medium to large (anterior HBI score) effect size was observed for the examined variables [22]. Furthermore, for the variable that was the only one to produce a statistically significant pairwise comparison result (Fig. 4A), the amount of explained variance once again differs more than 10% between the three groups and two groups (Fig. 4B) analysis modes, with the three-group model explaining 11% more variance than the two-group one.

The results of Tothill and Mosey's study showed that the total HBI for the cleft palate only group was −3 at the age of five; in our group of cleft palate only participants the mean score was −1.5 and the range was −12.00–0.00 [26]. The fact that participants with Pierre Robin sequence were not excluded in Tothill and Mosey's study is perhaps the reason for the better value of total HBI than expected.

Due to heterogeneity in the previous studies on the topic of this paper, an estimation of actual effect size is difficult when grouping subjects into the SHCP and SPC group criteria, as these differences have been investigated in few studies with small sample sizes and varied methodologies of obtaining measurements. Due to these limitations, the a-priori power of our study was not calculated. Furthermore, due to the relative rarity of children born with cleft palates, this study included a convenience sample, whose size is likely too small to discover all the effects of cleft and could overestimate the discovered effect sizes. Nevertheless, it provides valuable insight into some of the more accentuated between-group differences and illuminates some considerations pertaining to study design that must be made when studying the effects of clefts.

To conclude, the growth of the upper dental arch at the age of five is impeded in participants with CP in comparison to a control group. In the present study we show that when the maxillary arch dimensions, dentoalveolar relationships and spacing conditions in SHPC, SPC and healthy controls are compared, the three groups differ significantly on many variables, with the most pronounced differences expectedly arising from the measurements of maxillary arch dimensions. Of special interest for further research and exploration are the differences observed between the SHPC and SPC groups.

Acknowledgements

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Appendix

Table A.1

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