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The Slavcleft: A three-center study of the outcome of treatment of cleft lip and palate. Part 2: Dental arch relationships

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ABSTRACT

Purpose: Our aim was to evaluate the dental arch relationship in a preadolescent Slavic population with unilateral cleft lip and palate (UCLP) by using the Goslon Yardstick.
Materials and methods: Patients treated in Warsaw, Poland (n = 32), Prague, Czech Republic (n = 33) and Bratislava, Slovakia (n = 30) were included in this retrospective study. Each cleft center used a unique surgical protocol. Three raters scored blindly the dental arch relationship on plaster models. Intra- and inter-rater agreement were assessed with kappa statistics, and differences between the groups were evaluated with one-way analysis of variance. Intra-rater agreement was very good (k > 0.825), while inter-rater agreement was either good or very good (kappa >0.703).
Results: We found that patients treated in Warsaw showed a more favorable dental arch relationship (Goslon score = 2.58, SD = 0.77) than patients treated in Prague (Goslon score = 3.21, SD = 1.04). Patients treated in Bratislava showed an intermediate Goslon score (3.07; SD = 0.99).
Conclusion: This study showed that the dental arch relationships in patients treated in Warsaw with a one-stage repair were more favorable than in patients treated in Prague and Bratislava with a two-stage protocol and comparable to the best results obtained in the Eurocleft and Americleft studies.

1. Introduction

Cephalometric evaluation of the outcome of treatment of cleft lip and palate (CLP) is associated with challenges, as already discussed in the first part of the Slavcleft study (Urbanova et al. (2016). For instance, it is conceivable that differences in the ethnic background affect cephalometric findings; e.g., patients with a cleft who come from populations with a high prevalence of dolichocephalic facial pattern could show different craniofacial morphology from that of patients coming from populations with a high prevalence of brachycephalic faces. An assessment of dental arch relationships addresses this problem, as normal occlusion can be evaluated irrespective of ethnic backgrounds and craniofacial patterns. The classification of occlusion is based on the mutual relationship of maxillary and mandibular dental arches and can be independent of craniofacial morphology.

An additional advantage of using dental arch relationships to discriminate between patient groups is that in comparison, cephalometric measurements can be relatively insensitive measures of outcome. Thus, required sample sizes are large and

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most cleft centers would have difficulty collecting them. For example, to detect a difference between a mean of 2 centers of 1.5°, the required sample sizes would be >100 subjects per group. Assessing the mean Goslon score for dental arch relationships has been validated in the Eurocleft study and appears to be more feasible in this context. To detect a systematic difference of 0.75 of Goslon points between two centers and to examine pair-wise differences, a sample size of 34 patients is sufficient (Shaw et al., 1992).

Therefore, the objective of this study was to compare dental arch relationships in a sample of patients with complete unilateral cleft lip and palate treated in three centers (Warsaw, Prague, and Bratislava) using different surgical protocols. The H_0 hypothesis is that dental arch relationships in all groups are comparable.

2. Materials and methods

2.1. Subjects

Dental arch relationships were assessed on plaster casts of 95 children with complete unilateral cleft lip and palate (UCLP) who were treated in cleft centers located in Warsaw (Poland), Prague (Czech Republic), and Bratislava (Slovakia), using different surgical protocols; details are available in the first part of the Slavcleft study (Urbanova et al., 2016).

In summary, 32 patients from the Warsaw Cleft Center, affiliated with the Institute of Mother and Child (IMC), were treated with a one-stage repair of the cleft at 9 months (SD = 1.9; range 6.1–15.8 months). The mean age when records were taken was 10.6 years (SD = 1.3; range: 8–13.6). The gender proportion was: males 71.4%, females 28.6%.

A total of 33 children from the Prague Cleft Center, affiliated with the Faculty Hospital Royal Vineard underwent a two-stage repair: the lip was repaired at 7 months, while palatoplasty was done at 36 months. The mean age when records were taken was 9.1 years (SD = 0.9; range: 7.3–10.2). The gender proportion was: males, 65%, and females, 35%.

A total of 30 children from the Bratislava Cleft Center affiliated with the Clinic of Plastic and Reconstructive Surgery, Comenius University, were treated with a two-stage repair: the ip was repaired at 4 months, while palatoplasty was done at 11 months. The mean age when records were taken was 9.3 years (SD = 1.8; range: 6.1–12.8). The gender proportion was: males, 66%, and females, 34%.

2.2. Methods

The Goslon Yardstick (Mars et al., 1987) was used to rate the dental arch relationship. The 95 models were coded and placed in random order. Three raters experienced in the treatment of patients with cleft lip and palate (WU, IK, and PF) scored the models in the presence of the reference models. One of three raters (PF) was additionally experienced in using the Goslon Yardstick. Prior to the rating session, a calibration exercise was carried out.

The mean score of the first rating session was used to establish the distribution of the Goslon groups in the samples (Fig. 1). Categorization of the groups was as follows: group 1 when the mean score was ≤1.50; group 2 when the mean score was >1.50 and ≤2.50; group 3 when the mean score was >2.50 and ≤3.50; group 4 when mean score was >3.50 and ≤ 4.50; and group 5 when the mean score was >4.50.

Surgical experience was assessed according to Bearn et al. (2001); surgeons who performed annually 5 or fewer primary palatoplasties were considered low-volume. In Warsaw, a single surgeon performed all repairs and was determined as a high-volume operator; in Prague, all 5 surgeons were also high-volume; in Bratislava, 2 surgeons were high-volume and 3 surgeons were low-volume operators.

In order to assess intra-rater reliability, 30 randomly selected models were reassessed after a 60-min break.

2.3. Statistical analysis

Intra- and inter-observer agreement was evaluated with proportionally weighted kappa statistics. The kappa values were interpreted according to the method used by Altman (1991). One-way analysis of variance was carried out to compare the Goslon scores between samples.

Three regression models were made with the Goslon score as dependent variable and the following independent variables: (1) non-trivial principal components (PC1 to PC8) calculated in the

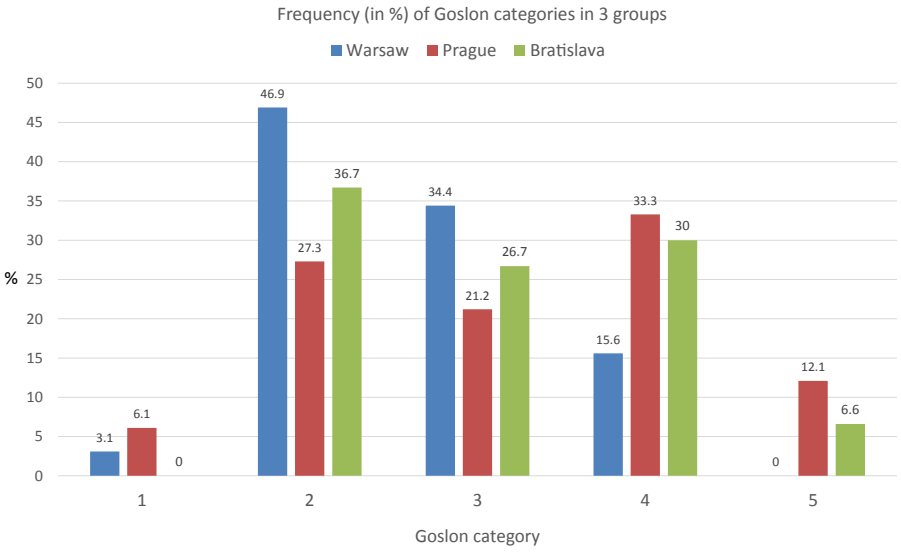

Fig. 1. Frequency of Goslon scores in the study groups.

first part of the Slavcleft study, (2) cephalometric variables sella-nasion-subspinale angle (SNA), subspinale-nasion-submentale angle (ANB), and inclination of the mandibular plane relative to the cranial base (NSL/ML) measured in the first part of the Slavcleft study, and (3) group, age when models were made, age at palatoplasty, and high-vs. low-volume surgeon who performed palatoplasty.

Statistical analyses were performed with STATA software v.13.

3. Results

3.1. Reliability

Both intra- and inter-rater agreement were good or very good according to the method used by Altman (1991). The kappa for intra-rater concordance ranged from 0.825 to 0.959, while the kappa for inter-rater agreement ranged from 0.703 to 0.807 (Table 1).

3.2. Treatment outcome

Table 2 demonstrates the mean Goslon scores for the Warsaw (mean, 2.58), Prague (mean, 3.21), and Bratislava (mean, 3.07) groups. The only statistically significant difference was between the Warsaw and Prague centers. Distribution of the Goslon grades in the samples is shown in Fig. 1. In the Warsaw sample, 50% of patients were scored 1 or 2, 34.4% patients, scored 3, and 15.6% were scored 4 (none was scored 5). In the Prague sample, 33.4% of participants were scored 1 or 2, 21.2% participants were scored 3, and 45.4% participants were scored 4 or 5. In the Bratislava sample, 36.7% of patients were scored 1 or 2, 26.7% participants were scored 3, and 36.6% were scored 4 or 5.

The regression model (Table 3) demonstrated that the group (W, P, and B) was a predictor of the Goslon score (coefficient = 0.45; 95% confidence interval = 0.046–0.853; p = 0.03). Neither age when palatoplasty was performed nor age when models were made was associated with the Goslon score.

3.3. Relationship between Goslon score and craniofacial morphology

Table 4 shows that none of the 8 non-trivial PCs was a predictor of the Goslon score. Similarly, no cephalometric variable was associated with Goslon rating (Table 5). It should be noted, however, that it was possible to analyze the relationship between Goslon and craniofacial morphology only in 29 of 95 participants, with the remaining subjects having cephalograms taken more than 1 year earlier or later then when diagnostic models were made.

4. Discussion

In the second part of the Slavcleft study, we compared the dental arch relationship in patients treated in Warsaw, Prague,

Table 1
Intra- and inter-rater agreement assessed with proportionally weighted kappa statistics.

Raters	Weighted kappa	95% CI
1	0.836	0.693 to 0.979
2	0.825	0.656 to 0.993
3	0.959	0.881 to 1.000
1 vs 2	0.733	0.641 to 0.824
1 vs 3	0.807	0.729 to 0.884
2 vs 3	0.703	0.606 to 0.799

CI, confidence interval.

Table 2
Goslon scores in groups compared with analysis of variance with post hoc Tukey–Kramer tests for pairwise comparisons.

Group	Mean Goslon score	SD	Inter-group differences (for p < 0.05)
Warsaw (n = 32)	2.58	0.77	W vs P
Prague (n = 33)	3.21	1.04	P vs W
Bratislava (n = 30)	3.07	0.99	

P, Prague; SD, standard deviation; W, Warsaw.

Table 4
Regression model with Goslon Yardstick score as dependent variable and 8 non-trivial principal components (PC 1 through PC 8) as independent variables.

	Coef.	SE	P value	LL 95% CI	UL 95% CI
PC 1	−4.512	4.336	0.311	−13.557	4.533
PC 2	5.489	5.622	0.340	−6.238	17.215
PC 3	2.721	6.383	0.674	−10.594	16.035
PC 4	19.110	9.340	0.054	−0.373	38.593
PC 5	−10.031	10.014	0.328	−30.921	10.859
PC 6	−6.824	10.212	0.512	−28.125	14.477
PC 7	3.088	10.381	0.769	−18.567	24.742
PC 8	18.228	12.402	0.157	−7.642	44.097
Constant	2.861	0.184	<0.001	2.476	3.246

CI, confidence interval; Coef., coefficient; LL, lower limit; SE, standard error; UL, upper limit.

Table 5
Regression model with Goslon Yardstick score as dependent variable and 3 cephalometric variables (s-n-ss, ss-n-sm, and NSL/ML angles) as independent variables.

	Coef.	SE	P value	LL 95% CI	UL 95% CI
s-n-ss (SNA)	0.005	0.058	0.933	−0.115	0.124
ss-n-sm (ANB)	−0.057	0.079	0.475	−0.220	0.106
NSL/ML	−0.009	0.037	0.815	−0.085	0.068
Constant	2.802	4.983	0.579	−7.505	13.110

CI, confidence interval; Coef., coefficient; LL, lower limit; SE, standard error; UL, upper limit.

Table 3
Regression model with Goslon Yardstick score as dependent variable and group, age when models were made, surgeon who performed palatoplasty, and age at palatoplasty as independent variables.

	Coef.	SE	P>|t|	LL 95% CI	UL 95% CI
Group (1, Warsaw; 2, Prague; 3, Bratislava)	0.450	0.202	**0.030**	0.046	0.853
Age when models were made	−0.017	0.100	0.869	−0.216	0.183
Surgeon who repaired palate	−0.066	0.051	0.202	−0.169	0.036
Age when palatal repair was done	0.085	0.071	0.236	−0.057	0.227
Constant	2.420	1.227	0.053	−0.031	4.871

CI, confidence interval; Coef., coefficient; LL, lower limit; SE, standard error; UL, upper limit. Bold refers to a statistically significant difference.
P = 0.025, R^2 = 0.156, adjusted R^2 = 0.104.

and Bratislava with different surgical protocols. The current findings are in consistency with the results of our cephalometric study (Urbanova et al., 2016). Patients treated in Warsaw showed relatively favorable outcomes, while those patients treated in Prague demonstrated relatively unfavorable outcomes. Thus, the H_0 hypothesis was rejected: the dental arch relationship in the 3 groups is different.

We used the Goslon Yardstick because it is a popular index for rating occlusal outcomes in CLP during the mixed dentition stage. Consequently, it is possible to relate outcomes achieved in the participating centers with outcomes achieved by other cleft teams, e.g., participants of the Eurocleft (Mølsted et al., 2005) and Americleft (Hathaway et al., 2011) studies. Thus, in comparison to outcomes reported in these two inter-center collaborations, dental arch relationships obtained in Warsaw are comparable to the best results achieved within the Eurocleft and Americleft studies, while the occlusal outcome achieved in Prague is comparable to an average occlusal outcome reported by the Eurocleft/Americleft teams.

The same patients treated in Warsaw were assessed with the Goslon Yardstick during the Warsaw–Oslo comparison (Fudalej et al., 2009). It was reassuring to see that the scores given in 2009 and in this study are almost identical (2.68 vs. 2.58, respectively).

In the first part of the Slavcleft study (Urbanova et al., 2016), we found that 3 of 8 non-trivial PCs discriminated patients from Warsaw, Prague, and Bratislava (PC3, PC6, and PC8), respectively. These PCs referred mainly to variation of the cranial base, maxillary alveolus, mandibular angle, and soft tissues (nose and lips). The regression analysis performed here demonstrated that none of these PCs was associated with the Goslon score. Thus, our findings disagree with reports that found an association between dento-craniofacial variables and dental arch relationship assessed with the Goslon Yardstick. For example, Morris et al. (1994) reported that among the variables overjet, overbite, incisal angulation, and canine and molar cross-bites, the presence of overjet alone explained 87% of the variance of the Goslon score. The Americleft investigation (Daskalogiannakis et al., 2011) showed a weak but statistically significant correlation between the Goslon score and s-n-ss (SNA) angle ($r = 0.284$) and s-n-sm (SNB) angle ($r = 0.253$). A significantly moderate negative correlation was found between the Goslon rating and ss-n-sm (ANB) angle ($r = -0.607$). The disagreement between our findings and the reports of Morris et al. (1994) and Daskalogiannakis et al. (2011) may be the result of the relatively few participants ($n = 29$) in our study in whom dental arch relationship and craniofacial morphology were evaluated. Both Morris et al. and Daskalogiannakis et al. studied significantly more subjects (40 and 148, respectively) than we did.

To our knowledge, only one inter-center study using the Goslon Yardstick has been published on a sample of patients having had CUCLP closure in one single operation (Fudalej et al., 2009). One reason for this may be that few cleft teams have adopted this protocol. In a survey of 201 European cleft teams, a one-stage closure was used by only 5% of the teams (Shaw et al., 2001). Nevertheless, one-stage repair of CUCLP could be a very interesting treatment alternative, particularly in developing countries in which there is limited access to cleft services. In these countries, some patients with CUCLP have only their lip repaired and do not return for palatoplasty for economic and compliance reasons. If the current findings such as favorable dental arch relationship after Warsaw one-stage repair of CUCLP can be generalized for other surgeons and other outcomes, the one-stage technique could be a valuable treatment option. Not only could the CUCLP be fully repaired during one operation, but also the burden of care for the child and family would be markedly reduced by eliminating the need for a second operation. Moreover, health care costs would be less.

5. Conclusion

This investigation showed that the dental arch relationships in patients treated in Warsaw with one-stage repair were more favorable than in those treated in Prague treated with a two-stage repair. Furthermore, it was demonstrated also that the dental arch relationships were comparable to the best results as evaluated in the Eurocleft and Americleft studies. The outcome scores obtained in Bratislava were found to be intermediate between the other centers. The current results corroborate the morphological findings assessed in the first part of the Slavcleft study.

Conflicts of interest

The authors declare no conflicts of interest.

Acknowledgment

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jcms.2019.03.023.

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