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ORIGINAL ARTICLE

Caries prevalence and enamel defects in 5- and 10-year-old children with cleft lip and/or palate: A case-control study

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Abstract

Objective. To determine the prevalence of dental caries and enamel defects in 5- and 10-year-old Swedish children with cleft lip and/or palate (CL(P)) in comparison to non-cleft controls. ***Materials and methods.*** The study group consisted of 139 children with CL(P) (80 subjects aged 5 years and 59 aged 10 years) and 313 age-matched non-cleft controls. All children were examined by one of two calibrated examiners. Caries was scored according to the International Caries Detection and Assessment System (ICDAS-II) and enamel defects as presence and frequency of hypoplasia and hypomineralization. ***Results.*** The caries prevalence among the 5-year-old CL(P) children and the non-cleft controls was 36% and 18%, respectively ($p < 0.05$). The CL(P) children had higher caries frequency (initial and cavitated lesions) in the primary dentition than their controls (1.2 vs 0.9; $p < 0.05$). A significantly higher prevalence of enamel defects was found in CL(P) children of both age groups and anterior permanent teeth were most commonly affected. ***Conclusions.*** Preschool children with cleft lip and/or palate seem to have more caries in the primary dentition than age-matched non-cleft controls. Enamel defects were more common in CL(P) children in both age groups.

Key Words: *Cleft palate, cleft lip and palate, dental caries, hypomineralization, hypoplasia*

Introduction

Cleft lip and/or palate (CL(P)) are fairly common deformities. The incidence in Sweden is 2/1000 newborns [1] and around 150–200 children are born each year with some form of oral clefts. The management is a challenge that demands extensive specialist care, involving multi-disciplinary team from medicine and dentistry. In addition, children with CL(P) rarely escape dental complications that can cause additional suffering and lead to an impaired oral-health related quality-of-life [2]. It is generally thought that children with CL(P) have an increased caries risk and numerous studies from different parts of the world have shown a higher prevalence of caries in comparison with a general non-cleft population [3–10]. However, conflicting results have been reported by others [11–14] and also comprehensive reviews have displayed diverging conclusions [15,16]. The current knowledge on caries prevalence in Swedish children with CL(P) is sparse, since the most recent investigation dates back to the mid-80s [17]. In the light of the global decline in childhood caries [18] it was thought to be of interest to evaluate the current burden of caries among children with CL(P) living in Sweden. In addition, the prevalence of enamel development disturbances has been reported to be high in children with CL(P), especially on incisors located adjacent to the cleft [19–21]. The aim of the present study was, therefore, to investigate the caries prevalence and frequency, as well as enamel alterations, in primary and permanent teeth of children with CL(P) in comparison to age-matched non-cleft controls. The null hypothesis was that no difference in caries prevalence would be displayed between the two groups.

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Table I. Distribution of different types of clefts in the CL(P) groups. Values in the table denote the number of subjects and the percentages are in brackets.

Type of cleft	5-year-olds	10-year-olds
Cleft lip (CL)	18 (22.5)	9 (15.3)
Cleft palate (CP)	12 (15.0)	17 (28.8)
Unilateral cleft lip and palate (UCLP)	37 (46.3)	23 (39.0)
Bilateral cleft lip and palate (BCLP)	13 (16.3)	10 (16.9)
Total CL(P)	80	59

Table II. Distribution of medical conditions in CL(P) and non-cleft controls. Values in the table denote the number of subjects and the percentages are in brackets.

Variable	CL(P) n = 139	Control n = 313
Asthma	16 (11.5)	22 (7.0)
Heart disease	2 (1.4)	5 (1.6)
Epilepsy	0 (0)	3 (0.9)
Diabetes	0 (0)	1 (0.3)
Attention Deficit/Hyperactivity Disorder	2 (1.4)	2 (0.6)
Autism	2 (1.4)	1 (0.3)
Pierre Robins sequence	1 (0.7)	0 (0)

Materials and methods

The project had a cross-sectional case-control design and was approved by the regional ethics committee in Linköping (Dnr 2011/252-31 and Dnr 2012/304-32).

Study groups

CL(P) group. All 5- and 10-year-old children born with CL(P) attending two regional cleft centers in Sweden (Linköping and Gothenburg) were eligible and invited to participate. The cleft center in Linköping is serving the entire south-east region of Sweden and the center in Gothenburg is serving the south-western part of the country. Children in the youngest age group (5-year-olds) were born between October 2006 and December 2008 and children in the older age group (10-year-olds) were born between December 2001 and December 2003. Parents and their children received written information about the study, sent by mail or handed over when they were visiting the cleft center. Non-responders were re-contacted per mail and/or per telephone 2–4 weeks after the first information. In total, 258 children were invited and 139 children (50%) were enrolled and clinically examined after informed consent. The reasons for the attrition were (i) no response to the invitation (n = 82), (ii) declined to consent (n = 35), and (iii) no cooperation (n = 2). The distribution of the children according to type of clefts is shown in Table I. Forty-four children were adopted from China. The children's medical background is presented in Table II.

Control group. Age-matched children born without CL(P) were randomly selected from six different public dental clinics located in the same geographic regions as the CL(P) group (Jönköping, Tranås and Mjölby in the southeast region; Borås, Ulricehamn and Falköping in the southwest region). The children were born between January 2002 and December 2003 and January 2007 and December 2008 and the dental examination was timed with the regular dental recall visit at the clinics. The parents agreed to their child's participation in the study. In total, 313 non-cleft children were examined. In this group, one child was adopted from China. The children's medical background is presented in Table II.

Clinical examination

All children were examined from May 2012 through January 2014. The examinations were performed at the children's local dental clinics or at the cleft centers in Linköping or Gothenburg. All children were examined by one of two pediatric dentists (ALS or AKN) and the inspections were performed in a fully equipped dental chair. The caries scoring was preceded by professional tooth cleaning with rubber cup and prophylactic paste and thorough drying with compressed air. Caries registrations were made according to International Caries Detection and Assessment System (ICDAS-II) as described by Pitts and Ekstrand [22]. The caries lesions were staged as 'initial' (ICDAS 1–2), 'moderate' (ICDAS 3–4) and 'extensive' (ICDAS 5–6). A primary incisive or canine earlier extracted because of caries was counted as two decayed surfaces and a primary molar was counted as three. Enamel alterations, assessed as hypomineralization (including opacities) or hypoplasia were registered on surface level in both primary and permanent teeth. Hypomineralization was registered when the enamel had normal thickness, intact surface and displayed alterations in enamel translucency of a variable degree. Both demarcated and diffuse opacity with white, creamy, yellow or brown color were recorded. Hypoplasia was recorded when localized reduction in enamel thickness was present as pits or grooves or when a more extensive part of the tooth surface was affected. No radiographs were taken. Before the start of the study, the examiners were calibrated using the ICDAS-II criteria and the registrations were validated through a re-examination of 15 children within a period of 1 month. The intra- and inter-examiner agreement produced an index of positive consensus 0.75 (examiner 1), 0.92 (examiner 2) and 0.97, respectively. The corresponding values for a negative consensus were 0.99 (examiner 1), 1.00 (examiner 2) and 1.00.

Outcome measures

The primary end-point was caries prevalence in the primary and mixed dentition, expressed as a percentage. Secondary end-points were caries frequency, expressed as decayed, missing or filled surfaces in primary (dmfs) and in permanent teeth (DMFS). Enamel defects were scored as the presence and frequency of hypoplasia or hypomineralization in primary and permanent teeth.

Statistical analysis

Descriptive statistics were computed using IBM-SPSS software (version 20, Chicago, IL). A non-parametric test was performed to evaluate the difference between the study and control groups (Mann-Whitney U-test). The same method was used to compare mean hypomineralization and hypoplasia scores between study and control groups. Analysis of variance (ANOVA) was used to compare caries prevalence between children with different cleft diagnosis as well as to compare caries prevalence in the study group between children adopted from China and children born in Sweden. A multivariate logistic regression analysis with the independent variables: CL(P), health status and born in China, was performed to determine how these factors contribute to the frequency of hypomineralization and hypoplasia in primary and permanent teeth. Level of statistical significance was set at 5% ($p < 0.05$).

Power analysis

Based on regional epidemiology, the caries prevalence among non-cleft children was estimated to vary around 20–40% in the two age groups. A power calculation with $\alpha = 0.05$ and $\beta = 0.2$ indicated that 176 patients in each group (case vs control) would be required in order to detect a 50% difference between the groups. It was, therefore, decided to increase the ratio of sample sizes to 1/2 (case vs control), thereby requiring 133 cases and 266 non-cleft controls.

Results

The distribution concerning age and gender of the participants in the different groups is presented in Table III.

5-year-olds

The caries prevalence was 36% in the CL(P) group compared to 18% in the non-cleft control group and this difference was statistically significant. The children with CL(P) also had significantly more caries lesions (d1-6mfs) than those in the control group, as shown in Table IV. The difference was also significant at the dentin level (d3–6mfs) and for extensive dentin lesions (d5–6mfs). The relative proportion of caries lesions located in primary incisors was 36% in children with CL(P) compared to 19% in non-cleft controls. The difference was statistically significant ($p < 0.05$).

Table III. Distribution of gender and age. Values in the table denote number of subjects.

Variable	5-year-olds		10-year-olds	
	CL(P)	Control	CL(P)	Control
Total	80	144	59	169
Boys	46	77	34	71
Girls	34	67	25	98
Mean age (SD)	5.4 (0.5)	5.2 (0.3)	10.4 (0.6)	10.1 (0.3)

Table IV. Caries frequency (mean values and standard deviation, SD) in 5-year-old children with CL(P) and non-cleft controls. Decayed missing filled surfaces refers to ICDAS.

Variable	CL(P) n = 80		Control n = 144		
	Mean	SD	Mean	SD	p
d1–2mfs	0.6	2.2	0.4	1.8	NS
d3–4mfs	0.7	2.2	0.5	1.9	0.04
d5–6mfs	0.8	2.1	0.5	1.8	0.01
d1–6mfs	1.2	2.6	0.9	3.2	0.01
ms	0.3	1.3	0.1	0.8	NS
fs	0.2	0.9	0.1	0.4	NS

Statistical method: Mann-Whitney U-test.
NS = no statistically significant difference.

10-year-olds

In this age group, the caries prevalence in the permanent teeth was 47% in the CL(P) group compared with 38% in the control group, but this difference was not statistically significant. The caries scores in primary and permanent teeth as well as the total mean values for the mixed dentition are shown in Table V.

When the two age groups were analyzed together the caries prevalence was 41% in the CL(P) group compared to 29% in the control group, a difference that was statistically significant.

Type of clefts and adoption

No significant differences in caries prevalence or frequency were found between children with different types of cleft. No difference in caries prevalence or frequency between children with CL(P) adopted from China and the rest of the CL(P) children were found.

Table V. Caries frequency (mean values and standard deviation, SD) in 10-year-old children with CL(P) and non-cleft controls. Decayed missing filled surfaces refers to ICDAS.

Variable	CL(P) n = 59		Control n = 169		
	Mean	SD	Mean	SD	p
d1–2mfs	0.6	1.4	0.9	2.1	NS
d3–4mfs	0.7	1.4	0.9	2.2	NS
d5–6mfs	0.7	1.4	1.1	2.3	NS
d1–6mfs	0.9	1.5	1.2	2.6	NS
ms	0	0	0.2	1.2	NS
fs	0.6	1.3	0.7	1.7	NS
D1–2MFS	0.4	0.8	0.4	1.3	NS
D3–4MFS	0.5	1.2	0.4	1.3	NS
D5–6MFS	0.3	0.7	0.3	1.2	NS
D1–6MFS	0.7	1.4	0.5	1.5	NS
MS	0	0	0	0	NS
FS	0.3	0.6	0.3	1.4	0.04

Statistical method: Mann-Whitney U-test.
NS = No statistically significant difference.

Table VI. Frequency of primary tooth surfaces with hypomineralization and hypoplasia (mean values and standard deviation, SD) in 5-year-old children with CL(P) and non-cleft controls.

Primary dentition	CL(P) n = 80		Control n = 144		
	Mean	SD	Mean	SD	p
Hypomineralization incisors	0.5	1.0	0.2	0.8	0.00
Hypomineralization canines and molars	0.3	0.8	0. 2	0.6	NS
Hypoplasia incisors	0.2	0.6	0.0	0.0	0.00
Hypoplasia canines and molars	0.4	1.1	0.1	0.2	0.00
Total hypomineralization and hypoplasia	1.5	2.0	0.8	1.6	0.00

Statistical method: Mann-Whitney U-test.
NS = No statistically significant difference.

A significant difference ($p < 0.05$) in total numbers of primary teeth surfaces with hypomineralization and also hypomineralization/hypoplasia between CL(P) adopted from China and the rest of the children with CL(P) were seen. No such difference was seen in the permanent dentition. In the multivariate logistic regression analysis, only the independent factor CL(P) significantly increased ($p < 0.05$) the risk for hypomineralization and hypoplasia in primary and permanent teeth.

Enamel defects

Among the 5-year-olds, the prevalence of hypomineralization/hypoplasia was 61% in the CL(P) group compared to 26% in the non-cleft control group ($p < 0.05$). The corresponding values for the 10-year-olds were 75% and 47%, respectively ($p < 0.05$). The frequency in relation to the location within the dentition is detailed in Tables VI and VII. Enamel defects were by far most common in permanent central incisors, while this pattern was less clear in the primary dentition.

Discussion

This study was performed to elucidate the caries burden in two comparatively large samples of age-defined children with CL(P). We used a comprehensive and validated caries classification system (ICDAS II) that provided detailed caries information on initial and moderate lesions as well as extensive cavities. Moreover, the inter- and intra-examiner agreement values were good and thus, the caries scores were considered as reliable. The results were clear-cut; 5-year-old children with CL(P) had significantly more caries than the non-cleft controls, while this was not the case in the mixed dentition of the 10-year-olds. Thus, the null-hypothesis could only partly be rejected. The results were in agreement with several previous studies [16,23,24] but in contrast to others [12,15,25]. Earlier data suggest that children with cleft lip and palate have more caries than children with isolated cleft lip [19,26,27], but this could not be verified in the present study. It should however be noted that this study was sub-optimal for this research question. Five-year-old children with CL(P) had more caries in the primary teeth than the 10-year-olds. This was likely explained by the high number of exfoliated caries-affected anterior teeth in the older age group. This is the first Swedish study since 1989, investigating caries frequency in children with CL(P). At that time, Dahllöf et al. [17] reported mean dmfs values of 7.0 and 3.9 in their cleft and non-cleft groups. Corresponding ICDAS values in the present study were 1.2 and 0.9, respectively.

An interesting finding was the high prevalence of enamel defects among the children with CL(P), with permanent teeth more affected than primary. The most common type was more or less demarcated opacities on permanent anterior teeth, which was in accordance with previous reports [20,21]. The high prevalence of hypomineralization and hypoplasia in anterior permanent teeth may be an esthetical concern and one of many risk factors for caries development in children with CL(P) [28].

The present conclusions must, however, be drawn with some caution since there are a number of factors that may have influenced the results. First of all, the high number of non-responders was a concern; 50%

Table VII. Frequency of permanent tooth surfaces with hypomineralization and hypoplasia (mean values and standard deviation, SD) in 10-year-old children with CL(P) and non-cleft controls.

Permanent dentition	CL(P) n = 59		Control n = 169		
	Mean	SD	Mean	SD	p
Hypomineralization incisors	1.7	2.0	1.3	2.1	0.01
Hypomineralization molars	0.8	1.4	0.5	1.2	0.03
Hypoplasia incisors	0.4	0.8	0.0	0.1	0.00
Hypoplasia molars	0.1	0.4	0.0	0.1	NS
Total hypomineralization and hypoplasia	3.0	3.0	1.8	2.7	0.00

Statistical method: Mann-Whitney U-test.
NS = No statistically significant difference.

of the invited children with CL(P) chose to decline to participate in this study and their level of oral health remains an open question. The exact reasons for the low interest to participate are unknown, but many children with CL(P) have a history of extensive and long-term treatment needs and one additional examination could possibly have been perceived as one too many. Unfortunately, no caries data were available from the non-responders, so one can only speculate on the selection bias. The limited attendance made the present investigation slightly underpowered, especially in 10-year-olds, which made any sub-grouping on type of clefts or its location less meaningful. Secondly, medical or neuropsychiatric conditions were relatively common in the CL(P) group and several of them, such as asthma and ADHD, may also impair the oral health [29,30]. Furthermore, 32% of the children with CL(P) were adopted from China, many of them with special needs, which also may have affected the results, albeit no significant difference in caries prevalence was found between adopted and non-adopted children in this study. Thirdly, no radiographs were taken and, with visual-tactile caries detection only, the true caries prevalence is clearly under-estimated. For example, in 5-year-old Swedish children, a 'gain' of 1.2 proximal caries lesions for primary molars was found when bite-wings were added to the clinical examination [31]. The use of radiographs in caries surveys is controversial and should not be used unless the children clearly benefit from the examination. In this context, children with CL(P) are exposed to frequent radiographic examinations and this was the main reason to refrain from additional radiographs. Nevertheless, the potential caries underscoring of proximal caries was most likely of similar magnitude in both the CL(P) and non-cleft group or possibly even somewhat higher in the CL(P) group.

The finding that the children with CL(P) did not have a higher caries prevalence or frequency in their permanent teeth in comparison to non-cleft controls was comforting and inspiring, being a result of regular and comprehensive preventive care. Otherwise, it is well established that high caries levels in the primary dentition are related to an increased caries risk in the permanent dentition [32]. Furthermore, caries treatment in young ages can cause dental fear and behavior management problems [33]. This can jeopardize the orthodontic treatment frequently needed in CL(P) children. In any case, our present findings reinforce the fact that preventive care should be introduced in pediatric and dental healthcare at an early age and a pediatric dentist responsible for oral health should be included in the cleft team.

Conclusion

Within the limitations of the present study, it was shown that children with CL(P) displayed more caries in the primary dentition than age-matched non-cleft controls. This difference was however not apparent in the mixed and young permanent dentition. Likewise, enamel defects were more frequent in CL(P) children, especially in anterior permanent teeth. Consequently, pre-school children with CL(P) are at risk for caries development and in need of additional professional preventive care in young age.

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