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Caries prevalence in children with cleft lip and palate – a systematic review of case–control studies

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Aim. To conduct a systematic review of literature in order to examine the evidence of an increased prevalence of dental caries in children with cleft lip and palate (CLP).

Methods. A search of the PubMed database was conducted through May 2006. Sex- and age-matched case–control studies with noncavitated and manifest caries lesions as endpoint were targeted ($n = 6$). The studies were assessed independently by two reviewers and scored A–C according to predetermined criteria for methodology and performance.

Results. Significantly more caries in CLP children were reported in two of the four studies in the permanent dentition and in three out of four publications dealing with primary teeth. None of the articles were, however, assessed with the highest grade 'A' and the level of evidence was therefore based on three papers graded 'B'. There was a tendency towards higher caries scores in preschool children, but as conflicting results were revealed, the evidence that children with CLP exhibit more caries than noncleft controls was inconclusive.

Conclusion. This systematic review of literature was unable to find firm evidence for the assumption that CLP children have an increased prevalence of dental caries.

Introduction

The incidence of cleft lip and palate (CLP) is reported to be 2 for every 1000 live births[1], making it one of the most common congenital malformations in Sweden. A healthy primary dentition with avoidance of early extractions is essential in order to preserve bone adjacent to the alveolar cleft and for maintaining space[2]. Dental caries is still a global public health problem and constitutes the main threat to children's oral health today[3]. It is commonly believed from studies conducted worldwide that children with CLP have an increased risk for caries, an elevated incidence and more untreated cavities, especially in the primary dentition[4–7]. Other studies have, however, not been able to confirm this and factors such as cleft type, inclusion of syndromes, the age of subjects, preventive care, and incidence levels on top of in-homogenous study designs may have contributed to the inconsistency. The aim of this study was therefore to undertake a systematic review of literature in order to assess the evidence whether or not children with CLP display elevated caries levels in the primary and permanent dentition.

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Materials and methods

Literature search

A search of the PubMed database was conducted from 1966 through May 2006, using 'cleft lip and or palate', 'dental caries', 'craniofacial disorders' and 'dental health' as key words. Clinical studies evaluating the prevalence of dental caries in children with CLP, 0–16 years of age, were included while textbooks, review papers, dissertations, abstracts, and case reports were excluded. Papers published in English, German, Swedish, Danish, or Norwegian were accepted. The key issue for inclusion was a cross-sectional case–control study design with the controls matched for at least sex and age and with caries assessed and scored by defined

criteria. Of the 90 references initially found, 43 were selected based on their abstracts. After full reading of these 43 papers by two independent examiners, six papers fulfilled the inclusion criteria (Tables 1 and 2[5,8–12]). In two cases, the examiners had diverging opinions and those papers were re-examined until a consensus was reached. The excluded papers and the main reason for exclusion are listed in Table 3[13–49]. The most common reasons for exclusion were that a control group was lacking (11 papers) or that the comparison group was not properly matched (11 papers). Four review papers, as denoted in Table 3, were hand-searched for relevant papers, but no additional eligible studies were found.

Evaluation of papers and level of evidence

The selected papers were subjected to a critical appraisal carried out independently by the authors. Data were extracted and each reference was assessed with score A–C according to predetermined criteria for methodology and performance. To obtain score 'A', a matching of the controls concerning age, gender, ethnicity, and socio-economy was required along with clinical examinations including bitewing radiographs according to defined criteria, carried out by more than one examiner and with inter- and intra-examiner reproducibility tests. To obtain score 'B', matching for gender and age was required along with either radiographs or reproducibility tests. The number of participants in the study was also considered. In the event of disagreement between the examiners, the paper was re-evaluated until a consensus was reached. The papers were compiled in tables and the results were calculated. The primary outcome measure was caries prevalence expressed as percent or number of decayed, missed, and filled teeth/surfaces in the primary and permanent dentitions. A secondary outcome measure was the prevalence noncavitated lesions within the enamel when available. The percentage difference in caries prevalence between the cases and controls in the various studies was calculated as well as the odds ratio for having cavitated (manifest) lesions. Based on the evaluated literature, the level of evidence (1–4, Table 3) was judged on

Table 1. Study groups, matching, and quality assessment of the six included case–control papers.

First author, year	Ref.	Country	Age	Other syndrome	CLP (*n*)	CTR (*n*)	Matched	Preventive dental care	Grading
Al-Wahadni, 2005	8	Jordan	10–15 years	No	13	13	Age	?	C
Bokhout, 1996	5	Holland	2.5 years	No	76	75	Age, area of birth	?	C
Dahllöf, 1989	9	Sweden	5–6 years	Yes	49	49	Age, sex	Yes	B
Hewson, 2001	10	Ireland	1.5–16 years	Yes	90	100	Trauma clinic; age, sex, geography	?	B
Lauterstein, 1963	11	USA	6.5–10.5 years	?	285	300	Paediatric practice	Water-F	C
Lucas, 2000	12	England	3–15 years	?	60	60	Trauma clinic; age, sex, ethnicity, social class	Yes	B

findings from papers graded as 'A' and 'B' according to the protocol of the Swedish Council on Technology Assessment in Health Care[49].

Results

Six papers met the inclusion criteria as shown in Table 1 and the results are compiled in Table 2 and Fig. 1. Only one of the evaluated papers stated approval from an ethical committee. Significantly more caries in CLP children were reported in two out of four studies of permanent teeth and in three out of four publications dealing with primary teeth. The mean percentage difference in caries between children with CLP and noncleft children was 41% and 7% in the primary and permanent dentitions, respectively. The odds ratio for a child with CLP having manifest lesions (dmft/dmfs/DMFT/DMFS>0) as compared to a non-CLP child could be calculated in four of the included papers. The mean odds ratio was 3.0 with a range between 1.3 and 6.3.

None of the papers included for final assessment were graded high (A). Out of the three papers graded B, two displayed significant differences in cavitated caries prevalence in the primary[9] and in the permanent dentition[10], while one article found no significant differences in neither the primary or in the permanent dentition[12]. Due to the limited number of acceptable papers and the inconsistent findings, it was not possible to find firm evidence that children with CLP exhibit more caries than noncleft controls (evidence level 4).

Discussion

The systemic search for literature with subsequent data extraction and quality assessment is an important and established tool in evidence-based dentistry and the methodology used in this paper was adopted from the Swedish Council on Technology Assessment in Health Care. The present research question was intentionally limited and straightforward and if evidence for elevated caries prevalence was to be unveiled, the next step was planned to explore the possible explanations. The most striking reflection when conducting this review was the high number of publications that was

Table 2. Endpoint measures (percent with caries or mean decayed, filled teeth/surfaces ± SD) in the six included papers.

					Primary dentition				Permanent dentition		
					Initial lesions		Manifest lesions (dft, #dfs)		Manifest lesions (DMFT)		
First author, year	Method	Criteria	X-rays	Reproducibility	Cases	Control	Cases	Control	Cases	Control	Odds ratio
Al-Wahadni, 2005	VIP	WHO	No	Kappa = 0.92	–	–	–	–	4.8 ± 5.1	2.1 ± 1.1 (S)	1.3
Bokhout, 1996	VIP	Koch	No	No	17%	4% (S)	0.6 ± 1.4	0.1 ± 0.5 (S)	–	–	6.3
Dahlöf, 1989	VIP	Koch	Yes	No	0.8 ± 1.1	1.3 ± 2.0 (NS)	7.0# ± 8.5	3.9# ± 5.1 (S)	–	–	2.2
Hewson, 2001	VIP	WHO	No	Kappa ≥ 0.75	–	–	2.5 ± 2.9	0.9 ± 2.0 (S)	1.7 ± 2.2	2.1 ± 3.1 (NS)	2.4
Lauterstein, 1963	VIP	?	Yes	No					8.0	7.5 (NS)	No data
Lucas, 2000	VIP	WHO	No	Kappa = 0.95	–	–	2.4 ± 3.4	2.9 ± 3.1 (NS)	1.2 ± 1.7	1.5 ± 2.3 (NS)	No data

VIP, visual inspection and probing; S, statistically significant difference between CLP and controls ($P < 0.05$); NS, no significant difference ($P > 0.05$).

Table 3. List of excluded references and the main reason for exclusion. CS, cross-sectional case–control design.

First author, year	Ref.	Study design	Main reason for exclusion
Ahluwalia, 2004	13	CS	No matched controls
Bearn, 2001	14	Retrospective	Caries not an outcome measure
Besseling, 2004	15	CS	No control group
Bethmann, 1967	16	CS/cohort	No matched controls
Bethmann, 1968	17	CS	No matched controls
Bian, 2000	18	CS	No control group
Bokhout, 1996	29	CC	Caries not an outcome measure
Bokhout, 1997	20	Prospective	Intervention
Chapple, 2001	21	CS	No control group
Dalben, 2001	22	Case report	No control group
Fonnelöp, 1978	23	CS	No control group
Harris, 2004	24	Systematic review	No original data, hand-searched
Hochstein von, 1970	25	CS	No matched controls
Hochstein von, 1971	26	CS	No matched controls
Huth von, 1979	27	CS	No matched controls
Johnsen, 1984	28	CS	No matched controls
Kaufman, 1991	29	Review	No original data, hand-searched
Kirchberg, 2004	2	CS	No matched controls
Lages, 2004	30	CS	No matched controls
Lin, 1999	31	CS	No matched controls
Lin, 2000	32	RCT	Intervention
Paul, 1998	33	CS	No control group
Pässler, 1973	34	Intervention	No control group
Richter, 1983	35	CS	No control group
Rivkin, 2000	36, 37	Reviews	No original data, hand-searched
Sandy, 1998	38	Methodology	Caries not an outcome measure
Sandy, 2001	39	CS	Caries not an outcome measure
Sell, 2001	40	Treatment protocol	Caries not an outcome measure
Steen, 1981	41	CS	No matched controls
Trulsson, 2003	42	Qualitative	Caries not an outcome measure
Turner, 1998	43	CS	No matched controls
Waurick von, 1979	44	Intervention	No control group
Weiss, 2005	45	Intervention	No control group
Williams, 2001	46	CS	No control group
Wong, 1998	7	Review	No original data, hand-searched
Zschieschack, 1999	48	Intervention	Historic controls

excluded due to the fact that controls were lacking or consisted of convenience samples, historical, or epidemiological data on community level. Interestingly, in a majority (60%) of these excluded studies reporting caries as outcome measure, the conclusion was that CLP children were more prone to be decayed. To be included in this review, a case–control study design with gender- and age-matched controls were required, but even this cut-off level had been partly renounced in three papers. It was also notable that only two references reported matching based on ethnicity and social class/geographical area. On the other hand, these control children were selected from 'trauma clinics' and it may be questioned if they are representative subjects from a caries point of view. It is well known that caries is associated with poverty and especially in preschool children[50] but yet, the included studies lacked data on social class indicators. We are fully aware of the problems with a 'perfect match', but the system of 'social twins' is a concept that could be adopted in most cultures for studies in the future.

The included reports varied with respect to subject age and caries diagnostic methods, which somewhat hampered the comparison of the results. Bitewing radiographs were used in two studies and the intra-examiner reproducibility was reported in three. Previous studies have shown that approximately 30% less caries is registered in clinical examinations without bitewings[51,52] and a true difference between cases and control might therefore be obscured. Only one study had more than a single examiner.

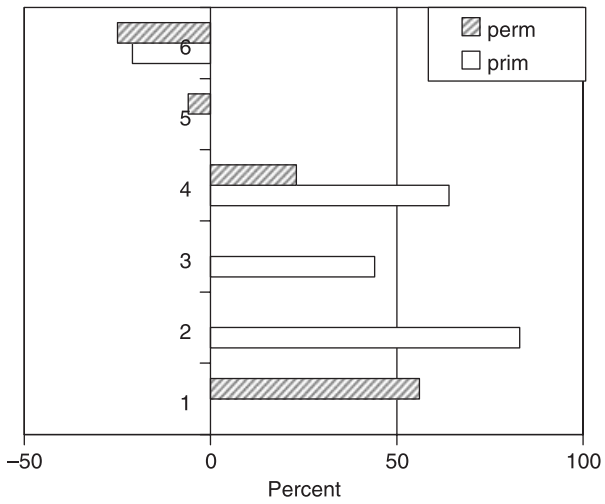

Fig. 1. Differences in caries prevalence in the primary and permanent dentitions expressed as percentage in six case–control reports. A positive value denotes more caries in cleft lip-palate children while a negative value represents higher caries prevalence among control subjects. The vertical numbers denote the included publications 1 = Al-Wahadni *et al.* 2005[8]; 2 = Bokhout *et al.* 1996[5]; 3 = Dahllöf *et al.* 1989[9]; 4 = Hewson *et al.* 2001[10]; 5 = Lautenstein and Mendelson, 1964[11]; 6 = Lucas *et al.* 2000[12].

The problem with one caries examiner is that it is unknown if this person 'under-' or 'over-scores' caries which is of importance and may influence the results. Furthermore, the examiner could not be blinded with the case–control study design and he or she was in many cases also the recall therapist of the children.

We chose to extract the caries data on tooth level when possible but, unfortunately, only the mean values on expense of frequency distributions were reported in several papers. In addition, the different components of the decayed, missed, and fill indices were too infrequently presented to form a basis for further analysis. The percentage difference in caries experience and subsequent odds ratio values were calculated as a rough comparison between the different papers, but it should be emphasized that these figures depended on the overall caries prevalence and must be considered with great caution. In two of the papers[9,12], a general preventive-orientated dental care was provided to both the cases and the controls, while in the study by Lauterstein and Mendelsohn[11], 40% of the children with CLP had fluoridated water in contrast to the vast majority of the control children. It is very likely that a comprehensive preventive care with daily administration of fluoride as the most important component is an effective measure to control caries, but the impact of preventive measures in CLP children was beyond the aim of this review.

In summary, in spite of a clear tendency of more caries in primary teeth among children with CLP, no firm conclusion could be made based on this evaluation. The quality of the eligible studies was low to moderate and the results displayed conflicting findings. There is an obvious need for an improved quality of future research within this area in order to formulate evidence-based treatment guidelines for the multiprofessional team that are taking care of children with CLP.

What this paper adds

- There are few well-designed studies concerning caries prevalence among children with CLP.
- There is a need for further studies to find out whether or not children with CLP have higher caries prevalence than those without CLP.

Why this paper is important to paediatric dentists

- Children with CLP are often managed by paediatric dentists and knowledge about their caries risk factors and prevalence is therefore important.

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