Oral Health Status and Salivary Properties in Relation to Gluten-free Diet in Children With Celiac Disease

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ABSTRACT

Background: Patients with celiac disease (CD) have a wide variety of symptoms, from being asymptomatic to having chronic diarrhea, abdominal pain, and extraintestinal symptoms. In the oral cavity, enamel defects and recurrent aphthous stomatitis are the most common symptoms. The aim of the study was to assess oral health, bacterial colonization and salivary buffering capacity of patients with CD at diagnosis were compared with patients with CD receiving a gluten-free diet (GFD) and healthy children.
Methods: Three groups were prospectively investigated: newly diagnosed CD, CD treated with GFD, and a control group. All of the children were examined by pediatric dentists, and saliva samples were collected for bacterial and pH analysis.
Results: Ninety children were enrolled in the study, 30 in each group. A higher prevalence of enamel hypoplasia (66%) was found in children with CD. Plaque index was significantly lower in the celiac-treated group, which correlated with oral health behavior: teeth brushing and frequency of eating between meals. Children receiving GFD brushed their teeth and used fluoride significantly more often than other children in the study. No difference between groups was found in snack consumption, mutans streptococci and lactobacilli counts in saliva, as well as pH and buffer capacity.
Conclusions: A lower degree of plaque was found in children with CD receiving GFD. This finding could not be explained by salivary properties or bacteria, but rather by better oral hygiene. The results should raise the awareness of pediatric gastroenterologists toward oral health–related issues in children with CD.

Key Words: celiac disease, dental health, plaque, saliva

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Celiac disease (CD) is a multiorgan disease with a strong genetic predisposition (1). Patients with CD have a wide variety of symptoms, from being asymptomatic, diagnosed by screening, to experiencing chronic diarrhea, abdominal pain, and extraintestinal symptoms (2). If untreated, patients with CD are prone to develop long-term complications such as osteoporosis, infertility, autoimmune diseases, and malignancies (2). CD has a wide variety of extraintestinal manifestations, including the oral tissues, in which dental enamel defects and recurrent aphthous stomatitis are the most common symptoms (3–5). Because enamel defects may present among the first symptoms of the disease, dentists may refer patients with such symptoms to be screened for CD even in the absence of gastrointestinal symptoms. Avsar et al (5) showed that better adherence to gluten-free diet (GFD) reduces the prevalence of caries. Acar et al (6) show that children with CD have a lower prevalence of salivary mutans streptococci (MS) and lactobacilli (LB) colonization. The aim of the present study was to assess oral health, bacterial colonization, and salivary buffering capacity of patients with CD at diagnosis compared with patients receiving GFD and with healthy children.

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METHODS

Sample Selection

The study protocol was approved by the institutional human subjects ethics committee of the Hadassah-Hebrew University, Jerusalem, Israel. Informed consent was obtained from all of the parents or legal guardians of the participating children.

Three groups of children were prospectively enrolled in the study: a newly diagnosed celiac group, which comprised children who had gastroscopy and duodenal biopsy for the diagnosis of CD; a celiac-treated group, which comprised children who had been on a GFD at least 6 months; and a control group, which comprised healthy children who attended the Pediatric Dentistry Department in the Hadassah School of Dental Medicine in Jerusalem for routine surveillance. By law, all children in Israel are entitled to free dental care. Exclusion criteria for all of the groups were diseases other than CD and known oral disease.

Clinical Examination

Medical history and medication data for all of the participants were gathered from medical records, interviews, and questionnaires. All of the children in the study were examined by 2 blinded (T.B. and O.L.) specialists in pediatric dentistry. Oral assessment included evaluating dental caries experience, grading of enamel defects, and charting of plaque index. Evaluation of caries experience was performed with the DMFT/dmft index, which describes the amount of dental caries in an individual. It numerically expresses the amount and the prevalence of dental caries in an individual. DMFT/dmft scores are obtained by calculating the number of decayed (D), missing (M), and filled (F) teeth in the dentition. It serves to illustrate the amount of the dentition that has become affected by dental caries up until the day of examination (7). Plaque index (according to Silness and Loe (8)) is used to evaluate the thickness of plaque at the gingival margin of the teeth.

Each surface of the tooth is given a score from 0 to 3, where 0 means no plaque, and 3 means abundance of soft debris. Plaque index is the total score divided by the number of teeth. Enamel defect charting and dental maturity grading were performed according to Aine (9) (Table 1). Examinations were conducted using a dental mirror.

Salivary Sampling

Whole saliva samples were collected from the ventral part of the tongue and the oral vestibulum using a sterile cotton pallet. Saliva analysis was performed using standard bioassay and plating procedures on a Caries Risk Test (CRT; Ivoclar Vivadent Inc, Amherst, NY) used to determine the MS and LB counts in saliva by means of selective culture media. The CRT kits were incubated at 37°C for 48 hours. Enumeration of bacterial growth was conducted as semiquantities ranking according to the manufacturer's instructions. Salivary buffer capacity was detected using pH indicators from CRT and was evaluated using a color scale provided by the manufacturer.

Oral Mucosal pH

Oral saliva was collected using a sterile pipette, and the pH of the collected saliva was assessed using a pH level indicator test strip (pH indicator strips, Mark KGaA, Darmstadt, Germany).

Oral Health Behavior

Parents and participants younger than 15 years were asked to complete a questionnaire translated from that used by Blomqvist et al (10) regarding their children's diet and oral health behavior, including in-between-meals snacks, main beverage consumed during the day, toothbrushing habits, and the use of fluoride-containing toothpastes. This questionnaire was validated in a Swedish study (11). The dietary habits component included the dietary behaviors that were found to be most predictive of caries risk in a clinical setting in the United States (12).

Statistical Analysis

Sample size needed was determined at the design stage of the study. The mean DMFT for the control group was known to be 2.5 ± 3 (mean $\pm$ SD), and for those with CD the mean DMFT was 4.5 ± 3 (Avşar et al (5)). The ratio between the DMFT scores of the 2 groups was 1.8. With the significance level set at 5% (1-tailed) and 80% power, it was calculated that 29 children would be needed for each study group. Accordingly, 30 participants each for the study and control groups, respectively, were recruited.

Data were analyzed using SPSS software (SPSS Inc, Chicago, IL). The analysis consisted of basic descriptive statistical analysis assays: analysis of variance (ANOVA) test was used to compare the DMFT/dmft, pH, and bacterial findings between the 3 groups. χ^2 analysis was used to compare plaque index and oral health behavior among the 3 groups. The Kruskal-Wallis test was used to check for correlation between DMFT/dmft and plaque index. Significance level was set at $P \leq 0.05$.

RESULTS

Population

The newly diagnosed celiac group comprised 30 children (12 boys and 18 girls) ages 1.4 to 15.5 years (mean age 6.9 ± 4.1 years). The celiac-treated group or GFD group comprised 30 children (13 boys and 17 girls) ages 2.5 to 18 years (mean age 9.5 ± 4.5 years). The control group comprised 30 children (15 boys and 15 girls), ages 1.25 to 15 years (mean age 6 ± 2.9 years).

There were no sex differences among the 3 groups ($\chi^2 = 1.69$; $df = 2$; $P = 0.43$), but the celiac-GFD group was significantly older than the other groups (1-way ANOVA; $df = 2$, $F = 6.755$, $P = 0.002$).

DMFT/dmft and Plaque Indexes

The celiac-diagnosed group's mean DMFT/dmft was 1.5 ± 2.2 (range DMFT/dmft 0–9 with a median of 0). The celiac-GFD group's mean DMFT/dmft was 2.0 ± 2.6 (range DMFT/dmft 0–8 with a median of 0.50). Control group mean DMFT/dmft was 3.4 ± 3.7 (DMFT/dmft range 0–11 with a median of 2). No significant difference ($\chi^2 = 1.69$; $df = 2$; $P = 0.43$) was found among the 3 groups, although there was a tendency toward a higher DMFT/dmft index in the control group.

There was a statistically significant difference in plaque index ($P = 0.02$) among the 3 groups (Table 2). The highest plaque index values were found in the celiac-diagnosed group, whereas the lowest values were found in the celiac-treated group. There was no correlation between the plaque index and DMFT/dmft scores. A correlation was found between plaque index and oral health behavior, especially with the frequency of between-meals snacks consumption ($\chi^2 = 9.521$; $df = 2$, $P = 0.009$).

Enamel Defects

No differences were found among the 3 groups regarding the existence of enamel defects (Fisher exact test, $P = 0.79$; Table 3). Children with enamel defects (Fig. 1) had a significantly higher

TABLE 1. Grading of the celiac disease dental enamel defects according to Aine (9)

Classification	Enamel defect
Grade 0	No defect
Grade I	Defect in enamel color. Single or multiple cream, yellow or brown opacities with clearly defined or diffuse margins; in addition a part or the entire surface of enamel is without shiny surface
Grade II	Slight structural defects. Enamel surface rough, filled with horizontal grooves or shallow pits; light opacities and discolorations may be found; in addition a part or the entire surface of enamel is without shiny surface
Grade III	Evident structural defects. A part or the entire surface of enamel rough and filled with deep horizontal grooves that vary in width or have large vertical pits; large opacities of different colors or strong discolorations may appear in combination
Grade IV	Severe structural defects. The shape of the tooth has changed: the tips of cusps are sharp pointed and/or the incisal edges are unevenly thinned and rough; the thinning of the enamel material is easily detectable and the margins of the lesions are well defined; the lesion may be strongly discolored

TABLE 2. Plaque debris index

	Celiac	Celiac control	Control	Total
Mean	1.879378	1.311776	1.421878	1.537677
Median	1.750000	1.233333	1.330000	1.500000
Minimum	1.0000	0.3330	0.0000	0.0000
Maximum	3.1600	2.3333	6.3333	6.3333
SD	0.5622492	0.5556649	1.1840080	0.8494024
N	30	30	30	90

One-way ANOVA $df = 2$, $F = 6.75$, $P = 0.02$. SD = standard deviation.

DMFT/dmft index compared with those without enamel defects (Mann-Whitney test, $P = 0.002$).

Oral Mucosal pH

No difference was found among the 3 groups regarding oral mucosal pH (1-way ANOVA; $df = 2$, $F = 1.76$, $P = 0.18$), but the pH was correlated with buffer capacity ($\chi^2 = 16.791$; $df = 2$, $P = 0.0002$). No correlation was found between salivary buffer capacity and DMFT/dmft ($\chi^2 = 16.791$; $df = 2$, $P = 0.121$).

MS and LB Counts in Saliva

No differences were found among the groups regarding the LB (Fisher exact test, $P = 0.134$) and MS counts (Fisher exact test, $P = 0.143$). No correlation was found between DMFT/dmft and MS counts ($\chi^2 = 2.512$; $df = 3$, $P = 0.47$), but a correlation was found between DMFT/dmft and LB counts ($\chi^2 = 11.023$; $df = 4$, $P = 0.026$).

Dietary and Oral Health Behavior

No differences were found among the 3 groups in consumption of snacks between meals (Fisher exact test, $P = 0.82$, Table 4). A significant (Fisher exact test, $P = 0.001$) difference was found in toothbrushing habits (Table 5). Thirty percent of the newly diagnosed children with CD did not brush their teeth at all compared with 6.7% in the control group and 0% in the celiac-treated group. Use of fluoride was correlated with use of toothpaste during brushing and was also significantly different ($P = 0.02$) among the groups. A correlation was found between brushing habits and DMFT/dmft. ($\chi^2 = 7.945$; $df = 2$, $P = 0.019$). Children who brushed their teeth twice daily had significantly less decay than those who brushed once per day or those who did not brush at all. No correlation was found between snack consumption during the day and DMFT/dmft ($\chi^2 = 4.282$; $df = 2$, $P = 0.118$).

FIGURE 1. Dental enamel defects in a 10-year-old boy with newly diagnosed celiac disease.

DISCUSSION

The present study aimed to assess oral health and elucidate the role of bacteria and saliva properties in the pathogenesis of the oral manifestation of celiac. In our cohort, we found higher rates of plaque in children who were newly diagnosed with CD. This finding was correlated with the oral health behavior and snacks consumption between meals in the newly diagnosed children.

Enamel abnormalities are often described in CD, with prevalence ranging from 10% to 97% (3,4,6). In accordance with the literature, in the present study's cohort, the overall rate of enamel hypoplasia in children with CD, in both treated and untreated groups, was 66%. Surprisingly, the rate of enamel hypoplasia in the control group was higher than expected. This finding may be an anomaly. We have no explanation as to the higher rate of enamel hypoplasia in Israeli children and it warrants further studies to assess the rate of enamel hypoplasia in Israeli population.

The present study found that children with CD receiving GFD had a lower degree of dental plaque. This was in accordance with the finding that this group had significantly better oral hygiene habits than the other groups. The lower rate of plaque could not be explained by the salivary properties that were assessed. The findings that DMFT/dmft values did not differ significantly among the groups are in accordance with Acar et al (6), but they did not distinguish between children receiving GFD and those recently diagnosed as having CD. Although the present study found only a tendency for higher DMFT/dmft values in the control group,

TABLE 3. Enamel hypoplasia grades according to study groups

	Grade I hypoplasia (%)	Grade II hypoplasia (%)	Grade III hypoplasia (%)	Total (% of total enamel hypoplasia)
Celiac newly diagnosed	11 (36.7)	4 (13.3)	2 (6.7)	17 (40)
Celiac treated (GFD)	11 (36.7)	4 (13.3)	1 (3.3)	16 (36.7)
Control	7 (23.3)	2 (6.7)	1 (3.3)	10 (30)
Total (% of total study cohort)	29 (32.2)	10 (11.1)	4 (4.4)	43 (47)

Fisher exact test, $P = 0.79$. GFD = gluten-free diet.

TABLE 4. Summary of dietary and oral health behavior in newly diagnosed celiac group, treated group, and control group

	Snack (times per day)		
	≥3	1–2	Only mealtime
Control			
Subjects (n)	5	22	3
% within study group	16.7%	73.3%	10.0%
Celiac			
Subjects (n)	7	19	4
% within study group	23.3%	63.3%	13.3%
Celiac control			
Subjects (n)	4	21	5
% within study group	13.3%	70.0%	16.7%
Total			
Subjects (n)	16	62	12
% within study group	17.8%	68.9%	13.3%

Fisher exact test, $P = 0.82$.

TABLE 5. Summary of toothbrushing in newly diagnosed celiac group, treated group, and control group

	Brushing (times per day)		
	0	1	2
Control			
Subjects (n)	2	10	18
% within study group	6.7	33.3	60.0
New celiac			
Subjects (n)	9	13	8
% within study group	30.0	43.3	26.7
Controlled celiac			
Subjects (n)	0	10	20
% within study group	0.0	33.3	66.7
Total			
Subjects (n)	11	33	46
% within study group	12.2	36.7	51.1

Fisher exact test, $P = 0.001$.

Priovolou et al (13) found significantly higher values of DMFT/dmft in the nonceliac group. These findings may be explained by the higher awareness of health-related habits in the celiac group and not by any physical difference among groups; however, children receiving GFD were significantly older than those in the other groups, probably because of the time needed to be on the GFD to be included in the present study. This may account for a higher level of maturity and better compliance for brushing on their part, resulting in a correlation between brushing habits and DMFT/dmft scores. The inequality in age among the groups is a limitation of the study.

The difference in the oral hygiene among the groups that resulted in less dental caries in the GFD-treated group may be attributed to differences in socioeconomic background. Data to assess this issue were not collected initially, but when looking at the origin of the patients, most families lived in the 3 major cities around the medical center with similar socioeconomic background. Furthermore, because dental care and education is available free by law to all Israeli children, it is unlikely that low economic status will preclude children to access adequate dental care; however, these issues should be addressed further in future studies.

Contrary to the assumption that high plaque index, an indicator of large bacterial counts, would result in higher caries, no relations were found between plaque index and DMFT/dmft scores. This may be explained by the fact that no differences were found between the MS and LB counts.

All children with CD are diagnosed by a pediatric gastroenterologist. Once the diagnosis is made, apart from initiating GFD, other issues should be addressed. Mostly the presenting signs and symptoms are improved with GFD, but commonly these children may need iron and calcium supplementation. Most physicians do not address oral health issues. Regardless of our findings that show that oral health is improved in celiac-treated children, we showed that children with newly diagnosed CD have significant more caries than the control group This by itself should prompt the treating physician to refer newly diagnosed children with CD to the dentist.

To the best of our knowledge, the present study is the first to examine oral heath in relation to GFD in children with CD. The results showed that newly diagnosed children with CD have more dental plaque and caries than the control groups, and children receiving GFD had lower dental plaque and better oral hygiene. These results should raise pediatric gastroenterologists' awareness toward oral health–related issues in children with CD. Larger prospective studies are required to standardize oral health awareness in newly diagnosed patients with CD.

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