The presence and distribution of dental enamel defects and caries in children with celiac disease

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The aim of this study was to investigate the presence and distribution of developmental enamel defects and caries in children with celiac disease (CD) and compare the results obtained with those of a control group of children without CD. A total of 64 subjects (mean age 8.2 years) selected from patients diagnosed and treated for CD were studied. Sixty-four age/sex-matched healthy children were enrolled as a control group. Permanent dentition enamel defects were recorded according to Aine's classification. The caries experience of the children was recorded according to the criteria of the World Health Organization (WHO). The prevalence of enamel defect in CD subjects was found to be significantly higher (42.2%) than in healthy subjects (9.4%) ($p<0.001$). Grade I type enamel defects were most commonly diagnosed in both groups (20.3% and 6.3%, respectively). The number of caries-free subjects in the control group was higher (38%) than in the CD group (17%). This study clearly showed that children with CD were at an increased risk of dental enamel defects compared with healthy subjects. Enamel defects were associated with an increased caries incidence.

Key words: celiac disease, children, enamel defect, caries, permanent teeth.

The existence of an association between gastrointestinal disorders and dental enamel defects has been known since the turn of the century. The North American Society for Pediatric Gastroenterology, Hepatology, and Nutrition included the presence of specific dental enamel defects as a risk factor for celiac disease (CD)[1]. Dental enamel defects, mainly characterized by pitting, grooving and sometimes by complete loss of enamel, were first reported in children with CD by Aine[2]. These defects are considered specific to CD if they occur symmetrically and are chronologically distributed in all sections of permanent teeth. Bossu et al.[3] analyzed on scanning electron microscope, two samples of enamel fragments from hypoplastic teeth, both deciduous and permanent, harvested from celiac children, and demonstrated that the enamel hypoplasia of deciduous and permanent teeth in CD is highly hypomineralized with shorter prisms, more irregularly distributed and less interprismatic substance than observed in the non-celiac enamel hypoplasia.

Other enamel defects, defined as disturbances in hard tissue matrices, including enamel hypoplasia, enamel opacities, and enamel discolorations that were not symmetrical or chronological in all four sections of the dentition were considered unspecific[2].

The prevalence of specific enamel defects in different European countries varies from 38% to 96%. A number of authors have described the greater prevalence of dental problems in patients with CD[2,4-6] and have postulated that such problems may constitute a diagnostic clue in presentations of the disease that prove difficult to identify[7-8].

The presence or absence of enamel disturbances is presumably dependent on the timing of enamel formation. Enamel mineralization disturbances secondary to CD do not occur before a period of gluten intake coinciding with enamel mineralization. A possible explanation for the enamel defects could be hypocalcemia or, more likely, a particular genetic condition that leads to a specific immune response to gluten[4,10-11].

The aim of this study was to investigate the presence and distribution of developmental enamel defects and caries in children with CD and compare the results obtained with those of a control group of children without CD.

Material and Methods

Study Population

This study was approved by the Ethical Committee of Ondokuz Mayıs University and written informed consent was obtained from each parent. Sixty-four children with CD (33 boys, 31 girls, age range 6-15 yrs) were selected from subjects who had been previously diagnosed and treated based on finding of total or subtotal atrophy on proximal jejunal mucosa and of normalization of the villous architecture together with disappearance of symptoms on gluten-free diet at the Pediatric Gastroenterology Department of Ondokuz Mayıs University between 2000 and 2005. Serological test was also performed to evaluate antigliadin and antiendomysial antibodies. In order to assess the causes of enamel mineralization disturbances, the presence of other disorders in CD subjects was documented. Similarly, the patients' parents were also queried regarding any gastrointestinal symptoms during infancy and early childhood.

Control Subjects

For each CD subject, a sex- and age-matched child was selected from patients receiving their dental treatment at Ondokuz Mayıs University, Faculty of Dentistry, Department of Pediatric Dentistry and included in the study as the control group.

Questionnaire

Gluten-free dietary habits of CD subjects, tooth-brushing habits (less than once, once, more than once per day), daily dietary sugar exposure of both groups (≤1, 2-3, >3 times per day), family income (low income: ≤$600/month, high income: >$600/month), and education levels (low education level: 0-8 years; moderate education level: 8-13 years; high education level: >13 years) of the parents were recorded by a structured questionnaire[12].

Dental Enamel Defect Parameters

Dental examination was always performed by one pediatric dentist (AA) in the Pediatric Dentistry Clinic of Ondokuz Mayıs University, Dental Faculty.

All patients in both groups had at least four permanent teeth in the maxilla and the mandible. All children received a toothbrush and brushed their teeth before examination. Subsequently, all subjects were placed in a conventional dental chair. The teeth were cleaned with an abrasive paste, washed, dried and then carefully observed in good artificial light. The enamel defects affecting the permanent teeth were graded 0 to IV according to Aine's classification[2]. The locations of the defects were noted, along with their number, the affected teeth, and the altered zones of each tooth (incisal, middle and cervical).

Specific enamel defects are defined as occurring symmetrically and chronologically in all four quadrants of dentition, whereas unspecific enamel defects occur symmetrically and chronologically but involve the superior and inferior hemi-arches on the same side[2].

Scoring of Caries Prevalence

The score of decayed, missing and filled teeth (DMFT) was calculated according to World Health Organization (WHO) criteria[13].

Statistical Analysis

The χ^2 or Student's t-test was used to determine significant differences between groups. The statistical level of significance was set at $p<0.05$.

Results

Demographics

The age and sex distribution of the 128 subjects is shown in Table I. The CD subjects studied were between the ages of 6-15 (mean age: 8.2±1.3). Ages at diagnosis, when the first biopsy was made, varied between 8 months to 14 years, but the majority (83%) had not been diagnosed with CD before the age of 2 (mean age: 5.7±2.6).

Twenty-nine percent of the CD subjects regularly brushed their teeth more than once a day, compared with 26% in the control group. Daily dietary sugar exposure was ≤1 per day in 24% of the CD subjects, compared with 38% in the control group. The mean family income of all subjects was $600/month. Twenty-two percent of the CD subjects and 27% of the control group had a high family

Table I. Demographics of Celiac Disease (CD) and Control Subjects

	N	Mean (SD) age (years)	Gender	
			Girls	Boys
CD subjects	64	8.2±1.3	31	33
Control Subjects	64	8.1±4.2	31	33
Total	128	8.1±2.8	62	66

income (>$600/month). Sixteen percent of the mothers and 21% of the fathers of the CD subjects and 13% of the mothers and 22% of the fathers of the control group were highly educated (>13 years). The differences in tooth-brushing habits, socioeconomic status and education levels of the parents between the CD subjects and control group were not statistically significant (p>0.05).

Forty-four of CD subjects reported that they adhered strictly to the gluten-free diet and 17/64 (28.2%) almost strictly.

Prevalence of Dental Enamel Defect

Table II shows enamel defect distribution as a function of grade in the CD and control groups. Systematic enamel defects were determined in 42.2% of CD subjects and 9.4% of the control subjects (Fig. 1). Statistically significant differences were observed between the two groups (p<0.001). Grade I type enamel defects were most commonly diagnosed in both groups (20.3% and 6.3%, respectively). Only one child with CD had Grade IV type enamel defect.

Table II. Grading of Enamel Defects in Celiac Disease (CD) and Control Subjects

	CD group	Control group
Grade	n (%)	n (%)
No defects	26 (40.6%)*	44 (68.8%)
Unspecific defects	11 (17.2%)	14 (21.9%)
Systematic defects	27 (42.2%)**	6 (9.4%)
I	13 (20.3%)	4 (6.3%)
II	8 (12.5%)	2 (3.1%)
III	5 (7.8%)	0
IV	1 (1.6%)	0
Total	64 (100%)	64 (100%)

* p<0.01.
** p<0.001.

The mean age at diagnosis was not statistically different between children with dental enamel defects (8.6±1.3 years) and children without dental enamel defects in the CD subjects (8.1±1.3 years).

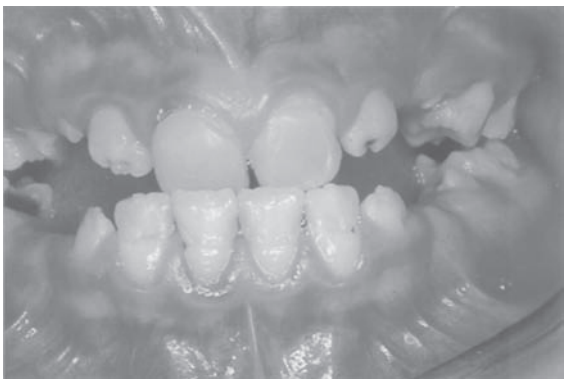

Fig. 1. Systematic dental enamel defects on middle third of mandibular incisors and incisal third of maxillary lateral incisors.

Locations of Dental Enamel Defect

As shown in Table III, the differences observed were statistically significant with respect to involvement of both the incisors (p<0.001) and molars (p<0.001) in the CD subjects compared with the control group (Fig. 2).

Table III. Location of Enamel Defects in Different Groups of Teeth in Celiac Disease (CD) and Control Subjects

Location of enamel defects	CD group n=38 (%)	Control group n=20 (%)
Incisors	35 (92.1%)*	13 (65%)
Canines	15 (39.5%)**	5 (25%)
Premolars	9 (23.7%)	4 (20%)
Molars	27 (71.1%)*	9 (45%)

* p<0.001.
** p<0.05.

The coronal distribution of the defects is shown in Table IV. In contrast to the controls, the CD subjects showed a significant involvement of the incisal two-thirds of the teeth (p<0.05).

Prevalence of Caries

The number of caries-free subjects in the control group was higher than in the CD group (38% and 17%, respectively) and the difference was statistically significant (p<0.001).

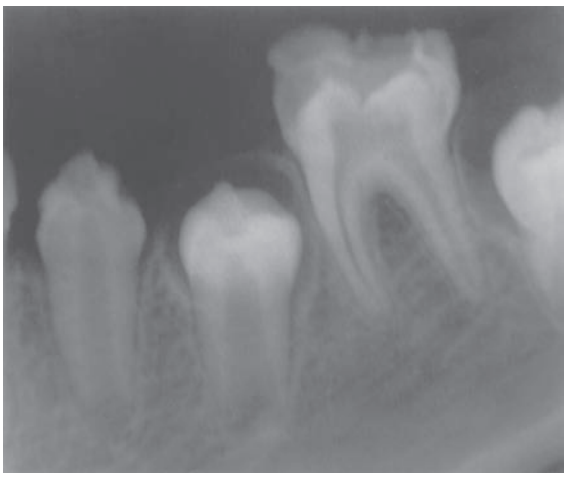

Fig. 2. Radiographic views of systematic enamel defect on first molar and unerupted premolars.

Table IV. Coronal Distribution of Enamel Defects in Patients with Celiac Disease (CD) and Controls

Coronal third	CD group n=38 (%)	Control group n=20 (%)
Incisal	11 (28.9%)	10 (50%)
Middle	8 (21.1%)	7 (35%)
Cervical	6 (15.8%)	3 (15%)
Incisal and middle	7 (18.4%)	0
Complete crown	6 (15.8%)	31 0

The mean DMFT index rates among the CD subjects who adhered strictly or almost strictly to the gluten-free diet and who did not follow diet recommendation were 3.6 (SD=3.26), 5.8 (SD=3.17) and 6.1 (SD=4.28), respectively, compared with a rate of 6.4 (SD=4.32) in the control group. There was a positive correlation between dental enamel defect and caries experience in CD and control groups (Fig. 3).

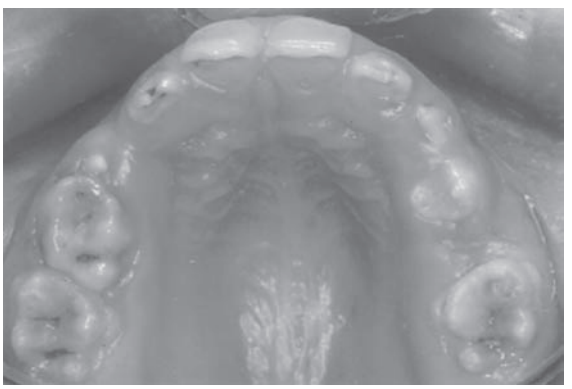

Fig. 3. Dental caries and systematic enamel defect on maxillary permanent first molars.

Association of the dental enamel defect with hypocalcemia at the age of diagnosis of CD hypocalcemia was found in 4 of 11 (36.4%) children in the unspecific defect group and in 10 of 27 (37%) in the systematic defect group. The mean serum concentration of calcium of the unspecific (9.02±2.4 mg/dl) and systematic (9.11±2.7 mg/dl) enamel defect groups was not significantly different.

Discussion

In earlier studies, it was clearly shown that ingestion of gluten and its consequences were responsible for the enamel defects in children with CD in many countries[5-6,8,14-16]. The higher incidence of enamel defects constitutes a fundamental clue in the clinical identification of the disease, with particular application to the growing number of patients with oligosymptomatic or asymptomatic CD[5-7,16].

This is the first study undertaken in Turkey of enamel defects in children with CD. It is important to point out that all of the patients investigated had been diagnosed with CD by intestinal biopsy and serological tests performed to evaluate antigliadin and antiendomysial antibodies, which are the markers of greatest sensitivity and specificity in screening for this disease[7,17-18].

In the present study, a greater number of enamel defects were found in children with CD than in controls (59.4% and 31.2%, respectively). This percentage is much lower than that reported (96%) in Finnish celiacs[2]. Our values are somewhat higher than in some studies. In the Swedish celiac group, 50% (20/40) had mineralization disturbances, which were all opacities except for two cases[13]. This study showed that neither the illness nor the treatment of Swedish children with CD appears to have had a significant influence on the mineralization of permanent teeth. Aguirre et al.[4] reported that 52.5% of 137 Spanish children with CD had enamel defects. In the study of Mariani et al.[11], enamel lesions were found in 28% of 82 Italian children with CD; however, only 15.8% of those had systematic dental lesions. Only 9 of 252 (3.2%) CD children showed enamel hypoplasia in a Swiss study that dealt only with hypoplasia and not at all with enamel opacities[16]. With regard to the normal controls, children also showed less enamel defects (31.2%), similar to Finnish controls (31.4%), than reported in other studies[5,15].

With respect to the occurrence of different types of developmental enamel defects in our study, most of the CD children had Grade I defects (20.3%), which was defect in color of enamel. This was similar to Italian children according to Aguirre et al.[4]. Aine[2], however, observed Grade II defects (23.3%) in most of Finnish children with CD. Grade IV type enamel defects were seen in only one patient (1.6%) in our study. Lahteenoja et al.[15] suggested that the prevalence of Grade III and IV enamel defects was 10.3% in pediatric patients. In the study of Aine[2], 30% of Finnish children with CD had grade IV enamel defects.

The enamel of permanent teeth is formed in early childhood, and structural defects in the enamel might indicate CD prevalence even at that time. Aguirre et al.[4] suggested that diagnosis of CD at an early stage may account for the lesser prevalence of enamel defects. On the other hand, Mariani et al.[11] suggested that the occurrence of enamel lesions does not depend on age at diagnosis. Similar to Aguirre et al.'s[4] study, in our CD children who had been diagnosed in the first two years, 17% of all the cases had the lowest number of enamel defects.

Characterized by small bowel mucosal villous atrophy, CD heals after introduction of gluten-free diet[7,18]. Strict gluten-free diet led to a normal enamel formation and reintroduction of gluten in early childhood resulted in defective enamel re-development[15]. Forty-four of the children with CD, who adhered strictly to the gluten-free diet, showed fewer enamel defects than both those who adhered almost strictly and those who did not follow the diet recommendation.

According to Aine[2], the central incisors are always affected in children with CD. Our results coincide with those reported elsewhere with respect to the important prevalence of enamel defects in incisors and molars, as a result of the time course of odontogenesis and its relation to the active phases of the disease. It should be taken into account that the incisors and molars are the first teeth to undergo calcification, and that molar calcification commences at birth. When gluten is incorporated into the child's diet, bowel alterations with malabsorption develop in response to the immune reaction induced by gluten. Either this reaction or malabsorption itself then generates the metabolic alterations that ultimately produce the enamel defects. This would explain the lesser deterioration of those teeth that calcify at a later stage[4]. The prevalence of enamel defects in premolars and canines was higher than rates reported by Aguirre et al.[4] (20.8% and 9.7%, respectively). The differences could be meaningful because 64% of children with CD were diagnosed before two years of life, compared with 17% of our cases.

Regarding the distribution of enamel defects on the coronal surface, the incisal third was the most affected surface in children with CD. Enamel defects on coronal surface other than incisal third were higher than those reported by previous investigations[4]. This again can be linked to the time course of the disease in relation to odontogenesis.

Intestinal damage leads to malabsorption in patients with CD and some authors suggested that a possible explanation for the presence of enamel defects in these children is that enamel hypoplasia is caused by hypocalcemia, although they did not explore the role of vitamin D[11]. There were no differences in mean serum calcium concentration between children with or without enamel defects in the present study, so it seems unlikely that enamel defects could depend only on calcium metabolism.

The mean DMFT index rate was fairly higher in the control group than among the children with CD who adhered strictly to the gluten-free diet. Similar observations were previously reported by some investigators[4,16,21]. It could be related to a low cariogenic diet and better dental care.

There was a positive correlation between dental enamel defect and caries experience in the CD and control group. This result was in accordance with the reports of Daneshkazemi et al.[19] and Montero et al.[20], who suggested that enamel defects were associated with an increased caries incidence.

Based on the results of our study, we concluded the following:

- Dental enamel defects are common among Turkish children with CD.
- Age at diagnosis of CD plays a role in determining the number of affected teeth.

- Dental enamel defects are not caused by hypocalcemia.
- Dental enamel defects affect mainly the incisors and molars.
- Dental enamel defects are a risk factor for caries in both CD subjects and control group.

Dentists could play an important role in recognizing patients with CD. The dentist will have to be alert for specific dental enamel defects, especially when there are symptoms suggesting CD in the medical history of a child. The limited results of this study need to be validated in the future with larger CD populations in our country.

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