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ORAL MANIFESTATIONS IN A PEDIATRIC PATIENT WITH COELIAC DISEASE: A CASE REPORT

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ABSTRACT

Celiac disease (CD) is among the most common gastrointestinal diseases, both in children and adults. Among clinical oral manifestations of CD, enamel hypoplasia [1-9], atrophic glossitis [10], recurrent aphthous stomatitis (RAS) [4] and a delay in dental eruption [11] have been reported. In this paper we describe the case of a 6 years old girl with severely affected teeth due to celiac disease

INTRODUCTION

During the past few decades, we have come to recognize that celiac disease is one of the most common gastrointestinal diseases, both in children and adults. The true prevalence of the disease in many Western countries is estimated to be as high as 1–3% and is increasing.[12]

The highest occurrence of celiac disease was found among females (65.38%), some studies showing an average female-to-male ratio of 2:1 in the prevalence of this condition. [13,14]

Unfortunately, celiac disease remains undiagnosed in most cases. It is believed that in the United States, more than 90% of all affected patients are unacknowledged.

The prevalence of celiac disease has increased notably in reference to the past, ranging now from 1:85 to 1:300, according to the population and the area considered [16,17-20]. The reason for detecting an increased number of cases may be the improvement in the accuracy of serological markers (measurements of anti-endomysium antibodies EMA, anti-gliadin antibodies

AGA and anti-transglutaminase antibodies tTG), used in the early stages of disease screening and diagnosis [21]. Afterwards, a small-bowel biopsy and a histological examination must be performed to confirm diagnosis [21].

It is important that pediatricians, gastroenterologists and internists have a multidisciplinary approach, because they must pay attention to extra-intestinal manifestations of CD (hematologic, dermatologic, neurologic, gynecological and oral), in order to make an early diagnosis [22].

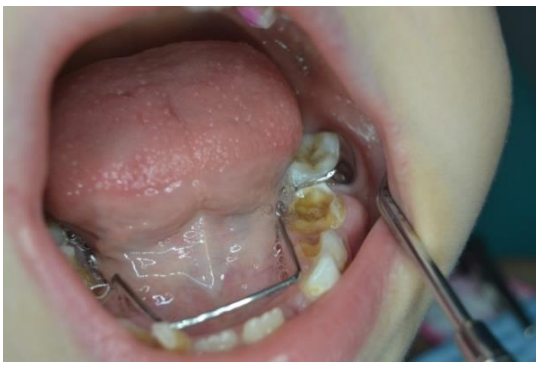
According to a study, 33.0% of coeliac patients were affected by enamel hypoplasia, 8.3% by recurrent aphthous stomatitis, 3.3% by atrophic glossitis and 20.0% by delay in dental eruption.[23]

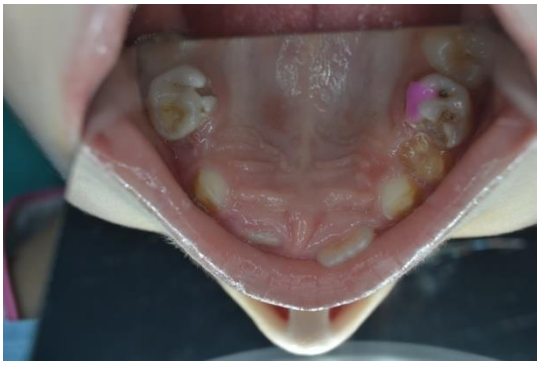
Disturbances in the amelogenesis of permanent and deciduous teeth is a well-defined finding in celiac disease, but there are considerable variations in its reported prevalence. Enamel defects are very common in older studies together with severe infant celiac disease and lower general health [24]. Celiac disease is not a causative factor, but malnutrition, hypocalcemia, and immunologic disturbances have been suggested as causative factors of enamel defects [25], and the severityseems to be associated with the duration of gluten-exposure [26].

MATERIAL AND METHOD

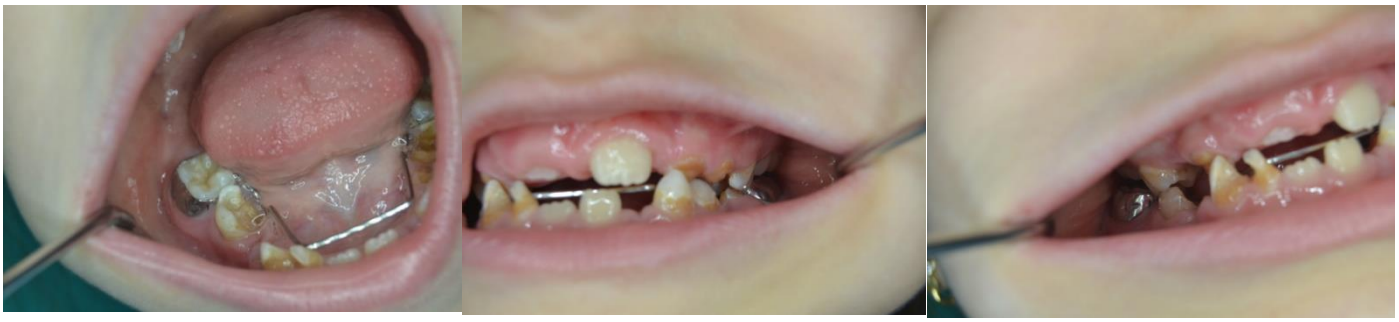
A 6-year-old girl was referred for specialist consult at the University's Pediatric Clinic, regarding multiple tooth lesions, tooth ache when eating and malocclusion.

Investigation of the medical history revealed that the patient was diagnosed at an early age with celiac disease. The parents were informed about the connection between the affected teeth and celiac disease and also about the need of a better oral hygiene, fluoride topical application and pits and fissures sealing. They were also informed about the increased risk of caries in coeliac patients





For the purpose of assessing dental enamel defects, Aine L. *et al.* (Table 1) classified the specific enamel defects in grades I–IV according to the severity of their clinical aspect.[27,28-30]



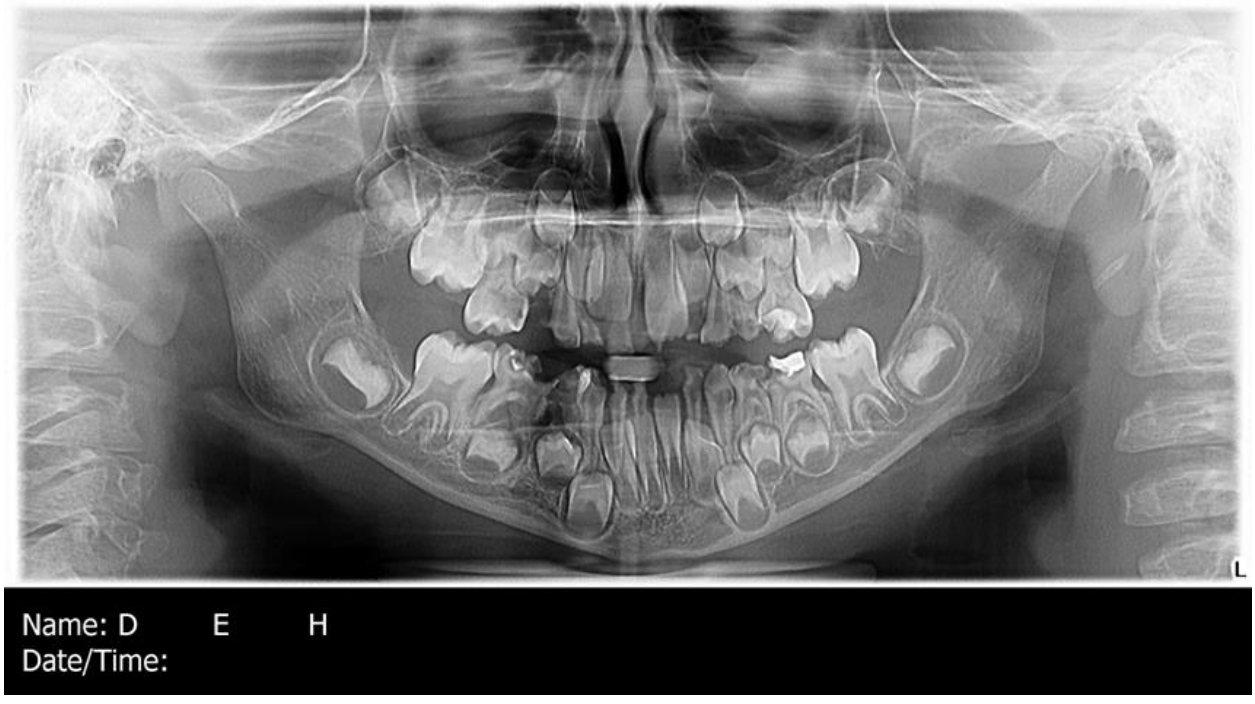


Table 1. Classification of dental enamel defects in coeliac disease according to Aine L. [27,28-30]

Grade 0	No Defects.
Grade I	Defect in enamel color. Single or multiple cream, yellow or brown opacities (marks) with clear or hazed boundary, part of the dental enamel may lack
Grade II	Slight structural enamel defects, rough surface with horizontal groves or pits, distortion of enamel color and transparency.
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 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 lesion may be strongly

RESULTS

At each dental visit, the status of hard tissues (enamel hypoplasia, dental caries) and soft tissues (recurrent aphthous stomatitis, atrophic glossitis, geographic tongue) was evaluated. The evaluation of a delay in dental change phases was performed through specific dental eruption tables and with a panoramic radiography. There were observed severe structural defects, dental enamel defects and multiple carious lesions. We didn't notice any other oral manifestations. There was no atrophic glossitis, delay in dental eruption or recurrent aphthous stomatitis as described in other cases. Also, it can be observed that some teeth were lost early: 54, 64 and others lost too much structure to be saved.

DISCUSSION

In previous studies performed upon enamel defects in CD, it has been demonstrated that enamel hypoplasia is more prevalent in coeliac patients as compared to healthy subjects [1-9].

Among coeliac patients, the enamel defects of deciduous teeth mostly occur in canines (45%) and second premolars (51%). In the permanent dentition, the teeth involved are central incisors (25%), lateral incisors (20%), first molars (24%), canines (9%), first premolars (8%), second

premolars (7%) and second molars (7%).[23]

A study has revealed the structural aspect of enamel defects of coeliac patients, both in deciduous and permanent teeth using scansion electron microscopy (SEM). The teeth were highly hypo-mineralized, with shorter prisms of the enamel, distributed irregularly and less interprismatic substance, compared to the enamel of non-coeliac subjects [31]. These defects, associated with the celiac disease, are explained by two etiopathogenic mechanisms: malabsorption-hypocalcemia and autoimmune response. The malabsorption due to the enteropathy determines an alteration of phospho-calcium metabolism and a consequent hypocalcemia [32,33]. Regarding the autoimmune response, the antigen, i.e. the gluten, binding to class II molecules of the major histocompatibility complex, produces an autoimmune response, mediated by lymphocytes, against the enamel organ through the release of anti-matrix antibodies. [33,4,35]

The high frequency of caries in coeliac patients is not a manifestation of celiac disease. The explanation could be the copresence of risk factors, like the fragility of hypoplastic enamel, alterations in salivary concentrations and reductions in salivary flow. The decrease of salivary flow, occurring in the active phase of disease and in concomitance with the gluten free diet, determines dryness/soreness of the mouth, soreness/burning sensation in the tongue [36], alterations of oral protective factors and it could increase the risk for oral mucosal infections and dental caries.

CONCLUSIONS

Because of the indirect effects the celiac disease has on the oral health, coeliac patients should be included in a preventive dental program that should be composed of professional oral hygiene, motivation-education for home oral hygiene, pits and fissures sealing, fluoride topical application. All caries or fractures of the hypoplastic enamel should be repaired.

Pediatric dentists should be aware of the high prevalence and of the fact that most of the patients are not diagnosed. Through the help of interdisciplinary collaborators, pediatric dentists have one of the most important roles in the management of the celiac disease

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