Enamel defects in children with coeliac disease

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ABSTRACT: Aim. This was to investigate the prevalence of enamel developmental defects in a group of children with a history of coeliac disease. Methods. A study group of children attending the Dept. Paediatrics (Leeds General Infirmary), born between 1985 and 1986 and subsequently diagnosed and treated for coeliac disease (CD) were recruited. A group of age/sex-matched children attending the Paediatric Dentistry department were used as a control group (Cont). Examinations were carried out for enamel defects and opacities (DDE index), dmf, dmfs, DMF and DMFS (BASCD method), and a full medical and dental history were obtained. Results. Significantly more children in the CD group had a greater number of enamel defects than controls for both primary (p=<0.02) and permanent (p=<0.001) dentitions. Opacities in both primary and permanent teeth were statistically significantly greater in the CD group than controls (p=<0.04 and p=<0.001 respectively). Dental caries in both primary and permanent dentitions was less in the CD group compared with the control group of children. Conclusion. Coeliac disease was associated with an increased prevalence of developmental enamel defects.

KEYWORDS: Enamel defects, Coeliac disease.

Introduction

Coeliac disease was first described by Gee in 1888 [quoted by Paveley, 1988], but an earlier description has been attributed to Aretaeus in the second century AD [Paveley, 1988]. Although there was awareness that coeliac disease was aggravated by the use of starch/bread, it was a Dutch paediatrician, Dicke, who eventually proved that it was wheat flour and the provoking substance gluten was responsible [Paveley, 1988]. The disease is defined as an immunological reaction with a morphological abnormality of the small intestine mucosa. According to the European Society for Paediatric Gastroenterology and Nutrition (ESPGAN) childhood coeliac disease is a permanent disorder with a prevalence varying between 1:500 and 1:3000 children [Auricchio et al., 1988].

Suckling and Thurley [1984] concluded that demarcated enamel opacities resulted from either a sudden severe insult to a discrete number of ameloblasts, during their maturation stage, or from a less severe, but longer lasting, disturbance during their secretory stage. These authors also concluded that diffuse opacities were the aftermath of a chronic, less severe insult during and/or post-secretary phase of the ameloblasts, so causing a delay in the completion of the maturation process. If, however, the cause was felt to be genetic, then enamel in both dentitions will be affected, opaque in colour and dull in appearance. Soon after tooth eruption, enamel that is affected by such disturbances will become discoloured and affected areas of enamel may wear excessively and in due course dentine may become exposed. If the cause is felt to be systemic, then hypocalcification may occur in a more or less belt-like appearance, depending on the period of time that the cause was affecting tooth development. The disturbance will usually present as symmetrical. Correspondingly, if the hypomineralisation is due to local factors then the distribution will not be symmetrical. Rather the pattern of distribution, within a mouth, will be random.

Calcification of human primary teeth begins at about 13-16 weeks of gestation for incisors, 15-18 weeks for canines and 14-24 weeks for molars [Lunt and Law, 1974]. Other workers have also looked at these calcification times but in broad terms with similar results. For permanent teeth calcification of the first permanent molar occurs just before birth [Christensen and Kraus, 1965].

Dental defects occurring with cases of coeliac disease were described by Smith and Miller [1979] and also by Rasmussen and Espelid [1980]. Aine et al. [1989] found that 69% of a group of adults with coeliac disease had enamel defects compared with 19% of a control group. Such enamel defects are...
symmetrically and chronologically distributed in a pattern that is strongly associated with time of onset and treatment of the condition [Maki et al., 1991].

Studies regarding the effects of various diseases, syndromes and premature birth on tooth development have been reported in the literature since the early studies of Logan and Kronfeld [1933] to the extensive studies of Seow et al., [1987, 1989] and Seow and Perham [1990]. Coeliac disease has also been implicated in the aetiology of enamel defects [Smith and Miller, 1979; Maki et al., 1991; Ventura and Martelossi, 1997]. However, not all researchers have found such a relationship [Andersson-Wenckert et al., 1984; Rasmusson and Eriksson, 2001]. Because of this difference in opinion of enamel defects and the availability of a population of British children with coeliac disease, this study was prompted whereby enamel defects were assessed for comparison with a normal control group.

**Materials and Methods**

The study populations were recruited from children attending the Leeds General Infirmary (LGI) and Leeds Dental Institute (LDI), Leeds, England. Children with a history of coeliac disease were identified through the Department of Paediatrics (LGI). Approval for the study was obtained from the Ethical Research Committee of the LGI.

**Inclusion criteria.** For inclusion in the study the following criteria were used:
- diagnosed as suffering from coeliac disease and attending LGI;
- all subjects had primary and/or permanent teeth present;
- no crown restorations present;
- no gross caries with damage to the structural appearance of the enamel or with rampant caries.

A control group of children were recruited from those attending the LDI department of Paediatric Dentistry and their inclusion criteria were:
- caucasian children born at full-term (gestation 38-40 weeks) at LGI and with no relevant medical history;
- all subjects had primary and/or permanent teeth present;
- no crown restorations present;
- no gross caries with damage to the structural appearance of the enamel or with rampant caries.

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**Dental examinations.** Each child was examined in the department of Paediatric Dentistry (LDI) in a dental surgery/operatory under standardised conditions, including a dental light. Dental caries was diagnosed and recorded by the BASCD method [Palmer et al., 1984] and enamel defects by the DDE Index [Clarkson and O’Mullane, 1989]. Teeth were dried for examination and plaque and debris cleaned away as necessary. No radiographs were taken.

A medical/dental history was taken for each child from the parents to ascertain past medical care, dental care and the use of fluoride as drops, tablets, toothpaste etc.

Prior to the dental examinations, the principal examiner (EF) was calibrated by one other author (KJT), for all parameters. Calibration was carried out on random patients attending the children’s clinic at LDI. Subsequently some 10% of study children were re-examined to determine the reliability of the examiner.

**Statistical methods.** All data was entered into an Elonex computer. Statistical tests were used to examine the null hypothesis that the percentage of affected teeth per child was equal for experimental CD when compared with controls. Intra-examiner reliability was examined using a reproducibility ratio. Differences between affected subjects in groups were assessed using Chi-square ($\chi^2$), with or without Yate’s correction and one degree of freedom. As the number of subjects was small, then Fisher’s exact probability test was used.

**Results**

The coeliac group consisted of 9 male (mean age 9.8 yrs) and ten female (mean age 10 yrs) children. In this group 13 subjects had primary teeth present and 15 permanent. The matched control group consisted of 9 male (mean age 10.0 yrs) and 10 female (mean age 9.8 yrs). All subjects, except two males and two females, had primary teeth present.

**Examiner reliability.** The intra-examiner reliability was found to 95% confidence for dental caries and 98% for DDE as affected surfaces and extent.

**Fluoride history.** None of the subjects reported any use of pre-natal fluoride supplementation. One male in the CD and two children (one male one female) in the Control group reported the post-natal use of fluoride supplements as drops or tablets. All subjects reported using fluoride toothpaste with mean ages of introduction being 14 mths CD and 16.5 mths for the control group. The amount of toothpaste used, which was mostly an adult paste, varied from a smear to the full length of the brush head but with no differences in distribution between groups.
Enamel defects. The results of the assessment for enamel defects, as opacities and hypoplasia, are shown in Table 1.

Dental caries. The distribution of dental caries by various parameters is shown in Table 2. Caries in both primary and permanent dentitions was less in the CD group compared with all other groups. The differences were highly statistically significant for primary and permanent teeth and surfaces (p=<0.001)

Discussion

The relationship of CD and enamel defects is based on the possible role of hypocalcaemia on the developing dentition(s). Rasmussen and Espelid [1980], and later authors [Smith and Miller, 1979; Maki et al., 1991], have described enamel defects of the permanent teeth of children suffering from CD. In the present study there was a significant difference (p=<0.006) between CD and control children for combined types of enamel defects in the primary dentition (92.3% CD versus 46.1% control). It was also true for just opacities (84.6% CD versus 46.1% control).

A speculation as to why this occurs can be formulated. As the first degree relatives of CD children have a higher prevalence of enamel defects [Maki et al., 1991] and as Shah et al. [1990] has previously reported a five-fold increase for inflammatory bowel disease in these people, it could be that there is problem with malabsorption that affects amelogenesis early, so that primary teeth develop defects. This contention is supported by the fact that intra-uterine calcification of first permanent molars in affected individuals occurs [Smith and Miller, 1979; Rasmussen and Espelid, 1980; Maki et al., 1991].

The present work seems to be the only study reporting on enamel defects of primary teeth in children with CD. Even though much of the primary dentition development occurs in utero, if the malabsorption does occur early in development so it is possible that the primary dentition can be affected. That the permanent dentition is affected by CD has been well established, but these teeth, with the exception of the cusps of the first permanent molars, calcify and mature post-natal when the condition is established and diagnosed. The CD group presented with a higher number of affected permanent teeth than with the controls (100% CD versus 38% controls, which was statistically significant (p=<0.001). This difference was also found for opacities (100% CD versus 23.8% controls, p=<0.001). This higher prevalence could be explained by the malnutrition that can occur with CD with consequent hypocalcaemia. Another possible factor that may be implicated is that of IgE mediated gluten sensitivity, as noted above [Robinson et al., 1992]. Hence it can be speculated that at the time of gluten introduction in the diet IgE could enter the maturing enamel and inhibit full maturation, thus giving rise to defects and opacities.

Comparison of our findings for permanent teeth in this study with those of others is difficult because not all studies were carried out in the same way. Thus, Smith and Miller [1979] and Rasmussen and Espelid [1980]
reported on hypoplastic defects only and in one and two subjects respectively. The study of Aine et al. [1989] used a much older population, with a greater chance that many of the study patients had suffered from CD before gluten was identified as the causative factor.

The higher prevalence of opacities in the CD group could be explained by malnutrition, when symptoms of CD first occur, with consequent hypocalcaemia. Another possible factor is the IgE mediated gluten sensitivity in these children. According to Robinson et al. [1992] albumin can enter the maturation process of the enamel and inhibit the full maturation (because ameloblasts cannot remove albumin). The latter event would give rise to opacities of the enamel. Surprisingly, the difference between the CD and its control group regarding hypoplastic defects was not significantly different. One possible explanation for this is individual variability of expression of the effect. Alternatively it could be that as CD is detected early these days there is less metabolic stress. Yet another possible explanation is the protective role of breast feeding on the onset and intensity of symptoms [Auricchio et al., 1988; Kelly et al., 1989]. Amongst other investigators Maki et al. [1991] have reported a changing pattern of CD presentation towards a milder onset of symptoms.

It is interesting that the prevalence of dental caries in the CD children was considerably lower than that of the control group. Prati et al. [1987] found that there was no difference in caries between a CD and normal control group. The finding of a significant difference in our study may be related to different socio-economic conditions between Italy and England. In addition, local general dental practitioners mostly referred the children comprising the control group in the Leeds study for treatment of high levels of dental caries. However, a comparison of the dental caries indices for the control group in this study were comparable to similar age groups surveyed in Leeds as part of the annual screening of school children for dental disease. The control group was therefore considered representative of the Leeds child population. It could be speculated that the significantly lower prevalence of dental caries in the coeliac group might well be related to the need for a carefully controlled diet.

**Conclusion**

Children suffering from coeliac disease had significantly more enamel defects of both primary and permanent teeth compared with a control group of age/sex matched children. Coeliac disease children also had a significantly lower prevalence of dental caries.

### References