Histology of primary incisor enamel in children with early onset celiac disease

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Abstract

The manifestations of celiac disease are a result of nutritional malabsorption. An early onset of such malabsorption may jeopardize the primary enamel which is not mineralized. Prepared sections of 10 primary incisors from 10 children with early onset celiac disease were examined using polarized light microscopy to determine if enamel defects were present. Study of the tooth sections dry in air and after water imbibition revealed a normal neonatal line. Furthermore, both prenatal and postnatal enamel were found to have normal pore volume distribution. The absence of enamel disturbances suggests normal nutritional absorption during the period of primary incisor enamel mineralization.

Celiac disease (CD) is a frequent cause of nutritional malabsorption in childhood, characterized by degenerative changes in small intestinal mucosa (Rasmussen and Espelid 1980; Lebenthal and Branski 1981). In Sweden, the incidence of CD is approximately 1:900 births (Berg and Lindberg 1979; Stenhammer and Johansson 1981). Gluten, a protein found in wheat, rye, barley, and oats, causes villous atrophy and the subsequent malabsorption in CD (Rasmussen and Espelid 1980; Lebenthal and Branski 1981). The classic clinical manifestations, including diarrhea, steatorrhea, abdominal distention, wasting of muscles, vomiting, irritability, and growth retardation, usually occur in the first 2 years of life (Lebenthal and Branski 1981; Andersson-Wenckert et al. 1984). However, growth failure alone may be the only demonstrable symptom (Verkasalo et al. 1978; Lebenthal and Branski 1981). A definitive diagnosis can be made only after histologic examination of 3 intestinal biopsies (Smith and Miller 1979; Rasmussen and Espelid 1980; Aine 1986). An enamel defect prevalence of 96% has been reported in CD children in Finland (Aine 1986). All investigations of enamel disturbances in subjects with CD have involved visual inspection in situ: only 2 teeth (permanent molars) have been examined histologically and reported (Smith and Miller 1979; Rasmussen and Espelid 1980). The early onset of CD symptoms indicates that primary tooth enamel may exhibit abnormalities; yet, to our knowledge, no histologic studies on primary teeth of children with confirmed CD are available. The purpose of this paper is to determine if histologic defects exist in the primary incisor enamel of children with early onset CD.

Materials and Methods

Exfoliated or extracted primary teeth were collected from 10 children with early onset of celiac disease residing in Umeå, Sweden. All children had a full-term, uneventful birth with normal presentation and weight. The sample included 4 maxillary central incisors, 4 maxillary lateral incisors, and 2 mandibular lateral incisors. The diagnostic criteria of 3 intestinal biopsies were met by all subjects. The table (next page) presents data concerning the children and their disease.

Sagittal buccolingual 80-100 μm-thick sections were prepared from teeth embedded in methylmethacrylate using a saw microtome (Leitz® 1600 low-speed saw microtome — Ernst Leitz Wetzler GmbH; Federal Republic of Germany; Norén and Engström 1987). After polishing, the sections were examined dry in air and after water imbibition in a polarized light microscope (Olympus BH® polarizing light microscope — Olympus Optical Co, Ltd; Japan) employing strain-free objectives.
Results

When examined dry in air the neonatal line could be seen as a distinct band with positive birefringence (Fig 1), thus distinguishing the prenatal enamel from the postnatal enamel. After water imbibition for 24 hr the neonatal line remained positively birefringent (Fig 2). The prenatal enamel appeared either pseudoisotropic or positively birefringent when examined dry in air (Fig 1) and changed to negative birefringence after water imbibition (Fig 2). The postnatal enamel appeared negatively birefringent dry in air and after water imbibition (Figs 1, 2). The overall degree of mineralization did not differ greatly between the prenatal and postnatal enamel.

A positive birefringent area in the postnatal enamel was commonly observed adjacent to the neonatal line. Water imbibition did not alter the positive birefringence of this area, although the apparent size of the porous lesion was reduced.

In a large proportion of the specimens (8), subsurface enamel defects were found in the cervical area of the postnatal enamel. The defects appeared as positively birefringent lesions beneath a negatively birefringent surface zone both in air and after water imbibition. These defects did not extend into the bulk of the postnatal enamel.

Discussion

Polarization microscopy offers a sensitive qualitative technique for investigating changes in the mineral content (pore volume distribution) of enamel (Theuns and Groeneveld 1977). Additionally, examination of tooth sections dry in air and after water imbibition allows estimation of enamel pore volume distribution (> < 5%; Theuns and Groeneveld 1977). The neonatal line provides a reasonably accurate demarcation between the prenatal and postnatal enamel of primary teeth (Norén 1983a, 1983b). Therefore, the chronology of metabolic disturbances reflected in primary enamel can be assessed to have occurred before or after birth. Defects in the prenatal or postnatal enamel of children with CD would indicate a period of malabsorption before or after birth, respectively (Nikiforuk and Fraser 1979).

The prenatal enamel in this study sample showed a mineralization pattern not unlike normal primary enamel (Norén 1983b). The postnatal enamel was also considered to have a normal degree of mineralization, although some of the sections showed a slight increase in porosity. These findings are in agreement with descriptions of the histomorphology of normal primary enamel (Norén 1983b). The relative absence of prenatal

![FIG 1 (left). Polared light photomicrograph of primary incisor enamel dry in air from a celiac child (subject 08). The prenatal enamel (A) appears positively birefringent, while the postnatal enamel (B) is negatively birefringent. The neonatal line (C) is seen as a positively birefringent band. Adjacent to the neonatal line is a positively birefringent area in the postnatal enamel (D).](image1)

![FIG 2 (right). Polared light photomicrograph of the same sample observed after water imbibition. Both prenatal enamel (A) and postnatal enamel (B) appear negatively birefringent; the neonatal line (C) remains positively birefringent. The postnatal enamel lesion adjacent to the neonatal line (D) continues to appear positively birefringent, despite its reduced area.](image2)
and postnatal enamel abnormalities in this small sample suggests normal absorption in CD patients during the time of primary incisor enamel formation. The presence or absence of enamel disturbances is presumably dependent on the timing of enamel formation and gluten introduction, as reports of clinically evident enamel defects in the primary teeth of children with CD exist (Smith and Miller 1979; Aine 1986). Primary incisor crowns are fully mineralized 3 months after birth (Lunt and Law 1974). Since only one subject (#08) in this study was younger than 3 months of age at the onset of CD symptoms, it is not surprising that we found an absence of incisor enamel defects.

These results support previous findings indicating that enamel mineralization disturbances secondary to CD do not occur before a period of gluten intake coinciding with enamel mineralization. Since gluten is usually introduced (and CD symptoms usually appear) within the first year of life (Walker-Smith 1979), primary canines and molars appear to be more susceptible to enamel abnormalities than incisors as they are mineralizing from shortly after birth until 11 months of age (Lunt and Law 1974). Smith and Miller (1979) reported a case of hypoplastic primary second molars in a 6 1/2-year-old boy with CD. Aine (1986), however, found clinically evident enamel defects in the primary molars of only 2 of 4 CD children with primary tooth defects. We are now examining histologic sections of primary canines and molars (in addition to a larger number of incisors) from CD patients to determine the frequency of enamel abnormalities in primary teeth of children with CD.

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