Microscopic structure of deciduous teeth from patients with hypophosphatemic x-linked rickets

Klinge RF, Risnes S, Li CF, Skogedal N and Storhaug K

Department of Oral Biology, Faculty of Dentistry, University of Oslo and National Resource Centre for Oral Health in Rare Medical Conditions (TAKO-centre), Lovisenberg Diakonale Hospital, Oslo, Norway

Introduction

Irregularities in microscopic structure, particularly in the dentin, have been demonstrated in teeth from patients with hypophosphatemic rickets (e.g. Chaussain-Miller et al., 2003). The aim of this study was to further investigate the structure of teeth from patients with this disease.

Materials and methods.

The material consisted of 10 deciduous teeth from 4 patients. In 3 of the patients the diagnosis was made and treatment started around 2 years of age and in 1 patient diagnosis was made and treatment started at 6 months of age. Two teeth were extracted for orthodontic reasons, while 8 teeth were extracted due to pulp necrosis and/or abscess formation. A central longitudinal ground section was cut for light microscopy and microradiography. Of the remaining mesial and distal parts, one was prepared for scanning electron microscopy (etched 30 sec in 1% nitric acid and coated with gold-palladium), while the other was decalcified and processed for light microscopy.

Results

The morphology of the teeth appeared normal. Opaque patches in the enamel were quite frequent. The structure of the enamel was generally normal. Mineralization was more variable as reflected in the presence of coated crystals with variable size.

The mantle dentin had a normal degree of mineralization, but the width varied. Particularly in the boys the circumpulpal dentin had an irregular mineralization pattern. Close to the pulp a more normal mineralization pattern was seen. The root dentin also had an irregular structure, both in the mineralization pattern and in the structure of the organic matrix in some teeth. The cementum layer seemed to be thinner than normal, and was mostly unaffected. For further details see fig. texts.

Figs. 1 and 2. Microradiographs of 73 (left) and 63 (right) from the same patient. 73 was extracted for orthodontic reasons, while 63 was extracted due to pulp necrosis when the boy was 9 -10 years of age. Hypophosphatemic rickets was diagnosed at 2 years of age and treatment started immediately. 73 shows a normal degree of mineralization peripherally. Further pulpally a wide zone of interglobular dentin can be noted, followed by a zone of more uniform mineral content. In the cuspal area enamel and dentin are abraded, exposing the peripheral dentin to the oral cavity. From the pulp horn a narrow canal extending almost through the entire dentin in the plane of section can be noted (arrow).
Apart from a more narrow or absent layer of unaffected dentin peripherally in the crown, 63 shows the same mineralization pattern as 73 in the crown, as well as in the root. Enamel (E).

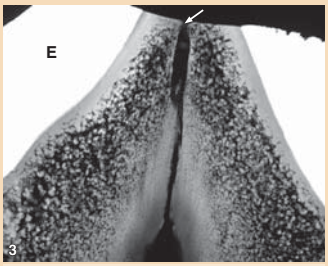


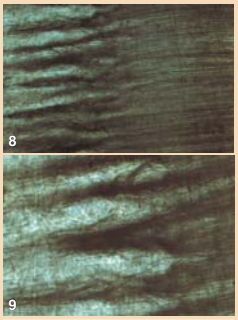
Fig. 3. Microradiograph of 55 extracted due to pulp necrosis when the boy was 10 years of age. Hypophosphatemic rickets was diagnosed at 2 years of age and treatment started immediately.
A mineralization pattern similar to that observed in figs.1 and 2 can be noted. This tooth also shows abrasion with exposure of the dentin to the oral environment. The extension of the pulp horn reaches the oral cavity in the plane of section (arrow). Enamel (E).

Fig. 4. Microradiograph of 61 extracted due to pulp necrosis and abscess formation when the girl was 4 years of age. Hypophosphatemic rickets diagnosed at 2 years of age, and treatment started immediately. Canal extending from the pulp horn (arrow) penetrates the dentin and results in communication with the oral cavity. A few interglobular areas are seen in the crown dentin. The root dentin is shown in fig. 7. Tertiary dentin has formed in the crown pulp. Linguocervically the superficial enamel appears hypomineralized.

Fig. 5. Microradiograph of 82 which was extracted due to pulp necrosis when the boy was 2 years of age. Hypophosphatemic rickets diagnosed right after birth and treatment started 6 months of age. The peripheral dentin seems unaffected, while numerous interglobular areas are seen in the circumpulpal dentin and in the root dentin. Extension of the pulp horn (arrow).

Fig. 6. Microradiograph of 51 extracted due to pulp necrosis when the girl was 6 years old. Hypophosphatemic rickets diagnosed at 2 years of age and treatment started immediately. The tooth was extracted due to pulp necrosis at the age of 6. The dentin has a uniform mineralization except for a narrow zone pulpally which shows numerous small interglobular areas. Root dentin from this tooth is shown in figs. 8 and 9.

Fig. 7. Micrograph of ground section from the middle part of the root of 61 photographed in transmitted light. The section is from the same tooth as shown in fig. 4. In the outer part of the dentin the tubules have a regular course and a few interglobular areas can be noted. In the inner part the course of the tubules is more irregular and numerous large interglobular areas are seen.



Figs. 8 and 9. Micrographs of ground section from the middle part of the root of 51 photographed in transmitted light. This section is from the same tooth as shown in fig. 6. In the outer half of the dentin (right) the tubules have a regular course, while in the inner half the tubules have an irregular course and appear be collected in bundles. Only a few interglobular areas are seen. The changes are most pronounced in the apical half of the root. Fig. 9 shows the transition between tubules with a regular and irregular course at higher magnification.

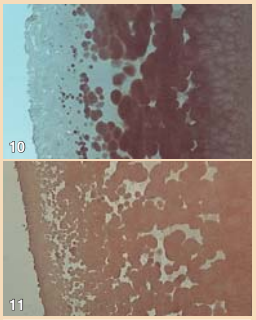


Fig. 10 and 11 Photomicrographs of H/E stained sections from the roots of 52 and 72. The teeth were extracted when the boy was 1½ years of age due to pulp necrosis and abscess formation and are from the same patient as the tooth shown in fig. 5. The peripheral dentin has a normal morphology, while interglobular dentin with numerous calcified globules with decreasing size can be seen more pulpally. A wide zone of predentin with irregular structure can be noted in fig.10.
In fig. 11 a narrow zone of dentin with a normal mineralization pattern, but a slightly more irregular course of the dentinal tubules can be seen. In this specimen the predentin is absent.

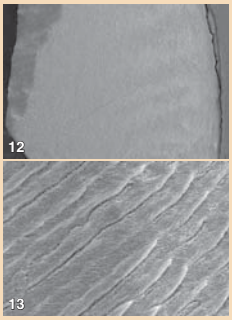


Fig. 12. Same tooth as in fig. 3 showing enamel with normal structure. Darker zone of less mineralized enamel in outer part. x 110.

Fig. 13. Higher magnification of fig. 12. Transition from less (darker, left) to more (lighter, right) mineralized enamel. x 2200.

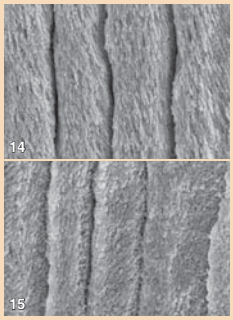


Fig. 14. Same tooth as in figs. 12 and 13. Showing prisms with normal to slighthly thicker crystals. x 5570.

Fig. 15. Same tooth as in fig. 2. Prisms with thick, coated crystals indicative of hypomineralization. x 5970.

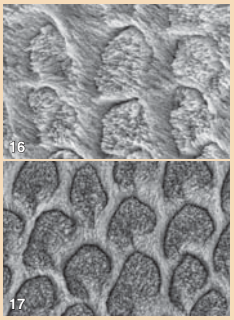


Fig. 16. Same tooth as fig. 15. Prisms with crystals of normal size. x 5440.

Fig. 17. Same tooth as figs 4 and 7. Prisms with slightly thicker crystals . x 5960.

Conclusion

The present observations indicate that the microscopic structure of the mineralized dental tissues is affected in hypophosphatemic rickets. The largest effect is seen in the mineralized phase of the dentin, but the organic phase may also be affected in some areas of the dentin. Our material is small, but variability in the degree of disturbances is evident. The observations also indicate thatt boys are more affected than girls.

Ref. Chaussain-Miller C, Sinding C, Wolikow M, Lasfargues J J and Garabédian M. (2003). J.Pediatrics. 142: 324-331.

Layout: J.U., Section for Photography, Faculty of Dentistry, University of Oslo