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STUDIES ON MINERALIZED DENTAL TISSUES

VIII. Histologic and microradiographic investigation of hereditary opalescent dentine.

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The expression *hereditary opalescent dentine* is used in the literature as a name for a certain disturbance in the development of the teeth characterized by an irregular structure of the dentine, with widely separated and curled dentinal tubules, by posteruptive obliteration of the pulpal cavity, by short roots, by a tendency to enamel fractures and pathological attrition, and furthermore by opalescence and colour changes in the crowns of the teeth. According to several investigators, hereditary opalescent dentine is a dominantly inherited disturbance of development but is not sex limited (*Noyes 1935, Finn 1938, Hodge et al. 1939, Roberts & Schour 1939, Lyons 1940*). *Roberts & Schour, 1939*, introduced the name *dentinogenesis imperfecta* for the dental changes listed above, and this term is sometimes also used to describe similar dental changes which occur concomitant with the disorder of the skeleton known as osteogenesis imperfecta. A biophysical study of the dental changes in the latter disorder has recently been published by *Bergman & Engfeldt (1954 b)*.

The histological changes occurring in hereditary opalescent dentine have been described previously by *Wilson & Steinbrecher 1929, Skillen 1937, Roberts & Schour 1939* and *Pindborg 1948*. X-ray absorption studies of the dentine from such teeth have

been performed by *Mc Cauley* 1942. He demonstrated that the x-ray absorption was less in diseased dentine than that in dentine taken from healthy teeth. According to *Hodge et al.* 1936, the water content of hereditary opalescent dentine is higher, while the amount of inorganic substances is lower than normal.

In the present investigation we have studied the morphological changes in teeth from material which was clinically diagnosed as hereditary opalescent dentine. We have also studied the amount and distribution of mineral salts in ground sections by means of microradiography. The findings have been compared with those from our earlier study of osteogenesis imperfecta.

MATERIAL

The material consisted of 38 teeth, all but 3 of which were deciduous teeth, from 10 children ranging in age from 3 to 10 years. The material was collected in the Eastman Institute of Stockholm during the years 1937—1950. All the teeth originated from patients in which hereditary opalescent dentine had been diagnosed. In nine of the ten cases there was a family history of similar dental changes. However, none of the patients had any physical or radiological evidence of skeletal disorders. A clinical description of the dental changes in these children will be given elsewhere.

METHODS

The teeth were fixed in 10 % formalin. Ground sections of a thickness between 30 and 150 μ were prepared from 17 teeth. The remaining 21 teeth were decalcified, embedded in paraffin, sectioned by a microtome, and the resultant sections were stained by Bock, Hansen, Azan and Mayer's haemalum-eosin. About 1 000 sections were studied.

The ground sections were used for microradiography, the technical details of which have been described by *Bergman & Engfeldt* 1954 a. A Machlett type OEG 50 x-ray tube served as the radiation source. The tube was energized with 24 kV and the radiation was filtered in 0.2 mm Be. The focus to emulsion distance was 10 cm. The microradiograms were registered on

Eastman Kodak Spectroscopic Plates No. 649, and were enlarged by photomicrography on Kodak 0 250 plates.

After microradiography, the ground sections were mounted with canada balsam on slides and were photomicrographed in transmitted light.

RESULTS

The dentine of all the teeth showed great deviations from the normal structure, but no pathological changes were found in the enamel.

Most of the primary dentine showed an irregular structure, with sparse and curled dentinal tubules except in a narrow zone immediately adjacent to the enamel and the root cementum (Fig. 1). This dentine, situated in the periphery ("mantle dentine"), as a rule had a lighter colour in haemalum-eosin stained preparations than had the circumpulpal dentine. The latter was often unevenly stained. In several cases, the irregular dentine exhibited a lamellar structure, which after haemalum-eosin staining showed up as thin blue lines separated by lighter areas (Fig. 2). Denticle-like bodies with onion-like structure were often seen. Sometimes these structures were enclosed in the dentine (Fig. 2) but in many cases they were observed in the pulp tissue (Fig. 3). A definite separation of the dentine into a thin layer of regular mantle dentine and a broader layer of irregular circumpulpal dentine was not observed in all teeth. Figure 4 shows that isolated streaks of regular dentine could be found in the region corresponding to the primary dentine of normal teeth while the surrounding tissue was irregular.

From most of the sections, one gained the impression that the odontoblastema had been quantitatively productive, but that the tissue produced was of inferior quality. With few exceptions the pulp cavity was definitely diminished by the encroachment of hard tissue which usually had the appearance of irregular dentine with curled dentinal tubules. In some cases, however, the tissue was morphologically more similar to bone because of the cellular elements enclosed in it (Fig. 5).

The structure of the pulpal tissue could only be studied in one tooth since it was necrotic in all the remaining teeth. The odonto-

blastema in this tooth clearly had become dedifferentiated, and the predentinal zone was unevenly wavy (Fig. 6). In this tooth, there was a pulpitis so it was not possible to exclude that local factors had caused the changes in the odontoblastema.

In connection with the narrowing of the pulpal cavity caused by the overproduction of irregular hard tissue there occurred spaces (Fig. 4) which were filled with necrotic tissue. Similar spaces or canals were also observed in the peripheral areas of the dentine (Figs. 7—10). Their appearance and location indicated that they had occurred during the early development of the tooth. Their main direction in general was parallel to the dentinal tubules (Fig. 9), although some deviations were observed (Fig. 8). These "canals", which showed features somewhat similar to those described for the dentine in osteogenesis imperfecta (Bergman & Engfeldt 1954 b), were sometimes surrounded by a thin zone of high mineral content (Figs. 9 and 10).

Fig. 1. Decalcified section of +03* (Pat. I. A.).

Resorbed deciduous tooth with a light peripheral zone and a dark, irregularly stained center. The light zone is regular, the dark zone irregular in structure. Htx-eosin. $\times 4$.

Fig. 2. Decalcified section of 03+ (Pat. I. G.).

Cusp tip showing lamellar structure of the dentine with built-in denticles. The white areas are artefacts. Htx-eosin. $\times 25$.

Fig. 3. Decalcified section of —04 (Pat. I. G.).

True adherent denticles. The right one is being built into the pulp wall by hard-substance-forming odontoblastema. Htx-eosin. $\times 40$.

Fig. 4. Decalcified section of —6 (Pat. L. A.).

The dentine is irregular except in the left cusp, where straight parallel dentinal tubules extend from the cusp tip to a point corresponding to the pulp horn of a normal tooth. Htx-eosin. $\times 2.5$.

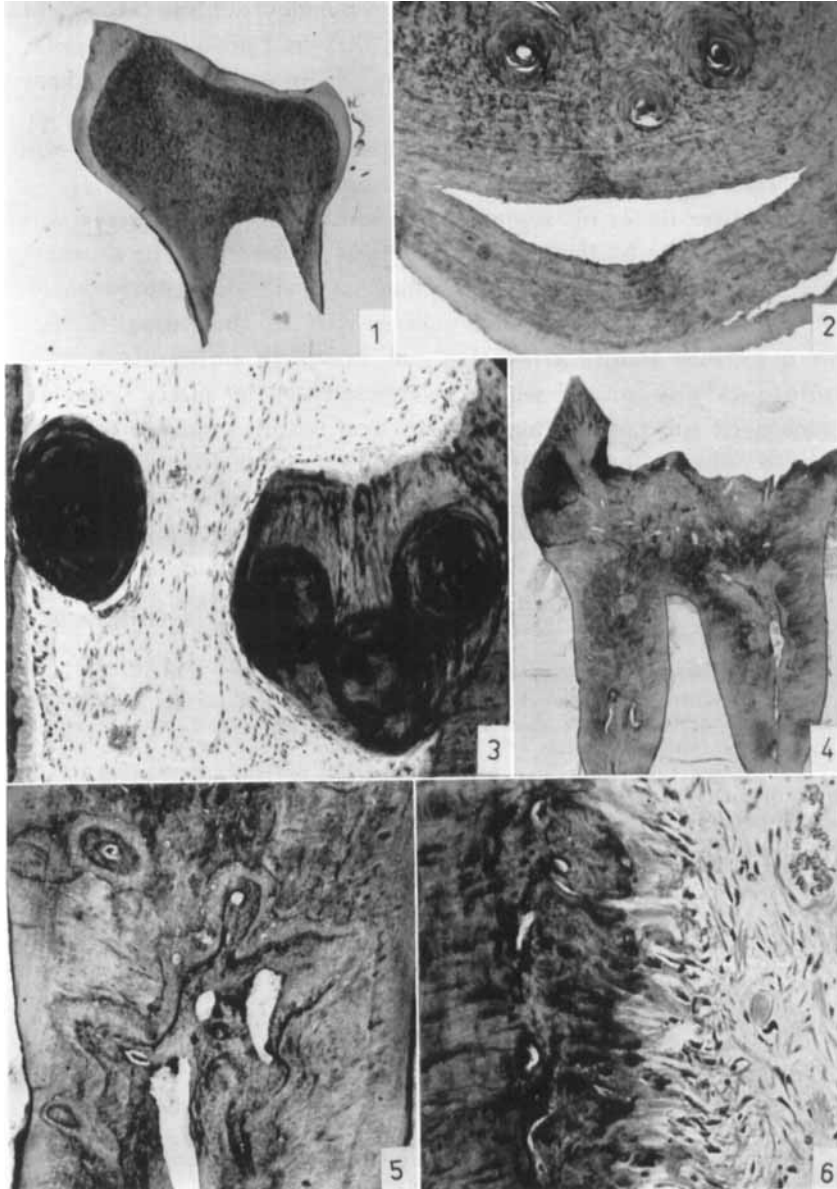
Fig. 5. Decalcified section of —6 (Pat. L. A.).

Root with traces of resorption and secondary apposition of bone-like tissue. Htx-eosin. $\times 40$.

Fig. 6. Decalcified section of —04 (Pat. I. G.).

Dedifferentiated, irregular odontoblastema. Predentine of inferior quality with uneven border. Htx-eosin. $\times 200$.

* In this paper the teeth are designated according to the *Haderup* nomenclature: + denotes the upper jaw, — the lower jaw. When the + or — is placed to the right of the tooth number, the right side is indicated and vice versa. 0 denotes a deciduous tooth. +03 thus means the left maxillary deciduous canine.



Figs. 1—6.

The structure of some of the sections suggested that the odontoblasts could start producing normal dentine (Figs. 11 and 12) after a period during which they had produced irregular dentine. The greater part of the dentine situated at the periphery was irregular (Fig. 11) and was unevenly mineralized (Fig. 12), while areas of the dentine situated next to the pulp cavity had a normal structure and were homogeneously mineralized.

A picture differing somewhat from that described above was observed in the tooth illustrated in Figs. 13 and 14. The development of the dentine in this tooth had apparently continued normally to the boundary which corresponds to the pulpal border in a normal tooth. After that, an unevenly mineralized hard substance was formed which was penetrated by many "canals". This hard substance revealed structural features similar to those of the pulpal bone which occurs in the teeth of scorbutic guinea pigs. Near the pulpal cavity this abnormal tissue was covered by a zone of regular dentine which was homogeneously mineralized. Thus a picture occurred which is reminiscent of the healing effect of vitamin C in experimental scurvy.

Fig. 7. Microradiogram of a 80μ ground section of 04— (Pat. B. K.)

Enamel with high x-ray absorption in the upper part of the picture. The main part of the picture shows dentine with "worm-eaten" appearance on account of spaces or canals with low absorption. $\times 50$.

The border line has a high x-ray absorption probably depending on the penetration of filling material into the dentine. Straight and curved spaces in the dentine. $\times 50$.

Fig. 8. Microradiogram of a 80μ ground section of 04— (Pat. B. K.).

Fig. 9. Microradiogram of a 100μ ground section of 02— (Pat. B. G.).

"Canals" with low x-ray absorption running parallel to the dentinal tubules. Mineral salt globuli with high x-ray absorption are visible in the borders of the "canals". $\times 100$.

Fig. 10. Microradiogram of a 100μ ground section of 04+ (Pat. B. G.).

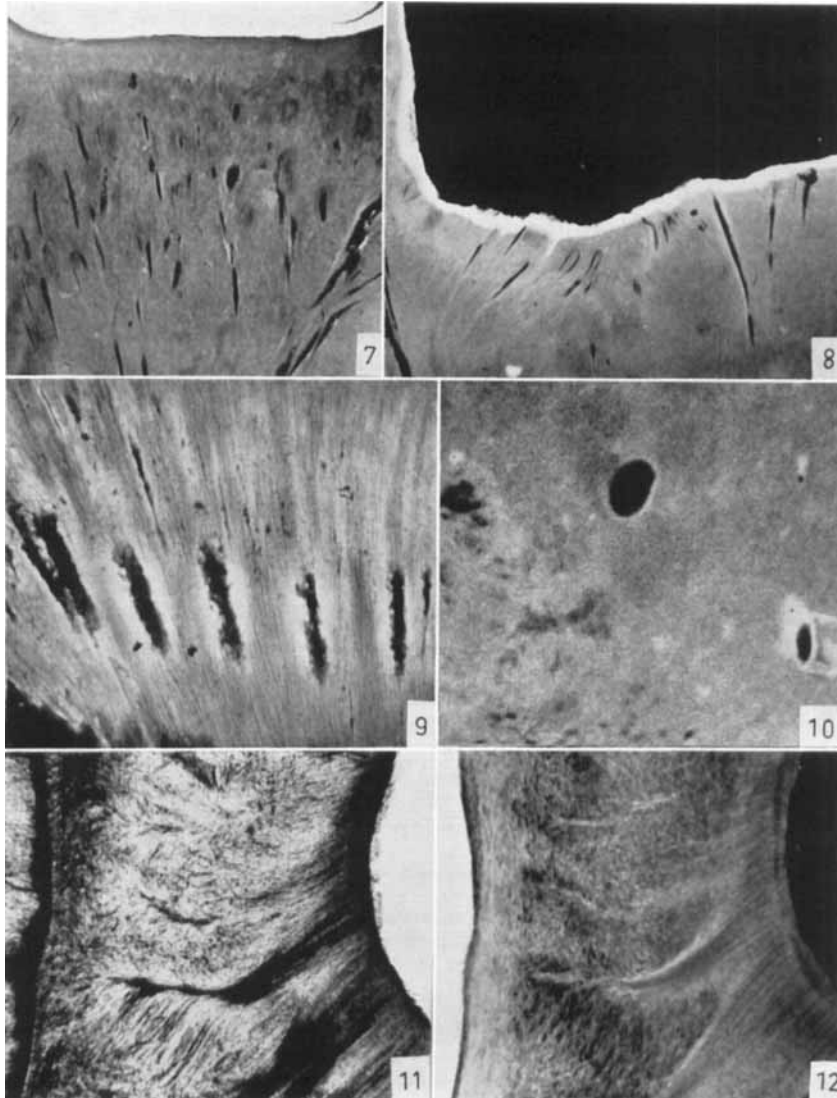
Two cross-cut coarse "canals", surrounded by a narrow zone of high x-ray absorption. $\times 200$.

Fig. 11. Ground, unstained section of 6— in transmitted light; 150μ thick. (Pat. I. G.)

Normal enamel in the left part of the picture. The main part of the dentine is irregular. Near the pulp the dentine is of normal structure. $\times 30$.

Fig. 12. Microradiogram of the ground section in Fig. 11.

The irregular dentine shows uneven, the regular dentine even mineralization. The narrow bands with low x-ray absorption near the pulp and at the dentino-enamel junction are normal features. $\times 30$.



Figs. 7—12.

As is seen in Figures 15 and 16 the dentine formed before birth showed a higher degree of mineralization than did the postnatal dentine. Although the postnatal dentine is regular in the areas illustrated, that in other parts of the same tooth exhibited irregular areas with uneven mineralization (Figs. 17 and 18). The microradiogram, Fig. 18, shows that areas with a low x-ray absorption correspond to the bundles of dentinal tubules shown in Fig. 17. However, the lowered x-ray absorption might not have depended solely on the fact that the dentinal tubules are gathered in bundles. A contributing factor might also have been that the bundles were observed in areas where the surrounding dentine possessed a low degree of mineralization (Fig. 20).

Anastomosing cells were often observed enclosed in the irregular dentine (Fig. 19).

DISCUSSION

It must be considered remarkable that only one vital pulp was found among the 21 decalcified teeth. A definite explanation of this fact can not be based on this material although there are two findings which might throw some light on the question. Thus, it is evident that the overproduction of hard substance which progressively reduces the pulpal cavity to a system of

Fig. 13. 100 μ unstained ground section of +02 in transmitted light (Pat. L. F.).

The primary dentine is normal in structure. The secondary dentine exhibits bone-like structure near the primary dentine and normal structure near the pulp. $\times 30$.

Fig. 14. Microradiogram of the section in Fig. 13.

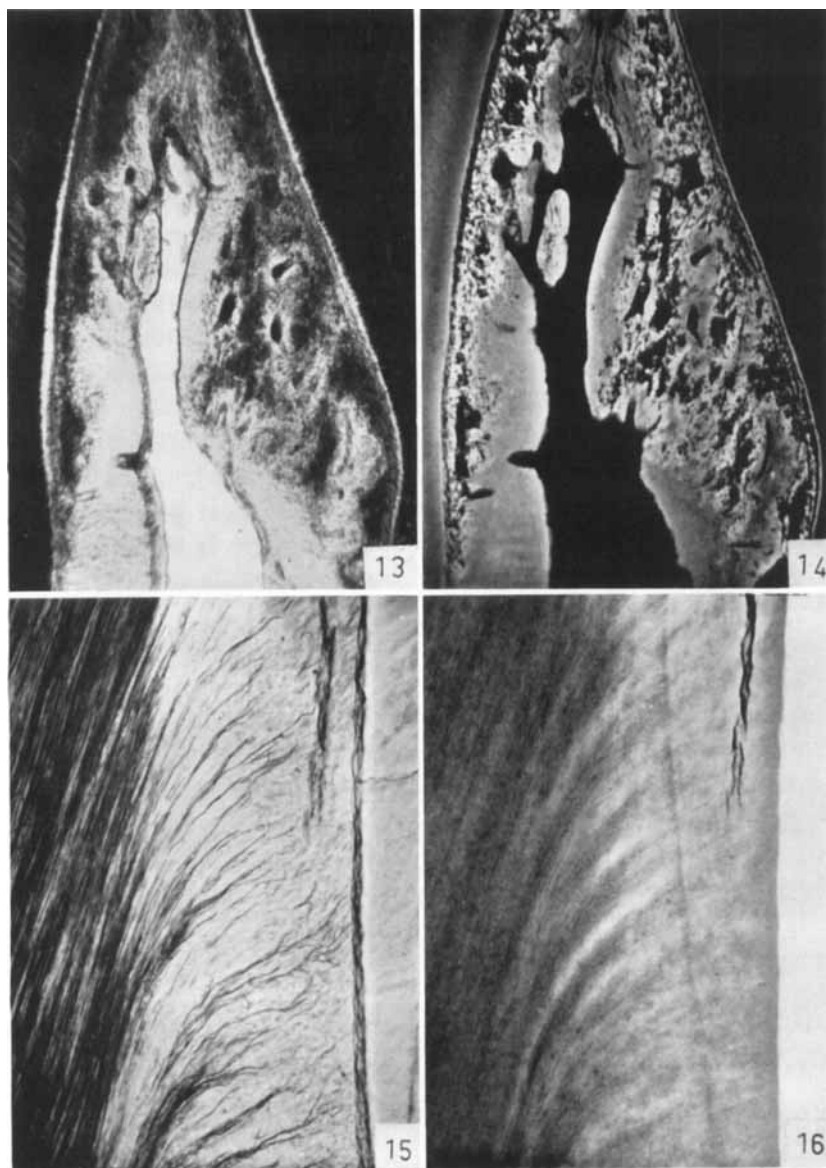
A narrow zone of high x-ray absorption separates the primary dentine from the bone-like, irregularly mineralized secondary dentine. The regular dentine, situated nearest the pulp, shows even mineralization. $\times 30$.

Fig. 15. 110 μ unstained ground section of -03 in transmitted light (Pat. B. G.).

To the right enamel, to the left dentine. The dentine is almost regular. $\times 100$.

Fig. 16. Microradiogram of the section in Fig. 15.

The neonatal line is visible as a thin line with a low mineral salt content. The prenatal dentine on the right side of the line has a high mineral salt content, the postnatal dentine shows lower mineralization. $\times 100$.



Figs. 13-16.

narrow spaces or canals must be followed by a disturbance in the blood circulation in the pulp. Moreover, this narrowing of the pulp cavity in combination with the frequently observed formation of denticles may in some cases have caused necrosis of the pulpal tissue. Another point which might have been of importance is the entry of infectious agents via the wide "canals" of the dentine. In a few sections it could be demonstrated that these "canals" may penetrate to a point immediately beneath the enamel. The characteristic tendency to enamel fractures, reported earlier, may in many cases lead to a direct connection between the pulp and the oral cavity through these rather wide "canals". Apical osteitis is reported to be rather common in patients with hereditary opalescent dentine. The reasons given above might at least partly explain the high frequency of such inflammations. When carious lesions exist in the dentine, even when the lesions are only superficial, a pulpal involvement is most probably present, and thus the prognosis for filling therapy is bad. Our material suggests that extraction of the damaged tooth is often to be preferred in hereditary opalescent dentine rather than conservative treatment.

Nothing definite can be said from our material concerning the mechanism underlying the appearance of the wide "canals" in hereditary opalescent dentine. The canals might have been formed through pulpal vessels which have been built into the dentine during its formation. However, it has not been possible to identify the necrotic material in the wide canals with certainty.

Earlier investigations have established that hereditary opalescent dentine is genetically determined. One might then expect

Fig. 17. Another part of the same ground section as in Fig. 15 in transmitted light. $\times 100$.

Fig. 18. Microradiogram of the ground section in Fig 17.

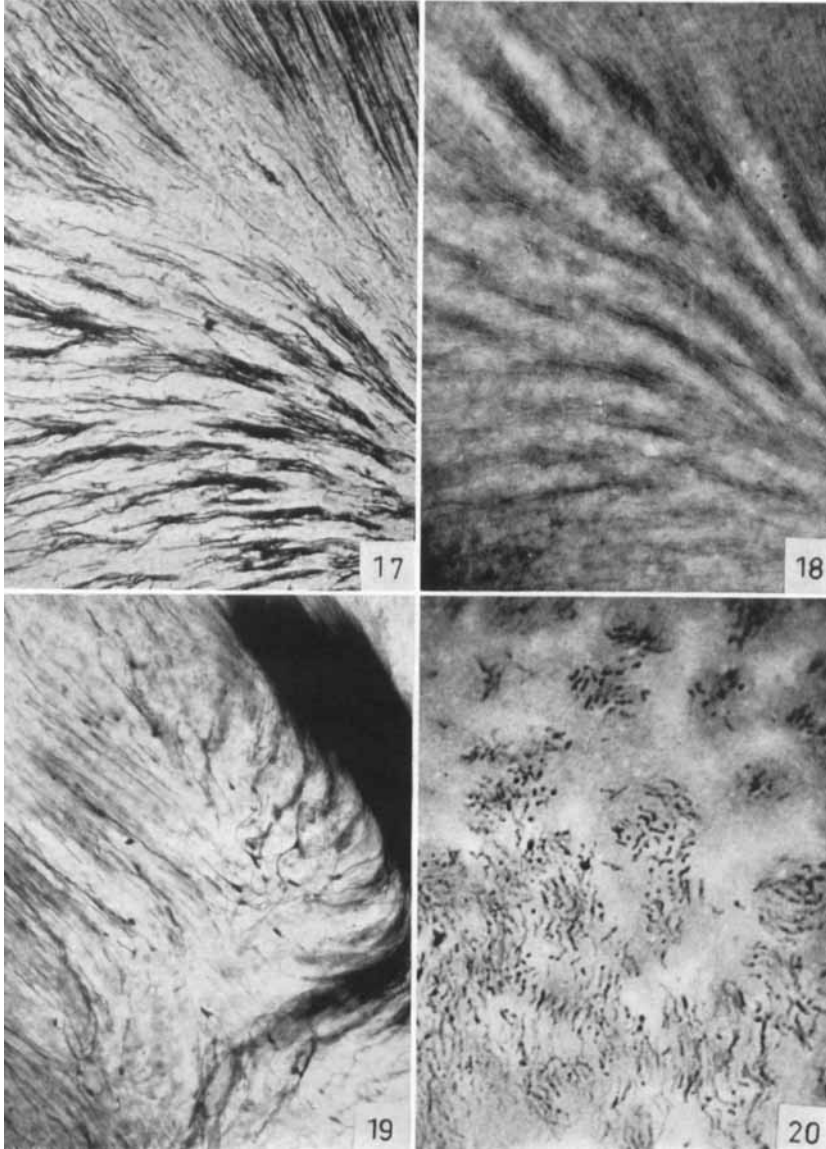
Areas with low x-ray absorption correspond to regions where dentinal tubules are abundant. $\times 30$.

Fig. 19. Detail of the section, shown in Fig. 11.

Irregular dentine with enclosed anastomosing cells. $\times 100$.

Fig. 20. Microradiogram of a 100μ ground section (Pat. C. K.).

Cross-cut dentinal tubules from irregular dentine. The tubules are collected in bundles, which in part run through areas of the dentine, where the ground substance has a low mineralization. $\times 200$.



Figs. 17-20.

the changes of the dentine to be identical in all teeth which develop in the jaws at the same time. Our material shows, however, that differences occur between different teeth in the same patient. These differences are difficult to explain, but possibly may reflect the local action of factors such as disturbances in blood circulation or local infections.

Another finding which seems to be difficult to explain is the fact that in the same tooth the dentine may show areas extending from the enamel to the pulp which are structurally normal while adjacent areas may exhibit completely irregular dentine. Especially in the cusp tips one often finds such isolated areas of regular dentine. Also perplexing is the fact that the dentine is irregularly formed up to a certain stage of development and after that time is normal. Possibly there may be a change in the mechanism which governs the function of the odontoblasts.

The dentinal changes in hereditary opalescent dentine reveal certain similarities to those occurring in osteogenesis imperfecta, and also to those in scurvy. Although it is rather clear that hereditary opalescent dentine is not primarily caused by vitamin C deficiency one cannot, however, exclude the possibility that such a deficiency might have modified the dentinal changes in certain cases. If this is true it may give us one explanation of the differences in dentinal changes between different patients, which are frequently extensive.

Although it might be expected that reaction to the damaging factor would show great variations from case to case, the question arises as to whether the changes observed in this material always are in response to the same factor. The variations in response observed in this study suggest the possibility that the dental changes which are classified clinically as hereditary opalescent dentine may actually represent more than one etiological factor.

SUMMARY

A total of 38 deciduous and permanent teeth from 10 children with the clinical diagnosis of hereditary opalescent dentine have been investigated with histologic and microradiographic techniques.

The enamel was normal, but the dentine showed pronounced pathological changes in all of the teeth. It was more or less irregular, and wide canals, different in character from the dentinal tubules, crossed the dentine. These wide canals very likely act as pathways for the entry of pulp infections. In the periphery of these canals there was frequently a high content of mineral salts. The pulp was mostly necrotic.

Microradiographic investigation showed that the dentine often was irregularly mineralized.

The histological changes in the dentine from different patients showed great variations. These variations suggest the possibility that the dental changes, which are classified clinically as hereditary opalescent dentine may represent more than one etiological factor.

RÉSUMÉ

ÉTUDES SUR LES TISSUS DENTAIRES MINÉRALISÉS

VIII. Observations histologiques et micro-radiographiques de la dentine opalescente du type héréditaire

Ont été étudiées suivant les techniques histologique et micro-radiographique 38 dents de lait et dents définitives au total, prélevées à 10 enfants chez qui avait été prononcé le diagnostic de dentine opalescente héréditaire.

L'émail était normal, mais la dentine présentait dans tous les cas des modifications pathologiques prononcées. Elle était plus ou moins irrégulière, traversée de canaux larges, nettement différents des tubuli de l'ivoire. Ces canaux permettent vraisemblablement le passage des infections vers la pulpe. A leur périphérie, on trouvait fréquemment un dépôt important de sels minéraux. La pulpe était généralement nécrosée.

Les observations micro-radiographiques ont montré que la dentine présentait souvent une minéralisation irrégulière.

Les modifications histologiques de la dentine variant considérablement d'un sujet à l'autre, on peut supposer que les modifications classées cliniquement sous la dénomination de dentine opalescente héréditaire sont attribuables à plus d'un facteur étiologique.

ZUSAMMENFASSUNG

STUDIEN AN MINERALISIERTEN ZAHNGEWEBEN

VIII. Eine histologische und mikroradiographische Untersuchung von „hereditary opalescent dentine“

38 Milchzähne und permanente Zähne von 10 Kindern mit der klinischen Diagnose "hereditary opalescent dentine" wurden histologisch und mikroradiographisch untersucht. Bei allen Zähnen war der Schmelz normal, während sich im Dentin ausgeprägte pathologische Veränderungen fanden. Das Dentin war mehr oder weniger irregulär und durchsetzt von dicken Kanälen, deren Aussehen sich von dem der Dentinkanäle unterschied. Diese dicken Kanäle können sicher als Eingangspforten für Pulpainfektion fungieren. An der Peripherie der Kanäle war der Mineralgehalt oft erhöht. Die Pulpa war meist nekrotisch. Die mikroradiographische Untersuchung zeigte, dass das irreguläre Dentin unregelmässig mineralisiert war.

Im histologischen Bild des Dentins traten bei den einzelnen Personen erhebliche Variationen auf. Diese Variationen lassen eventuell darauf schliessen, dass die Zahnveränderungen, die klinisch als "hereditary opalescent dentine" bezeichnet werden, mehr als nur einen ätiologischen Faktor repräsentieren.

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