Root resorption and signs of repair in Papillon-Lefèvre syndrome. A case study

Stefan Rüdiger and Tord Berglundh

Departments of Periodontology, School of Dental Medicine, University of Würzburg, Germany
and Faculty of Odontology, Göteborgs University, Sweden

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The aim of this investigation was to describe some tooth-related histological features of prepubertal periodontitis. Teeth extracted during treatment of two Papillon-Lefèvre syndrome patients were processed by means of the sawing and grinding technique. Light microscopy examination revealed little or no cementum in the coronal parts of the roots. Resorptions of various depths (0.02 to 1.5 mm) and to various extents (affecting up to 1/3 of the root surface) were observed in the 5 investigated teeth. Some resorptive defects on 1 of the examined incisors showed signs of spontaneous repair. Extrinsic fibers were inserted into the new cellular intrinsic fiber cementum which had formed directly on the bottom of the defect. Intact acellular extrinsic fiber cementum was found where fibers were still attached. Here, the characteristic of pristine cementum, a hyaline layer of peripheral dentin, could be identified. If resorption was not present, the cementum did not show any signs of hypoplasia. Thus, histological features of prepubertal periodontitis in the current material were (i) areas of extensive resorption, (ii) signs of spontaneous repair, and (iii) healthy cementum. □ *Histology; Papillon-Lefèvre syndrome; prepubertal periodontitis; root resorption; spontaneous repair*

Stefan Rüdiger, Clinic for Periodontics, Public Dental Health Service, Medicinaregatan 12C/5, S-413 90 Göteborg, Sweden. Tel. +46 31 773 3719, fax. +46 31 773 3847, e-mail. stefan.rudiger@odontologi.gu.se

Apart from bacteriological and immunological factors, anatomical defects on the root surfaces might predispose to prepubertal (1) and juvenile (2) periodontitis. Cementum hypoplasia has been observed in permanent teeth of subjects suffering from prepubertal (3) and juvenile (4) periodontitis. This developmental defect was believed to characterize the disease pattern by facilitating bacterial penetration (4). Also, a reduced cemental thickness on permanent teeth has been reported in Papillon-Lefèvre syndrome (PLS) cases (5).

External root resorptions occur under conditions of inflammation in both apical and marginal periodontitis (6). As long as the root is covered by the periodontal ligament, resorptions are apt to undergo repair (7, 8). If the root is exposed due to pocket formation before repair occurs, however, the resorbed areas become irreversibly denuded and may enhance plaque retention (9–11). In PLS patients, early resorption of deciduous teeth was observed radiographically (12), histologically (13), and in one reported PLS case, resorption of cementum in a permanent tooth was detected (14).

The aim of the present investigation was to further describe the tooth-related histological features of prepubertal periodontitis on extracted teeth in patients with PLS.

Material and methods

Patients

Two siblings, a boy aged 11 and a girl aged 8, suffering from PLS were treated for periodontal disease (for details regarding the patient sample, see Rüdiger et al. (15)). The upper left first molar, the lower right lateral incisor, and the lower right first molar of the boy and the lower left lateral incisor and lower left central incisor of the girl were extracted.

Histologic preparation of extracted teeth

The extracted teeth were placed in 3% formalin for fixation and, following dehydration in serial steps of ethanol, embedded in T 7200 VLC (Kulzer & Co GmbH, Wehrheim/Ts., Germany). Ground sections, 30 μm thick, in the mesiodistal plane (except for the upper left first molar: buccolingual plane) and parallel to the long axis of the teeth, were produced using the sawing and grinding technique (16). Four to 5 sections per tooth, about 0.5 mm apart, were prepared, including the crown and entire root(s), except for the distobuccal root of the upper left first molar of the boy. The sections were stained in toluidine blue.

Histological analysis

The histological analysis was carried out in a Leitz DM-RDE microscope (Leica, Wetzlar, Germany) for transmitted light and, when indicated, interference contrast and polarized light combined with a FITC fluorescein filter was used. The sections were analyzed with respect to (i) the presence, size, and shape of root resorptions, (ii) root cementum morphology, and (iii) character of remaining

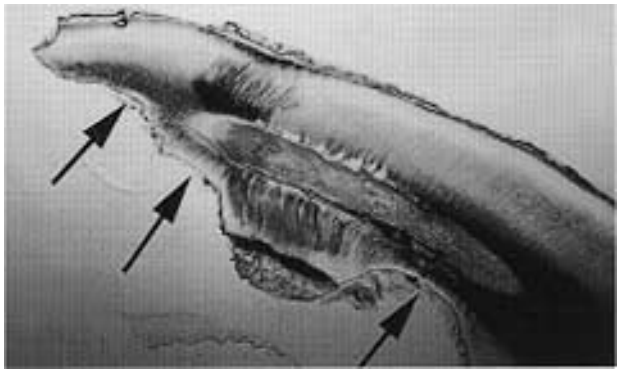

Fig. 1. Overview of the palatal root of the upper 1st molar showing extensive resorptions (*arrows*) on the interradicular aspect. In the apical 3rd of this surface, only a small area covered by cementum is left. Toluidine blue staining. Original magnification ×10.

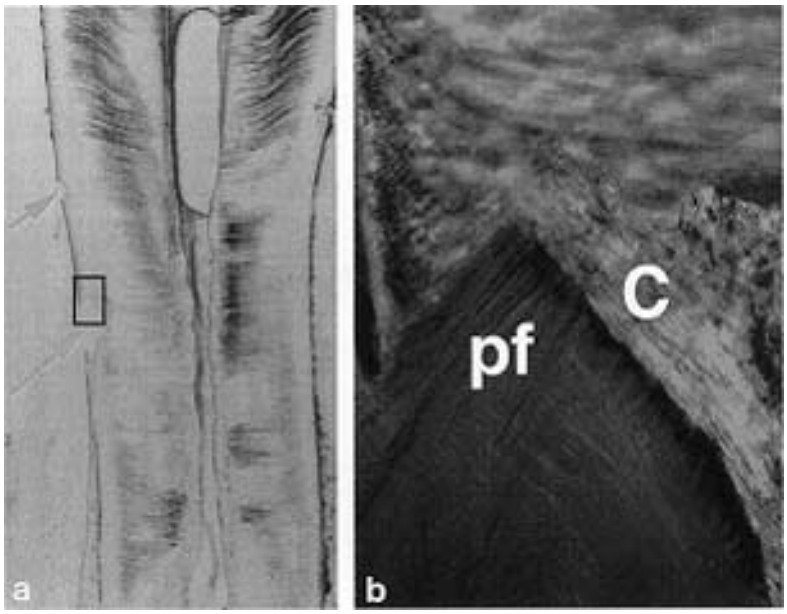

Fig. 2. Outlined area in (a) corresponds to (b). (a) Overview of the middle part of the root of an incisor. Resorption defects are indicated by arrows. Toluidine blue staining. Original magnification × 10. (b) One of the resorptive defects displays repair. Note cellular intrinsic and extrinsic fiber cementum (C), new periodontal fibers (pf) as well as a direct contact between newly formed cementum and remaining dentin. Toluidine blue staining: Original magnification ×200.

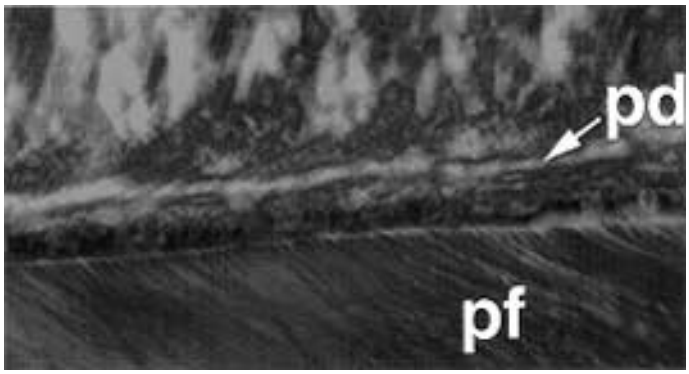

Fig. 3. Healthy root surface area in the apical 3rd of a lower diseased incisor. Periodontal fibers (pf) are still attached to the root surface. Acellular extrinsic fiber cementum covers the root surface. Note the bright hyaline layer of peripheral dentin (pd) between cementum and the granular layer of Tomes. Toluidine blue staining. Original magnification ×200.

Table 1. Histometric analysis of the root surfaces

Tooth	26	46	42	32	31
Mean % of resorbed/total root surface length	22	5	4	4	1
No. of resorption cavities/section	2 to 8	1 to 7	2 to 3	4 to 5	2 to 4
Resorption depth (mm) maximum	1.51	0.66	0.75	0.05	0.03
Resorption depth (mm) mean ± SD	0.65 ± 0.14	0.25 ± 0.16	0.26 ± 0.13	0.03 ± 0.01	0.03 ± 0.03

attachment on root surfaces. Thus, the following measurements were performed in all sections (Table 1):

- the mean percentage of resorbed areas of total root surface length
- number of resorption cavities determined as sharply delimited root surface defects
- maximum, mean, and standard deviation of the depth of the resorption cavities in mm

Results

A total of 20 histologic sections were examined. Plaque and calculus were found on all root surfaces and little or no cementum was left in the coronal parts of the roots.

Root resorption

Resorptions were observed in both incisors and molars, but occurred more frequently in molars. While all 9 sections from the 2 molars revealed resorptions, only 6 out of 11 sections of the 3 incisors showed this kind of defect. The number of resorptive defects per section varied between 0 and 5 for incisors, and between 1 and 8 for molars. The extent of the resorptive defects accounted for up to 27% of the total root surface on molars. The corresponding figure for incisors was 5%.

The most extensive and deepest resorptions were observed on the upper left first molar, especially in the furcation area. Up to one-third of the width of the root was resorbed (Fig. 1). All roots, except for the lower right lateral incisor, had resorptions on the apex. Resorption depths ranged between 0.02 mm in the incisors and 1.51 mm in the upper left 1st molar. In 3 out of 5 sections of the upper left 1st molar and in 1 out of 4 sections of the lower right 1st molar, the maximum resorption depth exceeded the maximum thickness of remaining cementum, i.e., extended into the dentin.

Repair

In 2 of the sections of the lower right lateral incisor, i.e. in 1 tooth out of 5, some resorption areas showed repair. The new cementum was in direct contact with the dentin and leveled out the irregular bottom of multilobular resorptive defects (Fig. 2). Extrinsic fibers inserted into the newly formed cementum which was of cellular, extrinsic, and intrinsic type. However, extrinsic fiber areas were rare. The cementum surface revealed a cementoid seam which was covered by a network of periodontal ligament fibers.

Healthy areas

In some root surface areas of the incisors, periodontal fibers were still attached to the root surface (Fig. 3). A very bright zone of peripheral dentin could be distinguished between the acellular extrinsic fiber cementum and the granular layer of Tomes. Fibers inserted directly into acellular extrinsic fiber cementum.

Discussion

The present histological investigation demonstrated that (i) wide and deep root resorptions appeared frequently in PLS patients, (ii) resorptive defects may undergo repair, and (iii) healthy cementum is present in unaffected areas.

Certain areas of the examined root surfaces of the present material were previously subjected to treatment, which included scaling and root planing (17). The histological findings must consequently be evaluated with care, in particular when judging signs of loss of root substance in the coronal portions. It may be anticipated that interradicular areas and areas displaying remaining attachment were not accessible to the performed non-surgical scaling procedures.

Root resorptions of various depth were found in the present material (Fig. 1). This observation is in agreement with previous reports on prepubertal periodontitis (11, 18–20). Schroeder & Rateitschak-Plüss (11) compared root surfaces of teeth extracted from 2 patients, 1 patient suffering from PLS and the other from rapidly progressive periodontitis. Resorption defects were only found in PLS. These findings (11) contradict data reported by López et al. (19), who could not demonstrate any difference concerning the prevalence of root resorptions between prepubertal periodontitis and other forms of periodontal disease.

The finding that resorptive areas may undergo repair is not new. Cemental repair in relation to prepubertal periodontitis was described by Sloan et al. (18) and Myers et al. (21). However, they reported on observations made on exfoliated primary incisors, but did not specify the type of newly formed cementum.

Cellular intrinsic fiber cementum grows more rapidly than acellular extrinsic fiber cementum (22), which might explain the predominance of intrinsic fibers in reparative cementum and the exclusive cellular character of the present material. Spontaneous reparative cementum as observed in our findings resembled therapeutically induced reparative cementum (23) in 2 ways. Firstly, extrinsic fibers inserted into the peripheral part of the new cellular cementum formed a network with intrinsic fibers. Secondly, peripheral dentin, a characteristic finding of pristine cementum (8), could not be observed.

In healthy areas of the present material, however, peripheral dentin was found. This hyaline layer between the granular layer of Tomes, a poorly calcified layer of dentin, and the cementum, has frequently been subjected to discussion because of its developmental origin. The lack of agreement on this matter is reflected in the unclear terminology of this anatomical structure, which is known as peripheral dentin, layer of Hopewell-Smith, and intermediate cementum (8).

In the present material, small cemental defects were observed. Developmental malformation reported to be a possible predisposing factor for early onset forms of periodontal disease, cannot be distinguished from resorptions using light microscopy (4). The observation of reparative cementum might provide indirect evidence that the root surface defects in the present material are resorption cavities.

Early onset periodontitis in the presence of PLS, as in this case, may be described as a very aggressive form of disease with an annual progression rate of up to 3 to 4 mm (15, 24). Resorptions, as observed in the present material, may be associated with periodontal breakdown. There is, however, no clear evidence of their etiologic role. It is suggested that root resorption is a conspicuous feature of early onset periodontitis in the presence of PLS.

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