Case Report

Progressive, Generalized, Apical Idiopathic Root Resorption and Hypercementosis

Marc Schätzle,* Sandro D. Tanner,† and Dieter D. Bosshardt*

Background: Root resorption is a multifactorial process that results in loss of tooth structure. The causes of root resorption may vary, leading to several types of resorptions. Some factors have been identified and may be categorized into physiological resorption, local factors, systemic conditions, and idiopathic resorptions. The objective of this report was to present a case of a 17-year-old white female with progressive, generalized, apical idiopathic root resorption followed up for 34 months.

Methods: Two panoramic radiographs, 14 and 34 months after initial clinical and radiological examinations, showed the rapid progression of apical root resorption. Two molars, teeth #15 and #16, which had to be extracted, and a bone sample from the distal aspect of tooth #15 were processed for histologic analysis.

Results: Two millimeters apical to the cemento-enamel junction, an abrupt increase in the cementum thickness was noted, amounting to 300 and 800 µm in teeth #15 and #16, respectively. The thickening of the cementum layer was due to an accelerated deposition of cellular intrinsic fiber cementum. An unusually high number of mineralization foci were observed in association with acellular extrinsic fiber cementum, and both free and fused cementicles were seen. In contrast to tooth #16, tooth #15 revealed extensive dentin replacement by a bone-like and a cementum-like tissue. Furthermore, ankylosis was demonstrated in tooth #15 and confirmed in the bone sample.

Conclusions: At present, there is no preventive or therapeutic regimen for the type of root resorption seen in this case report. Treatment usually consists of the extraction of teeth with advanced lesions. J Periodontol 2005;76:2002-2011.

KEY WORDS Analysis; case report; follow-up; progression; root resorption.

Root resorption can be described according to its location (internal or external), its distribution (localized or generalized), and affected dentition (primary or secondary dentition or both).

Internal root resorption of primary and secondary teeth has been reported to occur as a result of chronic inflammation/infection in the pulp, caries, trauma, pulpotomy, restorative procedures, orthodontic movement, and herpes zoster.1-5

External root resorption results from the activity of multinucleated clastic cells in the periodontal ligament. As regards bone, under normal conditions, there is a dynamic balance between osteoblastic and osteoclastic activities.6 An imbalance may result in external root resorption whereby cementum and dentin are removed.

In deciduous teeth, external root resorption normally occurs prior to the eruption of the permanent successors. Numerous other stimuli lead to external root resorption, such as reimplantation of teeth,6,7 periapical inflammation, tumors or cysts, excessive mechanical or occlusal forces,7 extreme orthodontic movements,8 tooth impactions,7 supernumerary teeth,9 and periodontitis.10

External root resorption has also been hypothesized to occur as a result of endocrine imbalances and systemic conditions (Table 111-22). In some cases, a familial pattern was noted.23-25

Root resorption can also occur without any determined intrinsic or extrinsic cause, and in these situations, the term idiopathic is applied. Consequently, idiopathic root resorption remains an appropriate term to be used when the condition exists without a known etiology.26

Two types of idiopathic external root resorption have been observed: cervical and apical. Cervical root resorption starts in the cervical area of the teeth and progresses toward the pulp. In the apical type, the resorption starts apically and progresses in the cervical direction whereby there is a rounding of the remaining root portion. Although permanent teeth are not usually thought of as undergoing root resorption, Henry and Weinmann27 found that most permanent teeth display microscopic evidence of external root resorption. Massler and Malone28 found radiographic

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evidence of root resorption in four or more teeth of all examined subjects. Minimal apical external root resorption may be present in all permanent teeth and has been attributed to a variety of causes. Extensive idiopathic external resorption is rarely reported. Only seven clearly identified cases of generalized idiopathic apical root resorption have been previously described (Table 2). Apparently, most of the recorded cases relate to young males with ages ranging from 14 to 35 years.

This report of a female patient with progressive, generalized, idiopathic external root resorption differs from previously reported cases. In contrast to most other reported case, this case is unusual in its pattern, progression, extent, and number of resorbed teeth, and the female patient could be followed up for 34 months.

**CASE DESCRIPTION**

A 17-year-old white female (born in October 1984) was referred to the Department of Maxillo-Facial Surgery of Lucerne for wisdom tooth extractions in March 2001. The patient provided informed consent to be profiled, and the study was conducted in accordance with the Helsinki Declaration of 1975, as revised in 2000.

Oral clinical examination yielded a full permanent dentition, the morphology and size of the crowns were normal, and the periodontal condition was unremarkable, with a general probing depth of ≤3 mm and a moderate degree of gingivitis. The tooth mobility was generally normal, although ankylosis was suspected in one of the molars. Pulp vitality as tested with carbon dioxide snow was normal.

A one-quarter Class III occlusion in the first molar intermaxillar relation and a Class I relation in the canine region was observed. The patient presented with an open and cross bite tendency in both lateral segments (Fig. 1).

At the initial clinical examinations, the orthopantomogram (Fig. 2A) revealed normal morphology and size of the crowns of all teeth and extensive root resorption of all permanent teeth in all quadrants. The maxillary molars were most affected, and the canines were least affected. Careful radiological examination of periapical regions showed resorptive cavities with replacement of the root by calcified tissue. All retained third molars appeared to be unaffected.

Her previous dental care involved preventive and restorative therapy. There was no history of trauma, infection, or tumor. There was no history of orthodontic treatment or radiation therapy. No possible local factors could be discerned from the radiographs. A systemic medical history of this patient was unremarkable, and laboratory hematological investigations of inflammation and endocrine markers were insignificant.

The possibility of heredity having a role in multiple root resorptions has been mentioned in the literature, but in this case possible genetic factors could not be ascertained. The patient’s father was examined radiologically and exhibited generalized chronic periodontitis with no signs of root resorptions.

The patient and her parents were informed that no etiology could be determined and that the external root resorption might be progressively worsening. No treatment could be recommended that would arrest the external root resorption, but several alternatives were discussed. As a precaution, the patient was encouraged to continue practicing meticulous oral hygiene. Whether a functioning dentition would persist with age and usage was not known.

In a follow-up examination, 14 months later in May 2002, the patient reported pain in the left maxilla. Reportedly, during the intervening time she had had no noticeable change in her general or medical condition. The pulp vitality tested with carbon dioxide snow showed some teeth with an abnormal response (Table 3). There was no change in her dental caries experience. Teeth #14 and #15 showed increased probing depth and a Class III furcation involvement. The mobility of all affected teeth was tested with an

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**Table 1.**

<table>
<thead>
<tr>
<th>Study</th>
<th>Endocrine or Systemic Imbalance</th>
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<tr>
<td>Sunde and Hals</td>
<td>Hypoparathyroidialism</td>
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<tr>
<td>Goultscin et al.</td>
<td>Hyperparathyroidialism</td>
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<tr>
<td>Tangney</td>
<td>Hypophosphatemia</td>
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<td>Eyring and Eisenberg</td>
<td>Hyperphosphatemia</td>
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<td>Smith</td>
<td>Paget's disease</td>
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<td>Bender and Bender</td>
<td>Gaucher's disease</td>
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<td>De Man</td>
<td>Stevens-Johnson syndrome</td>
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<td>Fayad and Steffensen</td>
<td>Parry-Romberg syndrome (hemifacialatrophy)</td>
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<td>Baxter et al.</td>
<td>Goltz syndrome</td>
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<td>Rudiger and Berglund</td>
<td>Papillon-Lefevre syndrome</td>
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<tr>
<td>Thoma et al.</td>
<td>Hereditary fibrous osteodysplasia</td>
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<td>Penido et al.</td>
<td>Anachoreosis</td>
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The new panoramic radiograph (Fig. 2B) showed that the resorptions had progressed significantly since the last visit. Further loss of root length had taken place in premolars and molars of both jaws. The pulps of the maxillary molars were partly replaced by calcified tissue. The painful teeth (#14 and #15) had to be removed along with tooth #16 due to lack of therapeutic alternatives. Teeth #15 and #16 were analyzed histologically. During the extraction, a bone sample

Table 2.
Case Reports of Idiopathic Apical Root Resorption

<table>
<thead>
<tr>
<th>Report</th>
<th>Year</th>
<th>Gender</th>
<th>Age (years)</th>
<th>Affected Teeth (#)</th>
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<tr>
<td>Rivera and Walton\textsuperscript{30}</td>
<td>1994</td>
<td>M</td>
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<td>-, 2, 3, 4, 5, 6, 7, 8</td>
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<tr>
<td>Counts and Widlak\textsuperscript{31}</td>
<td>1993</td>
<td>M</td>
<td>23</td>
<td>9, 10, 11, - 13, 14, -</td>
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<tr>
<td>Postlethwaite and Hamilton\textsuperscript{32}</td>
<td>1989</td>
<td>M</td>
<td>14</td>
<td>9, 10, 11, 12, 13, -</td>
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<td>Pankhurst et al\textsuperscript{29}</td>
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<td>Belanger and Coke\textsuperscript{26}</td>
<td>1985</td>
<td>M</td>
<td>14</td>
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<td>1981</td>
<td>M</td>
<td>27</td>
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<td>Soni and La Velle\textsuperscript{34}</td>
<td>1970</td>
<td>M</td>
<td>34</td>
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<td>Schätzle et al. (present report)</td>
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<td>17</td>
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<td>-, - 31, 30, 29, 28, 27, 26, 25</td>
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\(=\) no tooth.

**Figure 1.**
from the distal aspect of tooth #15 was removed and subjected to histologic analysis. An implant is contemplated as a replacement for tooth #14.

At the final examination in January 2004, 2 years and 10 months after the initial examination, the patient reported pain in the right maxilla. No change in her general or medical condition was reported. The pulp vitality of the remaining teeth showed no change compared to May 2002. However, at this time, teeth #2 and #3 showed increased probing depth and Class III furcation involvement.

The new panoramic radiograph (Fig. 2C) shows that the resorptions had progressed further and that the root length of all maxillary and mandibular molars had decreased substantially. In tooth #2, almost the whole coronal pulp chamber seems to be replaced by bone or bone like tissue. All impacted wisdom teeth also showed signs of root resorption. The removal of the painful teeth was planned.

Most of the teeth seem to have a poor clinical prognosis. It was anticipated that the remaining teeth would be extracted and replaced.

HISTOLOGIC ANALYSIS

Materials and Methods

Immediately following extraction, teeth #15 and #16 and a bony sample from the distal aspect of tooth #15 were fixed in 1% glutaraldehyde and 1% formaldehyde, buffered with 0.08 M sodium cacodylate (pH 7.3) for 24 hours at 4°C. After washing twice in 0.1 M sodium cacodylate buffer containing 5% sucrose and 0.05% CaCl₂, pH 7.3, the teeth were decalcified in 4.13% EDTA for 8 weeks at 4°C and extensively washed again in wash-buffer solution. The teeth were subdivided in a coronal-apical direction into numerous thin slices as described elsewhere. Some tissue samples were then post-fixed with potassium ferrocyanide-reduced osmium tetroxide, whereas others were left unosmicated. Osmicated samples were processed for embedding in epoxy resin and acrylic resin, whereas unosmicated samples were processed for embedding in the

Figure 2.
A) Orthopantomogram of subject at the age of 16 years and 6 months (March 2001). B) Orthopantomogram at the age of 17 years and 8 months (May 2002). C) Orthopantomogram at the age of 19 years and 4 months (January 2004).
acrylic resin only. Semithin sections (1 μm thick) were cut with a diamond knife on a microtome, stained with toluidine blue, and observed in a microscope.**

HISTOLOGIC RESULTS

*Tooth #16*

Tooth #16 had short roots and the apical foramen was open. At the advancing root edge, the Hertwig’s epithelial root sheath was poorly developed or not discernible. The odon- 

**blasts looked normal and a preden
tin layer was present (Fig. 3A). The cervical region of the root was covered by acellular extrinsic fiber cementum (AEFC). Numerous mineralization foci were present at its mineralization front, and the entire cementum layer appeared to consist of densely packed and coalesced mineralization foci (Figs. 3B through 3D). Cementicles in the periodontal ligament (Fig. 3B) and fused with the cementum layer (Fig. 3C) were observed. In addition, intensely stained and bizarrely shaped matrix patches were occasionally found in the periodontal ligament (Fig. 3D).

Two millimeters apical to the cemento-enamel junction (CEJ), an abrupt increase in thickness was observed in the cementum layer, i.e., the cementum thickness increased from 50 to 300 μm. The thick cementum layer was of the cellular type, i.e., cellular intrinsic fiber cementum (CIFC) (Fig. 3E). In the midroot region, a few resorption lesions were observed (Fig. 3F). They were partly repaired with CIFC. No signs of ongoing odontoclastic activity were detected, and ankylosis was absent.

*Tooth #15*

The roots of tooth #15 were almost totally resorbed. In a few areas around the cervical region, remnants of the roots were present. As in tooth #16, an abrupt increase in the thickness of the cementum layer occurred about 1.5 mm apical to the CEJ (Figs.

Table 3.
Tooth Vitality (May 2002)

| # Reichert Ultracut E, Leica Microsystems, Glattbrugg, Switzerland.   |

| **Leica Dialux 22 EB microscope, Leica Microsystems.** | **Table 4.**

Tooth Mobility (May 2002)
4A and 4B). The maximal thickness of the cementum layer amounted to 800 μm. Internal resorptive lesions were observed, and the dentin matrix was extensively replaced by bone-like (Fig. 4C) or a cementum-like tissues (Fig. 4D). Both replacement tissues were characterized by embedded cells, i.e., osteocytes or cementocytes, respectively. Although the bone-like tissue was characterized by osteons, the cementum-like tissue did not reveal a structural organization. Osteoclasts or odontoclasts were occasionally observed (Fig. 4E).

A bony particle harvested from the distal aspect of tooth #15 revealed a few dentin particles totally surrounded by bone matrix (Fig. 4F). Osteoclasts or odontoclasts were particularly observed in association with the dentin matrix (Fig. 4E).

DISCUSSION

This paper reports a case of idiopathic apical root resorption. The diagnosis was made by exclusion of local or systemic factors and diseases. In this case, there was no evidence of a possible etiological factor.

The reason for the open bite in both lateral segments and the increasing probing depth could only be speculated. The progression of the root resorption from the apical area occurred in combination with an apical migration of the junctional epithelium and/or a loss of collagen fibers inserting into the root cementum, which may have led to an increased probing depth.

Lateral tongue interposition due to early loss of deciduous teeth could be an
explanation for this open bite, but it might be also a result of idiopathic ankylosis and/or hypercementosis, which then led to the resorptive phenomena seen. The open bite in both lateral buccal segments may have caused excessive load on the incisors and canines. This could have resulted in resorption of the roots after the adaptive capacity of the periodontium was exceeded.8 In the present case, however, this circumstance may have had only a little impact on the resorptive processes due to the generalized involvement of the entire permanent dentition.

Histologic analysis of tooth #15 showed extensive resorption and ankylosis. In contrast, tooth #16 did not reveal histologic evidence of ankylosis. In contrast to tooth #16, tooth #15 had lost almost the entire root through root resorption. Replacement of the dentin by both a bone-like and a cementum-like tissue was a common finding in this tooth. The formation of repair cementum following root resorption is a common observation.43 The formation of bone, however, is less common and indicative of ankylosis.

Secondary retention is also a result of hypercementosis.44 But in contrast, Humerfelt and Reitan44 reported hypercementosis as parallel running incremental lines in the apical area. Ankylosis may occur between the alveolar bone and root surface following compression and atrophy of the periodontal ligament. If there were root resorption, the defects may be repaired by formation of cellular cementum, osteocementum, or bone.

The observation of small dentin particles surrounded by bone in the bone biopsy taken from the distal aspect of tooth #15 and the x-ray pictures support the assumption that tooth #15 was ankylosed. Ankylosis may occur after traumatic influences such as tooth luxations and retransplantations or autotransplantations7,8 or in association with hypercementosis.45-49

In the present case, the most striking histologic finding in both teeth was a thickening of the cementum layer about 2 mm apical to the cemento-enamel junction. The maximal thickness of cementum measured was 800 μm, which corresponds to about four times the normal width of the periodontal ligament. That the maximal thickness of cementum was greater in tooth #15 than in tooth #16 may be explained by the fact that the development of tooth #15 was more advanced. A cementum thickness in the range of 300 to 800 μm in the cervical root portion and at the developmental stages of the two examined teeth is extraordinary. Normally, the coronal half of the roots is covered with AEFC.50,51 This type of cementum grows very slowly and is thin in erupting or freshly erupted teeth.52 In the two teeth examined here, the increased thickness of the cementum layer was the result of an accelerated deposition of CIFC. This type of
cementum normally covers the apical half of the roots and the furcations. Therefore, we diagnose (or classify) the aberrant cementum formation as early generalized hypercementosis.

Possible factors causing hyperplastic cementum formation are tooth impaction or retention, chronic periapical inflammation, and developmental disturbances. There were no signs of inflammation observed in the periapical region of either tooth. However, ankylosis was observed.

Generalized hypercementosis may also be associated with general disease (e.g., osteitis deformans, acromemegaly, arthritis, rheumatic fever, or Paget’s disease) or occur in clinically healthy persons for yet unknown reasons. In the present case, it is very likely that there was an association between hypercementosis and ankylosis. What was the cause or effect of this association cannot be answered at the moment.

In this context, however, our findings in the periodontal ligament and the AEFC are of interest. The presence of cementicles in the periodontal ligament, although quite common in normal teeth, and the unusual high number of mineralization foci at the mineralization front and within the mineralized portion of AEFC suggest a disturbance in the regulation of mineralization. The periodontal ligament has a very important function in preventing mineralization and maintaining the periodontal ligament width. Therefore, developmental or functional disturbances and traumatic influences in the periodontal ligament have the potential to elicit the development of root resorption and ankylosis.

The periodontal ligament is a special soft connective tissue. It is the only ligament that spans between two distinct hard tissues, i.e., root cementum and alveolar bone. A remarkable feature of the periodontal ligament is the almost constant dimensions of its width throughout its lifetime. The integrity of the periodontal ligament is critical to the preservation of the normal functioning of the tooth. Thus, mechanisms exist that prevent mineralization of the periodontal ligament. The observed cementicles and the unusually high number of mineralization foci associated with the AEFC imply a disturbance in the regulation of mineralization within the periodontal ligament space. Furthermore, periodontal ligament cells are capable of regulating bone and cementum formation by producing paracrine factors that inhibit mineralized tissue resorption.

Although a number of treatment methods could be theoretically useful in trying to arrest this type of resorption while it occurs, none have been shown to be therapeutically effective.

Postlethwaite and Hamilton presented a case of a 14-year-old male teenager with multiple idiopathic resorptions involving all of the maxillary and mandibular incisors, canines, and premolar teeth. The pattern of resorption in that case, affecting only teeth with deciduous precursors, appears to be unique in the literature. They performed pulp extirpation and calcium hydroxide treatment on one half of the affected dentition for therapeutic study, possibly because the efforts were unsuccessful. Efforts to contact the authors failed.

The idiopathic apical root resorption does not seem to be mediated by or have its source internally from the pulp space. Tooth and bone resorption occurs through aberrant osteoclastic or odontoclastic activity in the periodontal ligament. Therefore, interceptive endodontic procedures including pulp removal and placement of calcium hydroxide or obturation with gutta-percha fail to address the target problem.

An experimental study in rats showed that the administration of very low doses of L-thyroxine during orthodontic tooth movement decreased the amount of root resorption by ~50%. The authors reported that the treatment with the hormone produced no new root resorption, no worsening of the existing resorption, and no adverse effects.

In two other animal studies, it was postulated that prostaglandin may be used as a root resorption inhibitor during orthodontic tooth movement. The amount of root resorption did increase with increased numbers and concentrations of prostaglandin injections. Also bisphosphonates, potent inhibitors of bone resorption, cause significant dose-dependent inhibition of root resorption in rats after orthodontic force application. But the administration of these potential root resorption inhibitors would rather temporarily inhibit the process than arrest it. Therefore, these possibilities were not considered. In addition, there is no clinical study performed in humans.

Although some potential factors (dietary habits and changes in the oral flora) have been suggested for idiopathic root resorptions, many have yet to be discovered. Thus, the future for the management or preventive therapies for idiopathic root resorption will depend on the identification of the specific cellular mechanisms that cause this disease.

At present, there is no preventive or therapeutic regimen for the type of root resorption observed in this case report. Treatment usually consists of the extraction of teeth with advanced lesions.

ACKNOWLEDGMENTS

The authors are indebted to Mrs. M. Aeberhard, Department of Periodontology and Fixed Prosthodontics, University of Berne, Switzerland, for excellent technical assistance. This study was supported by the Clinical Research Foundation (CRF) for the Promotion of Oral Health, University of Berne, Switzerland.
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Accepted for publication March 17, 2005.