Aggressive External Root Resorption of the Entire Dentition Accompanied by Osteolysis: A Case Report

Janicke Liaaen Jensen, DDS, PhD^a/Tore Solheim, DDS, PhD^b/Hanna Strømme Koppang, DDS, PhD^b/ Linda Zamoline Arvidsson, DDS, PhD^c

A healthy Caucasian male lost all of his teeth because of aggressive cervical, apical, and lateral root resorption over a period of 11 years. Microscopically, there was no sign of reparative cementum apposition, but cell-rich fibrous connective tissue filling the deep resorptions was seen. The resorbed teeth were surrounded by fibro-osseous tissue of osteolytic appearance, as noted during radiologic examination. Radiography showed the osteolytic processes to regress dramatically with the removal or loss of teeth. No underlying systemic or local factor was identified. In spite of endodontic and surgical treatment, none of the resorptions went into remission. *Int J Prosthodont 2012;25:459–464.*

Case Report

esorption of cervical or apical portions of the roots \mathbf{K} of one or more teeth may be seen after trauma, orthodontic treatment, or tooth bleaching or in relation to local or systemic pathologic conditions. If the cementum normally covering the root is resorbed or damaged, osteoclasts may initiate the process of root resorption. Developmental defects such as hypoplasia or hypomineralization of cementum have also been suggested as predisposing factors in root resorption. Resorption of an area of the root of a single tooth may be reversible or irreversible depending on the insult, and is not uncommon. In contrast, root resorption of multiple teeth or the entire dentition is exceedingly rare. Only a few cases of cervical or apical resorption of multiple teeth have been reported.¹⁻³ The etiology is unknown, and the condition has been found most frequently in otherwise healthy young female patients.1

An unusual case involving a combination of cervical, apical, and lateral root resorptions affecting the entire dentition and leading to complete edentulousness over a period of 11 years, in some instances resulting from almost total resorption of the roots, is described. The bone surrounding the most affected teeth showed extensive osteolysis, which regressed dramatically after loss or removal of the teeth. An otherwise healthy 54-year-old Caucasian man was first referred to the Faculty of Dentistry, University of Oslo, Oslo, Norway, in September 2001 because of swelling, pain, and aggressive root resorption of the molars in the mandibular left region. According to the patient, there was no family history of similar conditions. The root resorptions were confirmed by radiologic examination (Figs 1a to 1d). The surrounding bone showed osteolysis and slight expansion with thinning of the cortex using computed tomography scans (not shown). Retrospectively, these resorptions had been evident 2 years previously (1999). The mandibular left second molar was sacrificed, and the left first molar was hemisectioned. Histologic evaluation showed mild chronic osteomyelitis, and Dalacin (Pfeizer; 250 mg four times daily) was administered for 2 months.

Over the next couple of years, several teeth were lost or removed because of root resorptions (Figs 2 to 4). Consolidation of previously osteolytic bone occurred, but new areas of osteolysis were observed surrounding newly affected teeth. The patient was then lost to follow-up for some years but was referred again in 2009. According to the patient, the years in between had been uneventful. However, examination showed that several teeth had been removed. Figures 5a to 5f demonstrate the state of his dentition at that point. The mandibular right canine and first premolar as well as the left central incisor were removed as a result of extensive root resorption.

In September 2009, further resorptions at the maxillary anterior teeth and a possible cervical resorption of the mandibular right first molar were seen. In February 2010, the resorptions affected the entire

Volume 25. Number 5. 2012

459

^aProfessor, Department of Oral Surgery and Oral Medicine, Faculty of Dentistry, University of Oslo, Oslo, Norway.

^bProfessor, Department of Oral Biology, Faculty of Dentistry, University of Oslo, Oslo, Norway.

^cPostdoctoral Fellow, Department of Maxillofacial Radiology, Faculty of Dentistry, University of Oslo, Oslo, Norway.

Correspondence to: Dr Janicke Liaaen Jensen, Department of Oral Surgery and Oral Medicine, Faculty of Dentistry, University of Oslo, PO Box 1109, Blindern, 0317 Oslo, Norway. Fax: +4722852341. Email: jljensen@odont.uio.no



Figs 1a to 1d Panoramic and intraoral radiography at the initial consultation in September 2001 showed multiple extensive root resorptions in the maxillary right molar and mandibular left molar regions. The affected teeth were surrounded by osteolytic bone lesions.





Fig 2 Radiologic evaluation 6 months after the initial presentation showed further resorption of the maxillary right first molar and bone consolidation in the mandibular left molar region after removal of the teeth. The mandibular right second premolar also showed root resorption.

remaining set of teeth (Figs 6a and 6b). At that point, all maxillary incisors and the left canine were removed. The three mandibular teeth were spared as long as possible (Figs 7a to 7c), but were finally extracted in May 2011. A summary of the outcomes of the patient's dentition is given in Table 1.

Histology

At each surgical intervention, histologic evaluation of the teeth, soft tissue, and bone was performed. A summary of the microscopic investigations showed deep dentin resorptions bordered by normal-looking dentinoclasts. Most resorptions were associated with normal-looking mononuclear cells. Cell-rich connective tissue with only a few lymphocytes and plasma cells was seen in the resorptions, but there was no reparatory cementum (Fig 8).

The surrounding granulation tissue was heavily infiltrated with lymphocytes and plasma cells. Larger amounts of granulocytes were seen occasionally. This was interpreted to be a result of a secondary infection and not as an integral part of the process.

Specimens from areas adjacent to the teeth showed woven (immature) irregular bone trabeculae partly lined by osteoblasts in a fibrous connective tissue similar to that seen in the resorptions (Fig 9). Scattered lymphocytes and plasma cells were present but not sufficient to qualify for the diagnosis of chronic osteomyelitis. The fibro-osseous tissue bore

460 | The International Journal of Prosthodontics

© 2012 BY QUINTESSENCE PUBLISHING CO, INC. PRINTING OF THIS DOCUMENT IS RESTRICTED TO PERSONAL USE ONLY. NO PART MAY BE REPRODUCED OR TRANSMITTED IN ANY FORM WITHOUT WRITTEN PERMISSION FROM THE PUBLISHER.

Fig 3 Panoramic radiograph 1 year, 2 months after the initial consultation (2002). Note the extensive resorption of the mandibular right second premolar, left first premolar, and canine surrounded by osteolytic areas.

<image>

Figs 4a to 4d Intraoral radiographs 1 year, 10 months after the initial consultation (2003) showed **(a)** further resorption of the mandibular right second molar. **(b and c)** The mandibular right second molar showed internal and external root resorption with an adjacent osteolytic bone defect. **(d)** Cervical root resorption and an osteolytic defect were also noted apical to the mandibular left central incisor.

resemblance to that found in central ossifying fibroma and fibrous dysplasia.

General Considerations

The patient's postoperative course following the multiple surgical procedures was uneventful. Over the years, several medical evaluations were performed. Laboratory analyses included determination of values of ionized calcium, alkaline phosphatase, phosphorus, thyroid-stimulating hormone, creatinine, serum electrolytes, antinuclear antibodies, rheumatoid factor, liver enzymes, and complete blood cell counts. All findings were within normal limits.

Discussion

The present case, involving cervical, apical, and lateral root resorption associated with osteolytic changes and leading to loss of the complete dentition, is unusual. Some cases associated with high alkaline phosphatase levels, deafness, and pain have been

Volume 25. Number 5. 2012

461



Figs 5e and 5f Computed tomography scans of the mandible showed an osteolytic process with thinning of the cortex and mainly buccal expansion in the right canine area.



Figs 6a and 6b Panoramic and intraoral radiographs 8 years, 5 months after the initial consultation (2010) showed bone consolidation in the mandibular right premolar and canine areas. Root resorptions were noted on all remaining teeth.

462 The International Journal of Prosthodontics

© 2012 BY QUINTESSENCE PUBLISHING CO, INC. PRINTING OF THIS DOCUMENT IS RESTRICTED TO PERSONAL USE ONLY. NO PART MAY BE REPRODUCED OR TRANSMITTED IN ANY FORM WITHOUT WRITTEN PERMISSION FROM THE PUBLISHER.

Figs 7a to 7c Panoramic and computed tomography scans of the mandible 9 years, 7 months after the initial consultation (2011). Note the consolidation of bone in the right canine area and the osteolytic and slightly expanding area around the resorbed right molar in Figs 7b and 7c compared to Figs 5e and 5f.







reported.^{4,5} These diseases, ie, Paget disease, expansile skeletal hyperphosphatasia, and familiar expansile osteolysis, are associated with increased osteoclast function. No systemic factors were identified in this patient. Local factors contributing to root resorptions were also ruled out since he had no history of trauma, tooth bleaching, or orthodontic treatment. Histologic findings pointed toward a fibro-osseous bone disease. However, this was inconsistent with the normalization of bone after the loss or removal of teeth.

Possible therapeutic modalities for cases of multiple cervical resorptions were discussed in depth by Yu et al,⁶ including surgical exposure and restoration of affected sites, extraction, and submergence of affected roots. Early intervention with restoration of resorptive defects followed by progressive extraction and replacement with implant-supported prostheses was recommended. This case involved extensive root resorption that appeared to be not only cervical but also apical and lateral. Even though root canal treatment and restorative and surgical therapy were performed, the disease progressed and led to the gradual loss of the entire dentition. This has been a tremendous burden for the patient, who finds complete dentures hard to accept. A fixed implant-based dentition is likely to improve his quality of life. Since the osteolytic lesions of bone healed uneventfully after tooth loss or tooth removal, future implant treatment will most likely be successful, and the patient is therefore motivated for implant treatment. However, the authors plan to perform implant treatment in a stepwise manner, starting with a few implants in the mandible, and follow the osseointegration closely, as proposed by Bhatt and Holroyd.⁴

Conclusion

No underlying factor could be identified in this case of aggressive root resorption leading to loss of the entire dentition over a period of 11 years.

Acknowledgments

The authors thank photographer Håkon Størmer, Faculty of Dentistry, University of Oslo, for helpful assistance with the illustrations. The radiographic examinations in this case report were performed at the Department of Maxillofacial Radiology, Faculty of Dentistry, University of Oslo, and the TAKO-centre, Lovisenberg Diakonale Hospital, Oslo, Norway.

Volume 25. Number 5. 2012

463

Tooth no.*	Initial state (2001)	Year of initial sign of resorption	Year of removal
18	Retained		2004-2008 ^t
17		2001	2003
16	C, E	2001	2002
15		2003	2004-2008 [†]
14	А		
13		2003	2004-2008 [†]
12	C, E	2009	2010
11		2009	2010
21		2009	2010
22		2009	2010
23		2009	2010
24	А		
25	C, E	Unknown	2004-2008 [†]
26		Unknown	2004-2008 [†]
27		Unknown	2004-2008 [†]
28	А		
38	А		
37	C, E	2001	2001
36		2001	2002
35	А		
34	C, E	2001	2002
33		2002	2002
32	E	2002	2002
31		2002	2009
41	E	2009	2011
42		2010	2011
43		2009	2009
44		2009	2009
45	C, E	2002	2002
46	C, E	2009	2011
47		2003	2003
48	Δ		

Table 1 Outcomes of the Dentition

C = crown; E = treated endodontically; A = absent.

*FDI tooth-numbering system.

[†]The patient was not seen by the authors during these years.

References

- Liang H, Burkes EJ, Frederiksen NL. Multiple idiopathic cervical root resorption: Systematic review and report of four cases. Dentomaxillofac Radiol 2003;32:150–155.
- Soğur E, Soğur HD, Baksi Akdeniz BG, Sen BH. Idiopathic root resorption of the entire permanent dentition: Systematic review and report of a case. Dent Traumatol 2008;24:490–495.
- Patel S, Kanagasingam S, Pitt Ford T. External cervical resorption: A review. J Endod 2009;35:616–625.



Fig 8 Dentin was noted with deep ramifying external resorptions filled with a fibrous connective tissue rich in cells with round, vesicular nuclei. Few lymphocytes were seen. A couple of multinucleated giant cells (dentinoclasts) were found, but resorptions were mainly outlined by single nucleated cells. There was no sign of reparatory cementum apposition (hematoxylineosin, original magnification $\times 20$).



Fig 9 Fibro-osseous tissue surrounding the teeth showed irregular trabecular structures of cellular bone partly outlined by normal-looking osteoblasts and a few scattered osteoclasts. Trabeculae were surrounded by a fibrous connective tissue similar to that seen in the resorptions (hematoxylin-eosin, original magnification ×20).

- Bhatt N, Holroyd I. Generalized idiopathic root resorption: A case report. Int J Paediatr Dent 2008;18:146–153.
- Olsen CB, Tangchaitrong K, Chippendale I, Graham HK, Dahl HM, Stockigt JR. Tooth root resorption associated with a familial bone dysplasia affecting mother and daughter. Pediatr Dent 1999;21:363–367.
- Yu VSH, Messer HH, Tan KB. Multiple idiopathic cervical resorption: Case report and discussion of management options. Int Endod J 2011;44:77–85.

464 | The International Journal of Prosthodontics

© 2012 BY QUINTESSENCE PUBLISHING CO, INC. PRINTING OF THIS DOCUMENT IS RESTRICTED TO PERSONAL USE ONLY. NO PART MAY BE REPRODUCED OR TRANSMITTED IN ANY FORM WITHOUT WRITTEN PERMISSION FROM THE PUBLISHER.

Copyright of International Journal of Prosthodontics is the property of Quintessence Publishing Company Inc. and its content may not be copied or emailed to multiple sites or posted to a listserv without the copyright holder's express written permission. However, users may print, download, or email articles for individual use.