Dental Traumatology

Dental Traumatology 2008; **24**: 490–495; doi: 10.1111/j.1600-9657.2008.00582.x

Idiopathic root resorption of the entire permanent dentition: systematic review and report of a case

CASE REPORT

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Accepted 30 November, 2006

Abstract – A rare case of idiopathic root resorption affecting the entire dentition was described. Root resorptions of all of the maxillary and mandibular teeth were coincidentally diagnosed on patient's radiographic examination. The patient's medical and dental findings were non-contributory, other than the increased mobility of upper incisors. No significant familial findings could be identified. Detailed radiographic examination revealed resorption affecting the apical part of the roots, resulting in vertical shortening of the teeth. The character of the resorption was as if a tapering occurred throughout the lateral root surfaces. Involved teeth were endodontically treated and the patient was subsequently followed-up for 18 months. Following endodontic therapy, all periapical lesions demonstrated clinical and radiographic evidence of healing. Endodontic therapy resulted in a high degree of success to stop resorption process and maintain the dentition.

Root resorption of the adult permanent dentition is a pathological process either of internal or external origin. Internal resorption occurs when the natural protection of the predentine and odontoblasts in the root canal is damaged or removed due to transformation of embryonic connective tissue cells into giant multinucleated odontoclasts (1). Although the etiologic factor is usually unknown, it may occur because of chronic inflammation/infection in the pulp secondary to caries, trauma, pulpotomy, restorative procedures, orthodontic movement or herpes zoster infection (2–5). There are many reports of idiopathic internal root resorption in both erupted and unerupted teeth (6–11).

On the contrary, external root resorption of multiple permanent teeth is a rare phenomenon. It occurs when the balance between osteoblastic and osteoclastic activities that maintain the physiological state of the tooth root and bone is damaged resulting from the removal of precementum and cementoblasts on the root surface. External root resorption occurring in one or two teeth has been attributed to a number of causes including periapical inflammation, re-implantation of teeth, tumours or cysts, impacted teeth, excessive mechanical or occlusal stresses (including orthodontic treatment) (12). It can also occur because of endocrine imbalance and systemic pathologies such as hypoparathyroidism, pseudohypothyroidism, Paget's disease, hypophosphatasia, calcinosis and Turner's syndrome (12).

An unknown mechanism without depending on the above-mentioned conditions may cause resorption of cementum, dentin and/or enamel, and it has been termed as 'idiopathic'. Idiopathic resorption in multiple roots is far rare than the resorption seen in one or two teeth and occurs in a pattern completely different from that seen in physiologic resorption. The authors have theorized on causes such as microbiologically induced osteoclastic (13) or chronic inflammatory activity (14), familial disorders (15, 16), occlusal trauma (17), orthodontic movement (14), systemic disorders or drug use (18). Although infrequently reported, two types of idiopathic root resorption have been observed: apical and cervical (17). Cervical root resorption starts in the cervical area of teeth and progresses towards the pulp, whereas apical root resorption affects the apical portion of the tooth root and causes gradual shortening (19, 20).

The most common clinical feature of idiopathic root resorption is that patients are asymptomatic with an occasional complaint of tooth mobility. Resorptive defects are generally coincidentally found on routine radiographic examination (21). The location of the resorbed teeth in the documented cases has been variable. It has been reported to affect either the posterior (22, 23) or anterior quadrants (14) or all teeth (12). Moreover, the rate of resorption has been reported to be difficult to determine, since there is only one controlled study up to date with long-term follow-up evaluations (24).

Review of the literature from 1965 until now reveals 49 cases of idiopathic multiple root resorption in man. Of these, eight were internal and 41 were external root resorptions. In addition, nine of these 41 cases were qualified as multiple idiopathic cervical root resorptions and remaining 32 cases were idiopathic apical root resorption (Table 1). However, only two of the 32 cases were reported to affect the entire dentition (12, 25).

No particular treatment has been recommended for idiopathic multiple external root resorption. Some reports have indicated that extractions were necessary (12, 18), whereas others claimed teeth to be maintained (14, 15, 23). Previous reports have shown that endodontic treatment using calcium hydroxide decreased the progress of resorption (12, 26–28).

The aim of this report was to describe an unusual case of idiopathic apical root resorption involving the entire dentition and to review the possible pathogenesis of the disease along with the outcome after proper treatment modalities.

Case report

An 18-year-old Caucasian female had referred to the outpatient clinic of a dental school. She had been complaining about the increased mobility of her upper incisors. She had no other clinical symptoms and/or orodental problems. She was healthy with no history of any systemic disorders.

Intraoral examination

Clinical intraoral examination revealed a full permanent dentition except her upper left second premolar extracted one month ago due to advanced mobility. Her periodontal condition was unremarkable with 1 to 2 mm pocketing and no haemorrhage. The dentition and oral soft tissues appeared normal. No carious lesions were present, but the maxillary right first molar had an amalgam restoration. Teeth numbers 11, 12, 15, 17, 21, 22, 26 and 42 were moderately mobile (Grade 2). Other teeth had no abnormal mobility. All teeth, except numbers 11, 15, 17, 27, 41, 44–47 responded positively to electrical pulp test. On questioning, it was determined that there had been no history of orthodontic treatment and that the deciduous dentition had exfoliated at the expected times. No evidence of attrition on occlusal surfaces of the posterior or incisal surfaces of the anterior teeth could be observed.

Radiographic examination

Panoramic radiography of the patient disclosed extensive resorption of the roots of all teeth in four quadrants. An

Table 1. Reported cases of idiopathic multiple external root resorption between years 1965–2006

No.	Reference	Etiology	No. of teeth involved	Therapy
1	Present case (2006)	Idiopathic	Entire dentition	Endodontic
2	Cholia et al. (2005)	Idiopathic	Multiple	Various
3	Llena-Puy et al. (2002)	Idiopathic	Multiple	Endodontic
4	Di Domizio et al. (2000)	Idiopathic	Multiple	Extraction
5	Mocks et al. (1997)	Idiopathic	1	Unspecified
6	Snelgrove (1995)	Idiopathic	Entire dentition	Extraction
7	van der Wall (1994)	Idiopathic	Multiple	Unspecified
8	Rivera & Walton (1994)	Idiopathic	Multiple	None
9	Ford et al. (1994)	Avulsed tooth	1	Reimplantation
10	Giunta & Kaplan (1994)	Idiopathic	1	Endodontic
11	Mesaros & Wayman (1994)	Idiopathic	1	Unspecified
12	Counts & Widlak (1993)	Idiopathic	Multiple	Extraction
13	Postlethwaite & Hamilton (1989)	Idiopathic	Multiple	Endodontic
14	Yusof & Ghazali (1989)	Idiopathic, inflammation, trauma	Multiple	Various
15	Leache & Moreno Gonzalez (1989)	Medical intervention	Multiple	No dental therapy
16	Saravia & Meyer (1989)	Idiopathic	Multiple	Endodontic
17	Posadinu et al. (1988)	Idiopathic	–	–
18	Pankhurst et al. (1988)	Systemic	Multiple	Conservative
19	Brooks (1986)	Idiopathic	Multiple	None
20	Belanger & Coke (1985)	Idiopathic	Entire dentition	Refused treatment
21	Pilon & van de Poel (1985)	Idiopathic	Multiple	Unspecified
22	Surveyor (1985)	Idiopathic	1	Unspecified
23	Lynch & Ahlberg (1984)	Idiopathic	2	Endodontic
24	Wenzel & Horsted (1984)	Familial	Multiple	Unspecified
25	Williams (1983)	Idiopathic	1	Endodontic
26	Cowie & Wright (1981)	Idiopathic	Multiple	Unspecified
27	Barker & Lockett (1977)	Idiopathic + infection	2	Unspecified
28	Weston (1977)	Idiopathic	1	Unspecified
29	Miklos (1974)	Idiopathic	1	Unspecified
30	el-Hadary (1972)	Idiopathic	Multiple	Unspecified
31	Geffner (1968)	Idiopathic	Multiple	Unspecified
32	Weisman (1966)	Idiopathic	Multiple	Unspecified
33	Brown (1966)	Idiopathic	1	Unspecified

Fig. 1. Panoramic radiograph taken at initial submission of the patient showing generalized root resorptions and bone sclerosis adjacent to the resorbed roots particularly evident in the mandibular premolar and molar areas.

accompanying thickening of the cancellous bone was observed as a radiopaque frame (outline) surrounding the resorbed roots throughout the alveoli of both jaws (Figs 1 and 2). This thickening was particularly dense both at the upper and lower molar regions (Figs 1 and 2). Lamina dura of the involved teeth could not be followed and the radiographic character of the radiopaque frame surrounding the roots was like an inter-radicular scalloping. The length of the roots was otherwise within the normal limits and the character of the resorption was as if a tapering occurred throughout the lateral root surfaces.

Periapical radiograph of the highly resorbed upper right second premolar revealed that the tooth was markedly affected by resorption and was highly mobile, being mainly supported by the gingival tissue.

Laboratory findings

Haematological investigations were done to exclude any systemic disorders including the complete blood and electrolyte count, calcium, phosphorus, serum parathormone and alkaline phosphatase levels. All were found to be within normal limits (Table 2). There was no familial history of the condition and radiographic examinations of the patient's siblings (one brother and one sister) had not disclosed any abnormality.

Endodontic treatment

Root canal treatments of the resorbed teeth were introduced, beginning from the mandibular premolars and followed by upper teeth. Standard access cavities were made using a water-cooled diamond fissure bur in a high-speed handpiece. When the access cavities were opened, it was observed that all teeth, which had responded negative to electrical pulp test during clinical examination, were vital. Following anaesthesia (Jetocain amp, Adeka, Turkey) and extirpation of the pulps, the root canals were instrumented with Hedström files (FKG Dentaire, NovaxaSpa, Italia) using step-back technique. Between each file, the root canals were irrigated with 1 ml of 2.5% NaOCl. Following instrumentation, the canals were dried with paper points (Sure-endo, Sure Dent Corp, Gyeonggi-do, Republic of Korea) and filled

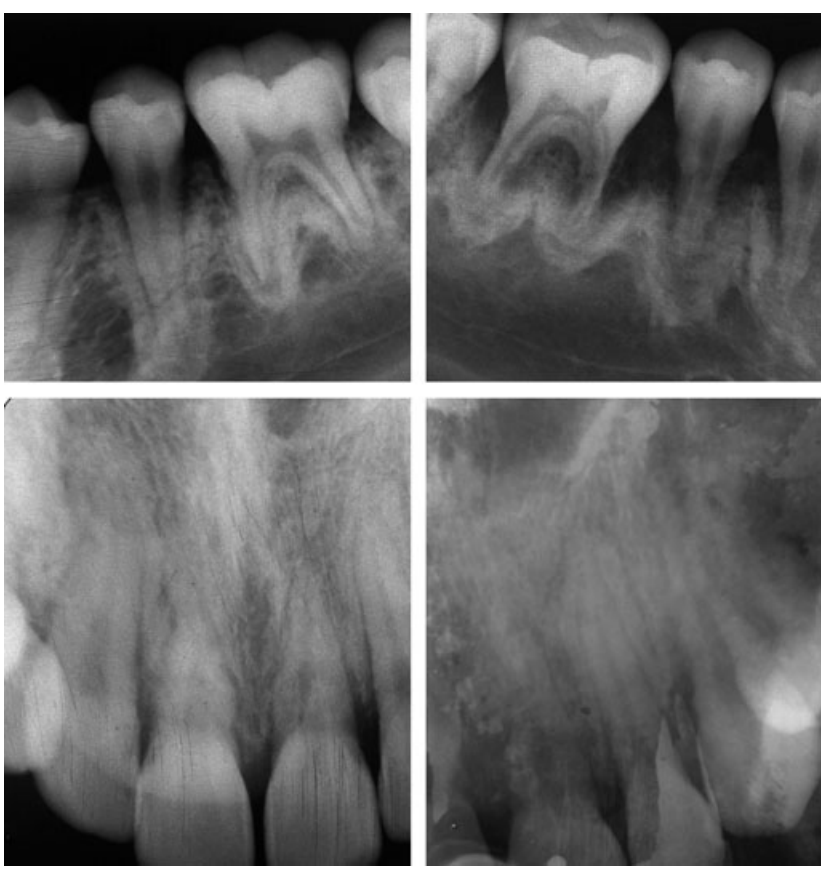

Fig. 2. Periapical radiographs demonstrating close view of bone sclerosis in the incisal and premolar–molar areas.

Table 2. Haematological values of the patient

	Eritrocyte count ($\times 106$ mm^{-3})	Leukocyte count (cells mm^{-3})	Calcium (ml dl^{-1})	Phosphatase (ml dl^{-1})	Alkaline phosphatase (U/L)	Parathormone (ng ml^{-1})
Patient's values	4.4	5500	9.3	4.2	74	0.4
Normal range	4–5	5000–10 000	8.5–10.5	2.5–4.5	42–128	0.4–1.4

with a slurry mixture of calcium hydroxide (Merck, KGaA, Darmstad, Germany) and glycerine. The patient was recalled for the following week and calcium hydroxide paste was renewed. This procedure continued with one-month intervals and occlusal adjustments checked and arranged if necessary. Mobility and resorption gradually decreased following these measures. After a period of 6 months, calcium hydroxide paste was replaced with permanent root filling with lateral compaction technique using standard #25 gutta-percha cones and Diaket (3M Espe; Seefeld, Germany) as the root canal sealer. Excess gutta-percha was cut with a hot instrument 1 mm-down from the canal orifices. After cleaning and etching procedures, the cavities were then restored with bonding (Adper Single Bond; 3M Espe, St Paul, MN, USA) application and resin composite restoration (Filtek Z 250; 3M Espe).

The upper right first and second premolar and first molar and left first premolar and molar teeth were extracted due to their high mobility and poor prognosis. Scanning electron microscopic examination of the extracted premolar tooth was done to exhibit the nature of resorption.

After completion of endodontic treatment of teeth numbers 11–13, 21–23, 34–36, 44 and 45, they were subsequently followed-up for 18 months and the patient is still under continuing observation and treatment (Fig. 4). Endodontic treatment was performed to the mandibular right first incisor and calcium hydroxide was left in the root canal for the follow-up of resorption process. Teeth numbers 32, 33 and 41 were asked for extraction due to the poor prognosis. However, the patient rejected this treatment option for the present because of aesthetic concerns.

SEM findings

One of the extracted teeth was processed for SEM evaluation. The apex of the tooth showed extensive areas with lacunar resorption (Fig. 3a). The borders of these lacunae were distinct. On the lateral surface of the root (Fig. 3b), there were numerous resorption cavities with varying sizes.

Discussion

Although idiopathic root resorption of single or multiple teeth is a more common entity, the involvement of the whole dentition is far rare. Stafne and Slocumb (1944) reported that only 19 of 179 (10.6%) idiopathic resorption cases involved more than one tooth (29). Moreover, only 9% of external root resorption cases have influenced the entire dentition with the inclusion of the present case (Table 1). The case presented here differs

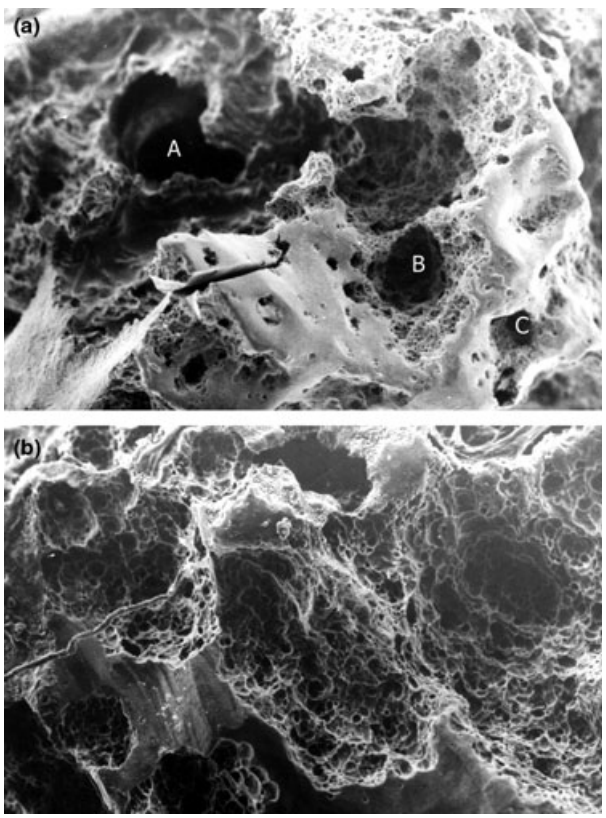

Fig. 3. (a) Extensive areas of lacunar resorption near the main foramen (A) and minor foramina (B–C). (b) Numerous resorption cavities with varying sizes.

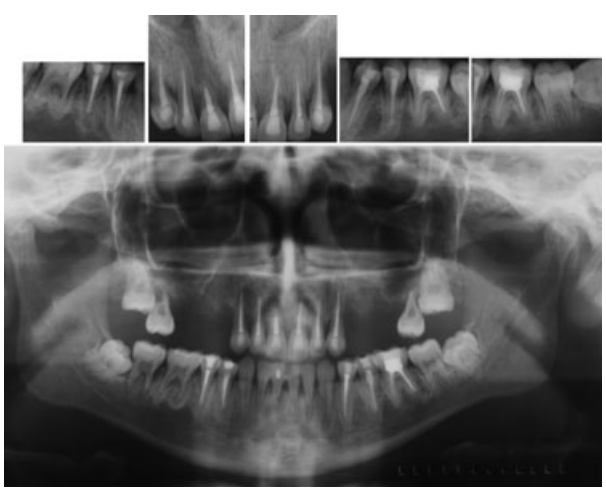

Fig. 4. Panoramic and periapical radiographs after completion of endodontic treatment of teeth numbers 11–13, 21–23, 34–36, 44 and 45 showing apical healing. Teeth numbers 14–16, 24 and 26 were extracted.

from the previously reported generalized resorption cases in that the resorption affected the apical region resulting in not only vertical shortening, but also tapering along the lateral surfaces of the roots. In addition, a thick radiopaque frame surrounding the alveolar bone around the resorbed roots was also an unreported feature up to date.

It has been reported that excessive occlusal loading can result in dystrophic changes in the periodontal ligament, alveolar bone, cementum and pulp. Furthermore, it has been judged to cause periapical inflammation, root resorption and increase in bone sclerosis (30). The thickening of the alveolar bone observed as the increase in density on radiographs, also named as 'bone sclerosis', has been attributed to develop as a reactionary response to either occlusal stresses or to low-grade inflammatory stimulus. Since there is no evidence of attrition or abrasion on occlusal surfaces of the teeth and accordingly no occlusal stresses, the bone sclerosis in the present case may be considered as an excessive production of bone as a compensatory response to the osteoclastic activity resulting in root resorptions. Osteosclerotic areas that do not seem to be associated with infection and/or inflammation are frequently observed on radiographs; some of which appear to be part of the reparative process, and others a compensatory response usually to an obscure (unknown) cause (31).

Previous case reports demonstrated that there was no inter-relation between any systemic disease and idiopathic root resorption (19, 29). In addition, Newman (1975) found that external root resorption cases could not be related to systemic factors (32). In the present case, the patient had received no previous orthodontic treatment and had no relevant medical history other than regular childhood illnesses. There was no known familial history of root resorption. Routine haematological tests were clear, indicating no obvious biochemical deficiencies as well. Since no previous radiographs could be found to compare with those taken at the time of diagnosis, both the onset and the duration of this phenomenon could not be determined.

While there is no definite treatment for this condition, the severity of resorptions and the presence of periapical lesions in the present case necessitated endodontic treatment. The teeth treated by calcium hydroxide and conventional root canal treatment demonstrated clinical and radiographic evidence of healing (Fig. 4). It has been already proved that pulp extirpation and calcium hydroxide treatment may arrest the condition by stimulating apical calcific repair (12). Endurance of the resorptions in the present case has proved once again that endodontic approach is considerably effective in the treatment of external root resorptions.

Conclusion

This case of idiopathic multiple root resorptions is unique because of severity of the resorptions affecting the entire permanent dentition and an accompanying reactionary bone sclerosis throughout the alveoli. Endodontic treatment with calcium hydroxide ceased the progress of resorptions and therefore, this treatment modality was recommended to retain the teeth with idiopathic external root resorption.

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