DOI: 10.1111/j.1365-263X.2007.00882.x

Idiopathic generalized apical root resorption: a report of three cases

AILBHE McMULLIN, PADHRAIG S. FLEMING & ANDREW T. DiBIASE

Maxillofacial Unit, Kent and Canterbury Hospital, Canterbury, UK

International Journal of Paediatric Dentistry 2008; 18: 312–316

Background. Idiopathic apical root resorption usually presents as a chance radiographic finding. It may be widespread, affecting the majority of teeth, with implications for the longevity of the dentition.

Case report. Three cases of significant idiopathic apical resorption resulting, respectively, in prevention, abandonment, and alternative methods of orthodontic treatment are described.

Conclusion. Significant idiopathic resorption may present as a chance radiographic finding, as pain, or excessive mobility. The prognosis for affected teeth is often poor with very limited scope for orthodontic movement due to the likelihood of uncontrolled resorption. Definitive prosthetic rehabilitation is often best deferred until adulthood due to the potential for further resorption during adolescence in addition to vertical growth considerations.

Introduction

Resorption is a condition associated with either physiologic or pathologic processes resulting in a loss of dentin, cementum, or bone[1]. Root resorption can be internal or external in origin and may be either transient or progressive. External root resorption can be associated with trauma, orthodontic forces, impacted teeth, necrotic pulpal tissue with peri-apical inflammation, pathology, as well as hereditary and systemic conditions[2,3].

By definition, an aetiological factor cannot be identified for idiopathic root resorption[4]. The condition has a predilection for young females[5] and was first described in 1930[6].

Involved teeth are initially vital but may subsequently become mobile while remaining asymptomatic[5]. Clinically, resorption is often accompanied by reduced alveolar height and compromised periodontal support. Histological examination of any soft tissue or bone removed reveals chronic nonspecific inflammation, and other laboratory test results indicate no significant abnormalities[7].

The pattern of resorption is variable, occasionally being generalized affecting the majority of teeth[8–11], while more usually being confined to less than three teeth[12]. The resorptive defects are typically progressive, resulting in gradual shortening and rounding of the roots, but may occasionally be self-limiting[8] and can be localized to apical or cervical root surfaces[13]. The cervical form is more common than apical and also more prevalent in females[14].

We present three cases of generalized idiopathic apical root resorption presenting to the orthodontic department for treatment planning. This finding had major implications on treatment planning due to the severity of the condition and the likelihood of exacerbating the problem with application of orthodontic forces.

Case reports

Case 1

A 15-year-old male adolescent presented with a class III malocclusion on a severe skeletal III base having been referred by his general dentist regarding the possibility of orthodontic treatment. His medical history revealed Down syndrome as well as a history of a repaired atrio ventricular septal defect and Fallot's tetralogy. He also had a history of trauma to the upper labial segment with the upper left central incisor being avulsed and replanted. Root canal treatment was subsequently performed by his general dental practitioner; this tooth has

Correspondence to:
Dr P. S. Fleming, Maxillofacial Unit, Kent and Canterbury Hospital, Ethelbert Road, Canterbury CT1 3NG, UK.
E-mail: padhraigfleming@hotmail.com

Fig. 1. Panoramic radiograph highlighting generalized apical root resorption.

Fig. 2. Panoramic radiograph showing generalized apical root resorption. The maxillary central incisors appear rootless and there is extensive pre-eruptive coronal resorption of the lower left second premolar.

remained functional with no evidence of ankylosis or reported symptoms.

On initial examination the lower arch was well aligned with mild crowding in the upper labial segment, palatal displacement of the upper right lateral incisor, and a buccally placed maxillary right canine. There was a reverse overjet of 10 mm and poor oral hygiene. Radiographic examination highlighted generalized crestal bone loss and root shortening of all of the teeth (Fig. 1). No cause for the resorption could be detected leading to a diagnosis of idiopathic apical root resorption, the severity of which precluded active orthodontic treatment. The patient remains under annual review.

Case 2

A 12-year-old girl presenting with a class III malocclusion, anterior open bite, and unerupted upper central incisors attended for orthodontic assessment. There was no significant medical history. On examination, the upper lateral incisors were mesially angulated, the lower left second primary molar was retained and infraoccluded.

On radiographic examination a radiolucent area associated with the unerupted lower left second premolar was noted (Fig. 2). The majority of teeth had reduced root length with the premolar and canine regions severely affected; the upper lateral incisors had an unusual root morphology; and the upper central incisors were unerupted and appeared rootless.

A compromise treatment plan was proposed in view of the significant apical resorption involving limited uprighting of the upper lateral incisors with provision of a partial denture, with the possibility of orthognathic surgery and implant-supported tooth replacements

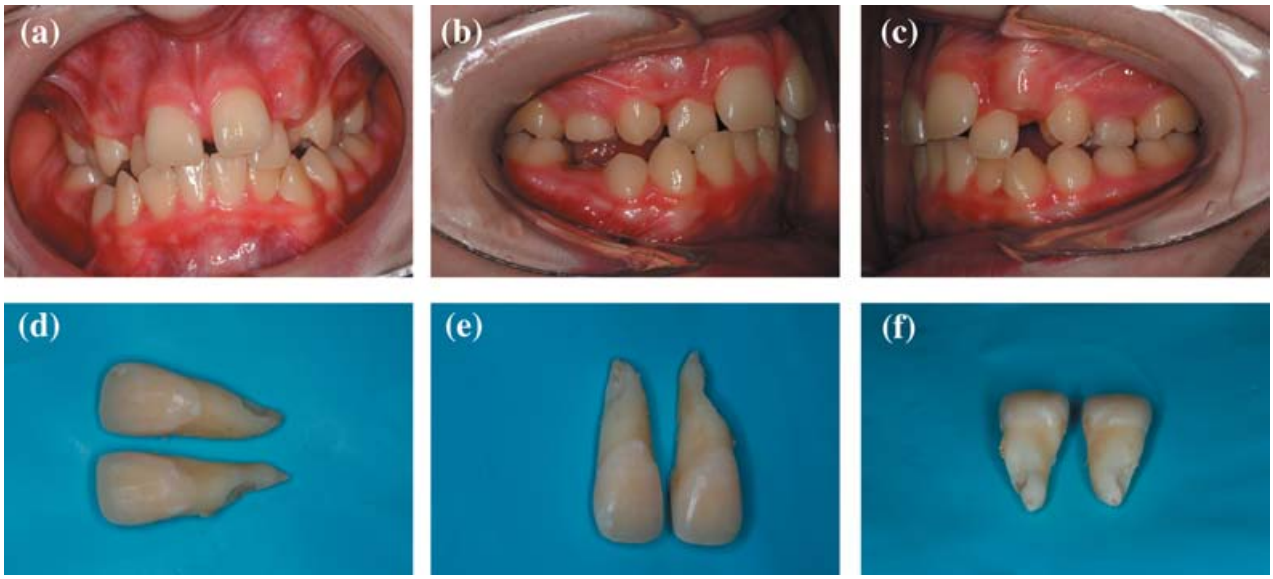

Fig. 3. (a–c) Class II, division 1 malocclusion with increased overjet, palatal displacement of both maxillary incisors, and moderate crowding in both dental arches. (d–f) Photographs of extracted maxillary lateral incisors showing advanced resorptive defects in the apical third of both roots.

in the future. An upper removable appliance was fitted to facilitate uprighting of the maxillary lateral incisors. However, within 6 months the treatment had to be abandoned as the lateral incisors had become excessively mobile, necessitating their removal. Prosthetic replacement of missing teeth is planned over the coming years with definitive restorations being postponed until cessation of the resorption can be ascertained from serial radiographs.

Case 3

A 12-year-old boy presented with a class II, division 1 malocclusion complicated by moderate crowding in the upper and lower arches, with palatal displacement of both maxillary lateral incisors and buccal exclusion of both maxillary canines (Fig. 3a–c). Radiographic examination showed apical root shortening of both maxillary lateral incisors as well as both mandibular second premolars). A compromised treatment plan was agreed involving loss of both maxillary incisors in the first instance to address the crowding in the upper labial segment while facilitating eruption of both maxillary canines (Fig. 3d–f). Upper and lower fixed appliance therapy is planned involving loss of both mandibular second premolars and canine substitution to simulate maxillary lateral incisors, with consolidation of spacing in the upper labial segment.

Discussion

Idiopathic external apical root resorption may be mimicked by shortened root development associated with radiation exposure[15], dentinal dysplasia, and taurodontism[16]. A similar pattern of resorptive defects have been described in combination with various systemic diseases, including hypoparathyroidism[17], renal disease[18], and hepatititis[19]. Case 1 describes presentation in association with Down syndrome and congenital heart disease; however, the association between Down syndrome and short root formation is typically thought to involve the lower labial segment[20].

The individual susceptibility to significant root resorption is gradually being unravelled[21,22]; with homozygosity for the IL-1B allele introducing a 5.6-fold increased risk[23]. Additionally, a gene encoding for a TNF receptor has also been implicated in the process[24]. Nevertheless concurrent presentation of idiopathic apical resorption and ectopic dental development is unreported; however, case 2 describes this condition in association with infraocclusion of a primary molar, and failure of eruption of both maxillary central incisors in the absence

of a mechanical obstruction. The presence of severe pre-eruptive coronal resorption in the mandibular premolar is also a highly significant and irreversible finding, suggesting that the resorptive process is not confined to the cementum and radicular dentine. While prosthetic replacements of severely affected teeth are likely to be necessitated, these have been postponed for two reasons. Ideally the severity and extent of the resorptive process should be ascertained prior to committing to extensive restoration, as deterioration of a previously unaffected tooth may have significant restorative implication. Furthermore, as dental implant-supported restorations are planned, such intervention should not be undertaken prior to completion of vertical facial growth[25,26], particularly in the upper labial segment[27]. Consequently, definitive treatment is unlikely to be commenced until the patient is 16–18 years old.

The third case represents a milder form of the condition with just four teeth affected. The presenting malocclusion would ideally have been managed with loss of maxillary first premolars and mandibular second premolars combined with upper and lower fixed appliances. As both maxillary lateral incisors were palatally displaced, ideal upper labial segment alignment would have necessitated significant bodily movement. It is known that the orthodontic treatment-related causes of root resorption include the total distance the apex moves, the time such movement takes, magnitude of applied forces, previous dental trauma, and anomalous root morphology, including previous root resorption[21,28]. Consequently, the presence of resorption on the lateral incisors dictated a compromise plan involving their extraction and canine substitution to simplify treatment mechanics, and limit the risk of uncontrolled resorption, while still addressing the malocclusion. Canine substitution will be facilitated by adding palatal root torque during orthodontic treatment and by extrusion of the tooth to facilitate gingival aesthetics[29]. Intermittent incisal edge reduction and composite reshaping will also be performed to enhance dental aesthetics.

The severity of the condition in the first two cases either precluded orthodontic treatment altogether or resulted in abandonment of treatment at the risk of exacerbating the condition. This situation is typical, as no reliable method of arresting the process has been described. A single case report has reported abating resorption with administration of calcium, vitamin D, and bisphosphonates to a patient with generalized cervical external root resorption. In this case, however, the patient's alkaline phosphatase levels were raised prior to treatment[14]; no such abnormalities were found in the cases reported in the present paper. According to Fuss *et al.*[30], to 'render proper treatment' of root resorption invariably relies on 'removing the aetiological factor'; consequently, given that the aetiology was unclear in all cases, appropriate management involved stabilization of the condition and avoidance of factors likely to compound the problem.

What this paper adds

- This paper illustrates three cases of idiopathic root resorption of varying degrees of severity, showing that treatment planning with realistic objectives can permit orthodontic treatment.
- The association between idiopathic apical resorption, pre-eruptive coronal resorption, and local ectopic dental development is highlighted for the first time.

Why this paper is important to paediatric dentists

- Paediatric dentists should be aware of the possible dental and orthodontic implications for children with idiopathic root resorption.
- The widespread and destructive nature of the condition highlights that an isolated finding of root shortening on an intraoral radiograph warrants general clinical examination and possible radiographic examination to assess the extent of the condition.
- Paediatric dentists should be aware of the potential for prosthetic rehabilitation but consider the pitfalls of embarking on early prosthetic correction in such cases.

Conclusion

Idiopathic root resorption is a destructive dental condition of unknown origin. It may progress gradually or rapidly and may affect the majority of teeth. Orthodontic movement of affected teeth should be attempted with caution. In less severely affected individuals, treatment planning may be modified to address the patient's concerns and malocclusion while safeguarding their general dental health.

References

1 American Association of Endodontists. *Glossary: Contemporary Terminology for Endodontics*. Chicago, IL: American Association of Endodontists, 1994.
2 Lydiatt DD, Hollins RR, Peterson G. Multiple idiopathic root resorption: diagnostic considerations. *Oral Surg Oral Med Oral Pathol* 1989; **67**: 208–210.
3 Barclay CW. Root resorption: aetiology, classification and clinical management. *Dent Update* 1993; **20**: 248–250.
4 Belanger GK, Coke JM. Idiopathic external root resorption of the entire permanent dentition. *J Dent Child* 1985; **52**: 359–363.
5 Kerr DA, Courtney RM, Burkes EJ. Multiple idiopathic root resorption. *Oral Surg Oral Med Oral Pathol* 1970; **29**: 552–565.
6 Mueller E, Rony HR. Laboratory studies of an unusual case of resorption. *J Am Dent Assoc* 1930; **17**: 326–334.
7 Di Domizio P, Orsini G, Scarano A, Piattelli A. Idiopathic root resorption: report of a case. *J Endod* 2000; **26**: 299–300.
8 Rivera EM, Walton RE. Extensive idiopathic apical root resorption: a case report. *Oral Surg Oral Med Oral Pathol* 1994; **78**: 673–677.
9 Postlethwaite KR, Hamilton M. Multiple idiopathic external root resorption. *Oral Surg Oral Med Oral Pathol* 1989; **68**: 640–643.
10 Moody GH, Muir KF. Multiple idiopathic root resorption: a case report and discussion of pathogenesis. *J Clin Periodontol* 1991; **18**: 577–580.
11 Soni NN, LaVelle WE. Idiopathic root resorption: report of a case. *Oral Surg Oral Med Oral Pathol* 1970; **29**: 387–389.
12 Stafne EC, Slocumb CH. Idiopathic resorption of teeth. *Am J Orthod Oral Surg* 1944; **30**: 41–49.
13 Yusof WZ, Ghazali MN. Multiple external root resorption. *J Am Dent Assoc* 1989; **118**: 453–455.
14 Iwanatsu-Kobayashi Y, Satoh-Kuriwada S, Yamamoto T, *et al*. A case of multiple idiopathic external root resorption: a 6-year follow-up study. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2005; **100**: 772–779.
15 Pietokovski J, Menchel J. Tooth dwarfism and root underdevelopment following irradiation. *Oral Surg Oral Med Oral Pathol* 1966; **22**: 95–99.
16 Desai RS, Vanaki SS, Puranik RS, Rashmi GS, Nidawani P. An unusual combination of idiopathic generalized short-root anomaly associated with microdontia, taurodontia, multiple dens invaginatus, obliterated pulp chambers and infected cyst: a case report. *J Oral Pathol Med* 2006; **35**: 407–409.
17 Cohen DA. Radiology Forum-Shortened roots in maxilla and mandible. *Oral Surg Oral Med Oral Pathol* 1991; **71**: 252.
18 Brin I. Eruption of rootless teeth in congenital renal disease. *Oral Surg Oral Med Oral Pathol* 1985; **60**: 61–64.
19 Pankhurst CL, Moniz C. Multiple idiopathic external root resorption. *Oral Surg Oral Med Oral Pathol* 1988; **65**: 754–756.
20 Brown RH. Crown and root lengths, and root-crown ratios of lower incisor teeth of mongoloid, non-mongoloid retarded and normal individuals. *J Periodontal Res* 1971; **6**: 140–145.
21 Pizzo G, Licata ME, Guiglia R, Giuliana G. Root resorption and orthodontic treatment. Review of the literature. *Minerva Stomatol* 2007; **56**: 31–44.
22 Al-Qawasmi RA, Hartsfield JK Jr, Everett ET, *et al*. Root resorption associated with orthodontic force in inbred mice: genetic contributions. *Eur J Orthod* 2006; **28**: 13–19.
23 Al-Qawasmi RA, Hartsfield JK Jr, Everett ET, *et al*. Genetic predisposition to external apical root resorption. *Am J Orthod Dentofacial Orthop* 2003; **123**: 242–252.
24 Al-Qawasmi RA, Hartsfield JK Jr, Everett ET, *et al*. Genetic predisposition to external apical root resorption in orthodontic patients: linkage of chromosome-18 marker. *J Dent Res* 2003; **82**: 356–360.
25 Iseri H, Solow B. Continued eruption of maxillary incisors and first molars in girls from 9 to 25 years, studied by the implant method. *Eur J Orthod* 1996; **18**: 245–256.
26 Fudalej P, Kokich VG, Leroux B. Determining the cessation of vertical growth of the craniofacial structures to facilitate placement of single-tooth implants. *Am J Orthod Dentofacial Orthop* 2007; **131** (4 Suppl.): S59–S67.
27 Thilander B, Odman J, Grondahl K, Friberg B. Osseointegrated implants in adolescents. An alternative in replacing missing teeth? *Eur J Orthod* 1994; **16**: 84–95.
28 Segal GR, Schiffman PH, Tuncay OC. Meta analysis of the treatment-related factors of external apical root resorption. *Orthod Craniofac Res* 2004; **7**: 71–78.
29 Kokich VO Jr, Kinzer GA. Managing congenitally missing lateral incisors. Part I. Canine substitution. *J Esthet Restor Dent* 2005; **17**: 5–10.
30 Fuss Z, Tsesis I, Lin S. Root resorption: diagnosis, classification and treatment choices based on stimulation factors. *Dent Traumatol* 2003; **19**: 175–182.