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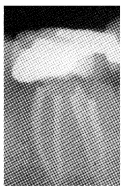

CASE REPORT

External apical root resorption: two case reports

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Abstract

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Aim To present two cases of external root resorption and in so doing highlight rarer causative factors as well as the difficulties in classifying root resorption.

Summary Root resorption in the permanent dentition is usually pathological. Local factors are the most frequent causes of resorption, most commonly excessive pressure and inflammation. However, many systemic abnormalities have been implicated in the resorption process. Resorption is often an incidental finding during routine examination. Otherwise, late in the disease process, tooth mobility or infection may arise. A rarer form of root resorption is external apical root resorption which may or may not be linked to systemic disease. In most cases, no causative factors are found. At present there is no curative treatment for external apical root resorption. The current management for these patients is symptomatic, minimal intervention and long-term monitoring.

Key learning points

- Apical resorption may be associated with a systemic disease or of an idiopathic origin. It may also occur in association with orthodontic treatment or with pathoses such as tumours, cysts, etc.
- In the absence of signs or symptoms of pulpal and/or periapical disease, endodontic treatment is not indicated.
- Long-term monitoring of affected patients is essential.

Keywords: Ehlers–Danlos syndrome, external apical root resorption, root resorption.

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Introduction

Physiological root resorption is a process involving resorptive activity followed by periods of attempted repair. This results in variable tooth mobility in deciduous teeth before

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exfoliation. In contrast, the process of root resorption in the permanent dentition is usually pathological (Bakland 1992).

Resorption of the teeth may occur as a result of different and varied causative factors such as inflammatory conditions, mechanical stimulation, traumatic injuries, neoplastic processes and association with systemic diseases; lesions may also be idiopathic (Pindborg 1970, Bakland 1992).

The classification of root resorption is difficult and many authors have proposed varying complexities of classifications, some of which are incomplete. It can be broadly classified as internal and external, with subdivisions (Table 1).

External apical root resorption affecting multiple permanent teeth appears to be rare with only 14 cases reported in the literature (Cholia *et al.* 2005). Only three of these cases report resorption affecting the entire permanent dentition.

Systemic abnormalities have been associated with external root resorption, as shown in Table 1 (Newman 1975, Cholia *et al.* 2005). However, to date, no association of external apical root resorption and Ehlers–Danlos syndrome (EDS) has been reported in the literature.

Two cases of external apical root resorption are presented. The first, related to systemic disease, affected multiple teeth in a 20-year-old male with EDS. The second case involved idiopathic external apical root resorption affecting most of the permanent dentition in a healthy 19-year-old male. By presenting these cases, rarer aetiological factors as well as the difficulties in classifying root resorption are highlighted.

Table 1 Classification and causative factors in pathological root resorption (**Bakland 1992**)

Site	Type	Causative factors
Internal		Trauma
		Infection
External	Surface	Trauma
	Inflammatory	Trauma
		Infection
	Replacement (ankylosis)	Avulsion and replantation
		Luxation
		Transplantation
	Pressure	Orthodontic tooth movement
		Excessive occlusal forces
		Impacted teeth
		Supernumerary teeth
		Tumours
		Cysts
	Related to systemic conditions	Hyperparathyroidism
		Paget's disease
		Papillon–Lefèvre syndrome
		Bone dysplasia
		Renal disease
		Hepatic disease
	Invasive (cervical)	Trauma
		Orthodontic tooth movement
		Periodontal treatment
		Intracoronal tooth bleaching
		Unknown
	Idiopathic	Unknown

Case reports

Case 1

A 20-year-old male was referred to Glasgow Dental Hospital and School, UK by his General Dental Practitioner, with a 2-month history of 'looseness' of the mandibular anterior teeth.

The patient also gave a long history of bleeding gums on brushing but otherwise was asymptomatic. There was no history of previous orthodontic treatment. His medical history included EDS and he was a nonsmoker.

Clinical examination revealed a fully dentate patient (wisdom teeth unerupted) with deficient oral hygiene. Periodontal examination displayed probing depths of ≤3 mm with mild bleeding on probing consistent with a provisional diagnosis of generalized mild chronic marginal gingivitis. The mandibular incisor teeth exhibited horizontal movement between 1 and 2 mm. Radiographs prior to the original referral were unfortunately unavailable for comparison. Radiographic investigation revealed very short mandibular incisor roots and sclerosis of the pulp canals (Fig. 1) and unerupted third molars also with absent roots. The remaining teeth, although normal in root length showed unusual root morphology described as follows: teeth 12 and 22 (FDI) showed a narrowing of the pulp canal in the mid portion. Tooth 23 showed a radiopacity superimposed on the root canal, at the level of the alveolar ridge, consistent with an intrapulpal calcification. The remaining canine teeth showed coronal pulps which appeared to be bifid at the level of the alveolar crest, with the central portion of the root clearly opaque and a relatively normal apical portion of the root canal (Fig. 2).

The mandibular incisor teeth and the maxillary and mandibular canine teeth responded normally to electric pulp sensibility tests which indicated that their pulps were clinically normal.

Figure 1 Case 1: Occlusal radiograph 27/07/00. Shows advanced apical root resorption of the four mandibular incisor teeth.

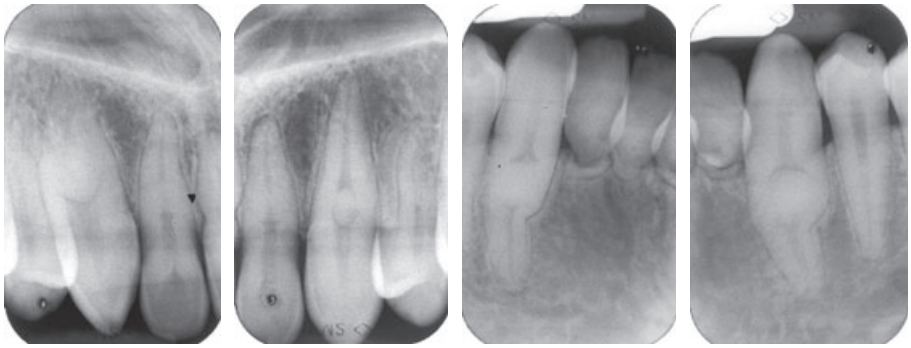

Figure 2 Case 1: Periapical radiographs 27/07/00. Clearer views of the advanced root resorption of the mandibular incisor teeth, showing sclerosis of the pulp canals and intrapulpal calcification of tooth 23. The maxillary right and lower left and right canine teeth show a coronal pulp which appears bifid just beyond the level of the alveolar crest, with the central portion of the root opaque and a relatively normal apical portion of the root canal beyond.

Definitive diagnoses were: generalized mild chronic marginal gingivitis, external root resorption of mandibular central and lateral incisor teeth and root malformation/intrapulpal calcification of maxillary and mandibular canine teeth, related to a systematic condition.

Informed and written consent was obtained before arrangements were made to construct an indirect reinforced composite splint (Ribbond®; Bisco Dental Products (Canada) Inc., Richmond, Canada), extending from 43 to 33, to stabilize the mandibular incisor teeth during initial treatment (Fig. 3). 'Ribbond' is a multi-directional fibre-reinforced composite material. This initial treatment also involved a course of nonsurgical hygiene phase therapy aiming to control plaque-induced inflammation.

Long-term treatment with implant-retained prostheses was planned, however the patient unfortunately had to relocate and could not attend further appointments.

Case 2

A 19-year-old male was referred by his General Dental Practitioner after sustaining trauma to tooth 11. A periapical radiograph showed a splint in place from tooth 12 to tooth 21, and also evidence of root resorption of the maxillary lateral and central incisors (Fig 4). This incidental finding triggered further investigation and panoramic radiography confirmed that many of the teeth had evidence of advanced root resorption (Fig. 5). A previous panoramic radiograph taken in 1999 revealed no evidence of root resorption (Fig. 6).

Figure 3 Case 1: Occlusal photograph 02/02/01, with Ribbond splint in place.

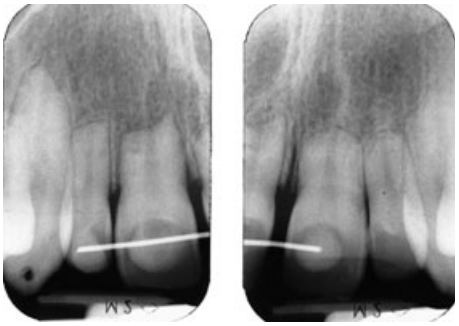

Figure 4 Case 2: Periapical radiograph 27/10/06. Splint attached to 12, 11 and 21. Evidence of external apical root resorption of four maxillary incisor teeth.

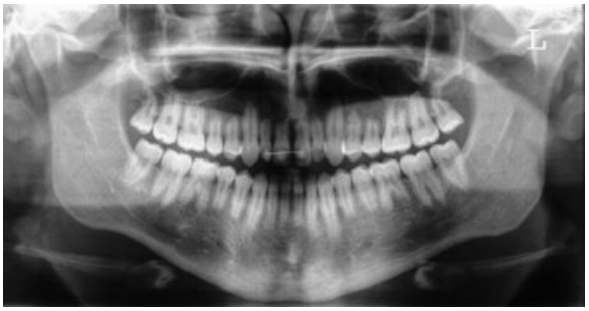

Figure 5 Case 2: Panoramic radiograph 27/10/06. External apical root resorption evident in the following teeth: 15 to 22, 24, 25, 34 to 36 and 43 to 46.

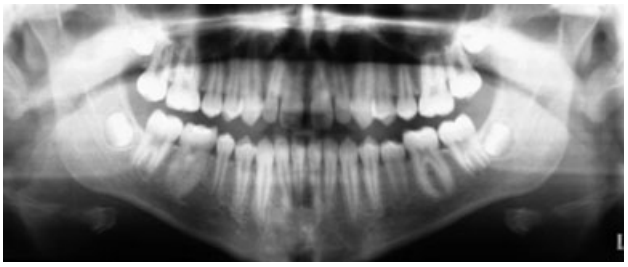

Figure 6 Case 2: Panoramic radiograph 27/07/99. Radiograph available from general dental practitioner records showing unrestored adult dentition with normal dental development (pre-molars have not completed development at this stage). No evidence of root resorption.

The patient was seen on referral in the Conservation Department at Glasgow Dental Hospital & School 2 weeks after the palatal luxation of tooth 11 for assessment and treatment of generalized root resorption of his permanent dentition.

There was no relevant medical or family history of systemic diseases that could be associated with this external apical root resorption. The patient also gave no history of previous orthodontic or orthognathic treatment.

On examination, oral hygiene was satisfactory with mild chronic marginal gingivitis in the maxillary anterior region. A splint was present stabilizing the maxillary incisor teeth. Tooth 11 had an enamel fracture.

Once the splint was removed, tooth 11, teeth 12 and 22 showed grade II mobility. The occlusion was a mild skeletal class III with an anterior open bite.

A panoramic radiograph showed (Fig. 5):

- A full permanent dentition present and erupted.
- A splint attached to 12, 11 and 21.
- Root resorption evident in the following teeth: 15 to 22, 24, 25, 34 to 36 and 43 to 46.

The patient was reviewed 2 months later to assess whether further clinical and/or radiological changes had occurred. There was no change clinically except for obvious discolouration of tooth 11. A 6-month follow-up was arranged.

The long-term patient management in this case will involve monitoring clinically and radiographically with interventions as and when signs and symptoms require as well as restorative treatment as required, most likely involving implants.

Discussion

The first documented case report of idiopathic external root resorption was by Muller & Rony (1930) and was described as generalized rapidly progressive cervical root resorption associated with functional hepatic disturbance. Since then, it has been postulated that there may be several causes of external root resorption (Table 1). External root resorption seems to be a relatively common incidental radiographic finding in isolated teeth, but much less common in a generalized form (Bakland 1992). Local causes are thought to be the most frequent causative factors, notably excessive pressure from orthodontic treatment (Karrer *et al.* 2000, Badauy *et al.* 2007) or chronic inflammatory processes (Ne *et al.* 1999*).*

In addition, many systemic abnormalities have been implicated in the causation of the resorptive process such as hypophosphatasia, hyperparathyroidism, Paget's disease, Papillon–Lefèvre syndrome, renal diseases, hepatic diseases and bone dysplasia (Newman 1975, Cholia *et al.* 2005).

The majority of attempts to classify root resorption make emphasis on the site, aetiology, mechanism of injury to the periodontal apparatus and the ability to provide treatment (Gunraj 1999, Ne *et al.* 1999, Fuss *et al.* 2003) and are generally inconclusive. Only a few reports have taken into consideration other causative factors like systemic conditions or hitherto unknown causative agents (Newman 1975, Bakland 1992).

The first case presented in this article did not have any obvious local causative factor but the medical history included EDS. There are no previous reports in the literature of association of external apical root resorption and EDS, although EDS type IV and type VIII have an association with periodontal disease (DeCoster *et al.* 2005, Hart & Atkinson 2007). In addition, formation of dysplastic dentine has been previously described in patients with EDS (Barabas & Barabas 1967).

The mechanism of the periodontal involvement in EDS has not yet been established although it has been postulated that a defect in type III collagen may undermine the integrity of the periodontal apparatus: collagen type III comprises 16%–18% of the total collagen in the periodontal ligament (Hartsfield & Kousseff 1990, DeCoster *et al.* 2005, Hart & Atkinson 2007). The periodontal ligament is a specialized connective tissue which acts as a barrier between the alveolar bone and cementum (Sasaki *et al.* 1990). Damage or loss of the periodontal ligament renders the denuded cementum surface chemotactic to dentinoclastic cells (Sasaki *et al.* 1990, Gunraj 1999). Subsequently, root resorption may occur. One possible biologically plausible explanation in case 1 may be that the defect in type III collagen characteristic of EDS may be associated with stimulation of clastic cells on the cementum surface, resulting in localized external apical root resorption. It may be useful to look at other EDS patients in order to check whether they are similarly affected.

Case 2 did not display any obvious local or systemic causative factors to explain the external apical root resorption pattern and as such can be categorized as idiopathic. By definition, idiopathic external apical root resorption (IEARR) is a diagnosis of exclusion.

Relying on the few cases reported in the literature it appears that IEARR affects younger patients from 14 to 39 years old. Male patients appear to be more frequently affected, with a male : female ratio of 11 : 4 (Cholia *et al.* 2005). The condition has a predilection for the posterior region of the mouth and the maxilla and mandible generally have a symmetrical pattern of distribution.

Orthodontic treatment is known to be associated with apical root resorption of different degrees. Individual pre-disposition might be a major reason for the variation in apical root resorption (Hartsfield *et al*. 2004). The degree and severity of EARR with known aetiological factors are multifactorial, involving host and environmental factors. Genetic pre-disposition accounts for at least 50% of the variation in EARR (Hartsfield *et al.* 2004), and this could be an important factor in the resorption process in patients with systemic conditions or those diagnosed with idiopathic external apical root resorption.

In patients affected by IEARR, the teeth and periodontal tissues have an otherwise normal appearance. The root resorption can be associated with clinically normal teeth as well as endodontically treated teeth. The alveolar bone level is usually normal. The patient is asymptomatic until very late in the pathological process when increased mobility may be reported. It is usually an incidental finding on radiographic investigation.

In cases of IEARR the treatment will depend largely on the presenting symptoms as well as the extent and the severity of the root resorption found. Long-term monitoring of the dentition using serial radiographs, sensibility testing and assessment of patient's symptoms is advocated and this is the option chosen for case 2.

Restorative dental treatment, including adhesive/conventional fixed prosthodontics, removable prosthodontics, periodontal measures and osseointegrated implants, may have a role in appropriate circumstances, in order to maintain function and aesthetics. Root canal treatment is usually not indicated in idiopathic root resorption cases in the absence of pulpal symptoms. Clearly further continued resorption will necessitate eventual loss of multiple teeth in this case and as such, implant planning will be required in the future. The success of osseointegration in sites where root resorption has been active is unknown (Marx & Garg 1998), and is worthy of future research.

Summary

In this article, two cases of IEARR have been reported, one of which appears to be the first reported case in a patient with EDS.

Classification of root resorption is difficult and some proposed classifications are incomplete and can lead to confusion. It is important to include association with systemic diseases and cases of idiopathic origin in any comprehensive classification of root resorption.

Furthermore, a diagnosis should be reached, if at all possible, in the majority of cases, ruling out sinister causes, association with systemic diseases, previous orthodontic treatment, etc. in order to treat the causative factor in the first instance.

Treatment of IEARR is limited, based on long-term monitoring and guided by patients' signs and symptoms. Root canal treatment is not advocated in absence of pulpal symptoms.

Disclaimer

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represent best practice, or the views of the IEJ Editorial Board, or of its affiliated Specialist Societies.

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