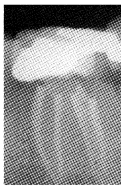
International Endodontic Journal

doi:10.1111/j.1365-2591.2010.01820.x



CASE REPORT

Multiple idiopathic cervical resorption: case report and discussion of management options

V. S. H. Yu[1], H. H. Messer[2] & K. B. Tan[1]

[1]Faculty of Dentistry, National University of Singapore, Singapore; and [2]Melbourne Dental School, University of Melbourne, Melbourne, Vic., Australia

Abstract

Yu VSH, Messer HH, Tan KB. Multiple idiopathic cervical resorption: case report and discussion of management options. *International Endodontic Journal*, **44**, 77–85, 2011.

Aim To present a case of cervical root resorption affecting all teeth and resulting in multiple tooth loss.

Summary A healthy 33-year-old Chinese male, with no contributory medical or family/social history, presented with generalized cervical root resorption. Lesions varied in severity amongst teeth and even involved an impacted third molar. All cervical root surfaces were affected and lesions often extended coronally, undermining enamel. CBCT demonstrated that the lesions were more extensive and more widely distributed than was seen using conventional radiography. Bone extended into many resorptive defects but without clinical evidence of ankylosis. Periodontal probing, pulp testing, percussion sound and mobility were within normal limits.

Key learning points

- Aetiology of the generalized idiopathic cervical root resorption is uncertain.
- Management is complex, and options have included surgical exposure and restoration of affected sites, extraction and submergence of affected roots.
- A staged approach involving early intervention with restoration of resorptive defects, followed by progressive extraction and replacement with implant-supported prostheses is recommended.

Keywords: CBCT, conventional radiography, human, idiopathic, management, multiple invasive root resorption, teeth.

Received 16 July 2010; accepted 8 October 2010

Introduction

Cervical root resorption is an uncommon condition of largely unknown aetiology. In contrast to invasive cervical resorption, which typically affects only one tooth and may be

Correspondence: Victoria Soo Hoon Yu, Faculty of Dentistry, National University of Singapore, 11 Lower Kent Ridge Road, Singapore 119083 (e-mail: rsdyshv@nus.edu.sg).

related to trauma or other local factors (Heithersay 1999, 2004), multiple idiopathic cervical root resorption affects varying numbers of teeth. The condition was first identified 80 years ago (Mueller & Rony 1930), and since that time only approximately 25 cases have been reported (Liang *et al.* 2003, Iwamatsu-Kobayashi *et al.* 2005, von Arx *et al.* 2009); the number of involved teeth ranges from three to more than 20 per subject (Iwamatsu-Kobayashi *et al.* 2005). The distribution varies from a single region (e.g. mandibular incisors) to most teeth within one arch or more generally distributed throughout the entire dentition. A similar condition involving apical resorption has also been described (Cholia *et al.* 2005).

Most affected individuals are healthy, with noncontributory medical histories. A predilection for young females has been reported (Macdonald-Jankowski 2005). An attempt has been made to link multiple idiopathic cervical root resorption to feline invasive cervical resorption (von Arx *et al.* 2009, DeLaurier *et al.* 2009). Management poses serious problems and the condition is generally progressive and leads to loss of affected teeth. Attempts to manage early lesions by surgical exposure and restoration have been undertaken, but ultimately lesions recur and crowns either fracture or teeth are extracted in anticipation of fracture. Root canal treatment does not arrest the resorption (Moody *et al.* 1990). The small number of cases has prevented a systematic study of either aetiology or ideal management.

In this case report an extreme case of severe multiple idiopathic cervical root resorption in a healthy young adult male, involving every tooth and leading to the loss of 14 teeth, including 10 over a 5-year period is presented.

Case report

A healthy 33-year-old Chinese male attended the specialist prosthodontic clinic at the National University Health System (Singapore) with the intention of seeking implant replacement for lost teeth. In addition to radiographs obtained at the time of the visit in December 2009, previous orthopantomograms (OPG) were available from 2004 and 2007.

Medical and family/social history

The medical history was noncontributory and the patient was healthy, with no record of systemic disease or abnormal blood picture. In a discussion of the widespread cervical resorption visible on radiographs, he reported that no other family member (father, mother, one older brother and one older sister) had a similar condition. The family had no pets and no close contact with cats.

Dental history and clinical assessment

Overview

The patient was aware of his condition and sought implant replacement for missing teeth 13–22 and 35–44. All lost teeth had been extracted because of resorption, including the four mandibular incisors before 2004, mandibular premolars by 2007, and five maxillary anterior teeth (13–22) between 2007 and 2009. The patient had a record of regular dental care. Previous attempts at treatment by various dentists included at least one fixed partial denture and removable prostheses. The patient was unable to specify the longevity of each intervention and specific records of these attempts were not available to the authors.

Clinical assessment

The patient was caries free, and had never had a restoration other than a fixed bridge to replace lost mandibular incisors. All remaining teeth were pulp tested using refrigerant spray and electrical pulp test, and all responded within normal limits. Teeth were not tender to percussion and gave a normal sound on percussion. Mobility was normal. Periodontal probing was consistently 3 mm or less and no bleeding on probing was detected. Oral hygiene was good and gingival tissues were generally healthy. Given the radiographic evidence of severe resorption, percussion and mobility testing were performed carefully and gently.

Radiographic assessment (including CBCT)

Orthopantomogram radiographs were available from 2004 and 2007, and a further OPG was taken, along with periapical radiographs of all remaining teeth (Fig. 1). A CBCT scan was obtained as part of the pre-implant assessment.

All remaining teeth showed radiographic evidence of cervical resorption, including tooth 38 which was unerupted (impacted) (Fig. 2). The severity of resorption varied amongst teeth, from a small distal lesion on tooth 38 to severe resorption involving more than half of the total tooth length. Lesions extended coronally beneath enamel, which appeared to remain intact even when it was extensively undermined. Based on conventional periapical films, the resorptive defects did not appear to involve the pulp space.

The pattern of resorption typically involved a large area of the affected root surface. A distinctive feature of the lesions was the ingrowth of alveolar bone into the root defects,

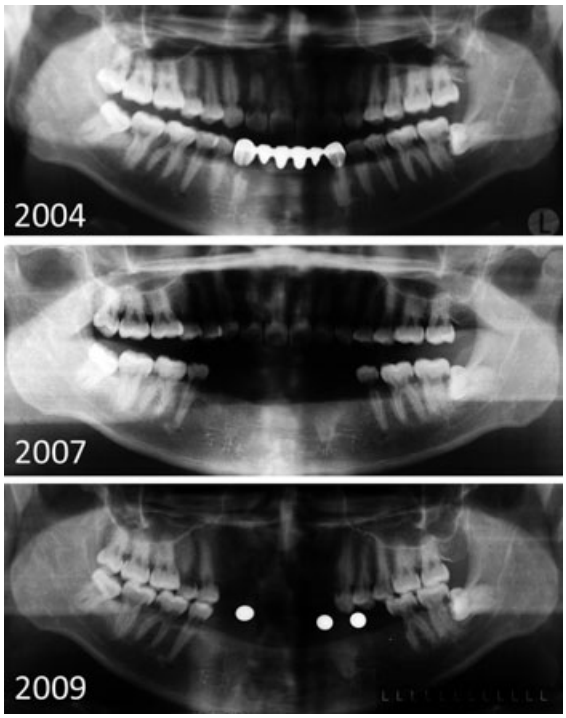


Figure 1 OPGs showing the extent and progression of resorptive lesions and tooth loss over a period of 5 years.

Figure 2 Periapical radiographs taken at the most recent visit. A small lesion is present on the distal cervical margin of impacted tooth 38 and larger lesions on first and second molars. Alveolar bone has extended into the resorption cavities, especially in teeth 46 and 47, where the bone has extended even into the coronal defect beneath enamel. Despite the bony ingrowth, the teeth did not show clinical evidence of ankylosis.

even extending into coronal defects and resulting in increased alveolar bone height (Fig. 2). Despite this ingrowth, ankylosis did not appear to occur, with a normal percussion sound and physiological mobility present in all teeth (within the limits of gentle testing).

A comparison of OPGs from 2004, 2007 and 2009 (Fig. 1) showed a variable pattern of lesion progression. Some lesions remained essentially unchanged (teeth 45M, 46D), whilst others increased in size (multiple teeth) and, surprisingly, several appeared to regress (26D, 47D). Over the period 2004–2009, however, 10 additional teeth had been extracted, indicating a generally progressive pattern.

Cone beam computed tomography demonstrated that lesions were generally larger than was evident from periapical films or OPGs. On the conventional periapical image tooth 25 did not appear to be affected (Fig. 3). However, an initial mesial lesion was discernible on the CBCT. Buccal and lingual/palatal lesions were also more readily visible on CBCT images (Fig. 4), and apparent extension into the pulp chamber could also be seen (although without any clinical evidence of pulpal pathology) (Fig. 5). The extent of lesions was demonstrated by axial slices in the cervical region (Fig. 6).

Discussion

The present case represents an extreme example of multiple cervical root resorption, involving every tooth and leading progressively to the loss of 14 teeth, at a relatively young age. Only a very small number of cases of generalized resorption involving all areas of the mouth have been reported previously (Table 1). Fewer involved teeth with a more limited

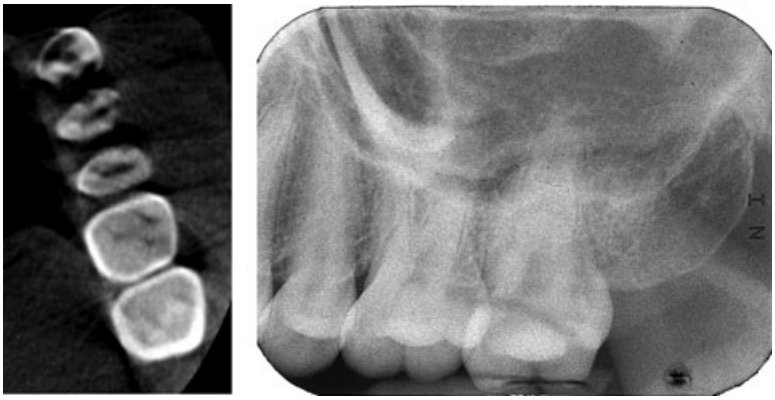


Figure 3 Periapical radiograph and CBCT image of tooth 25. A small lesion on the mesiobuccal surface is seen in the CBCT image but not the periapical radiograph.

Figure 4 Periapical radiographs of the maxillary molar region and a CBCT slice through the first molars. Extensive lesions on the buccal and palatal surfaces of tooth 16 and the buccal surface of tooth 26 were not detected by conventional radiography but are clearly present in the CBCT image.

distribution (summarized in Liang *et al.* 2003, Iwamatsu-Kobayashi *et al.* 2005) are presumably a partial expression of the same condition.

Resorption is progressive, although not uniformly so (this case report, Liang *et al.* 2003). Both Iwamatsu-Kobayashi *et al.* (2005) and von Arx *et al.* (2009) reported histological evidence of cemental repair of lesions, implying arrest. The pattern of resorption in this and other similar cases differs in character from that described for invasive cervical resorption affecting predominantly single teeth. In invasive cervical resorption the lesion tends to have a small point of entry on the root surface and most of the resorption occurs within the bulk of dentine, avoiding both the root surface and the pulp chamber (Heithersay 1999, 2004). The lesions in this case are more in the form of an external

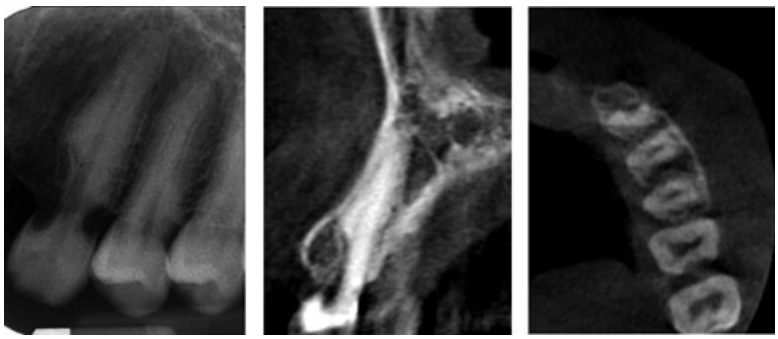


Figure 5 Periapical radiograph and CBCT images (bucco-lingual and axial slices) of tooth 23. The CBCT images appear to show an extensive pulp exposure on the buccal surface with bone growing into the lesion and expansion of the buccal plate. This is not apparent on the periapical radiograph. Clinically the tooth tested within normal limits to pulp tests and the gingiva was normal in appearance with a probing depth of less than 3 mm.

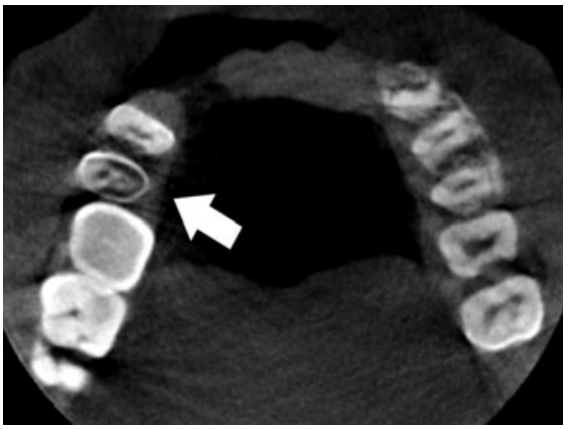


Figure 6 CBCT axial slice through the cervical region of the maxillary teeth. Tooth 15 (white arrow) shows the resorptive defect extending beneath enamel and involving most of the circumference of the tooth.

Table 1 Previously reported cases of generalized cervical resorption

Case	Author	*n*[a]	Age	Gender	Race	Systemic condition?
1	Kerr *et al.* (1970)	24	68	F	Caucasian	Low Ca, high P
2	Kerr *et al.* (1970)	17	30	F	Caucasian	High ALP [b]
3	Hopkins & Adams (1979)	18	20	F	Not reported	No
4	Moody *et al.* (1990)	17	27	M	Caucasian	No
5	Matsui *et al.* (1998)	17	31	M	Japanese	No
6	Liang *et al.* (2003)	13	19	F	Latina	No
7	Liang *et al.* (2003)	12	50	M	Caucasian	Cholecystectomy
8	Iwamatsu-Kobayashi *et al.* (2005)	21	49	F	Japanese	High ALP
9	Neely & Gordon (2007)	19	63	M	Caucasian	Hypothyroid
10	von Arx *et al.* (2009)	14	68	M	Caucasian	Not stated
11	von Arx *et al.* (2009)	20	66	M	Caucasian	Not stated
12	This case	31	33	M	Chinese	No

Generalized is defined as more than 10 involved teeth in at least three quadrants.
[a]Number of involved teeth.
[b]ALP: serum alkaline phosphatase.

defect affecting a large area of the cervical root surface. The defect becomes filled with bone, although without ankylosis. Invasive cervical resorption typically involves only one tooth, and only rarely more than one, implying local factors such as trauma or bleaching in its aetiology (Heithersay 2004). Although the aetiology of invasive cervical resorption is also uncertain, the two conditions should be considered separate entities.

Based on the very small number of cases reported to date, generalized cervical root resorption does not appear to demonstrate any gender, age, familial or ethnic predilection, and most commonly is not associated with any systemic condition. The lack of age and gender predilection is different from the conclusion of Macdonald-Jankowski (2005), who found a majority of cases of multiple lesions in young females, using the simpler inclusion criterion of more than one involved tooth. Clinically, the resorptive lesions were not accompanied by signs of overt inflammation (gingivitis, bleeding on probing, increased pocket depth), which has also been confirmed histologically (Iwamatsu-Kobayashi *et al.* 2005, von Arx *et al.* 2009). The involvement of an impacted third molar in this case indicates that exposure to the oral environment is not a predisposing factor. Thus, the suggestion that lesions result from microbiologically induced osteoclastic activity (Moody & Muir 1991) is not tenable. Attempts to link the condition to feline odontoclastic resorptive lesions (von Arx *et al.* 2009, DeLaurier *et al.* 2009), implicating feline herpes virus FeHV-1, are a useful insight, but it is premature to consider the virus as the cause of the condition. Whilst a blood test was not conducted on the current patient, he did not have any significant contact with cats. The possible association of herpes virus with pulpal and periapical pathology (Sabeti & Slots 2004) raises the possibility that the virus may be linked to root resorption, although why it should lead specifically to an attack of cervical dentine in healthy teeth of a very small number of patients is unknown.

Treatment planning for generalized, progressive resorption is a challenge. The large number of involved teeth and their distribution amongst multiple quadrants do not allow simple management. The rarity of cases and the lack of a recognized treatment protocol have resulted in *ad hoc* management on an individual basis. Typically, involved teeth are extracted when the lesions progress to the point where a crown fracture is likely or has actually occurred (Moody *et al.* 1990, Liang *et al.* 2003), eventually leading to the extraction of all teeth and fabrication of complete dentures (Kerr *et al.* 1970). The range of options undertaken in individual cases includes (i) observation, with episodic extraction and prosthetic replacement of fractured teeth; (ii) early intervention with surgical exposure, curettage of lesions and restoration with glass ionomer cement (sometimes with root canal treatment of involved teeth); (iii) crown resection with root submergence to preserve alveolar bone, and prosthetic replacement; (iv) extraction and replacement with implants; (v) extraction of all teeth, with full dentures; (vi) medical management using bisphosphonates to arrest resorption.

The present case illustrates the unsatisfactory outcome of nonintervention until teeth fracture or are in danger of fracturing. A fixed prosthesis to replace lost mandibular incisors failed when abutment teeth subsequently underwent resorption. A removable partial denture to replace lost maxillary incisors resulted in extensive loss of alveolar bone, making subsequent implant placement more difficult. Similarly the mandibular anterior ridge, although showing limited vertical loss, will require bone augmentation before implants can be placed.

Surgical intervention to expose lesions, with curettage and restoration with amalgam, glass ionomer cement (GIC) or resin composite has been reported in several cases (Moody *et al.* 1990, Iwamatsu-Kobayashi *et al.* 2005, Neely & Gordon 2007, von Arx *et al.* 2009). Neely & Gordon (2007) reported maintenance of a case over a period of 22 years, involving at least eight surgical procedures during that time. Ultimately, however,

resorption recurred and led to progressive tooth loss. There are a number of problems with this conservative approach: CBCT shows that lesions are more widely distributed than appears on conventional radiographs, including buccal and lingual/palatal surfaces; many lesions will be inaccessible; and ongoing periodontal management will be needed because gingival reattachment is unlikely. In addition, lesions tend to recur.

A more aggressive approach involves crown resection with root submergence (Matsui *et al.* 1998), or extraction and replacement with implants or removable prostheses. Whether root resorption ceases following submergence is not known, but this procedure could preserve the alveolar ridge for a long period. However, the presence of submerged roots would complicate the design of a removable partial denture, and fixed partial dentures using abutment teeth with resorbing roots are not recommended. Submerged roots could potentially interfere with subsequent implant placement.

Extraction of teeth and replacement with implant-supported prostheses is a more radical intervention, but may serve the long-term interests of the patient by maintaining healthy bone support. A possible strategy could be a staged approach, with strategic extraction and implant placement to support removable prostheses, with forward planning to anticipate future tooth loss in the prosthesis design. When indicated, full clearance of the remaining teeth could be undertaken, with subsequent placement of implants to support overdentures.

In one case report, resorption ceased after the patient was placed on bisphosphonate therapy for an orthopaedic condition (Iwamatsu-Kobayashi *et al.* 2005), over a period of 6 years. Whilst long-term therapy with bisphosphonates or other resorption-inhibiting drugs may appear extreme, with the potential for the well-known side effects of bisphosphonate therapy, the resorptive condition has a serious impact on patient wellbeing. Hence drug therapy could be considered if appropriate medical advice is obtained. On the basis of only one case report, however, it is probably premature to consider this approach.

The most systematic approach to management is as follows: If lesions are detected early, surgical exposure and restoration with GIC will probably maintain most teeth for many years. As resorption progresses and more teeth are involved, staged extraction and implant placement to support removable partial prostheses should follow. If most teeth are involved and at risk of crown fracture, then full clearance and implant-supported overdentures will become necessary.

In the present case, CBCT was useful in demonstrating the extent of the lesions more precisely, especially on buccal and lingual/palatal surfaces. Clearly, the entire cervical root surface is resorbed rather than a preferential attack on the approximal surfaces as suggested by conventional radiography (Liang *et al.* 2003). We recommend its routine use in evaluating cases of multiple idiopathic cervical resorption.

Disclaimer

Whilst this article has been subjected to Editorial review, the opinions expressed, unless specifically indicated, are those of the author. The views expressed do not necessarily represent best practice, or the views of the IEJ Editorial Board, or of its affiliated Specialist Societies.

References

von Arx T, Schawalder P, Ackerman M, Bosshardt DD (2009) Human and feline invasive cervical resorptions: the missing link? – Presentation of four cases. *Journal of Endodontics* **35**, 904–13.

Cholia SS, Wilson PH, Makdissi J (2005) Multiple idiopathic external apical root resorption: report of four cases. *Dentomaxillofacial Radiology* **34**, 240–6.

DeLaurier A, Boyde A, Jackson B, Horton MA, Price JS (2009) Identifying early osteoclastic resorptive lesions in feline teeth: a model for understanding the origin of multiple idiopathic root resorption. *Journal of Periodontal Research* **44**, 248–57.

Heithersay GS (1999) Invasive cervical resorption following trauma. *Australian Endodontic Journal* **25**, 79–85.

Heithersay GS (2004) Invasive cervical resorption. *Endodontic Topics* **7**, 73–92.

Hopkins R, Adams D (1979) Multiple idiopathic resorption of the teeth. *British Dental Journal* **146**, 309–12.

Iwamatsu-Kobayashi Y, Sato-Kuriwada S, Yamamoto T *et al.* (2005) A case of multiple idiopathic external root resorption: a 6-year follow-up study. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, & Endodontics* **100**, 772–9.

Kerr DA, Courtney RM, Burkes EJ (1970) Multiple idiopathic root resorption. *Oral Surgery, Oral Medicine & Oral Pathology* **29**, 552–65.

Liang H, Burkes EJ, Frederiksen NL (2003) Multiple idiopathic cervical root resorption: systematic review and report of four cases. *Dentomaxillofacial Radiology* **32**, 150–5.

Macdonald-Jankowski D (2005) Multiple idiopathic cervical root resorption most frequently seen in younger females. *Evidence Based Dentistry* **6**, 20.

Matsui K, Echigo S, Kimizuka S, Ito M (1998) A case of multiple idiopathic root resorption – treatment by impacting tooth root. *Journal of the Japanese Stomatological Society* **47**, 68–75. (cited in Iwamatsu-Kobayashi *et al.* 2005).

Moody GH, Muir KF (1991) Multiple idiopathic root resorption A case report and discussion of pathogenesis. *Journal of Clinical Periodontology* **18**, 577–80.

Moody AB, Speculand B, Smith AJ, Basu MK (1990) Multiple idiopathic external resorption of teeth. *International Journal of Oral and Maxillofacial Surgery* **19**, 200–2.

Mueller E, Rony HR (1930) Laboratory studies of an unusual case of resorption. *Journal of the American Dental Association* **17**, 326–34.

Neely AL, Gordon SC (2007) A familial pattern of multiple idiopathic cervical root resorption in a father and son: a 22-year follow-up. *Journal of Periodontology* **78**, 367–71.

Sabeti M, Slots J (2004) Herpesviral-bacterial coinfection in periapical pathosis. *Journal of Endodontics* **30**, 69–72.