



Multiple idiopathic external apical root resorption : A case report and systematic review of literature

1.Dr.Rajeev Talmohite, 2.Dr. Nikhil Mahajan, 3.Dr. Shubhangi Rajbhoj,

4.Dr. Sucheta Vanjari, 5.Dr. Chaitrali Hanumantrao Lakade

¹MDS, Reader, Department of Oral Medicine and Radiology, Sinhgad Dental College And Hospital India

²MDS, Reader, Department of Orthodontics, Sinhgad Dental College And Hospital Pune, India.

³MDS, Reader, Department of Periodontology, Sinhgad Dental College And Hospital Pune, India.

⁴MDS, Lecturer, Department of Periodontology, Sinhgad Dental College And Hospital Pune, India

⁵MDS, Lecturer, Department of Periodontology, Sinhgad Dental College And Hospital Pune, India

Corresponding author

Dr.Rajeev Talmohite, MDS, Reader, Department of Oral Medicine and Radiology, Sinhgad Dental College And Hospital India

rajeev_talmohite@yahoo.co.in

DOI: 10.48047/ecb/2023.12.si4.1702

Abstract: External root resorption of the permanent teeth is a multifactorial process. Well-recognized causes of apical root resorption in permanent teeth include orthodontic therapy, trauma, periapical or periodontal inflammation, tumors, cysts, occlusal stress, impacted teeth, systemic conditions, endocrine imbalances and dietary habits. When none of these causes are present, it is termed idiopathic root resorption which may be either cervical or apical. Multiple idiopathic apical root resorption is a rare condition which is usually detected as an incidental radiographic finding. However, it may cause pain and mobility in severe cases. The aim of this case report was to present a case of multiple idiopathic apical root resorption which is a rare condition in a 28-year-old adult female.

Keywords: Multiple Apical Root Resorption, Idiopathic root resorption, Cervical root resorption.

Introduction

Deciduous teeth are exfoliated as a result of root resorption. This is a physiological process, thought to arise from the pressure of erupting teeth. In contrast, root resorption in permanent teeth is pathological. The process of root resorption involves a complex interaction of inflammatory cells, resorbing cells, hard tissue, cytokines and enzymes such as collagenase, matrix metalloproteinase and cysteine proteinase. Localized damage or loss of periodontal ligament renders the denuded cementum surface chemotactic to clastic cells such as osteoclasts, macrophages and monocytes. This can result in root resorption. In cases where multiple teeth are involved, Lo'e and Waerhaug¹ have suggested that the dental tissues become part of the osseous system and thus subject to remodelling. Several different types of pathological root resorption are recognized (Table 1)² and it can be difficult for the clinician to differentiate between them. Resorption can be broadly classified as either internal or external, and usually involves one tooth. External root resorption can further be defined according to the site affected as cervical, apical or intraradicular.

Table 1 : classification and etiological factors in pathological root resorption

Site	Type	Aetiology
Internal		Trauma Infection
External	Surface	Trauma
	Inflammatory	Trauma Infection
	Replacement (Ankylosis)	Avulsion and re-implantation 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 Intracoronary tooth bleaching
	Idiopathic	Unknown

Idiopathic external root resorption has been found to be an infrequent phenomenon that affects either or both apical and cervical regions of one or several teeth, but most commonly occurring in the apical region. The etiology of both idiopathic internal and external root resorption remains elusive. It may occur spontaneously, in the absence of either local or systemic factors. External

root resorption which develops in the absence of a plausible cause is termed idiopathic. By definition, idiopathic external root resorption is a diagnosis of exclusion.

The first reported case of idiopathic cervical root resorption was by Mueller and Rony in 1930³. Recently, systematic review has become increasingly important in current evidence-based research. The goal of a systematic review is to minimize both bias and error, serving as an aid to clinical reasoning. The aim of this report was to describe the history, clinical findings and radiographic appearance of multiple idiopathic apical root resorption and to undertake a systematic literature review of this condition.

Case report:

A 28yrs old female referred to our institute Dr. D.Y. Patil Dental College and Hospital, Navi Mumbai in the Dept. of Oral Medicine and Radiology with the chief complaint of pain and increase in mobility in relation to mandibular right first molar teeth. There was no history of trauma, hospitalization or medical endocrine and systemic disease and orthodontic therapy.

Clinical and Dental examination revealed tooth 46 was grade 3⁰ mobile. Periodontal probing depth was 3mm which was normal. There was no restoration or carious lesion seen. Oral hygiene was relatively acceptable. Normal soft tissues were noted without any supra or subgingival calculi or abnormal pocketing.

Occlusion demonstrated class I molar relationship with normal overjet and overbite. No occlusal interferences or detectable tooth mobility was found. Involved teeth responded normally to both electrical and heat pulp tester. Percussion and palpation were unremarkable.

Intra-oral periapical radiograph of mandibular right posterior quadrant was taken which showed complete root resorption of distal root of 46 and thinning of mesial root. Saucer shaped bone loss noted just below the crown of 46. Tooth 47 showed blunting apical 3rd of the root suggestive of root resorption of distal root . (Fig.1)



Fig. Intraoral IOPA of 46 region

On OPG similar findings noted in 36, 37 region, hence IOPA of all four quadrants were taken. IOPA of left mandibular first molar also showed similar findings i.e resorption of distal root of 36 and Thinning of mesial root. 38 and 48 were unerupted and showed incomplete formation of roots suggestive of pathological resorption or incomplete development of roots.



FIG 2: OPG



FIG3: IOPA OF 36 REGION



FIG: HAND-WRIST RADIOGRAPH

Hand–wrist radiograph was taken to check for changes in the bone as in osteoporosis and hyperparathyroidism but no detectable changes evident.

CBVI added information on the third dimension of the lesion by showing presence of mesio-lingual root in 36 and complete absence of the mesio-buccal and distal roots, similar findings was seen in 46 where complete loss of distal root was noted and thin friable mesial roots were seen.

Haematological investigations including complete blood count as well as serum calcium, serum phosphorus and serum alkaline phosphatase advised and found to be within the normal range, so endocrine diseases such as Hyperparathyroidism, Hypoparathyroidism, hypophosphatemia, hyperphosphatemia and Paget's disease were ruled out. In Papillon-Lefevre syndrome, the history of premature tooth loss associated with hyperkeratosis is remarkable, of which none of them were recognized in the patient.

Based on history, Clinical examination, radiographic findings and haematological investigation diagnosis of multiple idiopathic root resorption was made. Teeth 46 and 36 were extracted under local anesthesia in Oral Surgery department. The patient was asked to follow proper oral hygiene instructions and periodic follow up was suggested. In six month follow up no detectable changes were seen in any other teeth, hence a fixed prosthesis was given for replacement of 46 and 36.

Discussion

With no absolute etiological factor identified we considered this as a case of multiple idiopathic apical root resorption. All teeth had vital pulps and there was no periodontal or periapical inflammation. Multiple root resorption was found incidentally in the panoramic view and the patient was totally asymptomatic. No local etiologic factor was detected and clinical appearance of the teeth and periodontium were normal.

In contrast to the higher clinical incidence of pathological root resorption in females, some published papers reported a predilection of idiopathic apical root resorption for men. In attempt to rule out the possible role of genetic as an etiologic factor, panoramic radiographs were prepared for the patient's parents and siblings which were inconclusive. Laboratory test results indicated no abnormality. Differentiated blood count, serum calcium, serum phosphorus and serum alkaline phosphatase were within normal range. Due to the severe degree of resorption, grade 3⁰ mobility of 36 and 46, extraction of same teeth were advised.

Few cases of multiple idiopathic apical root resorption (MIARR) exist in the literature .The first well documented report was in 1930 and since then more cases were presented.

Muller and Rony³, who postulated that external root resorption has several other causes (Table 1). It appears to be a relatively common incidental radiographic finding in isolated teeth, but uncommon in a generalized form. Local causes are thought to be the most frequent, caused by excessive pressure and inflammation. Mechanisms include large orthodontic forces, occlusal trauma, impacted teeth, re-implanted teeth, periradicular infection or tooth bleaching. Many systemic abnormalities have been implicated, which include hormonal disturbances⁴,

Hypophosphatasia⁵, hyperparathyroidism⁶, Paget's disease⁷, Papillon-Lefevre syndrome⁸, renal disease⁹, hepatic disease¹⁰ and bone dysplasia¹¹. It should be noted that arrested root development in radiotherapy¹², dental dysplasia¹³, hypothyroidism¹⁴ and Stevens-Johnson syndrome¹⁵ can have a similar radiographic presentation to external root resorption. Stafne and Slocumb¹⁶ in a study of 179 root resorption cases failed to find any definite associations with systemic disease. Newman reported a similar finding in a study of 47 individuals with idiopathic root resorption. In contrast, Gunraj¹⁷ has suggested that changes in the host cellular immune system may be implicated as a possible etiologic factor.

The case presented in this article did not have any obvious local or systemic causative factors for their root resorption patterns.

A recent literature search for Multiple idiopathic external apical root resorption (MIEARR) identified 11 published case reports describing 15 patients^{18,19,20,21,22,23,24,25,26} to which we have added our case presented in this article. These 16 cases indicate that MIEARR affects a wide age range of patients, from 14 years to 39 years old. In contrast to multiple idiopathic external cervical root resorption (MIECRR), males appear to be more frequently affected by MIEARR than females, with a male:female ratio of 11:5. In addition, MIEARR appears to have a predilection for premolar and molar regions. In contrast, there was no site specific relationship reported for MIECRR. Other common features of the MIEARR cases appear to be:

- Normal clinical appearance of teeth and periodontal tissues;
- Root resorption associated with vital teeth and endodontically treated teeth;
- Lack of periodontal and periradicular inflammation;
- Alveolar bone levels within normal limits;
- Absence of local aetiological factors;
- Patients asymptomatic until very late in the pathological process where increased tooth mobility reported;
- Commonly found as an incidental finding on radiographs; and
- Intramaxillary and intermaxillary symmetrical pattern of root resorption.

With no absolute aetiological factors identified, treatment of MIEARR depends largely on the presenting symptoms and the extent and the severity of root resorption. The usual treatment is the extraction of teeth of poor prognosis and long-term monitoring of the remaining dentition using serial radiographs, periodontal measures, sensibility tests or patient symptoms. This was the option chosen for the majority of our cases. Edentulous saddles may be restored using adhesive or conventional fixed bridges, removable partial dentures or osseointegrated implants. Abutment teeth must be carefully assessed for root resorption. The success of long-term osseointegration in sites where root resorption has been active is unknown. In severe cases the only option available may be extraction of all teeth and construction of a complete denture.

A more invasive approach involves endodontic treatment of the affected teeth. This has been well documented for inflammatory root resorption, where calcium hydroxide is the current intraradicular medicament of choice²⁷. However, a common finding in MIEARR is that teeth remain vital even after extensive root resorption. It has been suggested that Ledermix (Triamcinolone acetonide and Demeclocycline calcium; Lederle Laboratories, UK)²⁸ inhibits the

proliferation of dentinoclasts and it may prove effective when mixed with calcium hydroxide. An experimental approach may be calcitonin as an intracanal medicament. Calcitonin inhibits osteoclast motility and retraction and could be potentially useful in modifying the resorptive process. Postlethwaite and Hamilton²⁹ planned to extirpate the pulps and apply intraradicular calcium hydroxide to half of the affected teeth in their case of MIEARR. Unfortunately they have not reported the outcome. Rivera and Walton stated that MIEARR does not seem to be mediated by or have its source from the dental pulp. Therefore, in the absence of pulpal symptoms, endodontic therapy cannot be indicated for MIEARR. In the future bioactive molecules capable of modifying the process of root resorption may become available which target the periradicular resorptive process³⁰.

Conclusions

From the published literature and these four new cases, MIEARR affects a wide age range of individuals, with males affected more frequently than females. There appears to be a predilection for premolar and molar teeth, in a symmetrical pattern of expression. The current mainstay of management for affected individuals remains long-term monitoring, but occlusal therapy and restorative dental treatment have a role in appropriate circumstances.

However, endodontic therapy is not indicated at the present time. The aetiology of MIEARR remains unknown, but it is hoped that the discovery of the molecular and cellular mechanism of root resorption will yield new methods of treatment.

REFERENCES

1. Lo'e H, Waerhaug J. Experimental replantation of teeth in dogs and monkeys. *Arch Oral Biol* 1961; 3: 176–184.
2. Bakland LK. Root resorption. *Dent Clin North Am* 1992; 36: 491–507.
3. Muller E, Rony HR. Laboratory studies of unusual case of resorption. *J Am Dent Assoc* 1930; 17: 326–334.
4. George DI, Miller RL. Idiopathic resorption of teeth. *Am J Orthod* 1986; 89: 13–20. Newman WG.
5. Possible etiologic factors in external root resorption. *Am J Orthod* 1975; 67: 522–539.
6. Belanger GK, Coke JM. Idiopathic external root resorption of the entire permanent dentition: report of a case. *J Dent Child* 1985; 52: 359–363.
7. Smith BJ, Eveson JW. Paget's disease of bone with particular reference to dentistry. *J Oral Pathol* 1981; 10: 233–247.
8. Ru'diger S, Berglundh T. Root resorption and signs of repair in Papillon-Lefe`vre syndrome: a case study. *Acta Odontol Scand* 1999; 57: 221–224.
9. Moskow BS. Periodontal manifestations of hyperoxaluria and oxalosis. *J Periodontol* 1989; 60: 271–278.
10. Pankhurst CL, Eley BM, Moniz C. Multiple idiopathic external root resorption: a case report. *Oral Surg Oral Med Oral Pathol* 1988; 65: 754–756.
11. Olsen CB, Tangchaitrong K, Chippendale I, Graham Dahl HM, Stockigt JR. Tooth root resorption associated with familial bone dysplasia affecting mother and daughter. *Pediatr Dent* 1999; 21: 363–367.
12. Pietokovski J, Menchel J. Tooth dwarfism and root underdevelopment following irradiation. *Oral Surg Oral Med Oral Pathol* 1966; 22: 95–99.
13. Logan J. Dentinal dysplasia. *Oral Surg Oral Med Oral Pathol* 1962; 15: 317–333.

14. Sunde OE. Dental changes in a patient with hypoparathyroidism. *Br Dent J* 1961; 111: 112–117.
15. DeMan K. Abnormal root development probably due to erythema multiformae (Stevens-Johnson syndrome). *Int J Oral Surg* 1979; 8: 381–385.
16. Stafne EC, Slocumb CH. Idiopathic resorption of teeth. *Am J Orthod Oral Surg* 1944; 30: 41–49.
17. Gunraj MN. Dental root resorption. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1999; 88: 647–653.
18. Belanger GK, Coke JM. Idiopathic external root resorption of the entire permanent dentition: report of a case. *J Dent Child* 1985; 52: 359–363.
19. Pankhurst CL, Eley BM, Moniz C. Multiple idiopathic external root resorption: a case report. *Oral Surg Oral Med Oral Pathol* 1988; 65: 754–756.
20. Soni NN, La Velle WE. Idiopathic root resorption. *Oral Surg Oral Med Oral Pathol* 1970; 29: 387–389.
21. Postlethwaite KR, Hamilton M. Multiple idiopathic external root resorption. *Oral Surg Oral Med Oral Pathol* 1989; 68: 640–643.
22. Yusof WZ, Ghazali MN. Multiple external root resorption. *J Am Dent Assoc* 1989; 118: 453–455.
23. Rivera EM, Walton RE. Extensive idiopathic apical root resorption: a case report. *Oral Surg Oral Med Oral Pathol* 1994; 78: 673–677.
24. Di Domizio P, Orsini G, Scarano A, Piattelli A. Idiopathic root resorption: report of a case. *J Endod* 2000; 26: 299–300.
25. Cowie P, Wright BA. Multiple idiopathic root resorption. *J Can Dent Assoc* 1981; 47: 111–112.
26. Brooks JK. Multiple idiopathic apical external root resorption. *Gen Dent* 1986; 34: 385–386.
27. Trope M. Clinical management of the avulsed tooth. *Dent Clin North Am* 1995; 39: 93–112.
28. Pierce A, Heithersay GS, Lindskog S. Evidence for direct inhibition of dentinoclasts by a corticosteroid/antibiotic endodontic paste. *Endod Dent Traumatol* 1988; 4: 44–45.
29. Postlethwaite KR, Hamilton M. Multiple idiopathic external root resorption. *Oral Surg Oral Med Oral Pathol* 1989; 68: 640–643.
30. Goldberg M, Six N, Decup F, Lasfargues JJ, Salih E, Tompkins K, et al. Bioactive molecules and the future of pulp therapy. *Am J Dent* 2003; 16: 66–76.