Extensive Idiopathic External Apical Root Resorption on a 13 Year Old Child

Hegde S * / Shubha AB ** / Kakti A *** / Rao BD ****

External root resorption in permanent teeth can occur as a result of a multitude of local and systemic factors. Root resorption that is idiopathic or occurring without any identifiable underlying cause is an unusual phenomenon, especially in children. This article describes a rare case of extensive external apical root resorption affecting multiple teeth and occurring concomitantly with localized periodontitis in a 13 year-old child. No significant systemic, local or familial findings could be identified as a plausible cause for the root resorption.

Keywords: *idiopathic root resorption, external root resorption, localized periodontitis*
J Clin Pediatr Dent 36(3): 289–292, 2012

INTRODUCTION

External root resorption (ERR) in the primary dentition is a physiological process that is essential for the timely exfoliation of primary teeth and their eventual replacement by permanent successor teeth. This process is stimulated by forces generated during eruption of the permanent teeth, the increase in masticatory forces that accompanies growth and the inherent resorptive potential of primary teeth.[1]

In contrast, root resorption in permanent teeth is infrequent and pathological, and has been related to occlusal trauma, heavy orthodontic forces, replantation, ankylosis, cysts, tumors, inflammatory processes of pulp or periodontal origin, and tooth bleaching procedures.[2] Many systemic abnormalities have also been implicated, including hormonal disturbances,[3] hypophosphatasia,[4] hyperparathyroidism,[5] Paget's disease,[6] Papillon-Lefèvre syndrome,[7] renal disease,[8] hepatic disease[9] and bone dysplasia.[10]

The process of root resorption is an elaborate interaction of inflammatory cells, resorbing cells, hard tissue, cytokines and enzymes such as collagenase, matrix metalloproteinase and cysteine proteinase.[11] The periodontal ligament normally acts as a barrier between the alveolar bone and cementum. Any localized damage to or loss of this periodontal ligament renders the denuded cementum surface chemotactic to clastic cells such as osteoclasts, macrophages and monocytes[11,12] which can result in root resorption.

An uncommon presentation is that of external root resorption with no identifiable underlying cause, local or systemic, what is referred to as idiopathic root resorption. This form of root resorption can exhibit a familial trait, with siblings of similar age frequently affected. It may be associated with early loss of predecessors of the affected teeth.[4,13] Most cases exhibit a certain degree of apical resorption not attributable to any concrete cause; severe cases may be associated with mobility or loss of the permanent teeth.[5,9] The clinical forms of idiopathic external root resorption are no different from those of ERR of known etiology and more cases have been reported in the dental literature of multiple idiopathic external cervical root resorption (MIECRR)[14,15] than of multiple idiopathic external apical root resorption (MIEARR). ERR is usually a finding of routine radiographic explorations, with the exception of highly aggressive and advanced presentations where clinical signs in the form of pulp involvement or even tooth mobility can be observed.[16]

In this article we present a case of extensive idiopathic external apical root resorption affecting multiple teeth and occurring concomitantly with localized periodontitis in a 13 year-old child.

Case Report

A 13 year-old boy visited the Department of Pediatric Dentistry, Pacific Dental College, Udaipur, Rajasthan, India, with a complaint of loss of two of his upper front teeth, following a mild traumatic incident two days earlier. The patient did not bring with him the said teeth. From the

* Sapna Hegde, MDS, Professor and Head, Department of Pediatric Dentistry, Pacific Dental College and Hospital, Udaipur, India.
** Shubha A B, MDS, Reader, Department of Pediatric Dentistry, Pacific Dental College and Hospital, Udaipur, India.
*** Ateet Kakti, MDS, Former postgraduate student, Department of Pediatric Dentistry, Pacific Dental College and Hospital, Udaipur, India.
**** Dinesh Rao B, MDS, Professor, Department of Pediatric Dentistry, Pacific Dental college and Hospital, Udaipur, India.

Send all correspondence to: Dr. Sapna Hegde, Professor and Head, Department of Paediatric Dentistry, Pacific Dental College and Hospital, Debari, Udaipur - 313024, Rajasthan, India

Tel: +91 98281 44002
Fax: + 91 294 2491508

E-mail: drsapnahegde@yahoo.co.in

history it was inferred that the teeth had become progressively mobile prior to the traumatic injury. There was no history of early exfoliation of the primary dentition or of any previous orthodontic or periodontal treatment or any traumatic injury earlier to the current one. The previous medical and family histories were non-contributory.

The child appeared well nourished with a normal build. Intraoral examination revealed a permanent dentition with all teeth erupted up to the second molars and a rather unsightly space in the anterior region owing to loss of the maxillary central incisors. All teeth were discolored by mild fluorosis. Occlusal analysis revealed no pathological intermaxillary interferences or worn surfaces. The oral hygiene was rather unsatisfactory, with plaque deposits evident on the mandibular incisors and molars. Additionally, the maxillary lateral incisors displayed Grade II mobility resulting in pathologic tooth migration, while the mandibular central incisors exhibited Grade I mobility. Periodontal exploration indicated the presence of severe gingival inflammation and gingival recession in relation to the mandibular incisors (Figure 1). Radiography (Figures 2 and 3) revealed extensive alveolar bone loss in the region of the incisors, and absence of the lamina dura as well as moderate loss of alveolar bone in the molar areas.

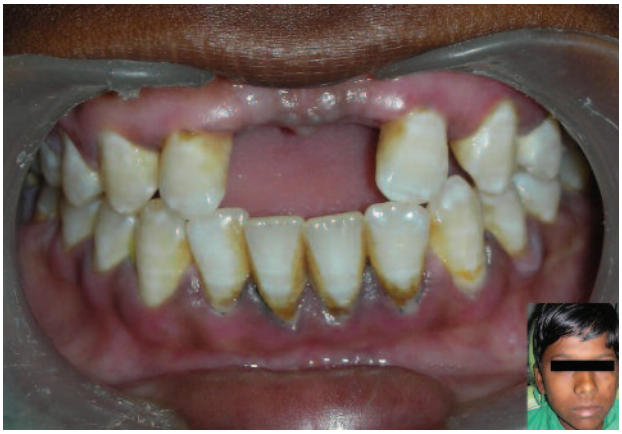

Figure 1. Intraoral photograph of patient (inset) showing missing maxillary central and migrated lateral incisors, gingival inflammation and recession affecting mandibular incisors

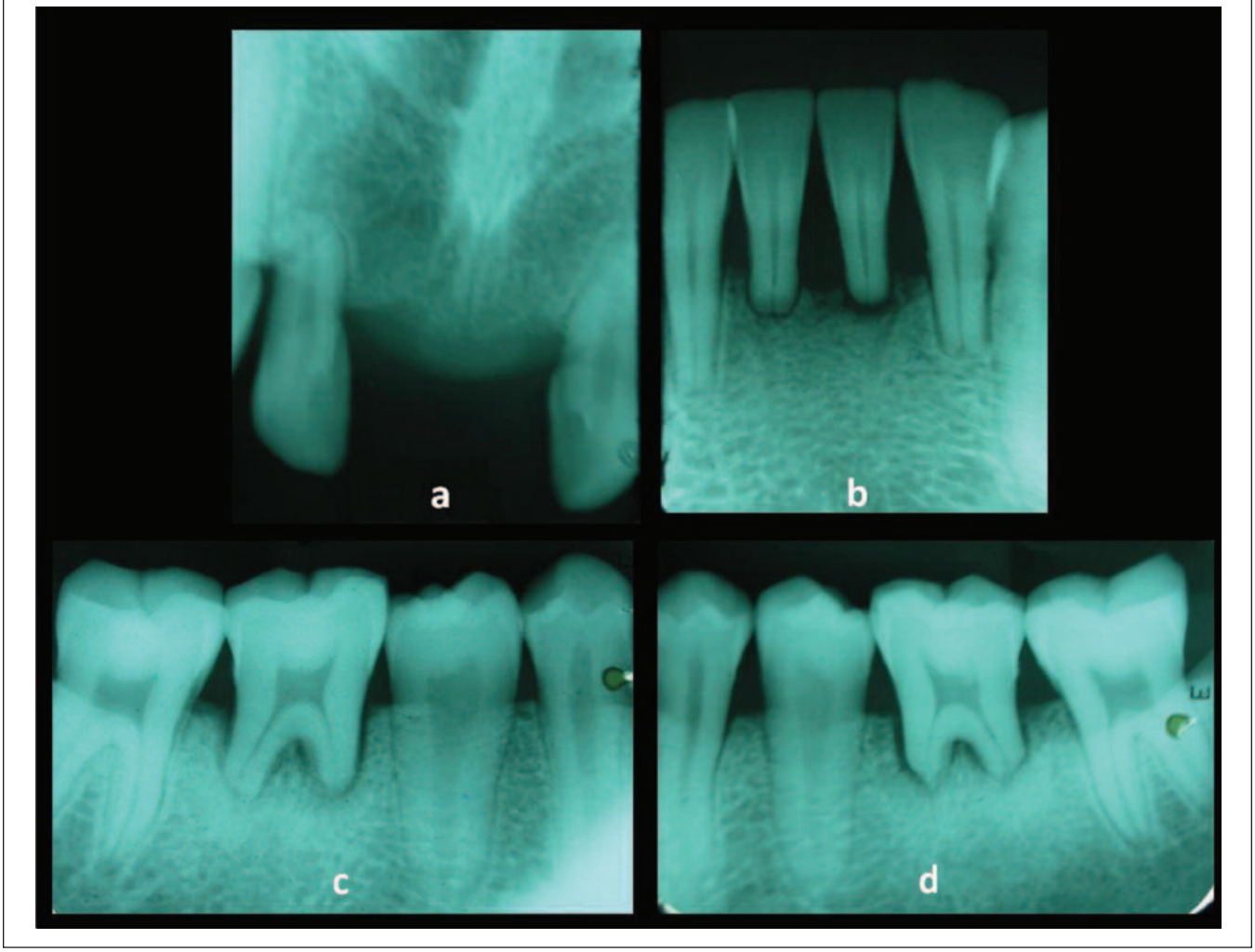

Figure 2. Intraoral periapical radiographs exhibiting apical root resorption and alveolar bone loss involving (a) 12 and 22, (b) 31, 32, 41 and 42, (c) 46 and (d) 36

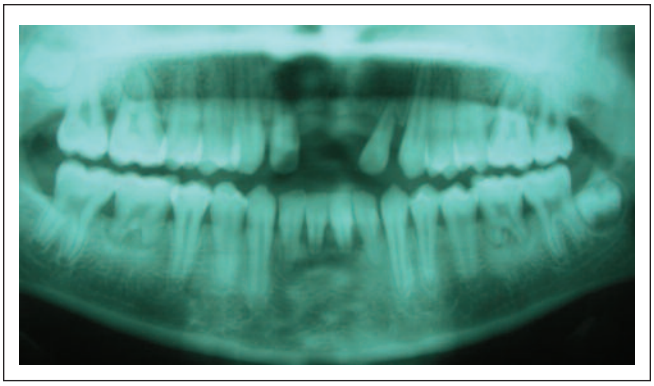

Figure 3. Orthopantamograph revealing symmetrical pattern of apical root resorption affecting multiple teeth.

Although these findings pointed to localized periodontitis, orthopantamograph and full-mouth intraoral radiographs (Figures 2 and 3) also highlighted a symmetrical pattern of moderate to severe apical root resorption of the 12, 22, 31, 32, 36, 41, 42 and 46, resulting in gradual shortening and rounding of the roots. In the mandibular first molars the root resorption had progressed to almost the furcation. The alveolar bone did not present any obvious signs of resorption except for some crestal bone loss in the region of the incisors and molars. Complete sockets of the lost teeth could not be appreciated radiographically. Electric pulp testing indicated that all teeth were vital. Bacterial culture of the gingival exudate from the affected areas did not yield any significant finding, except for *P. gingivalis*, which was consistent with the periodontitis observed. Hence root resorption was now suspected to be the major contributing factor to the tooth loss, with the trauma only providing the reason for sudden loss.

Another interesting finding was that the maxillary canine, all premolar and 2nd molar roots were almost completely formed which could indicate that the patient had erupted most of his teeth well before the standard age of eruption. And, although the premolars and mandibular canines also appeared to show the initial signs of resorption in the form of a slight blunting of the root apices, making a clear-cut distinction was difficult at the time since a few of these teeth could have been undergoing apical closure.

Some features of the present case (bone loss, loss of teeth following minor trauma) might suggest Papillon-Lefévre syndrome, but the absence of palmar-plantar hyperkeratosis ruled out this condition. Skull and long-bone radiographs were normal, thus excluding Paget's disease and bone dysplasia.

In view of the absence of local factors that could have contributed to this extensive root resorption, comprehensive hematological and urinary examinations were carried out to detect the presence of or to exclude any systemic conditions including vitamin deficiencies. The examination ruled out any avitaminosis and normal values were obtained for the blood count except for mild eosinophilia. The serum alkaline phosphatase level was within normal limits. This finding together with the absence of significant history or clinical and radiographic skull and long-bone findings excluded hypophosphatasia. Inorganic phosphorus, C-reactive protein, rheumatoid factor, antinuclear antibodies, thyroid and parathyroid hormone levels were within normal limits.

Results of liver function tests (serum creatinine, total serum bilirubin, SGOT, SGPT) were normal as were the parathyroid hormone, 24-hour urine calcium and serum spot calcium levels, thereby excluding hepatic disease, hyperparathyroidism and renal disease. The urinary pH was found to be normal.

With no plausible local or systemic factors in an otherwise healthy patient with normal appearance and asymptomatic vital teeth and no significant family history, the present case was diagnosed as multiple idiopathic external apical root resorption occurring concomitantly with localized periodontitis.

The patient was treated by a thorough oral prophylaxis and given instructions and training in oral hygiene maintenance. The edentulous area in the maxillary anterior region was restored using a removable partial denture without retentive clasps. Definitive restorations were postponed until cessation of the resorption can be ascertained from serial radiographs. The patient currently remains on periodic review and maintenance therapy.

DISCUSSION

The term 'idiopathic external root resorption' was introduced by Belanger and Coke[5] to describe those situations where root resorption is not associated with any plausible local or systemic cause. Our search of the literature reveals that the first such case was published in 1930 by Mueller and Rony.[17] Other cases with variable degrees of involvement have been described since then.[12,18]

Idiopathic external root resorption is an unusual phenomenon. Its etiology continues to remain elusive. Pinska and Jarzynka[19] were the first to suggest genetic susceptibility in their report of a family with generalized root resorption. Newman[4] found a tentative genetic association in his study of 37 families, but the results were not statistically significant because of the small sample size. The most compelling evidence for a genetic association with MIEARR came from Saravia *et al*[13] who described 14-year-old monozygotic twins presenting with identical clinical and radiographic patterns of MIEARR. Gunraj[12] however, has suggested that changes in the host cellular immune system may be implicated. More recently, it has been found that homozygosity for the IL-1B allele leads to a 5.6-fold increase in individual susceptibility to significant root resorption.[20] Additionally, a gene encoding for a TNF receptor has also been implicated in the process.[21]

Idiopathic external root resorption is, by definition, a diagnosis of exclusion. Hence, the diagnostic procedure should focus on the exclusion of all local factors that are contributory to ERR. All such factors were ruled out in our patient, along with systemic disorders associated with phosphorus-calcium metabolic alterations. The family history was not significant.

From the available literature it may be inferred that MIEARR is less common than MIECRR and affects males more frequently, with a predilection for molar and premolar regions.[5,9,13,22] Although most reported cases involve adults,[22,23] a few have described the condition in children as well.[18,24]

Since there is no identifiable cause, the treatment of MIEARR depends largely on the presenting symptoms and the extent and severity of root resorption. Treatment usually consists of extraction of teeth with poor prognosis and long-term monitoring of the remaining dentition. Lost teeth may be replaced using fixed or removable prosthesis or osteointegrated implants after carefully assessing the abutment teeth for root resorption.[23] However, dental implant-supported restorations must be undertaken only after completion of vertical facial growth. In our case, the edentulous space created by the loss of the maxillary central incisors was restored by a removable partial denture without clasps to prevent any excessive forces on the abutment teeth.

Adjustment of occlusal interferences if any may be indicated. A more invasive approach involves endodontic treatment of the affected teeth. It has been suggested that Ledermix which inhibits the proliferation of dentinoclasts[25] may prove effective as an intracanal medicament when mixed with calcium hydroxide. Another approach may involve the use of calcitonin[26] as an intracanal medicament. Calcitonin inhibits osteoclast motility and retraction and could be potentially useful in modifying the resorptive process. However, a common finding in MIEARR is that teeth remain vital even after extensive root resorption as was observed in the present case. Also, MIEARR does not seem to be mediated by or have its source from the dental pulp.[22] Therefore, in the absence of pulpal symptoms, endodontic therapy may not be indicated for MIEARR[23] and hence was not considered as a treatment option in the present case.

CONCLUSION

External root resorption of permanent teeth can be a result of several factors, local and systemic. Diagnosing root resorption as of idiopathic origin should, therefore, focus primarily on the exclusion of all factors that are otherwise contributory to ERR. We believe that until such time that definitive therapy for this condition has been firmly established by further research, appropriate management should involve only stabilization of the condition and avoidance of factors likely to compound the problem.

REFERENCES

1. Gonzalvo L, Ruiz de Temino P. Clinical review of root resorption. Endod, 10: 113–142, 1992.
2. Goultschin J, Nitzan D, Azaz B. Root resorption: review and discussion. Oral Surg Oral Med Oral Pathol, 54: 586–590, 1982.
3. George DI Jr, Miller RL. Idiopathic resorption of teeth: a report of three cases. Am J Orthod, 89: 13–20, 1986.
4. Newman WG. Possible etiologic factors in external root resorption. Am J Orthod, 67: 522–539, 1975.
5. Belanger GK, Coke JM. Idiopathic external root resorption of the entire permanent dentition: report of a case. J Dent Child, 52: 359–363, 1985.
6. Smith BJ, Eveson JW. Paget's disease of bone with particular reference to dentistry. J Oral Pathol, 10: 233–247, 1981.
7. Rudiger S, Berglundh T. Root resorption and signs of repair in Papillon-Lefe`vre syndrome: a case study. Acta Odontol Scand, 57: 221–224, 1999.
8. Moskow BS. Periodontal manifestations of hyperoxaluria and oxalosis. J Periodontol, 60: 271–278, 1989.
9. Pankhurst CL, Eley BM, Miniz C. Multiple idiopathic external root resorption: a case report. Oral Surg Oral Med Oral Pathol, 65: 754–756, 1988.
10. Olsen CB, Tangchaitrong K, Chippendale I, Graham HK, Dahl HM, Stockigt JR. Tooth root resorption associated with familial bone dysplasia affecting mother and daughter. Pediatr Dent, 21: 363–367, 1999.
11. Ne RF, Witherspoon DE, Gutmann JL. Tooth resorption. Quintessence Int, 30: 9–25, 1999.
12. Gunraj MN. Dental root resorption. Oral Surg Oral Med Oral Pathol Oral Radiol Endod, 88: 647–653, 1999.
13. Saravia ME, Meyer ML. Multiple idiopathic root resorption in monozygotic twins: case report. Pediatr Dent, 11: 76–78, 1989.
14. Liang H, Burkes EJ, Frederiksen NL. Multiple idiopathic cervical root resorption: systematic review and report of four cases. Dentomaxillofac Radiol, 32: 150–155, 2003.
15. Iwamatsu-Kobayashi Y, Satoh-Kuriwada S, Yamamoto T, Hirata M, Toyoda J, Endo H, Kindaichi K, Komatsu M. A case of multiple idiopathic external root resorption: a 6-year follow-up study. Oral Surg Oral Med Oral Pathol Oral Radiol Endod, 100: 772–779, 2005.
16. Lydiatt DD, Hollins RR, Peterson G. Multiple idiopathic root resorption: diagnostic considerations. Oral Surg Oral Med Oral Pathol, 67: 208–210, 1989.
17. Mueller E, Rony HR. Laboratory studies of an unusual case of resorption. J Am Dent Assoc, 17: 326–334, 1930.
18. Snelgrove RA. Generalized idiopathic apical resorption as an incidental finding in an adolescent. A case history. Dent Update, 22: 276–278, 1995.
19. Pinska E, Jarzynka W. Spontaneous resorption of the roots of all permanent teeth as a familial disease. Czas Stomatol, 19: 161–165, 1966.
20. Al-Qawasmi RA, Hartsfield JK Jr, Everett ET, Flury L, Liu L, Foroud TM et al. Genetic predisposition to external apical root resorption. Am J Orthod Dentofacial Orthop, 123: 242-252, 2003.
21. Al Al-Qawasmi RA, Hartsfield JK Jr, Everett ET, Flury L, Liu L, Foroud TM et al. Genetic predisposition to external apical root resorption in orthodontic patients: linkage of chromosome-18 marker. J Dent Res, 82: 356-360, 2003.
22. Rivera EM, Walton RE. Extensive idiopathic apical root resorption. A case report. Oral Surg Oral Med Oral Pathol, 78: 673–677, 1994.
23. Cholia SS, Wilson PHR, Makdissi J. Multiple idiopathic external apical root resorption: report of four cases. Dentomaxillofac Radiol, 34: 240–246, 2005.
24. McMullin A, Fleming PS, DiBiase AT. Ideopathic generalized apical root resorption: a report of three cases. Int J Paediatr Dent, 18: 312–316, 2008.
25. Pierce A, Heithersay G, Lindskog S. Evidence for direct inhibition of dentinoclasts by a corticosteroid/antibiotic endodontic paste. Endod Dent Traumatol, 4: 44–45, 1988.
26. Pierce A, Berg JO, Lindskog S. Calcitonin as an alternative therapy in the treatment of root resorption. J Endod, 14: 459–464, 1988.