



Multiple Idiopathic Apical Root Resorption Involving All Permanent Teeth: A Case Report

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ABSTRACT

Multiple idiopathic apical root resorption (MIARR) is a rare condition that affects multiple teeth of unknown etiology. This phenomenon is usually identified in clinical examinations or routine radiographs, is painless, and is sometimes accompanied by tooth mobility. Involvement of all permanent dentition is extremely rare. It is mostly seen in females and is usually asymptomatic. Diagnosis and treatment of these resorptive lesions are challenging for clinicians, and usually, the only available treatment is the extraction of all involved teeth. The prognosis for affected teeth is often poor; no treatment or prevention method is known for this phenomenon. The present report describes a 28-year-old male patient who had all his permanent teeth involved. This case is unique in that all teeth are involved in the absence of any systemic disease or local factors.

Keywords: Idiopathic root resorption; Multiple root resorption; Root resorption.

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Introduction

External apical root resorption is a rare condition that has been reported both single and multiple. Pathologic root resorption is related to several systemic and local factors. Orthodontic treatments, trauma, periapical or periodontal inflammation, tumors, cysts, occlusal stress, impacted teeth, supernumerary, transplantation, and reimplantation are local factors and systemic causes include hyperparathyroidism, hypoparathyroidism, hypoparaphosphatemia, Gaucher's disease, Paget's bone disease, Goltz syndrome, Papillon-Lefèvre syndrome, anachoresis, Turner's syndrome have been hypothesized to alter osteoclastic activity and lead to external root resorption [1]. The idiopathic type refers to the absence of identified etiologic factors. Shortness of the roots of the teeth has also been observed in disorders such as scleroderma, Down syndrome and Laurence–Moon–Bardet–Biedl syndrome [2,3]. Genetic variants linked specifically with root resorption, but not overt systemic/syndromic manifestations, have been rare to date. Altered Osteoclast/Odontoclast Activity is considered the main cause of these resorptive lesions [4].

The most common clinical manifestation of idiopathic root resorption is that patients are asymptomatic and sometimes complain of tooth mobility. These resorptive lesions are usually accidental radiographic findings and are detected during a routine examination. Two types of idiopathic root resorption have been identified, including cervical and apical types. Cervical root resorption begins in the cervical region of the tooth and progresses to the pulp. While apical root resorption begins in the apical area of the tooth and progresses to the coronal region, eventually leading to gradually rounded and shortened roots [5]. In a review study, Cholia et al. reported that idiopathic root resorption of the maxillary molars is slightly more common than in the mandible with single-tooth involvement, although these differences were not statistically significant. Research showed an inclination to females [6]. The first case of MIARR was reported in 1930 [7]. The root resorption process involves complex reactions between inflammatory cells, resorptive cells, hard tissue, and enzymes including collagenase, matrix metalloproteinase, and proteases. The periodontal ligament is a special connective tissue that acts as a barrier between the alveolar bone and the cementum. Local damage or PDL loss causes the damaged surface of the cementum to act as a chemotactic agent for clastic cells, including osteoclasts, macrophages, and monocytes, which in turn will eventually lead to root resorption

[6]. Root resorption is defined as a pathological process initiated by specific clastic cells. Etiological factors influencing external root resorption include biomechanical forces, surgery, chemicals, trauma, endodontic microorganisms, and their toxins, developmental defects, neoplasms, and hormonal disorders [8,9]. Mavridou et al. found that gender is not a risk factor for root resorption, and this phenomenon is strongly dependent on the tooth position [10]. The most common manifestations of multiple idiopathic root resorption (MIRR) are the normal clinical view of the teeth and their supporting structures, and the incidence of resorption in the vital teeth is similar to that of endodontically treated teeth, and there is generally no infiltration into the pulp space. The absence of periodontal inflammation and normal alveolar bone height are other common clinical findings. The asymptomatic patient usually reports tooth mobility in the late stages of the disease [8].

MIRR does not seem to be related to the patient's gender, age, race, genetic predisposition, or medical history. In these patients, it is difficult to predict which teeth will be involved in the future. The number of affected teeth usually varies from 3 to 31 teeth and an average of 14 teeth will be involved. These lesions will progress regardless of the type of intervention, whether surgical or non-surgical [11]. In most MICRR lesions, the resorption cavity is filled with a mass of fibrovascular tissue and a large number of monocytes and macrophages. Besides, multinucleated osteoclasts are present in Howship lacunae [12,13]. The present report aims to describe a male patient with MIARR and apical root resorption was found in all of his teeth. This case is unique in the sense that generalized apical root resorption was present in all teeth in the absence of any systemic disease or history of trauma.

Case Presentation

The patient was a 28-year-old male who was referred to the Department of Oral Medicine of the Dental School of Tehran University of Medical Sciences, with complaints of esthetic and chewing problems. The patient reported mobility in some teeth. Intraoral and extraoral examinations were performed. Intraoral examination showed 27 permanent teeth. All third molars were extracted and were not missed, and the left maxillary central tooth was exfoliated due to severe mobility (Figure 1). The patient did not report a history of endodontic, orthodontic, or periodontal treatment. There were no reports of a history of trauma to the jaw or teeth, and no premature tooth loss was found in the patient's family history. The patient was system-

ically healthy and there were no abnormal findings in laboratory tests and Ca, P, and alkaline phosphatase analyses. According to clinical and radiographic examination, the patient had fair oral health, and skeletal Class III malocclusion was confirmed (Figure 2). Teeth had a normal response to pulp sensitivity tests with ethyl chloride and percussion test. Mobility and probing depth were examined, and the first molar and upper central molar showed grade-3 mobility on both

sides and other teeth exhibited grade 1-2 mobility. Panoramic and full-mouth periapical images were obtained (Figure 3,4). Root resorption of varying degrees was found in the panoramic view of all teeth' apexes. Considering the degree of mobility and root resorption of all teeth, the proposed treatment plan included full extraction and implant placement after orthognathic surgery.

Table 1. Review of MIARR cases (A systematic search was done in the PubMed, Scopus and Google Scholar database using the following keywords: "multiple root resorption" OR "multiple tooth resorption" OR "numerous root resorption" OR "numerous tooth resorption" OR "multiple teeth resorption" OR "numerous teeth resorption" OR "idiopathic root resorption" OR "idiopathic tooth resorption" OR "idiopathic teeth resorption").

authors	Year	type of resorption	Number of Affected teeth	gender	clinical findings	age	Treatment
Mueller et al.	1930	radicular	9 teeth	female	no clinical sign	36	extraction
Carr et.al	1958	apical	5 teeth	female	no clinical sign	29	regular follow up
Pinska et.al	1966	apical	All teeth	female	no clinical sign	14	regular follow up
Soni et.al	1970	apical	9 teeth	male	no clinical sign	34	regular follow up
Cowie et.al	1981	apical	9 teeth	male	no clinical sign	27	regular follow up
Belanger et.al	1985	apical	26 teeth	male	no clinical sign	14	regular follow up
Brooks	1986	apical	4 teeth	male	no clinical sign	17	regular follow up
Pankhurst et al	1988	apical	13 teeth	male	no clinical sign	30	regular follow up
Postlethwaite	1989	apical	20 teeth	male	no clinical sign	14	regular follow up
Counts et.al	1993	apical	18 teeth	male	no clinical sign	23	regular follow up
Eric M	1994	apical	molar and pre-molars(16 teeth)	male	no clinical sign	24	regular follow up
Rivera et.al	1994	apical	21 teeth	male	no clinical sign	24	regular follow up
Di Domizio et al.	2000	apical	15 teeth	famale	no clinical sign	26	regular follow up
SS Cholia	2005	apical	All posterior teeth in both arch	male	no clinical sign	28	resin bite-raising appliance
SS Cholia	20005	apical	All molar teeth	male	mobility	37	long term follow up
Schaatzle et al.	2005	apical	28 teeth	famale	no clinical sign	17	regular follow up
F. Moazami	2007	apical	18 teeth	male	no clinical sign	27	regular follow up
Sogur et.al	2008	apical	All teeth	famale	mobility	18	Endodontic
Hegde S	2012	apical	8 teeth	male	mobility	13	removable partial denture
M Kanungo	2013	apical	3 teeth	male	Pain	16	regular follow up



Figure 1. Intraoral view.



Figure 2. Lateral cephalogram showing skeletal Class III malocclusion.

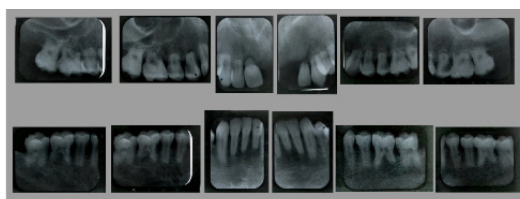


Figure 3. Full mouth periapical radiographs showing apical root resorption.

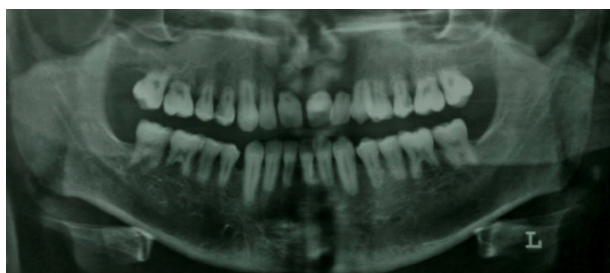


Figure 4. Panoramic image showed apical root resorption involving all permanent teeth.

Discussion

Although idiopathic root resorption is common in one or more teeth, total dentition involvement is very rare. The case presented in the current report was an example of severe MIARR involving all permanent teeth that occurs at a relatively young age and will lead to tooth loss. Multiple idiopathic external apical root resorption (MIEARR) is an unusual condition that can occur in the apical or cervical region. Root resorption is a complex process involving interactions between inflammatory cells, resorptive cells, hard tissue, cytokines, and enzymes [14]. When resorption affects multiple teeth, Loe & Waerhaug suggested that the tooth tissue is transformed into bone tissue in this process, which is a form of remodeling [15]. So far, few MIARR cases have been reported in the literature (Table 1). The first case of idiopathic external root resorption was introduced by Muller in 1930. The authors believed that cervical damage to the teeth was associated with underlying hepatic impairment and that their therapeutic intervention included dietary changes and improved liver function [7].

According to previous studies, the average number of teeth affected by MIARR is 18 teeth and the mean age of patients is 23.2 years [5], in which all permanent teeth were involved. Schatzle et al. reported a generalized and progressive apical root resorption case in a 17-year-old girl. They observed the thickening of the cementum layer and deposition of cellular cement after extracting three involved teeth and performing histopathological examinations. They identified this phenomenon as early generalized hypercementosis [16]. Sogur et al. also reported the overproduction of bone around the resorptive roots, which is a compensatory response to osteoclastic activity [17]. It has also been reported that excessive occlusal loading can lead to dystrophic changes in the periodontal ligament, alveolar bone, cementum, and pulp, as well as periapical inflammation, bone sclerosis, and root resorption [18]. In a study on 179 resorptive roots, Stafne et al. found no association between root resorption and systemic disease [19]. Newman et al. reported similar findings in a study of 47 patients with idiopathic root canal resorption [20]. Some investigators believe that external factors, including viral infections (feline herpes virus type 1 (FeHV-1)), can indirectly stimulate osteoclastogenesis through immune cells and the RANK-RANKL pathway. This process can lead to the destruction of the protective surface layer of cementum by the accumulation and stimulation of osteoclasts and the increase of collagenase production, exposing bone morphogen-

ic proteins (BMP) to osteoclasts, which will eventually lead to root resorption [14]. Since this phenomenon does not originate from the pulp space, treatment methods such as endodontic treatment and pulp tissue removal do not seem to have an indication. No treatment or prevention is known for this type of root resorption and follow-up is usually the only solution that eventually leads to tooth exfoliation. Some articles believe that tooth extraction is necessary under these conditions [21] while others believe that teeth should be preserved [22]. There are reports that calcium hydroxide-based endodontic treatment can slow down the resorption process [23,24]. Indeed, surgical repair without root canal therapy was considered the treatment of multiple idiopathic root resorption using mineral trioxide aggregate (MTA) and can be recommended for treating superficial lesions of the cervical resorption.

This surgical approach could also be combined with a root canal to treat more extensive lesions [25]. However, it was found that the root resorption process continued even in the MTA-treated teeth [26]. Recurrence of idiopathic root resorption was also reported due to the aggressive nature of osteoclast activity [27]. Various growth or sex hormones and environmental factors, including oral flora changes, can affect the resorption process [28]. In a case report, Samara et al. performed implant placement after extraction of the affected teeth, and complete osteointegration occurred and the implants showed normal function after 4.5 years of follow-up [29]. Also, the implant was proposed as one of the treatments for these patients. Further studies are needed to identify the nature of this disease and its treatment methods.

Conclusion

While root resorption is a normal physiological process required for resorption and exfoliation of primary teeth, root resorption of adult teeth is considered pathologic. Pathological root resorption can be broadly classified into internal (originating within the dental pulp) or external (attacking the outer root surface) processes, mediated by osteoclasts/odontoclasts. The present article was a report of a male patient with multiple idiopathic root resorption involving all permanent teeth, which is a very rare case, and its diagnosis and treatment plan are a challenging process for dentists. Therefore, regular dental visits and follow-up radiographs are very important for early detection. Further studies are recommended to discover the exact cause of this phenomenon and its effective prevention

methods.

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Conflict of Interest

There is no conflict of interest to declare.

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