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Case Report

Multiple idiopathic invasive cervical root resorption

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Abstract

MICRR is a rare disease affecting more than 3 teeth at a time and is known to gradually involve other teeth. The etiology of this progressive disease is unknown and is usually detected as an incidental radiographic finding. This article reports one such case involving 5 teeth in a 25 year old healthy male patient who reported the chief complaint of discomfort and vague dull pain in the left mandibular posterior tooth for 3 months. Intraoral examination revealed initial occlusal pit caries on mandibular left first molar (#36) with grade I mobility, which was nontender on percussion with no vestibular findings. A panoramic radiograph showed a well defined radiolucent lesion in the cervical region on the mesial and distal surfaces of tooth #36 and the distal cervical areas of mandibular left first (#34) and second premolar (#35). Tooth #36 was extracted due to poor prognosis and subjected to histopathological investigation, scanning electron microscopy (SEM), and energy dispersive X ray (EDX or EDS) analysis. Histopathological evaluation showed cervical resorption with surface resorptive irregularities with vacuolar changes, and SEM revealed areas of lacunar resorption with irregular borders. Results of EDX analysis showed atomic % of calcium (CA) and phosphate (P) of 4.8% and 3.4%, respectively, with a CA: P ratio of 1.41:1.

Keywords: Cervical root resorption, cone-beam computed tomography, dental caries, energy-dispersive X-ray, scanning electron microscopy

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INTRODUCTION

Multiple idiopathic cervical root resorption (MICRR) is a rare, insidious, and often aggressive form of external tooth resorption that occurs at the cementoenamel junction (CEJ), below the gingival epithelium, with an unknown etiology.[1,2] MICRR affects multiple teeth in the same arch, or it can be widely distributed throughout the dentition. It is usually detected during a routine dental examination or as an incidental finding on radiographs.[3] Previous case reports indicate that there is no relationship between MICRR and age, gender, ethnicity, or systemic conditions.[1] Clinically, the lesions may often be asymptomatic with no signs of inflammation. The resorption cavities may be hard, noncarious, and often exhibit sharp knife-edge borders and may have a pinkish hue which represents vascular granulation tissue.[4,5] In a vital tooth with minimal destruction, there is rarely any pulpal involvement, which distinguishes MICRR from external inflammatory root resorption, where pulpal

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necrosis or infection is a prerequisite. MICRR usually leads to a large irreversible loss of tooth structure as it may progress rapidly over a short period of time.[1] MICRR lesions are difficult to manage because they are frequently located subgingivally on the interproximal surface, and restoration of such teeth is difficult.[1,3] This paper presents a rare case of MICRR in an adult healthy young male involving several teeth with a thorough investigation of the extracted one.

CASE REPORT

A 25-year-old male South Asian patient reported to the outpatient department of oral medicine and radiology with discomfort and a vague dull pain in the mandibular left molar tooth for 3 months, which was occasional. There has been no history of swelling or pus discharge in 3 months but had undergone extractions of two teeth in the same region 2 years ago. The patient's past medical history was clear and noncontributory. Informed written consent was obtained from the patient. On intraoral examination, the mandibular left first molar (#36) showed initial occlusal pit caries and grade I mobility. The tooth was nontender on percussion with no vestibular findings. The local and overall periodontal conditions were good. Initial caries was also found in the maxillary right second (#17) and third molar (#18), maxillary left first (#26), and second molar (#27) teeth. Mandibular left second (#37) and third molar (#38) and mandibular right first molar (#46) were missing. On advising a radiographic investigation, the patient presented a 2-year-old panoramic radiograph which revealed a well-defined radiolucency on the distal aspect of tooth #36. Additionally, it also showed similar extensive cervical radiolucent lesions on tooth #37 and #38, which were extracted. A second detailed clinical examination in the area of interest was performed to confirm that no deep caries was evident as noticed on the first clinical local examination. An attempt to probe the distal surface of tooth #36 resulted in bleeding, but there was no evidence of a periodontal pocket or a catch on the distal surface. A newly acquired panoramic image revealed a similar well-defined radiolucent lesion in the cervical region on the mesial and distal surface of tooth #36 and the distal cervical areas of the mandibular left first (#34) and second premolar (#35) that was not visible on the old panoramic radiograph [Figure 1]. No other teeth in the oral cavity were involved. The CBCT image study reconfirmed multiple radiolucent cervical lesions on teeth #34, 35, and 36 well below the area of contact and above bone level [Figure 2]. A well-defined, sharp-edged radiolucency was noted on the distal surface of the mandibular left first molar (#36) involving enamel, dentin,

Table 1: Timeline

Time	Event	Patient symptoms
0	Patient visited clinic, history, clinical, and radiographic examination	Discomfort and vague dull pain in the mandibular left posterior molar tooth
+1 week	Revisit and additional investigations	Symptoms present
+2 weeks	Extraction of mandibular left first molar tooth (#36)	NSAIDs were prescribed
+3 weeks	1st follow-up (clinical examination)	No pain or swelling (symptom free) Healing of socket
+1 month	2nd follow-up Curettage and restoration of mandibular left first premolar (#34) and mandibular left second premolar (#35)	No pain or discomfort (symptom free)
+6 months	Patient failed to report for follow-up 3rd follow-up	

NSAIDs: Non-steroidal anti-inflammatory drugs

Table 2: Diagnostic methods and results

Tooth number	Electric pulp testing	Percussion	Palpation	Mobility	Radiographic findings	Histopathology	SEM and EDX analysis
#36	Response	Tender (moderate)	Mild	First distinguishable sign of movement greater than normal	OPG: Well-defined radiolucent lesion in cervical region on mesial and distal surface CBCT: Radiolucent cervical lesion on well below the area of contact and above bone level	Cervical resorption with surface resorptive irregularities and vacuolar changes	SEM: Revealed areas of lacunar resorption with irregular borders EDX: Ca and P had atomic % of 4.8% and 3.4%, respectively, with a Ca: P ratio of 1.41:1
#35	Response	Nontender	Normal	Within normal limit	OPG: Well-defined radiolucent lesion in distal cervical areas CBCT: Radiolucent cervical lesion well below the area of contact and above bone level	-	-
#34	Response	Nontender	Normal	Within normal limit	OPG: Well-defined radiolucent lesion in the distal cervical areas CBCT: Radiolucent cervical lesion well below the area of contact and above bone level	-	-

CBCT: Cone-beam computed tomography, SEM: Scanning electron microscopy, EDX: Energy-dispersive X-ray

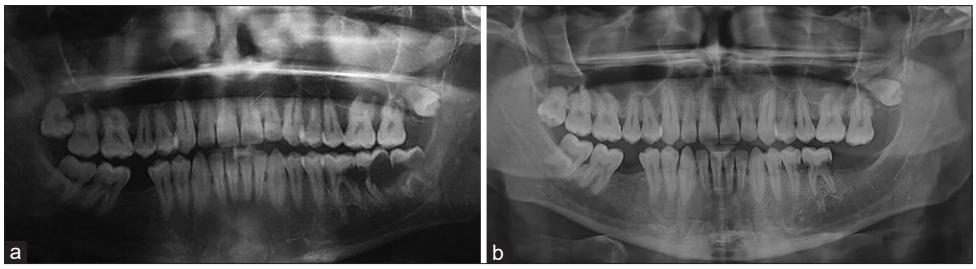

Figure 1: (Pretreatment): (a) Previous orthopantomogram showing extent and progression of resorptive lesion in relation tooth #36, 37, and 38, (b) orthopantomogram exposed on the day of examination showing resorptive lesion involving tooth #34, 35, and 36

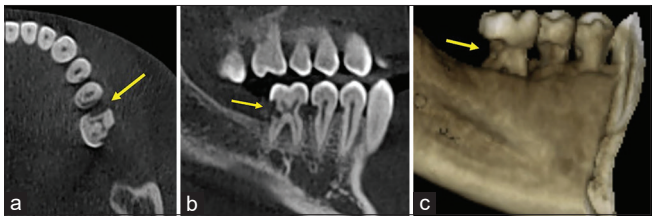

Figure 2: (Pretreatment): Cone-beam computed tomography images studied under (a) axial, (b) sagittal sections, (c) 3D reconstruction

cementum, and pulp, while the mesial lesion involved only enamel and dentin at the CEJ. Distal cervical radiolucency involving enamel and dentin was seen at the CEJ in teeth #34 and #35. The patient gave no history of trauma, orthodontic treatment, bleaching, surgeries, or periodontal procedures. The medical history of the patient was rechecked to rule out the possibility of usage of any medications or treatment for any systemic illness or diseases such as HBV infections. The patient's accompanying attendees, who were his father and younger brother, were clinically evaluated and panoramic images were acquired, which failed to show any such multiple cervical root resorbing lesions, thereby ruling out familial etiology.[3,6] The patient had no pets and gave no history of close contact with cats, hence the possibility of feline odontoclastic resorptive lesion was ruled out.[7] Since none of the conventional causes of cervical resorption were present in this case, it was considered to be idiopathic cervical root resorption. Based on the degree of hard tissue destruction, tooth #36 was extracted and the other teeth were planned for gingival curettage with restorations. The extracted tooth fractured during the procedure, and the sample was collected in three pieces – one crown and two roots [Figure 3]. Furthermore, the crown sample was subjected to scanning electron microscopy (SEM) examination along with energy-dispersive X-ray analysis (EDX or EDS) and histopathology. After extraction of tooth #36, the patient was recalled after 1 month for treatment of other teeth. Gingival curettage and restoration with Biodentine root restorative material (Septodont, USA) were performed on teeth #34 and 35. Unfortunately, the patient did not return for subsequent follow-ups [Table 1].

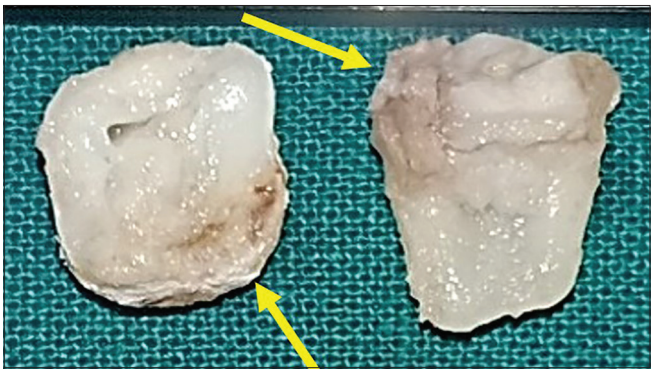

Figure 3: Extracted mandibular left first molar tooth with the crown and roots separated (arrow)

Investigations

Scanning electronic microscopy (ZEISS GeminiSEM 560, Oberkochen, Germany) was used to analyze the surface of the diseased portion of the crown sample exactly 1 mm pulpal from the crown edge, which revealed areas of lacunar resorption with irregular borders [Figure 4]. Crown of the extracted tooth was disinfected using chlorhexidine 2% and preserved in saline. The affected areas of the tooth were marked and subjected to energy-dispersive X-ray (EDX or EDS) analysis (EDAX AMETEK, USA). The weight % and atomic % of elements present were calculated using EDX analysis. Results showed that elements CA and *P* had atomic % of 4.8% and 3.4%, respectively, with a CA: P ratio of 1.41:1 [Figures 5 and 6]. Histopathological evaluation of the decalcified section of the sample with hematoxylin and eosin staining showed cervical resorption [Figure 7a] with surface resorptive irregularities with vacuolar changes [Figure 7b] and no evidence of any inflammatory cells. Underlying dentin with dentinal tubules [Figure 7c] was also evident (×100 magnification) [Figure 7 and Table 2].

DISCUSSION

Resorption of teeth/roots has long been of great interest to various specialties of dentistry, and several

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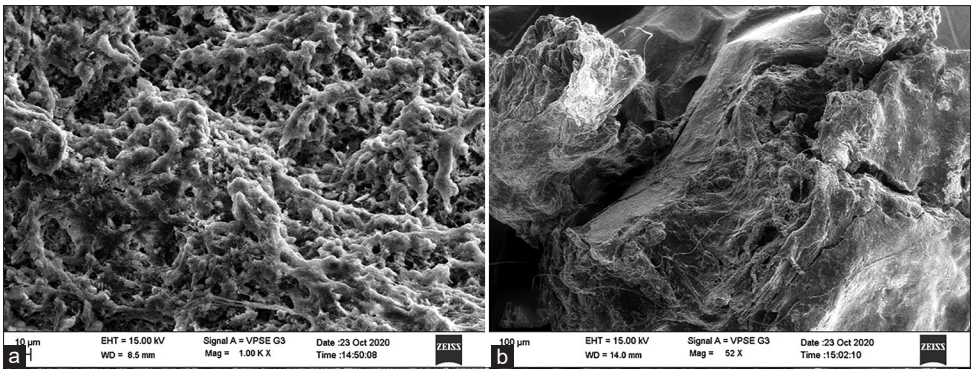

Figure 4: Scanning electron microscopy images of diseased part of the extracted tooth. (a) 10 µm, (b) 100 µm

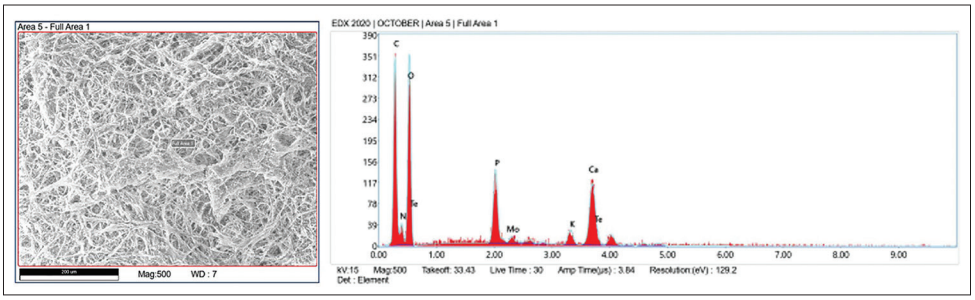

Figure 5: EDAX images of the diseased part of extracted tooth

Element	Weight %	Atomic %
C K	26.4	37.4
N K	8.3	10.1
O K	40.0	42.6
P K	6.3	3.4
K K	2.1	0.9
Ca K	11.3	4.8
Mo L	1.3	0.2
Te L	4.3	0.6

Figure 6: EDX analysis

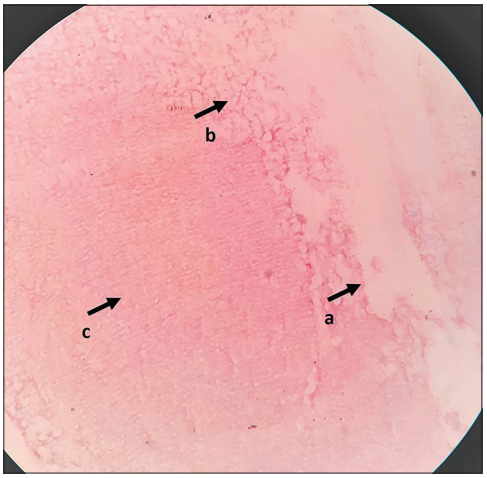

Figure 7: Histopathological image showing surface resorption (a) vacuolar changes (b) underlying dentin with dentinal tubules (c)

attempts to classify and gain a thorough understanding have been made.[8-10] Broadly, resorption can be classified as physiological or pathological. Primary teeth show physiological resorption and rarely pathological, but in permanent teeth, invariably all resorptions are considered pathological. A complete understanding of the exact mechanisms of resorption in all forms of resorption is still elusive, especially for the multiple tooth involvement variant. Solitary tooth/root resorption is common and has been reported extensively in the literature along with substantial explanations of pathophysiology.[8-12] MICRR is a rare pathological entity that often begins at the CEJ and progresses to the entire cervical region, affecting multiple teeth.[5] In the present case, 5 teeth were affected, which led to the loss of 3 teeth. It is often difficult to differentiate between cervical root resorption and cervical/interproximal caries.[13] Proximal caries occurs at the contact point or just below it. Cervical root caries is often associated with gingival recession and bone loss, followed by food lodgment clinically and thereby initiation of caries in the root.[13] Whereas in MICRR, lesions occur well below the contact point at the cervical third below the free gingiva with no evidence of any bone loss. Cervical root caries are frequently seen on radiographs as ill-defined saucer-shaped or notched radiolucency below the CEJ but above the bone height.[14,15] Interproximal caries starts as a radiolucent notch below the contact point and later progresses as a cone-shaped with the base toward the periphery.[14] MICRR, on the other hand, is seen radiographically as multiple radiolucent areas in the cervical region, well below the point

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of contact with the sharp edge of the cavity borders.[13] On radiographic examination, the present case also showed similar multiple radiolucent cervical lesions involving three teeth (#34, #35, and #36) well below the area of contact and above bone level. Although the etiology of MICRR remains obscure, various authors have proposed potential predisposing factors for MICRR. But a damaged or deficient cementum layer appears to be necessary for the initiation of the process.[5] In the present case, resorption was not associated with any of the conditions generally accepted as causative factors, such as trauma, intracoronal bleaching, orthodontic therapy, surgical and periodontal procedures damaging the cemental layer, bruxism, etc.[5] Other possible causes of MICRR include microbiologically induced osteoclastic activity, narcotic drug use, and HBV infection leading to hepatic disorder.[7,16,17] As a result, the etiology of cervical root resorption remains unknown. Since the resorptive process occurred in the absence of

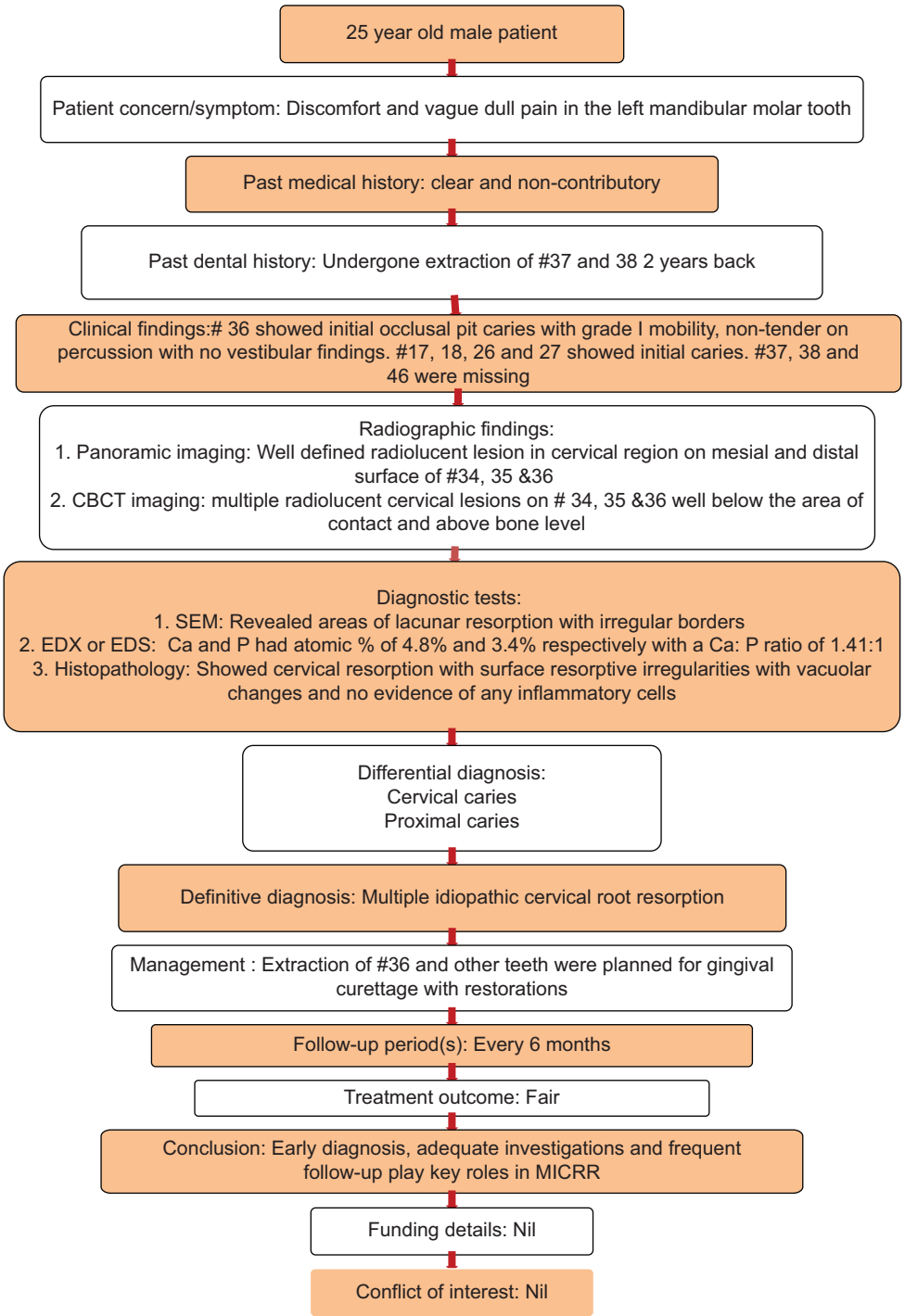

Figure 8: PRICE 2020 flowchart

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a local or systemic factor, it was considered "idiopathic resorption of teeth." In most of the reported cases, the affected teeth were subjected to histological examination and SEM analysis.[1,5] To the best of our knowledge, none of the reported cases/studies has performed EDX analysis on MICRR. Energy-dispersive X-ray analysis (EDX) is a technique used to identify the elemental composition of materials. The calcified tooth structure is composed primarily of Ca and *P* ions that make up hydroxyapatite crystals with the chemical formula $Ca10\ (PO_4)_6(OH)_2$.[18] By EDX analysis, the relative amounts of Ca, P, and Ca: P ratio in the diseased part of the affected tooth were determined. This helped in the evaluation of molecular-level changes that have occurred in the microelemental composition of the affected teeth. In the present study, the EDX analysis of the affected tooth showed Ca atomic % of 4.8% and P of 3.4%. The Ca: P ratio was 1.41:1. EDX analysis done on normal teeth by a few researchers found the atomic % of Ca and *P* to be 52.50%–21.19% and 25.7%–12.58%, respectively.[19,20] The Ca: P ratios were 2.47:1 and 2.04:1, respectively. While an adult carious tooth was subjected to EDX analysis where the atomic % of Ca and *P* was 16.98% and 9.61% with the ratio of 1.76:1.[20]

Based on the above findings, it can be hypothesized that normal healthy adult teeth have a Ca: P ratio >2, whereas carious lesions have a Ca: P ratio between 1.5 and 2. In the present study, the Ca: P ratio was 1.41:1, which was <1.5 of carious teeth. This gross reduction in Ca: P ratio in the present MICRR case indicates that the lesions are more aggressive with increased demineralization. However, since no standardized protocol for management is available for MICRR, a case-dependent treatment approach is always advised.[1] This may include observation with early intervention such as surgical exposure, curettage of the lesion followed by restoration and root canal therapy if necessary, extraction of severely affected teeth, and replacement with a partial or complete denture or implant.[3,4] In the present case, tooth#36 was extracted due to poor prognosis. Curettage of the lesion followed by restoration was done in teeth #34 and #35. From most of the reported case series, it can be understood that restorative interventions do not prevent disease progression, and tooth loss over time appears to be inevitable.[3,4] The present article reports a rare case of MICRR. This pathologic condition is similar to common dental caries and is often difficult to diagnose. The article goes into great detail about how to diagnose and differentiate these lesions, as well as a discussion of advanced investigations. The scientific literature has failed to propose a standardized treatment protocol for MICRR. This may be considered a limitation. Hence, in the present case, a conservative approach was also followed. This case report was prepared according to the PRICE 2020 Guidelines Figure 8.[21]

CONCLUSION

MICRR is often a challenging entity for the clinician. A detailed clinical examination and adequate investigations should be carried out. The position of the lesion clinically and radiographically aids in diagnosis. Advanced investigations such as SEM, EDX, and histological evaluation aid in diagnosis and help in improving the oral health of the patient.

Key learning points

All cervical radiolucency cannot be considered as caries. Thorough knowledge of such rare diseases should be of prime importance to the clinician. It is important to diagnose the condition in its early phase and find a solution to stop further resorption. With progression to the late stage, lesions are too extensive to treat, and extraction may be the only solution.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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