Importance of Considering Rare Disorder as Eosinophilic Granuloma in Differential Diagnosis of the Common Oral Disease

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Abstract

Introduction: Eosinophilic Granuloma (EG) is one of the subsets of langerhans cell histiocytosis. Oral presentation may comprise multiple alveolar lesions accompanied by bleeding, gingival inflammation and teeth mobility, which may encourage patients to seek a dentist.

Case Presentation: In this report, a case of an 18 year old male patient with multifocal lesions of EG in both jaws is presented.

Conclusions: Although LCH remains an enigma for clinicians to date because of its ambiguous presentations, but timely diagnosis could be critical for prognosis. And dentists could have an important role in diagnosis of rare disorders like EG.

Keywords: Eosinophilic granuloma, Oral Manifestation, Maxilla, Mandible

1. Introduction

LCH, previously termed Histiocytosis X, is a rare disease characterized by the abnormal proliferation of the bone marrow-derived histiocytes (1, 2). It can present both local and systemic manifestations involving bone, skin, mucosal tissue and internal organs (2). LCH includes three subtypes: EG, Hand-Schuller-Christian disease and Letterer-Siwe disease (3). EG is the most benign disorder of this triad and it involves localized lesions predominantly of the long bones (3, 4). It accounts for 60% - 70% of all cases of LCH and can be seen as solitary or multifocal lesions (1, 4). Multifocal lesions have a more severe and even fatal course (5). LCH is slightly more prevalent in young males and commonly involves mandible more than maxilla, when the jaws are affected (5, 6). In this report an 18 year old male patient with oral complications as the first sign of EG is presented.

2. Case Presentation

An 18 years-old male with complaint of proceeding tooth mobility in both jaws with occasional pus-like discharge, gingival bleeding and pain sensation during mastication, referred to the Yazd periodontology department.
Swelling of the left side of the maxilla was observed during the first oral examination (Figure 1). The lesion was characterized by a destructive area on the posterior side of the mandible (Figure 1D) and an exophytic mass on the anterior part of the mandible (Figure 1C). A destructive area was also observed on the buccal aspect of the mandible (Figure 1E).

Figure 2 shows a panoramic view of the patient's left upper arm, which also presented with an osteolytic bone lesion (Figure 3). Radiographic views of the left upper arm demonstrated lesions consistent with Langerhans cells (Figure 3). As multiple bone areas were involved, surgery or curettage was not the first treatment choice. The patient was referred to a hematologist for further treatment.

After two months, the patient was re-examined by the periodontist, and there was no significant change in oral condition except for a new ulcerative lesion in the palatal of the left maxilla (Figure 1B).

Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

3. Discussion

LCH is a rare disorder and even rarer with jaw involvement, usually involving children and young adults (1). It is inclined to occur in bones more than other organs like liver, skin, bone marrow, lymph nodes, and lung. Jaw involvement usually happens in 10% - 20% of bone involvement with tendency to mandible (1, 6). Common oral presentations of LCH are periodontal involvement and teeth loosening (7). A list of probable diagnosis for multifocal destructive lesions includes osteomyelitis, Ewing sarcoma, osteosarcoma, Giant cell granuloma (or Brown tumor), multiple odontogenic keratotic cyst, multi-lacunar cyst, leukemia, and lymphoma (2). All of these cases can lead to advanced periodontitis in affected areas. Accord-
ing to low age and no evidence of hyperparathyroidism, Brown tumor was ruled out. Leukemia usually occurs with prodromal signs like fever and myalgia (8).

In spite of its acute process, Lymphoma, in jaw especially non-Hodgkin type, usually presents as an exophytic mass in socket of extracted teeth (8). A generalized form is one of the differential diagnoses. It was noticed that osteomyelitis occurred in individuals with susceptible situations like poor controlled diabetes, immunosuppression or old age (9). None of these conditions exists in this case; therefore, it is an unlikely diagnosis. Multiple odontogenic keratocysts usually occurs in patients with basal cell nevus syndrome, but can occur independent to syndromes (9). The most likely diagnosis was LCH and after incisional biopsy from different involved areas, LCH was confirmed.

Due to being a rare disease with high clinical variability, it was a failed attempt to determine definite prognosis for LCH. Although LCH has a benign nature, it has some alternating phases of relapse and remission (2).

Because of multifocal lesions in jaws, in this case, surgery was not a sufficient modality of treatment; therefore, oncologist provide a CHOP chemotherapy for him in multiple sessions and follow-ups were scheduled.

3.1. Conclusions

Although LCH remains an enigma for clinicians to date because of its ambiguous presentations, but timely diagnosis could be critical for prognosis. And dentist could have an important role in diagnosis of rare disorder like EG.
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Footnotes

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References


