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Eosinophilic Granuloma of the Mandible: A Case Report

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Abstract

Eosinophilic Granuloma is the benign and localised form of "Langerhans Cell Diseases". Langerhans Cell Diseases are characterized by a clonal proliferation of pathologic cells with the characteristics of Langerhans cells in single or multiple organs.

The localized form of Langerhans Cell Diseases occurs in older children, adolescents and young adults. Eosinophilic Granuloma is a destructive osseous lesion characterized by presence of a vast number of eosinophils and histiocytes. The prognosis for this form of the disorder is fine.

We present a case of a 16 years old male patient. The radiolusent lesion associated with impacted left mandibular third molar region was detected during the patients routine dental examination. However there was radiolucent, non-sclerotic bordered lesion related with left mandibular third molar region in panoramic radiograph, there was no clinical signs or symptoms. Treatment of the lesion was surgical curettage with removal of impacted tooth. Definitive diagnosis was Eosinophilic Granuloma after the histopathologic analysis. Postoperative period was uneventful.

Keywords

eosinophilic granuloma; langerhans cell disease; maxillofacial region; surgical treatment

Abbreviations

EG: Eosinophilic Granuloma; LCD: Langerhans Cell Disease; LCA: Leukocyte Common Antigen

Introduction

Chronic localised form of Langerhans Cell Disease (LCD) or in other words Eosinophilic Granuloma (EG) refers to solitary or multiple bone lesions, and clinical manifestations are ranging from teeth loosening to diffuse involvement in the entire jaw. The most common symptoms are pain and soft tissue swelling [1].

The etiology and pathogenesis of disease remains obscure, although the cell of origin is now known. Although infection, inflammation, or immunologic response are mentioned as possible etiologic factors, how Langerhans cell disease develops normal Langerhans cells or their precursor cells is unknown. EG can occur in the pelvis, ribs, skull, long bones, vertebrae, and facial bones most commonly, but skeletal lesions may be seen in any bone. The incidence of the disease is reported as 7.9 % in the jaws. The body and angle of the mandible are the most commonly affected sites [2].

Radiologically, the lesions can be seen solitary or multiple radiolucent lesions. Lesions often affect

the alveolar bone, causing the teeth to appear as if they were floating in space. Bone lesions with a sharply circumscribed, punched-out appearance may occur in the central aspect of the mandible or maxilla [3].

Immunohistochemical and ultrastructural studies indicated that the dendritic antigen presenting Langerhans cells as the proliferative cell of this disorder and the tumor cells express CD1a antigen and S-100 protein [3]. Electron microscopic studies have shown Birbeck granules in affected cell cytoplasmas specifically [5].

Treatment choice of the chronic localised form of the Langerhans cell disease (EG) can be vigorous surgical curettage, low-dose radiotherapy or intralesional steroid injections. The prognosis of the solitary lesions is good, but recurrence rate in multifocal EG is high.

We aim to emphasize that conservative approach for the treatment of EG is effective and sufficient.

Case Presentation

A 16 year-old male patient was referred to the Department of Oral and Maxillofacial Surgery for evaluation of radiolucency associated with impacted left mandibular third molar region. The lesion was detected during routine dental examination (Figure 1). There was no clinical signs, history of trauma, pain, paresis, paresthesia or lymphadenopathy. The patient was early diagnosed as EG because of the lesions on his pelvic bone.

Under local anesthesia, impacted left mandibular third molar was removed and the lesion was curetted carefully for excisional biopsy. Microscopic examination presented that plenty of eosinophils and sporadically lymphocytes accompanying with langerhans cells have smooth, large and prominent nucleoli with pink cytoplasma in loose connective tissue stroma (Figure 2).,

Immunohistochemical examination showed strongly positive for CD1a and S100 markers, and negative for LCA (Leukocyte Common Antigen) (Figure 3 and 4).

The follow-up and healing of the patient was uneventful. Panoramic radiograph at postoperative sixth months showed no recurrence (Figure 5).

Figures

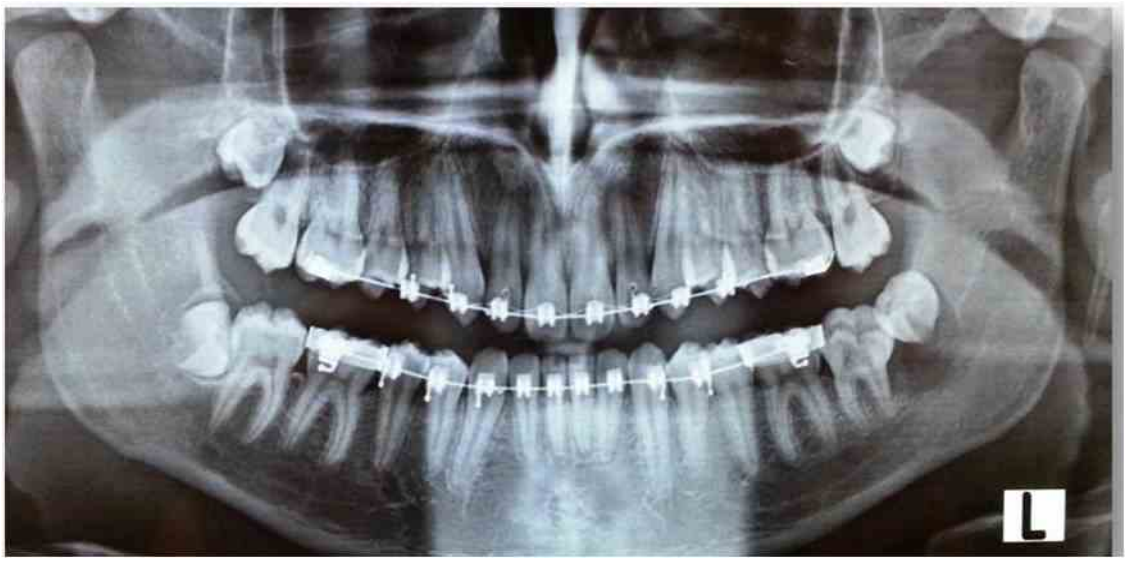

Figure 1: A panoramic radiograph showed an ovoid shaped, unilocular, non-sclerotic bordered, well-defined radiolucent lesion related with left mandibular third molar region.

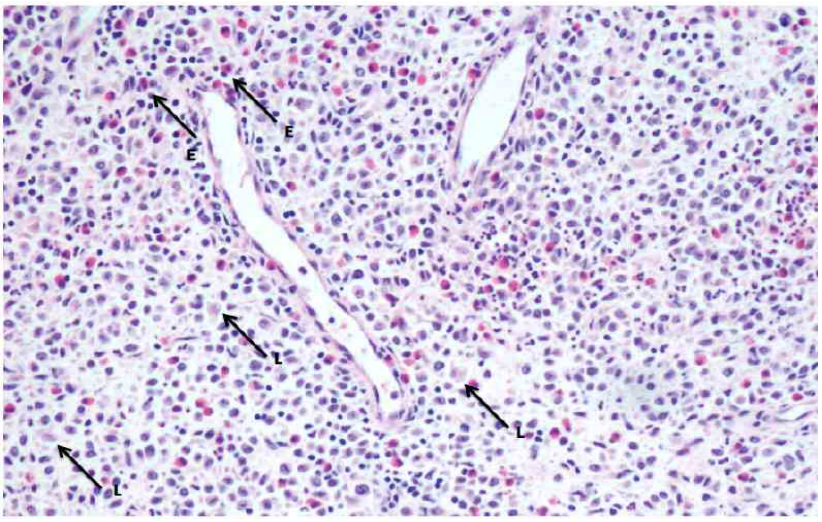

Figure 2: Langerhans cells and eosinophils were main components of the lesion. (HE x 200)
L: Langerhans cells have notched nucleus and their cytoplasmas have painted with light color.
E: Eosinophils are distinguished by 2 notched nucleus and bazophilic cytoplasm

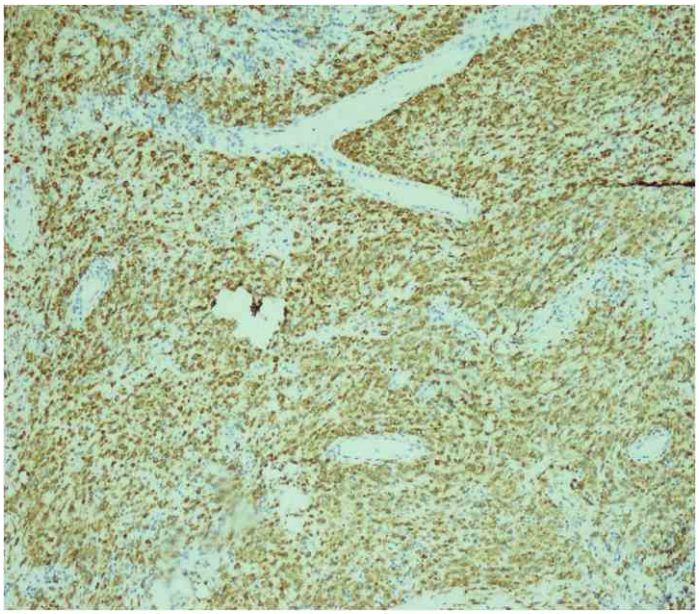

Figure 3: Strong CD1a positivity in Langerhans cells. (CD1a x 100)

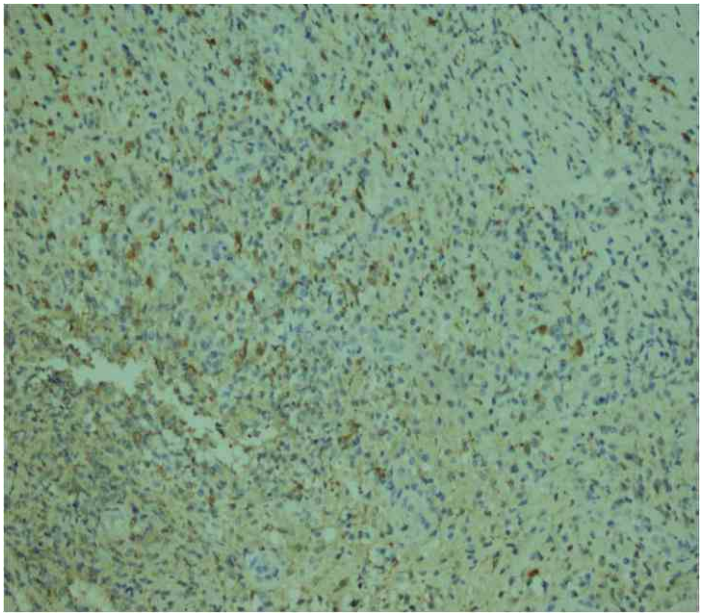

Figure 4: S100 positivity in Langerhans cells. (S100 x 200)

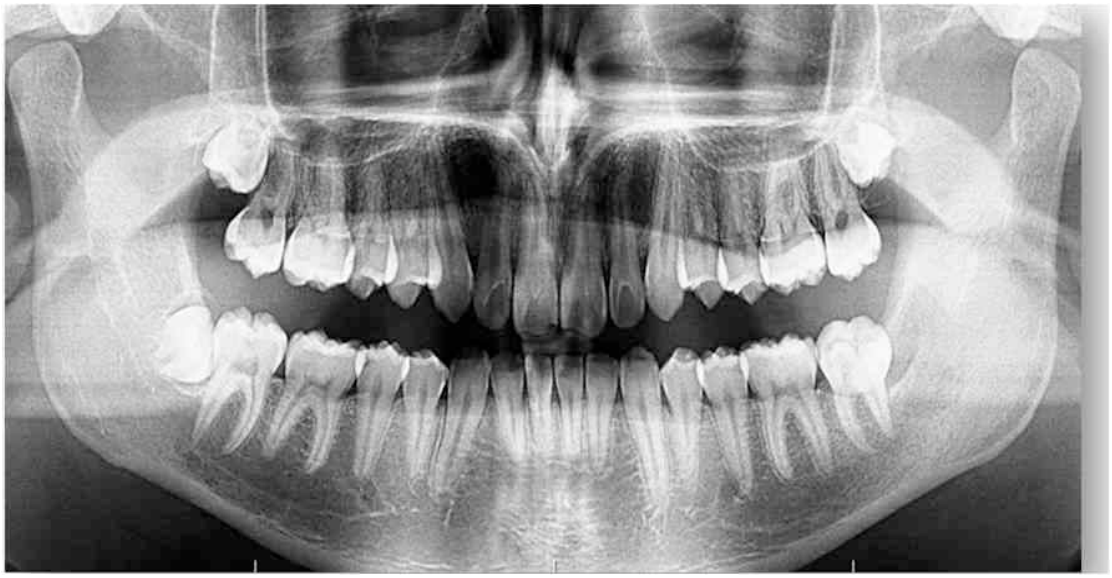

Figure 5: Patient was followed-up for six months during which no recurrence was observed.

Discussion/Conclusion

EG is an infrequent disease belonging to the Langerhans cell disease group and its incidence is just 1% among all tumor-like lesions of bone [6].

EG of bone is also rare in the maxillofacial region. However, it is difficult to diagnose this disease owing to the atypical clinical and radiographic findings. Differential diagnosis has been made with osteomyelitis, odontogenic cysts, giant cell granuloma, ameloblastoma, vascular malformations and malignancies [1,7].

There are a variety of therapeutic approaches including surgery, radiotherapy, intralesional corticosteroid injections and follow-up for spontaneous regression to eosinophilic granuloma.

Local expansion can cause destruction of bone and sometimes fracture of the jaw. Therefore surgical curettage is the most preferred treatment option to solitary and accessible lesions. Involved teeth are generally sacrificed at the time of surgical therapy due to the absence of bony support [1,8].

Although much less preferred, low-dose radiotherapy is used for lesions that are inaccessible to surgical treatment. Secondary malignant transformation or the deterioration of growth centers like the lens and dental follicles, especially in children are the potential risks of low-dose radiation therapy [5].

Mostly, limited surgical excision, steroid injections and radiotherapy provide desired response to eosinophilic granuloma. But chemotherapy has been applied in cases when there were widespread disease including visceral involvement or after failed interventions [9].

Complete removal of the lesion is not necessary in most cases. Although the mechanism of action is unknown, intralesional corticosteroid injections are effective to reduce the lesion size. The relief of pain and resolution of lesions are usually swift after injections. Radiographic evaluations have supported this application as a possible treatment alternative [5].

If the lesions are posing a direct threat to functioning structures or causing major cosmetic defects, the possibility of spontaneous regression of these lesions should be considered. It may be cautious to follow them up without any therapeutic intervention [10].

Dental professionals should be aware of this disease because early clinical signs may occur in the jaws and can progress extensively.

Patients must be evaluated for additional bone or visceral involvement after detection of the original lesion. Long-term follow up is necessary to rule out the possibility of recurrent disease [3].

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