

CASE REPORT**Eosinophilic Granuloma in Jaw Bone: A Rare Pediatric Case Report****Karthiga Kannan¹, Naif Alwithanani², Mohamed Salama², Manoj Kumar³, Roshan Uthappa⁴, Mazood Ahamed²****ABSTRACT**

BACKGROUND: *Eosinophilic granuloma (EG), one of the three clinical forms of Langerhans cell histiocytosis (LCH), is a benign inflammatory reaction to an unknown etiologic agent. It most commonly occurs in children and young adults. The most frequently involved bones are the skull, the ribs and the femurs. Alongside the cranium, the maxilla and mandible can also be affected.*

CASE DETAILS: *Herein, we report a case of eosinophilic granuloma in a ten years old boy involving posterior quadrants upper and lower jaws as a destructive lesion involving gingiva, periodontium and alveolar bone. Involvement of other bones is ruled out by nucleotide imaging study.*

CONCLUSION: *EG should be considered as a differential diagnosis whenever there is a bony destructive lesion involving alveolar bone of the Jaws. Early diagnosis and surgical intervention will resolve the lesion.*

KEYWORDS: *Eosinophilic granuloma, langerhans cell histiocytosis, oral manifestations, alveolar bone and surgical curettage*

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INTRODUCTION

Langerhans cell histiocytosis (LCH) is a rare disease characterized by intense and abnormal proliferation of bone marrow-derived histiocytes (Langerhans cells) (1). **Lichtenstein** classified LCH into three clinical forms depending on the age of the patient when the lesions first appear and their distribution: 1) Chronic focal LCH (**eosinophilic granuloma**) the most frequent and benign of the clinical forms which appears as a uni- or multifocal lesion in a single or various bones with or without soft tissue involvement, without systemic involvement and presenting at any age, 2) Chronic diffuse LCH (**Hand-Schüller-Christian disease**) which usually appears in children or young adults with the characteristic triad of exophthalmos, osteolytic lesions of the cranium and diabetes insipidus. Acute disseminated LCH (**Letterer-Siwe disease**) affects children under three years old involving

multiple organs and systems such as liver, lung, lymph nodes, skin, bone and bone marrow and has a fatal outcome in a short time (2,3,4).

In the majority of the cases, oral manifestations may be the first sign of LCH, and on some occasions, the oral cavity may be the only area affected. Therefore, the initial diagnosis in many cases is made by the odontologist (5). To establish an accurate diagnosis, apart from radiographic examination, these lesions should be biopsied. With curettage or roentgen therapy, the lesions begin to regress in a few weeks and should be completely healed in a few months. Without therapy, regression of the lesions is much slower (6).

CASE REPORT

A ten years old boy reported to the clinic with the complaint of pain in the gums of the mandible and

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the maxilla, associated with mobility of the teeth. Intraoral examination revealed granular, friable and swollen gingiva with severe recession exposing the roots of the teeth in all the four quadrants of the jaws more in the right posterior region (Figures 1 and 2). On palpation, the gingiva was soft, tender and bleeding. The clinical differential diagnosis included Acute Necrotizing Gingivitis (ANUG), Acute Necrotizing Periodontitis (ANUP) and leukemic gingival involvement. Intraoral periapical radiographs (Figure 3) revealed well defined, osteolytic lesions in the posterior quadrants of the maxilla and the mandible giving the teeth floating appearance. Complete hemogram showed a mild raise in the ESR. The differential leukocyte count revealed a moderately raised eosinophil count. Liver function and renal function tests were normal. Histopathological examination of the incisional biopsy specimen showed proliferation of histiocytic cells and infiltration of eosinophils and neutrophils along with increased vascularity rendering a final diagnosis of eosinophilic granuloma. The possibility of systemic involvement was ruled out by a radiographic skeletal survey and Tc 99 m MDP bone scintigraphy (Figure 4). After the surgical curettage, the lesions completely regressed and did not show any signs of recurrence during the follow up period.



Figure 1:-Upper right jaw quadrant showing extensive gingival recession exposing teeth roots.



Figure 2:Right lower jaw quadrant showing erythematous swollen and friable gingiva.

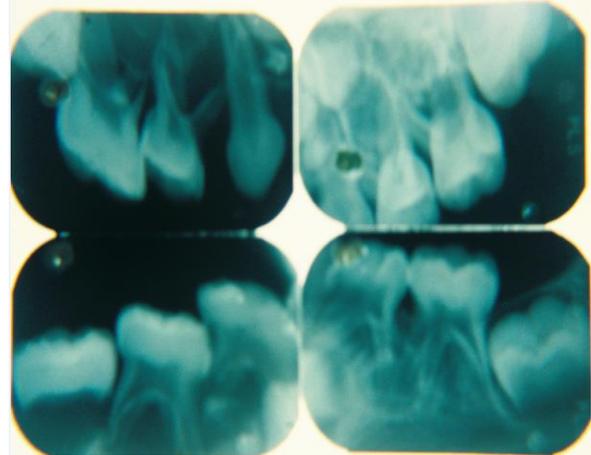


Figure 3:Intra oral radiographs showing extensive alveolar bone loss

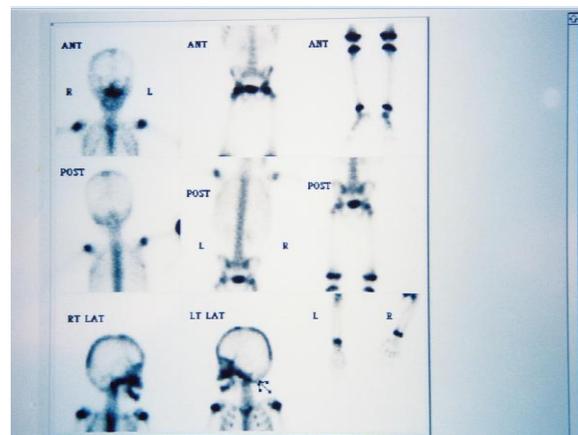


Figure 4:Bone scintigraphy ruled out other bone involvement

DISCUSSION

Eosinophilic granuloma of the bone is a disease of children and young adults with only 5.0% occurring in persons over 30 years of age. It has been reported that males are being most frequently affected than females with 75% of the lesions occurring as solitary lesions (7).

Even though the lesion can manifest in almost any bone, the involvement of the jaws is reported in 77% of LCH series (8). Dagenais et al., in a review of 29 cases of LCH, states that mandibular lesions are more frequent in all three forms of LCH, and the majority of bone lesions were presented in the posterior quadrants (9).

Clinically, the disease appears either to be symptomless or to exhibit manifestations of a very minor degree with the commonest complaints being slight pain and swelling involving alveolar bone of the jaw. Eosinophilic granuloma is often clinically confused with focal infection, since patients may have a slight fever, leukocytosis with eosinophilia and increased sedimentation rate (10).

Radiographically, eosinophilic granuloma typically presents as punched out osteolytic lesions with or without periosteal reaction (10). As 10% of patients with unifocal eosinophilic granuloma of the bone will develop multifocal and extra osseous disease, other imaging studies such as orthopantomography, computed tomography (CT), magnetic resonance imaging (MRI), radiographic skeletal survey and bone scintigraphy can be useful in evaluating the involvement of other cranial and facial bones. However, in the present case, we ruled out the possibility of dissemination of the disease by radiographic skeletal survey and bone scintigraphy.

Diverse therapeutic options are available such as surgery, radiotherapy, chemotherapy and steroid injections, alone or in combination. However, in general, no therapy is required for localized osseous eosinophilic granuloma. It is because, in many instances, the biopsy itself is enough to initiate healing and spontaneous resolution. However, steroid injection, curettage, excision or radiation can be reserved for multifocal lesions or disseminated disease. However, in our case, since the lesions were localized to the jaws, we efficiently and successfully treated by surgical curettage of the

lesions along with extraction of the involved teeth exhibiting marked resolution. When surgical curettage leaves large bony defects autologous bone, grafting can be made in an attempt to reduce the risk of pathological fracture and to facilitate bone regeneration (7). However, in the present case, as the thickness of the residual bone was sufficient enough to undergo self-regeneration, we did not opt for grafting.

To conclude, Eosinophilic granuloma is a benign lesion and the outlook for complete recovery is excellent. A few of the cases may progress into the more serious and chronic form of Hand-Schuller-Christian disease (7). Thus, it is important to include eosinophilic granuloma in the differential diagnosis of bony jaw lesions in young patients because of the possible dissemination of the disease if left untreated.

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