Eosinophilic Granuloma of Mandible: A Diagnostic Challenge
Bajracharya B,1 Poudel P,1 Bajracharya D,1 Bhattacharyya S,1 Shakya P2

1Department of Oral Pathology
Kantipur Dental College,
Basundhara, Kathmandu, Nepal.
2Department of Oral Surgery
Peoples Dental College,
Shorakhutte, Kathmandu, Nepal.

Corresponding Author
Dipshikha Bajracharya
Department of Oral Pathology
Kantipur Dental College,
Basundhara, Kathmandu, Nepal.
E-mail: drdipshikhabaj@gmail.com

ABSTRACT
Eosinophilic Granuloma is the mildest and localized form of Langerhans Cell Histiocytosis and is characterized by clonal proliferation of Langerhans cells. It is a rare disease, accounting for less than 1% of all the osseous neoplasms. It has predilection for the axial skeleton and incidence in jaws is just 7.9%. It lacks pathognomonic clinical and radiographic trait and hence is difficult to make a correct diagnosis without histopathological and immunohistochemical examination. This report describes a case of Eosinophilic Granuloma of mandible in 30 years old male who presented with complain of unhealed extraction wound and was clinically diagnosed as chronic suppurative osteomyelitis. The final diagnosis of Eosinophilic Granuloma was made only after histopathological and immunohistochemical evaluations.

KEY WORDS
Eosinophilic granuloma, Immunohistochemistry, Langerhans cell histiocytosis

INTRODUCTION
Langerhans cell histiocytosis (LCH) is a rare disease of reticuloendothelial system resulting from clonal proliferation of Langerhans cells. This disease of unknown etiology has diverse manifestations.1 Eosinophilic Granuloma (EG) is localized and mildest form of LCH, accounting for less than 1% of all osseous neoplasms.2 It has predilection for the axial skeleton, and has higher incidence among young children. 7.9% of all the cases of osseous EG involves the jaws, most commonly in the body and the angle of the mandible.3 It may present clinically with wide variety of non-specific signs and symptoms such as pain, loose tooth, edema, gingival bleeding and ulcers. Radiographic appearance of EG is quite variable and can be mistaken for osteomyelitis, radicular cyst and malignancies.4 In our report, we present an unusual case of EG of the mandible and discuss the importance of histopathological and immunohistochemical examination in its diagnosis.

CASE REPORT
A 30 year old male presented to the department of Oral Medicine, People’s Dental College, on 26/11/2014, with complain of unhealed extraction wound in the right side of lower jaw since four months. His medical history was not significant. Clinical examination revealed missing 47, 48 and unhealed extraction wound in the same area. 45 and 46 were mobile and tender. There was tender gingival swelling w.r.t 45, 46, 47 and 48. There was no regional lymphadenopathy. The patient was given several trials of antibiotics but there was no improvement. He then underwent extraction of 45 and 46. Based on the clinical findings, diagnosis of Chronic Suppurative Osteomyelitis was given and Intra Osseous Squamous Cell Carcinoma and central Giant cell granuloma were kept as differentials. As there was no improvement even after one week of extraction, complete blood count, CBCT and biopsy were advised.
Blood biochemistry was within normal limits. CBCT revealed lytic areas with ill-defined ragged border and irregular margin. The lesion extended from distal aspect of 44 to the right ramus and inferior aspect of coronoid process of mandible. It was 59.20 mm antero-posteriorly, 32.80 mm superio-inferiorly and 13.20 mm bucco-lingually. Buccal and lingual cortical plates showed thinning and perforation but without significant expansion (fig: 1). The internal structure of the lesion was homogeneous with no evidence of calcification. CBCT gave the impression of intraosseous carcinoma and chronic suppurative osteomyelitis. Histopathological examination revealed sheets of histiocytes having vesiculated nucleoli. The supporting stroma was loose with abundant eosinophilic infiltration (fig: 2, 3). Areas of hemorrhage were noted. Based on these findings histopathological diagnosis of Eosinophilic granuloma was given. But as EG is rare in jaws, to confirm the diagnosis, immunohistochemistry was done using horse radish peroxidase polymer. It showed positivity for CD 1A and S-100 whereas CD45 was negative (fig: 4, 5). These findings were consistent with Eosinophilic Granuloma. Hence diagnosis of Eosinophilic granuloma of mandible was confirmed.

The lesion was managed by surgical curettage. The post treatment period remained uneventful and patient was kept under regular follow up. Five months after the treatment, clinical examinations disclosed completely healed oral lesions and CBCT showed bone formation in previous lytic areas (fig: 6). Hence, patient responded favourably to therapy and showed no signs of recurrence.

DISCUSSION

The Writing Group of the Histiocyte Society introduced the term “Langerhans cell histiocytosis”, replacing the term “histiocytosis X”. LCH encompasses three disorders: Eosinophilic granuloma, Hand-Schüller-Christian disease and Letterer-Siwe disease. The term ‘Eosinophilic Granuloma of bone’ was first introduced by Lichtenstein and Jeffe in 1940. Accounting for 60% to 70% of all LCH cases, EG can present as either solitary (50% to 75%) or multifocal bone lesion. They arise from clonal proliferation of Langerhans cells. These cells can be identified under the electron microscope by the presence of Birbeck granules, which are racket-shaped cytoplasmic inclusions. The reason for proliferation of these cells in LCH is still unknown. However, various etiological factors have been suggested, including immunological reactions, bacteria, viruses and genetic factors.

EG of bone is a rare disease with incidence rate of one to two per million population per year. It occurs predominantly in children. 60% of EG patients having solitary lesions are less than 10 years in age. In our cases, the patient was 30 years old, which is an unusual age for EG. It has predilection for males (male:female = 2:1). EG can occur as unifocal or multifocal lesion, the former being more common. Bones of the axial skeleton are usually affected.

In the maxillofacial region, EG is mostly asymptomatic and is usually discovered incidentally while performing radiographic examination for other indications. When symptomatic, it may present in a various ways including pain, swelling, gingival inflammation, ulceration, and even pathological fracture. It can also present as periodontal destruction with recession of gingival and loss of alveolar bone. Common dental symptoms are mobile teeth, premature exfoliation and delayed healing after tooth
Case Note

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REFERENCES


