EOSINOPHILIC GRANULOMA WITH PREVIOUS HISTORY OF AGGRESSIVE PERIODONTITIS: A CASE REPORT

Mosavat F,[1] Mohammed Charlie S,[2] Razmara F [3]

1. *Assistant Professor, Department of Oral & Maxillofacial Radiology, School of Dentistry, Tehran University of Medical Science, Tehran, IRAN*
2. *Resident, Department of Oral & Maxillofacial Radiology, School of Dentistry, Tehran University of Medical Science, Tehran, IRAN*
3. *Assistant Professor, Department of Oral & Maxillofacial Surgery, School of Dentistry, Tehran University of Medical Science, Tehran, IRAN*

Abstract

Langerhans cell histiocytosis (LCH) refers to a relatively rare condition resulting from neoplastic proliferation of Langerhans cells. Histiocytosis X, encompassed three disorders: eosinophilic granuloma, Hand-Schüller-Christian disease, and Letterer-Siwe disease. Eosinophilic granuloma is the mildest type and is one of the rarest bone tumors representing less than 1% of them. In 90% of the reported cases it appears in children under the age of ten. The most common oral findings are soreness, swelling, ulceration, necrosis of the gingival tissues, and destruction of alveolar bone with teeth mobility and teeth loss. Aggressive Periodontitis is known as a disease with rapid progression and loss of bone support and may lead to loss of dental elements on account of the patient previous history of localized aggressive periodontitis and later on the development of EG and their similar clinical manifestation and radiological appearance had led us to believe this to be an intriguing case to be reported.

Kerwords: Bone diseases, Eosinophilic granuloma, Periodontitis.

Introduction

Langerhans Cell Histiocytosis (LCH) is an uncommon disease caused by neoplastic, rapid multiplication of Langherhans cells.[1] The introduction of "Langerhans cell histiocytosis" was made by the Writing Group of the Histoicyte Society, a substitution for histiocytosis X, which Lichtenstein had presented in 1953 which comprises three disorders: Letterer-Siwe disease, eosinophilic granuloma, and Schüller-Christian disease.[2] Lichetestien had also proposed the term "Eosinophilic Granuloma of bone" with the help of Jaffe in 1940. Eosinophilic Granuloma (EG)is considered as one of the least common bone tumors with less than 1% of abundance. Only 10% percent of the patients with this disease have been reported to be over ten-years old. The amplitude varies in genders and males take precedence in the number in which they show signs of this condition which happens to be twice the female population with this disease. It is a restricted and least severe manner of histiocytosis-X group of diseases. The aforementioned classification has been made upon the similitude between histopathologic appearance of the histiocytic and eosinphilic proliferation.[3] Skeletal lesions of histocytosis-X are not fastidious and can appear in any bone. Though the most common occurrences has been in the ribs, skull, facial and long bones, pelvis, and vertebrae. The reports have shown that the frequency in jaws is 7.9%. The angle and body of the mandible undergo more severe symptoms.[4] The appearance of oral lesions is a well known factor in all shapes of histocytosis X and they may occur as solitary lesions either in the maxilla or the mandible. Destruction of alveolar bone with teeth mobility and teeth loss, swelling, soreness, necrosis of gingival tissues, and ulceration are the most expected oral symptoms that may occur. Histologic examination is a necessity to distinguish diseases such as carcinoma, lymphoma, sarcoma, multiple myeloma and Paillon-Leferve syndrome, osteomyelitis, Piaget's disease and aggressive periodontitis (AP).[5] EG and AP can be similar according to their radiographic feature and both of the mentioned disease cause alveolar bone destruction even so epicenter of bone loss in AP starts from alveolar crest and develop apically down. Additionally tooth involvement in aggressive periodontitis is on the basis of tooth eruption sequence although the order of tooth involvement in EG is not in this form.Radiological researches demonstrated the existence of lytic lesion in the mandible along side floating teeth. Due to these discoveries, the diseases that were taken into consideration were primary bone tumor, lymphoma, multiple myeloma and osteomyelitis. On the other hand, the biopsy from the lesion revealed distinctive features of EG. We highlight this irregular case since up to now there was no case reported from a patient with history of AP afterward superimposition of EG and stress on the significance of histopathological examination in the diagnosis of this very uncommon disease.

Case report

A 29 -year-old male patient with poor oral hygiene was referred to the Radiology Department of Tehran University of Medical Sciences with complaint of pain in right region of lower face in the last 3 months. Having no previous history of systemic disease, he had been diagnosed with localized aggressive periodontitis (LAP) 6 years earlier. at the time of the diagnosis the condition of all four first molars plus the maxillary, mandibular right second molar and mandibular left second premolar teeth were affected through bone destruction and deep periodontal pockets subsequently, leading to their extraction. [Figure 1A]

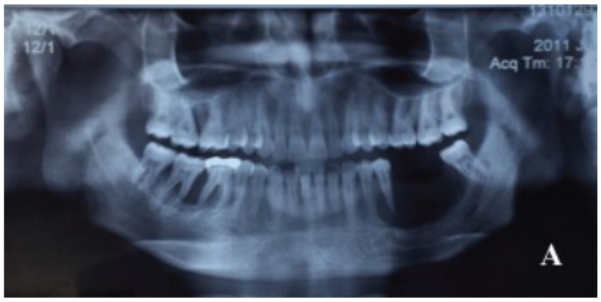

Figure 1A: Panoramic images of the patient – When the patient was diagnosed with Localized aggressive periodontitis

However upon the return of the patient, severe deterioration is visible as displayed. [Figure 1B]

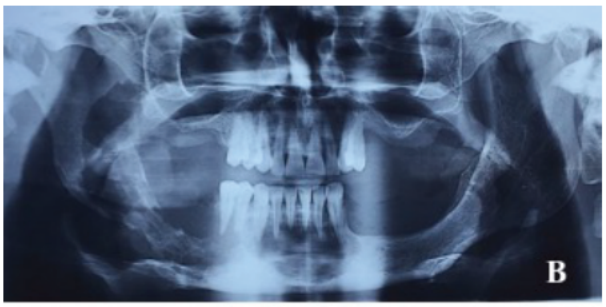

Figure 1B: Panoramic images of the patient – Bone destruction of the mandible caused by Eosinophilic granuloma.

Intraoral examination revealed periodontal pocket with slight mobility of lower anterior teeth. Other associated symptoms were fatigue, weight loss and low degree fever over the past 2 years. No regional lymphadenopathy was observed. On routine work up biochemical factors and haemogram were within normal limits except for slight leukocytosis ($11^3/mm^3$ normal range 3.5-10). Radiographic examination including orthopantogram and Cone Beam Computed Tomography (CBCT) were used. Panoramic view revealed a multifocal well demarcated yet no corticated radiolucency lesion. One extended from distal root of left canine to distal root of right canine. Root resorption and displacement of the involved tooth was not found. Moreover vertical alveolar bone loss in anterior region of mandible produced 'floating in air' appearance. The other large destructive region extended from mesial root of right first premolar up to sigmoid notch. The involved right inferior border of the mandible and its superior extension to alveolar crest could be easily visualized. This lesion produced a saucer shape erosive defect on the alveolar bone of right second premolar region. Punched out appearance is seen within the lesion. An axial CBCT showed destruction of lingual and buccal cortex with no significant expansion in cross sectional images. Inferior alveolar nerve canal cortical borders remained intact, which is an uncommon finding. [Figure 2 & 3]

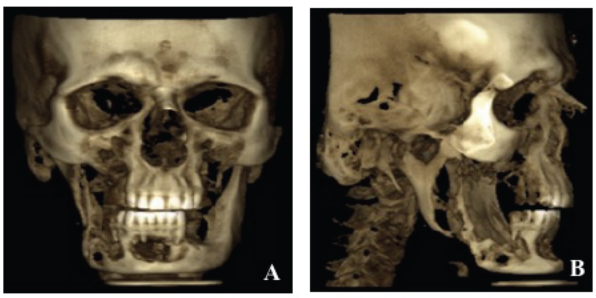

Figure 2: Three dimensional CBCT. A, the lesion is located in anterior part of the mandible, the distal lesion demonstrated mild expansion of buccal plate. B, the anterior posterior extension of the lesion from second premolar.

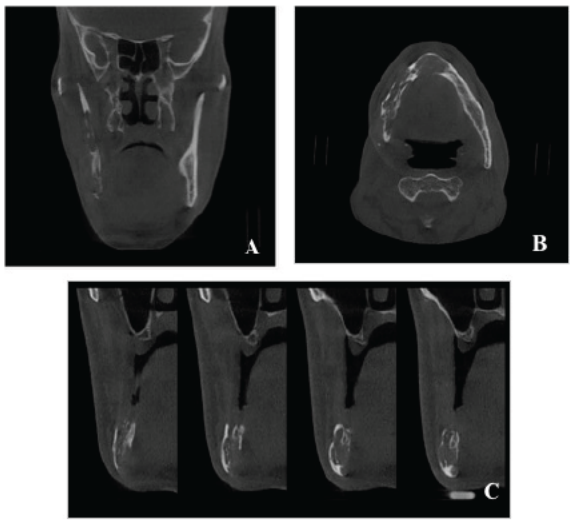

Figure 3: CBCT images of the lesion – A: Coronal image demonstrating perforations of the medial and lateral aspect of the ramus without expansion. B: Axial image shows perforation of the buccal and lingual plates without significant expansion. C: Cross sectional images shows the cortical borders of the inferior alveolar nerve canal remained intact, mouth eaten appearance of lesion can be seen.

A biopsy of the lesion was carried out from the body of the ramus for histological evaluation. Microscopic examination revealed portion of neoplastic tissue with langerhans cell infiltration (polyglonal cells with eosinophilic cytoplasm and oval nuclei with longitudinal grooves), mitotic figures and increased number of eosinophilic neutrophils and plasma cells. Thus a diagnosis of EG was made. [Figure 4]

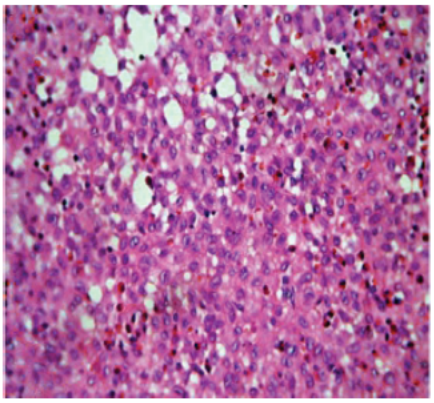

Figure 4: Sheet of aggregation of histiocytes and numerous eosinophils (H & E staining 100X)

Additionally whole body bone scintigraphy with a Tc-99m was performed to evaluate entire skeletal system. Beside the right side of the mandible which had shown hotspot, renal activity, minimal soft-tissue activity and Urinary bladder had increase in radiotracer uptake which are normally present. No other site of bone involvement was found. The patient underwent surgical resection of the mandible from distal root of left canine to right ramus and titanium reconstruction plate was placed as a spacer. The

patient had been followed-up after 9 months, showing no sign of recurrence. [Figure 5]

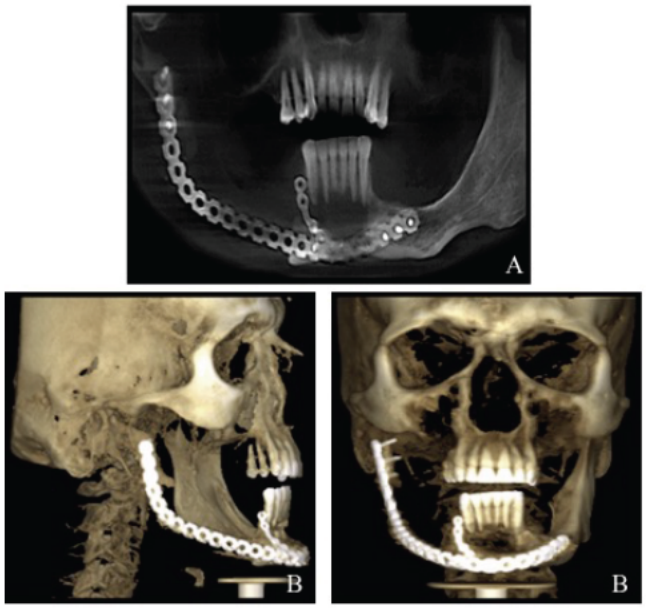

Figure 4: A, reformat panoramic .B, three-dimensional view. CBCT shows segmental mandibulectomy and the reconstruction plate.

Discussion

Lichtenstein and Jeffe were the primary representatives of the term "EG of bone". This is a condition in which its cause of occurrence is still a mystery and appears from clustery proliferation of Langerhans cells.[6] EG of bone can occur both as monostotic and polyostotic lesions. According to Hartman, 18 the monostotic shape of eosinophilic granuloma is predominant that polystotic form. Mandible, ribs, vertebrae, the skull (specially parietal bone) and femur are the bone-made areas that are more likely to be influenced by eosinophilic granuloma.[7] In the occasion that the jaw is affected, maxilla is a less frequent area than the mandible. When more than one lesions appear, the period that takes for the new osseous lesions to occur is 1-2 years.[2] Lesions in the long bones are rarely located in the epiphydis with the ratio of 2% and frequently appear in the diaphysis (58%). According to the observation done by Miyamoto *et al*, 7.9% involved the jaw with the angle and body being the main focus area of the disease. In the areas of neck and head, EG mostly influences soft tissues connected to the involved bones.[8]

In the clinical demonstration of eosinophilic granuloma age is a differentiating factor. This disease could have no symptoms at all or it could cause swelling, loosening of teeth, pain, and limitation of mouth opening. In adults, the disease usually spreads through one organ (as mentioned before) may have to no symptoms. Retrogression could be an impromptu event. In this unique cases a male of 29 years—which is an unusual age for this disease—was diagnosed with EG.

Eosinophilic granuloma is illustrated as an osteolytic lesion with identifiable boundaries in radiograph images. However, the bounds seem scattered and barely specifiable in certain cases. If the lesions have invaded the alveolar, the images may feature "scooped out-like" figures. In quite a lot of occasions, radiography has showed discriminatory disintegration of alveolar bone, inducing the image to appear "tooth floating-like".[9]

The stage in which the disease is now progressing in, is an important variable to the the radiographic characteristics. Barely demarcated boundaries are expected in the primary phase though the borders become more defined in the second stage of the progression. In an advanced stage, a broad peel of sclerotic tissue may appear. The osteolytic process is observable, being famous for its appearance in the medullary cavity, on occasion, in the cortex and the cortical bone could be punctured or the periosteal bone reaction may happen.[8] In this certain case, the medullary were the mandibular lesion was located, with a perforation of buccal and lingual cortex but no figuration of periosteal bone formation was taking place. [Figure 3]

The multivariate radiographic images cause more difficulty in the inconstant diagnosis of eosinophilic granuloma. Osteomyelitis, an odontogenic cyst, or a malignancy are the conditions that are like for the lesion to be mistaken for it.The scooped out feature causes other alternatives to rise, such as; Periodontal disease, lymphoma, Ewing's sarcoma, and metastatic disease.[10] In the present case, suspicion went toward a myltiple myeloma since the jaw lesion showed lytic and punched out appearance of the mandible.
Periodontal diseases that are famed in aggression are clinically heterogeneous and contain diseases that may blur the line when it comes to distinguishing them from other types of periodontitis, and multiple types of disease significantly confined clinical appearance with considerable ruination at an early age.[11] Constant attachment loss and bone destruction restricted in first molar and incisor teeth combined with interproximal attachment loss on at least two permanent teeth, causes manifestation of of LAP, according to radiographic images.[12]

On account of the patient previous history of localized aggressive periodontitis and later on the development of EG and their similar clinical and radiological appearance had led us to believe this to be an intriguing case to be reported.

References

1. Sherwani RK, Akhtar K, Qadri S, Ray PS. Eosinophilic granuloma of the mandible: a diagnostic dilemma. BMJ Case Rep 2014;2014:bcr2013200274.
2. Lichtenstein L. Histiocytosis X; Integration of eosinophilic granuloma of bone," Letterer-Siwe disease", and" Schuller-Christian disease" as related manifestations of a single nosologic entity. AMA Arch Pathol 1953;56(1):84-102.
3. Ando A, Hatori M, Hosaka M, Hagiwara Y, Kita A, Itoi E. Eosinophilic granuloma arising from the pelvis

in children: a report of three cases. Ups J Med Sci 2008;113(2):209-16.
4. Guruprasad Y, Chauhan DS. Solitary eosinophilic granuloma of mandibular condyle: literature review and report of a rare case. J Maxillofac Oral Surg 2015;14(Suppl 1):209-14.
5. Vandana KL, Desai R, Banupurmath CR, Kartik M. Eosinophilic granuloma with oral manifestations: A case report. J Indian Soc Pedo Prev Dent 2003;21(3):105-7.
6. Lichtenstein L, Jeffe HL. Eosinophilic granuloma of bone: With report of a case. Am J Pathol 1940;16(5):595-604.
7. Hartman KS. Histiocytosis X: a review of 114 cases with oral involvement. Oral Surg Oral Med Oral Pathol 1980;49(1):38-54.
8. Huysse W, Hogendoorn P, Bloem J, de Schepper A. Eosinophilic granuloma of the skull. JBR BTR. 2006;89(3):134-135.
9. Ardekian L, Peled M, Rosen D, Rachmiel A, Abu el-Naaj I, Laufer D. Clinical and radiographic features of eosinophilic granuloma in the jaws: review of 41 lesions treated by surgery and low-dose radiotherapy. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1999;87(2):238-42.
10. Rees J, Paterson AW. Langerhans cell histiocytosis in an adult. Br J Oral Maxillofac Surg 2009;47(1):52-3.
11. Araujo M. Localized juvenile periodontitis or localized aggressive periodontitis. J Mass Dent Soc 2002;51(2):14-8.
12. Teughels W, Dhondt R, Dekeyser C, Quirynen M. Treatment of aggressive periodontitis. Periodontol 2000. 2014;65(1):107-33.

Corresponding Author

Dr. Shabnam Mohammed Charlie
Resident,
Department of Oral & Maxillofacial Radiology,
School of Dentistry,
Tehran University of Medical Sciences,
Tehran, IRAN
Email Id: - shabnamch@yahoo.com