SCIENTIFIC ARCHIVES OF DENTAL SCIENCES

Volume 2 Issue 11 November 2019

Case Report

Mandibular Eosinophilic Granuloma Remission and Healing Post Biopsy

Assad Mehmood Khan[1]* and Tumadhir Ahmed Al-alwan[2]

[1]*Associate Consultant Maxillofacial Surgery, King Abdul-Aziz National Guard Hospital, Al-Ahsa, KSA*

[2]*General Dentist at Happiness Dental Clinic, Al-Ahsa, KSA*

***Corresponding Author:** Assad Mehmood Khan, Associate Consultant Maxillofacial Surgery, King Abdul-Aziz National Guard Hospital, Al-Ahsa, KSA.

Received: September 26, 2019; **Published:** October 11, 2019

Abstract

Langerhans cell histiocytosis (LCH), previously known as histiocytosis X, is a rare disease of unknown etiology. It is a clonal proliferation of dendritic cells of the immune system, which can affect multiple organ systems and range in behavior from a benign inflammatory process to a much more aggressive process. Not many cases have been documented in the Jaws. This case is an example of Langerhans cell histiocytosis in the right side mandible, which resolved spontaneously following an incisional biopsy without any further surgical intervention.

Keywords: Langerhans's cell histiocytosis; Eosinophilic Granuloma; Mandible; Spontaneous Resolution

Introduction

Histiocytosis X was introduced by Lichten-stein in 1953 (Lichtenstein, 1953) of a group of proliferative granulomatous disorders with histiocytic appearance of their cells. The disease, LCH, as originally identified, was grouped into three categories. (1) Letterer-Siwe syndrome (acute disseminated), (2) Hand-Schuller-Christian syndrome (chronic disseminated) and (3) eosinophilic granuloma (chronic localized). A workshop on childhood histiocytosis changed the name of this collection to Langerhans' cell histiocytosis, because of the main proliferative component of the cellular population. Langerhans' cell histiocytosis is also found to be a clonal disorder [1]. Localized Langerhans cell histiocytosis (LLCH), previously known as eosinophilic granuloma, is an intrabony lesion with an eosinophilic infiltrate confined to the bone without visceral involvement. It comprises of 60% - 70% of all cases of LCH and can be found as solitary or multifocal osseous defects. The skull, mandible, and ribs are most commonly affected in children. In adults, vertebrae and long bones are most commonly involved. The occurrence of the disease is seen to be higher in the first 3 decades of life, with males being affected twice as common as females [2].

The etiology is unknown, and different theories suggest environmental, infectious, immunologic, and genetic involvement [3].

Symptoms vary from mild to moderate pain and in some cases pain free, swelling, and tenderness over the site of the lesion. Radiographs are suggestive of radiolucent areas with well demarcated borders. The overlying buccal cortical bone expansion and resorption may occur. Big mandibular lesions may end up with pathological fractures. The jaw lesions can a have differential diagnosis, of periodontal diseases, apical cysts, ameloblastoma, central giant cell granuloma, vascular malformations, and malignancies [4].

The treatment of Langerhans' cell histiocytosis has changed and varied from the past. A review of the literature shows and confirms a lack of consensus. Patients with Langerhans' cell histiocytosis have been treated with antibiotics, steroids, radiation and in some cases, with cytotoxic chemotherapy. Spontaneous resolution and remission of the lesions and leading into healing of the disease has also been documented [5,6]. Some case reports suggest complete resolution of the disease with intra lesional steroids [4,8-10]. Surgical treatment varies from resections and reconstruction to more conservative approaches where remission of the disease process can be achieved by just biopsy.

Case Report

An 11-years-old boy was referred to the maxillofacial surgery clinic on March 2011 with a 4 to 5 months history of pain and

tenderness in the posterior mandible. His pain was dull and compressive in character and had recurrent episodes roughly two to three times per week. He also complained of slow growing swelling in the buccal vestibule of posterior mandible.

The patient was previously examined by his dentist in secondary care hospital and was referred to us for assessment and possible treatment. He did not have any systemic diseases, and clinical examination revealed a right mandibular posterior vestibular swelling with buccal cortical expansion extending to the angle of the mandible and ascending ramus. The swelling was tender, non-fluctuant, with no erythema, no trismus, no discharging sinus. Intra orally there was normal mucosal and gingival color. Missing third molars in all quadrant.

Radiographically a mandibular multilocular well defined lesion was seen in the posterior mandible and ascending ramus (Figure 1 and 2).

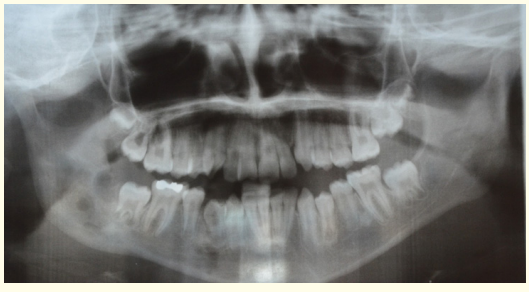

Figure 1: *A lytic multilocular radiolucent lesion at posterior mandible.*

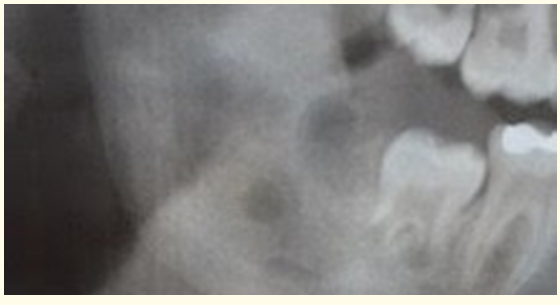

Figure 2: *Magnified view of OPG of the right side posterior mandible showing lytic multilocular lesion*

An incisional biopsy was performed under General anesthesia consisting of two specimens one having buccal cortical bone with lesion and the other from the lesion itself with minimal curettage.

Histologic examination of the biopsy specimen showed the presence of multinucleated histiocytes, numerous eosinophils, consistent with Langerhans cells, and the features were compatible to eosinophilic granuloma (Figure 3a and 3b). No additional lesions were found elsewhere in the body.

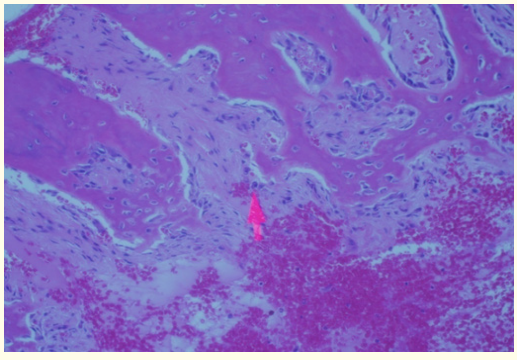

Figure 3a: *Paraffin embedded sections from right mandibular cortical bone Stained with H&E MP: 40X Showing eosinophilic infiltration.*

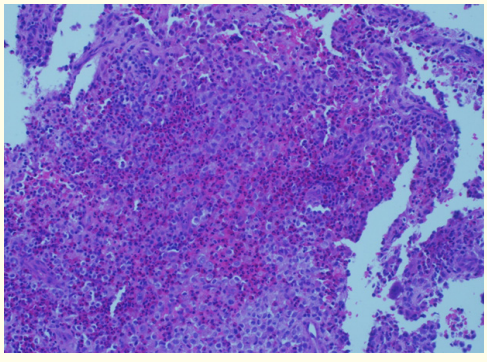

Figure 3b: *Paraffin embedded H&E stained section of right mandibular swelling showing heavy infiltration with eosinophils MP: 40 X.*

It was decided that the mandibular lesion would be monitored with OPG scans and should be kept on follow up for at least 2 to 3 years. This decision was undertaken because the mandibular lesion was solitary in nature and there was no systemic involvement. Keeping in mind the knowledge that intra osseous lesions of LCH can heal and resolve spontaneously after taking a biopsy. So, conservative approach was undertaken.

An OPG after 2 year revealed almost complete ossification of the lesion (Figure 4), which was again followed up a year later (Figure

5) and showed complete resolution of the lesion along with eruption of right mandibular 2nd molar.

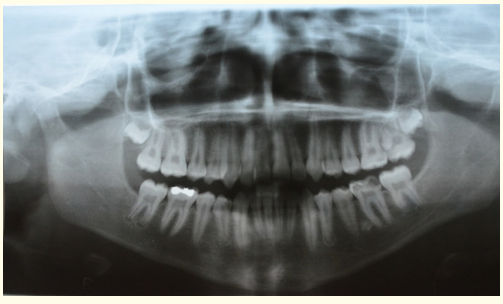

Figure 4: *This OPG is after 2 years of biopsy showing complete resolution of lesion and ossification at right posterior mandible.*

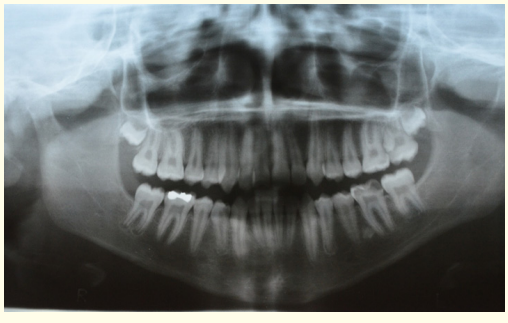

Figure 5: *OPG after 3 years of biopsy. Showing complete resolution and healing of the lesion with eruption of mandibular right second molar.*

Discussion

LCH can present itself in many clinical ways, like unifocal disease (solitary eosinophilic granuloma), multifocal disease (including cases of Hand-Schuller-Christian syndrome) and multifocal multisystem disease including cases of Letterer-Siwe syndrome [11-14]. Prevalence of male to female ratio is 2:1.

Langerhans' cell histiocytosis can be present at any age but usually is common in the young age group. In maxillofacial region, LCH can manifest in a variety of ways including swelling, pain, gingivitis, ulceration, mobility of teeth with periodontal involvement and even pathological fracture [15]. In a study by Angeli., *et al.* 25 patients with head and neck lesions from 4 different centers were reviewed. All the solitary skull and mandibular lesions were treated with biopsy and curettage and showed remission and healing of the disease process [16].

The consensus on solitary bone lesions is that they only require biopsy and curettage. Others report the use of intralesional steroids [9,17]. Spontaneous resolution and regression of the disease process have been reported by several clinicians and reported accordingly [19-21].

The case presented here advocate and affirm the use of conservative approaches to management of solitary bone lesions of the facial skeleton. Also a study from Japan had a similar approach to mandibular lesions [22].

The theory and logic behind spontaneous remission and healing of the lesion is difficult to explain. It can be assumed that curettage and biopsy of the lesions could stimulate and elicit an inflammatory response, ending in the maturation of the Langerhans cells with healing and resolution of lesion.

Conclusion

The case presented in this case report supports the idea that a conservative approach appears to be suitable and reliable in treating solitary bone lesions of LCH. Keeping in mind that all patients need thorough investigation to eliminate involvement of other systems.

Bibliography

1. Willman CL, Busque L, Griffith BB, Favara BE, McClain KL, Duncan MH, Gilliland DG. Langerhans' cell histiocytosis (histiocytosis X), a clonal proliferative disease. N Engl J Med. 1994;331(3):154-160.

2. SJ Key, CJ O'Brien, KC Silvester, SJ Crean. Eosinophilic granuloma: resolution of maxillofacial bony lesions following minimal intervention: Report of three cases and a review of the literature. J Craniomaxillofac Surg. 2004;32(3):170-175.

3. I Alajbeg, V Vucicevic Boras, R Femenic, A Cekic-Arambasin, M Anicic, J Kelecicc, IZ Alajbeg. Unrecognized oral manifestations of Langerhans cell histiocytosis which progressed to systemic disease. Oral Oncol. 2006;42(1):10-13.

4. LR Jones, BB Toth, A Cangir. Treatment for solitary eosinophilic granuloma of the mandible by steroid injection: report of a case. J Oral Maxillofac Surg. 1989;47(3):306-309.

5. C Nauert, J Zornoza, A Ayala, TS Harle. Eosinophilic granuloma of the jaw: diagnosis and management. Skeletal Radiol. 1983;10:227-235.

6. T Namai, H Yusa, H Yoshida. Spontaneous remission of a solitary eosinophilic granuloma of the mandible after biopsy: a case report. J Oral Maxillofac Surg. 2001;59(12):1485-1487.

7. M Cohen, J Zornoza, A Cancjir, JA Murray, S Wallace. Direct injection of methylprednisolone sodium succinate in the treatment of solitary eosinophilic granuloma of bone: a report of 9 cases. Radiology. 1980;136(2):289-293.

8. RM Egeler, RC Thompsom, PA.Voute, ME Nesbit. Intralesional infiltration of corticosteroids in localized Langerhans' cell histiocytosis. J Pediatr Orthop. 1992;12(6):811-814.

9. IM Watzke, W Millesi, C Kermer, Gisslinger. Multifocal eosinophilic granuloma of the jaw: long term follow-up of a novel intraosseous corticoid treatment for recalcitrant lesions. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2000;90(3):317-322.

10. TF Putters, JGAM Visscher, A van Veen, FKL Spijkervet. Intralesional infiltration of corticosteroids in the treatment of localised langerhans' cell histiocytosis of the mandible: Report of known cases and three new cases. Int J Oral Maxillofac Surg. 2005;34(5):571-575.

11. Schuller A. Uber eigenartige Sch adeldefekte im Jugendalter. Fortschr Rontgenstrahlen 1915;23:12-18.

12. Christian H. Defects in membranous bones, exophthalmos and diabetes insipidus; an unusual syndrome of dyspituitarism. Med Clin North Am. 1920;3:849-871.

13. Hashimoto K, Bale GF, Hawkins HK, Langston C, Pritzker MS. Congenital self-healing reticulohistiocytosis (Hashimoto-Pritzker type). Int J Dermatol. 1986;25(8):516-523.

14. Nezelof C, Frileux-Herbet, Cronier-Sachot J. Disseminated histiocytosis-X: analysis of prognostic factors based on a retrospective study of 50 cases. Cancer. 1979;44(5):1824-1838.

15. Holzhauer AM, Abdelsayed RA, Sutley SH. Eosinophilic granuloma: a case report with pathological fracture. Oral Surg Oral Med Oral Pathol Endod. 1999;87(6):756-759.

16. Angeli SI, Hoffman HT, Alcade J, Smith RJ. Langerhans' cell histiocytosis of the head and neck in children. Ann Otol Rhinol Laryngol. 1995;104(3):173-180.

17. Egeler RM, De Kraker J, Voute PA. Cytosine arabinoside, vincristine and prednisolone in the treatment of children with disseminated Langerhans' cell histiocytosis with organ dysfunction; experience at a single institution. Med Pediatr Oncol. 1993;21(4):265-270.

18. Ladisch S, Gadner MD. Treatment of Langerhans' cell histiocytosis-Evolution and current approaches. Br J Cancer. 1994;70(23):S41-S46.

19. Broadbent V, Pritchard J. Histiocytosis-X. Current controversies. Arch Dis Child. 1985;60(7):605-607.

20. Broadbent V, Pritchard J, Davies EG, Levinsky RJ, Heaf D, Atherton DJ, Pincott JR, Tucker S. Spontaneous remission of multi-system Histiocytosis X. Lancet 1984;1(8371):253-259.

21. Raney RB, D'Angio GJ. Langerhans' cell histiocytosis (histiocytosis X). Experience at the Childrens Hospital of Philadelphia, 1970-1984. Med Ped Oncol. 1989;17(1):20-28.

22. Kitano M, Landini G, Semba I, Aurago A, Sugihara K, Mukai H. Eosinophilic granuloma showing rapid regression. Report of a mandibular case with application of a modified PNA staining method for demonstration of Langerhans' cell histiocytosis. Acta Pathol Jpn. 1990;40(8):588-595.

Volume 2 Issue 11 November 2019