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Primary Xanthoma of the Mandible – A Case Report

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***Abstract**: Xanthomas are extremely rare benign lesions occurring in the bone. They are even rarer in the jaw bonescommonly affecting the mandible more. Primary xanthomas are ones which occur without any systemic manifestations. The radiographic findings are variable hence diagnosis is usually made with biopsy. Histo-pathologically, they are characterised by abundant histiocytes, foamy macrophages and multinucleated giant cells. Here we report a case of primary xanthoma of the mandible without any systemic diseases.*

***Keywords:** Xanthoma, Histiocytes, foamy macrophages, Multi-nucleated giant cells*

I. Introduction

Xanthoma of the jaw bones is a very rare benign lesion. Xanthoma derived from the Greek word "Xanthos" meaning yellow.[1] They affect various parts of the body; skin, tendons, flat bones, cerebral parenchyma etc.[2] The most frequent location of bone xanthoma is the diaphysis of long bones, especially the tibia. Other locations may be particularly seen in the facial skeleton, mastoid air cells and mandibular bone.[3]In the absence of systemic diseases, the lesion is called a Primary Xanthoma. Primary mandibular xanthomas are extremely rare. In this case report we describe a case of xanthoma in a 14year old boy localized in the mandibular anterior region.

II. Case report

A 14 year old male patient reported with a chief complaint of pain in the lower anterior region since one month. No carious teeth or any infection was noted. Overlying mucosa was intact. Therefore a radiograph was advised. Since OPG revealed a radiolucency in the lower anterior region of 41, 31, 32 and 33 region, it was decided forapicoectomy of 41, 31. The lesion removed was intact and sent for histopathological examination.

Histopathologically, the lesion showed connective tissue devoid of epithelium. The lesional tissue showed abundant number of round to ovoid cells with pale eosinophilic cytoplasm suggestive of foam cells-macrophages. The connective tissue showed abundant collagen fibres interspersed with fibroblasts, inflammatory cell infiltrate chiefly lymphocytes, plasma cells and macrophages. Multinucleated giant cells which vary in size and shape containing a few and up to 25 nuclei with areas of bone tissue, entrapped osteocytes, lining of osteoblasts and blood vessels, some of which showed endothelial proliferation and areas of hyalinization were observed. The overall picture was suggestive of Primary Xanthoma of the mandible. Immunohistochemical study was done with CD68 which showed strong positivity of the foamy macrophages. CD 68 is a glycoprotein and expressed on monocytes and macrophages.

III. Discussion

Xanthomas are extremely rare bone tumours. Cases are often reported in the long bones and axial skeleton. They commonly represent the manifestation of diseases that affect the lipid, cholesterol or glucose metabolism.[4,5] In cases associated with systemic diseases, the lesions are usually multiple. The xanthomas are included in the xanthomatous lesions group, which are composed of macrophages with a large, eosinophilic and granular cytoplasm called xanthomatous or foamy cells.[4,5]

Bone xanthomas most commonly occur in male patients, mainly between the third and the fifth decades.[6] The most commonly affected sites are the bones of the hand, although cases located in the long and pelvic bones, rib, cranium and vertebrae have also been reported. In the jaws, xanthomas are extremely rare, and most of them are reported as primary and occur exclusively in the mandible.[7]It is difficult to establish the exact number of xanthomas of the jaws reported in literature since several lesions are described only as xanthomatous lesions or inadequate terms and synonyms are used.[8]

The pathogenesis of the xanthomas consist of lipid leakage from the blood vessels in the site of the lesion with subsequent phagocytosis of this material by the macrophages. The non-degraded cholesterol

accumulates within the cytoplasm, leading to the presence of foamy macrophages. Minor trauma could be the etiologyinvolved in the development of xanthomas.[9]

The radiographic picture of xanthoma is variable. Radiographically the lesion is well-defined radiolucent with a sclerotic border. Our case also showed a similar picture. Histopathologically, the differential diagnosis include other bone lesions which show xanthomatous changes (showing foamy macrophages). It is necessary to evaluate the patient for systemic diseases, so as to rule out other diseases like Langerhans cell histiocytosis, Benign Fibrous Histiocytoma and Fibrous Dysplasia.[7, 10]In our case patient did not show any of these disorders.

Surgical removal is an adequate treatment. Xanthomas associated with lipid or metabolic disorders tend to recur. Therefore a treatment for the systemic disease should beadministered.[9]

FM de Moraes Ramos –Perez et al in their article"Primary xanthoma of the mandible" concluded that xanthomas of the jaws seem to be primary and occur exclusively in the mandible.[7]

HasnaeGuerrouj et al reported "Xanthoma of bone - A Case Report in a 56 year old patient and observed that after treatment with fenofibrate and statins, it gradually regressed.[3]

A report of 3 cases of Xanthoma of bone with review of literature by Kris J Alden et al who mentioned that the lesions were treated with curettage and bone grafting and there was no evidence of local recurrence. These lesions were seen as occipital, iliac crest and tibial lesions.[11]

Sayed Ali et al in their article, Bilateral Primary Xanthoma of the humeri with pathologic fractures: A case report presented acase of hyperlipidemia in a 57 year old women with intra-osseous xanthoma with atypical features like multifocality, wide zone of transition and pathologic fractures.[12]

Zhuo Wang et al proposed that Xanthoma should belong to bone tumours of undefined neoplastic nature, the etiology of which needs more data collection and further analysis.[13]

Habib Ansarin reported "Xanthoma Dissemination with Tumour like lesion on face" with widespread yellow brown, papulonodular lesions on the face, flexors and trunk as Xanthoma disseminatum.[14]

Mohammad Ismail Hussain, reported a case of Primary Xanthoma of femur, where the patient did not have any lipid disturbance but had hypothyroidism.[15]

Tom Daley et al in the article"Central Xanthoma of the jaws - A clinico-pathologic entity?" suggested that these lesions should be considered as a unique rare clinico-pathologic entity and questioned whether the entity is a benign neoplastic process or a persistent reactive process?

IV. Conclusion

The purpose of reporting this case is to add to the existing literature as these entities are rarely reported. These lesions, whenever they occur, are commonly seen in the mandible. The case studied here presents with classical histopathological findings of foam cells and multinucleated giant cells. Immunohistochemical study done with CD68 showedpositive findings for foam cells reactivity.

V. Figures

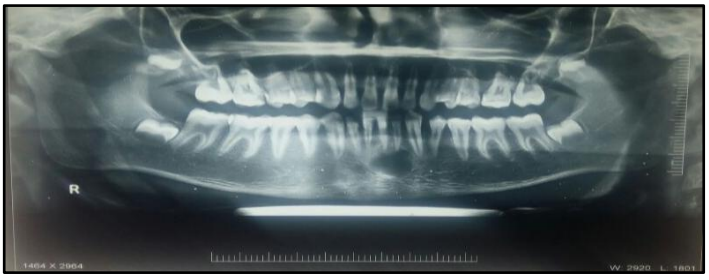


Fig 1 - OPG revealed a radiolucency in the lower anterior region of 41, 31, 32 and 33 region

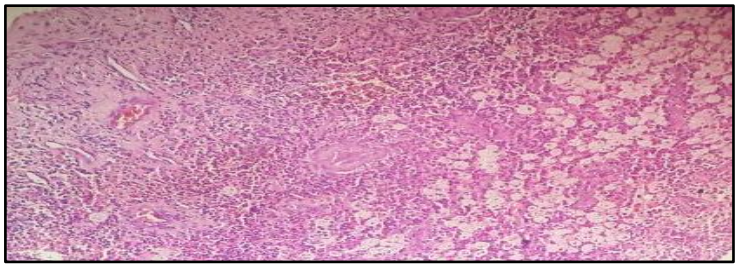


Figure 2- Low power view showing connective tissue with foamy macrophages and inflammatory infiltrate

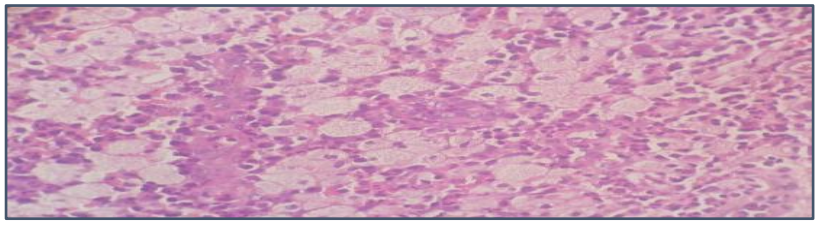
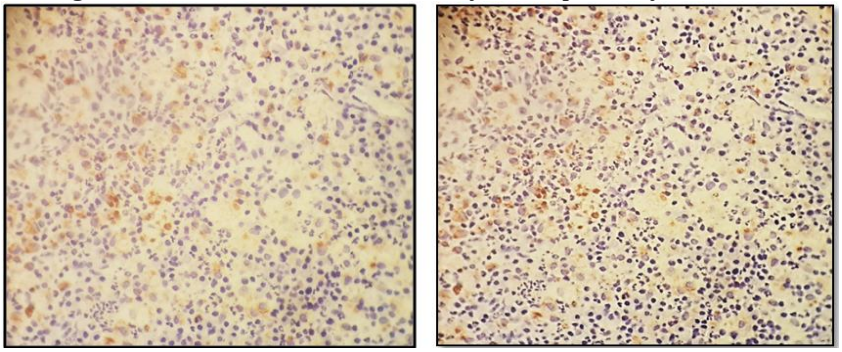


Figure 3 – High power view showing connective tissue with abundant foamy macrophages, multinucleated giant cells and inflammatory infiltrate

Figure 4 and 5 - Immunohistochemistry shows positivity with CD68



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