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Three Rare Cases of Mysterious Granulomas of Mandible in Adolescents - A Diagnostic Dilemma

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ABSTRACT

Due to relatively nonspecific clinical findings associated with a variety of granulomatous diseases, a microscopic diagnosis in it leads to diagnostic dilemma for the clinician. The true nature of this disease is controversial and remains unknown but it may be a reactive lesion, a developmental anomaly or a benign neoplasm. Often an extensive clinical, microscopic and laboratory evaluation may be required to identify the source of the granulomatous inflammation. In this case report we discuss bilateral idiopathic mandibular lesion in an 8 year old girl.

Keywords: Granulomatous inflammation, Benign, Mandible

INTRODUCTION

One of the distinctive forms of chronic inflammation is Granulomatous inflammation [1]. Granulomas are distinct structures composed of epithelioid-shaped macrophages, multinucleated giant cells, lymphocytes and fibroblasts. However, the clinical findings are often variable and indistinct when associated with granulomatous inflammation. Granulomatous inflammation has a multifactorial etiology and may arise as a reaction to environmental or genetic factors, infectious organisms or maybe idiopathic, for which there is no known trigger [2].

A typical differential diagnosis related to mandibular swelling includes:

- Ameloblastoma which is seen in older age, in posterior part of mandible and is multilocular.
- Odontogenic myxoma which is usually associated with missing or impacted tooth, multilocular with honey comb appearance.
- Dentigerous cyst.
- Cherubism with epicenter located in the posterior aspect of the mandible and maxilla, multiple and bilateral.
- Aneurysmal bone cyst which causes profound expansion and aspiration produces blood.
- Central giant cell granuloma.

Granulomas related to giant cells are usually an intraosseous lesion consisting of cellular fibrous tissue that may contains multiple foci of hemorrhage, aggregations of multinucleated giant cells and occasionally trabeculae of woven bone [3]. It was described by Jaffe in 1953 that separated giant cell lesions of jaw from other jaw lesions. He considered it to be a locally reparative reaction of bone, which can be either due to inflammatory response, local trauma or hemorrhage [4].

Clinically, it can vary from benign to rather aggressive lesion and demonstrates varying histopathological features [5]. Non-aggressive and aggressive variants are compared according to clinical and radiographic behavior. Non-aggressive lesions are slow growing, almost asymptomatic growth that does not perforate the cortical bone or induce root resorption and has low tendency to recur whereas aggressive lesions which are usually seen in younger patients are painful, rapidly growing with expansion or perforation of cortical bone, radicular resorption and high tendency to recur. He interprets that large functional surface area is occupied by giant cells and larger relative giant cells in aggressive lesion [6]. It occurs most commonly in children and young adults and has a female predilection. Lesions are located more commonly in the mandible mostly involving molar and premolar area and frequently cross the midline. Its presence in the mandibular body area, the entire ramus, condyle and coronoid leads to a therapeutic challenge

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for surgeons [7].

The treatment modalities include surgical excision either by curettage or en bloc resection and alternative non-surgical approaches such as intralesional corticosteroid injections, calcitonin injections and subcutaneous alpha interferon injections. Here, we report a case of aggressive unknown granulomas in mandible (which is very closely related to central giant cell granuloma) with emphasis on clinical, radiological and management of the lesion.

CASE REPORT I

An 8 year old girl came with complain of pain and swelling in lower left region of face since few months **(Figure 1)**.

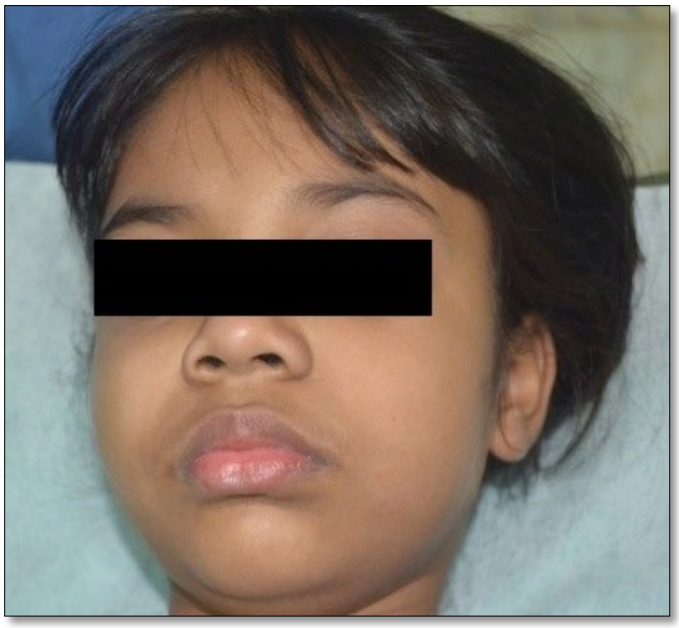


Figure 1. Pre-operative picture of patient.

It relieves on taking medication but reoccur as soon as medication stops. Patient has no relevant past medical or dental history. On oral examination nothing suspicious was found. Patient had good oral hygiene. Patient was advised for OPG. OPG revealed presence of radiolucency wrt to 36 and 46. Patient complained swelling only but OPG revealed similar lesion on the other side also which was not known to patient or parents as it was asymptomatic **(Figure 2)**.

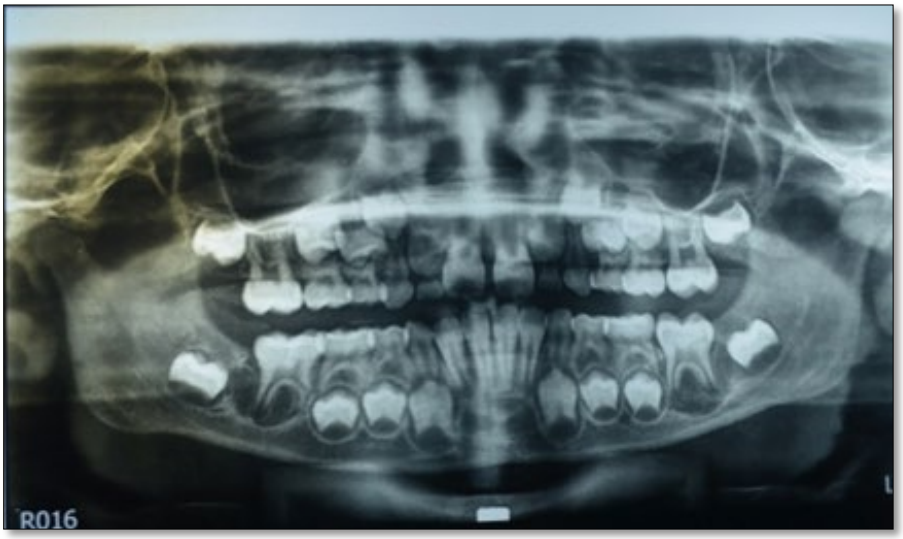


Figure 2. Pre-OP OPG.

Thereafter 3D-CT scan was done for precise evaluation and assessment of the lesion which showed bone destruction/perforation of cortical plate in that region, representing its aggressive nature **(Figure 3)**.

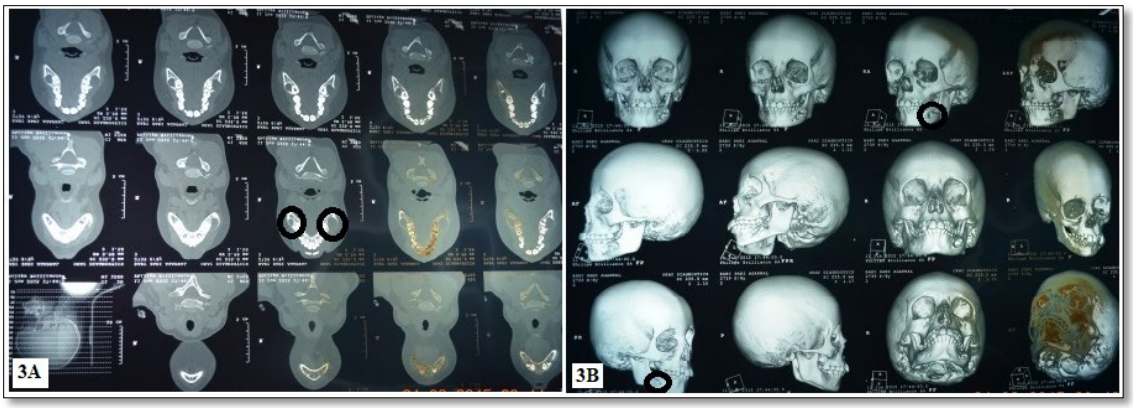


Figure 3. 3D CT scan.

Patient had no carious exposure, periodontal disease or any history of trauma. Looking at the aggressive nature of the lesion and bilateral involvement, it was planned to surgical excise the region and removes the tooth related to it, i.e., molar, after which only histopathological examination will ascertain its nature **(Figures 4A-4C)**.

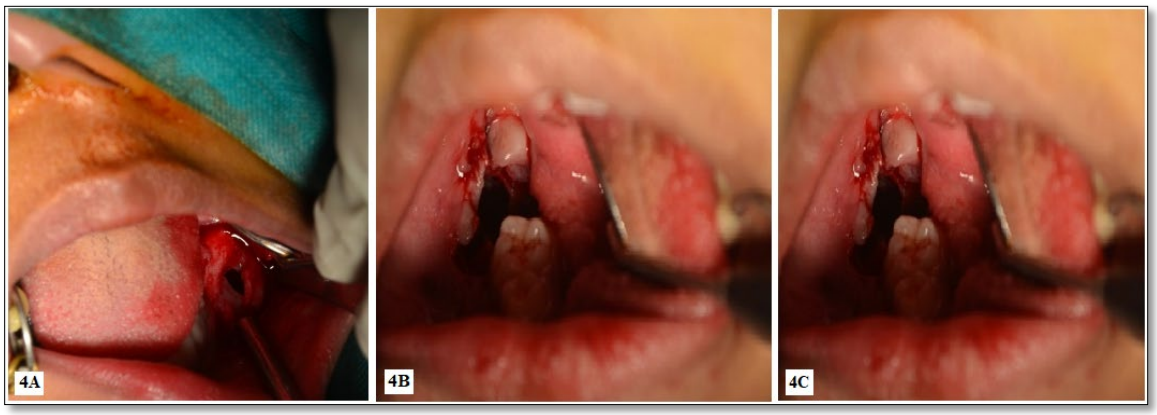


Figure 4. Surgical enucleation and excision of the lesion.

Following the careful enucleation, the tooth with the lesion (which was attached at and around the cemento-enamel junction) was sent for the histopathological examination **(Figure 5)**.

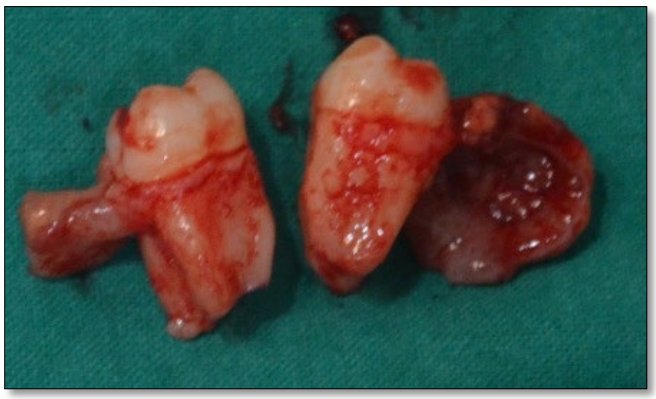
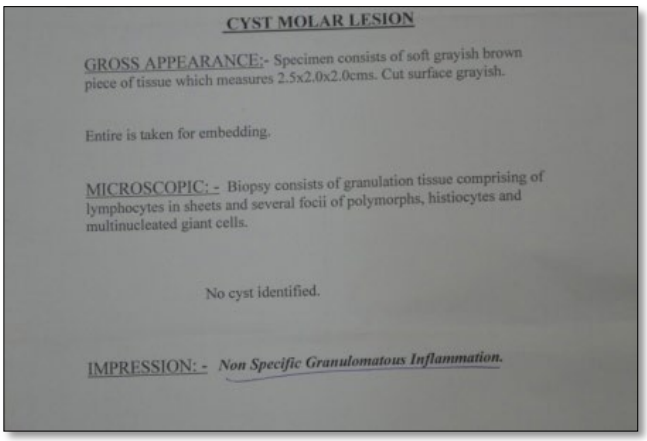


Figure 5. Both lesions associated with 36 and 46 molars send for biopsy.

The report revealed presence of lymphocytes, histiocytes and multinucleated giant cells. No cyst was identified. But diagnosis was not confirmed. The result was non-specific granulomatous inflammation **(Figure 6)**.



CYST MOLAR LESION

GROSS APPEARANCE:- Specimen consists of soft grayish brown piece of tissue which measures 2.5x2.0x2.0cms. Cut surface grayish.

Entire is taken for embedding.

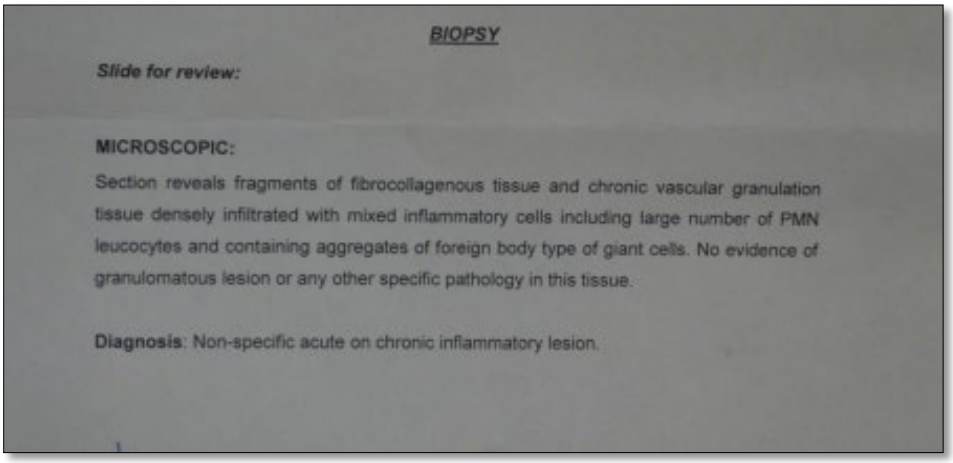
MICROSCOPIC: - Biopsy consists of granulation tissue comprising of lymphocytes in sheets and several focii of polymorphs, histiocytes and multinucleated giant cells.

No cyst identified.

IMPRESSION: - *Non Specific Granulomatous Inflammation.*

Figure 6. Surgical enucleation and excision of the lesion.

To cross check and confirm the report, specimen was sent to other labs also, but all failed to give a definitive diagnosis and similar presentation of giant cells, lymphocytes was reported **(Figure 7)**.


BIOPSY

Slide for review:

MICROSCOPIC:

Section reveals fragments of fibrocollagenous tissue and chronic vascular granulation tissue densely infiltrated with mixed inflammatory cells including large number of PMN leucocytes and containing aggregates of foreign body type of giant cells. No evidence of granulomatous lesion or any other specific pathology in this tissue.

Diagnosis: Non-specific acute on chronic inflammatory lesion.

Figure 7. Second biopsy report.

Patient was recalled after 6 months and no swelling or radiolucency was present. But actual cause was still unknown **(Figure 8)**.

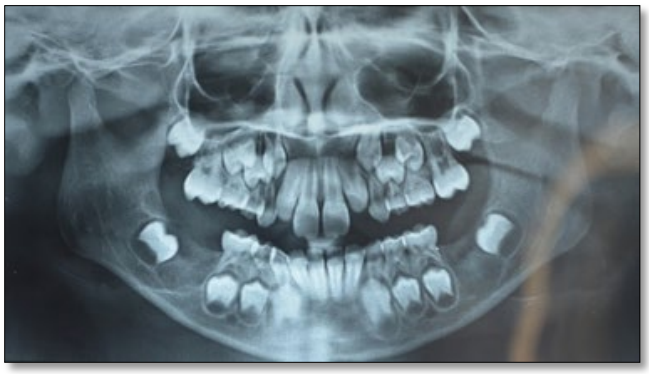


Figure 8. Follow up after 6 months.

After 2 years of follow up, the 37 and 47 have physiologically mesially migrated at the site of extracted first molars uneventfully **(Figure 9)**.

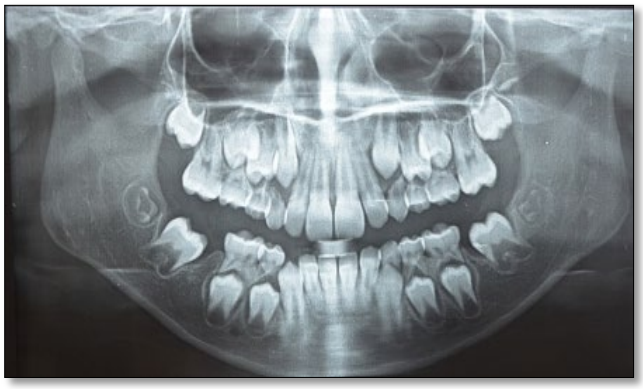


Figure 9. Follow up after 2 year.

CASE REPORT II

Similar case with exactly similar clinical presentation, radiographic finding, histopathological reporting and immediate recovery after similar intervention procedure irt to 46. The only difference was that this lesion was unilateral **(Figures 10-14)**.

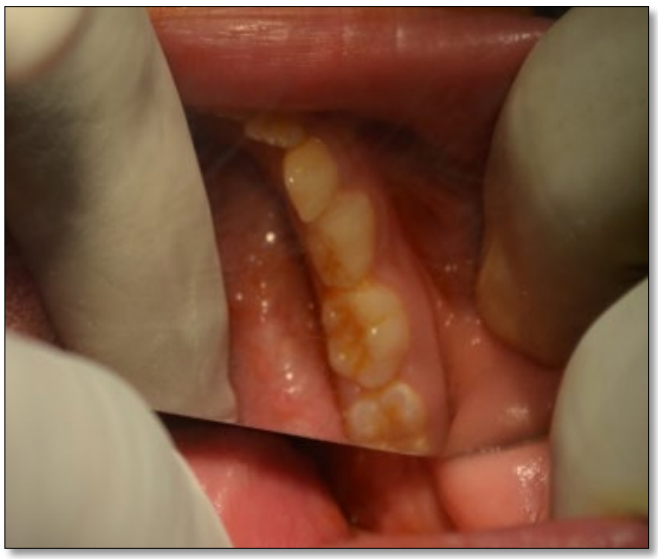


Figure 10. Pre-operative picture of patient.

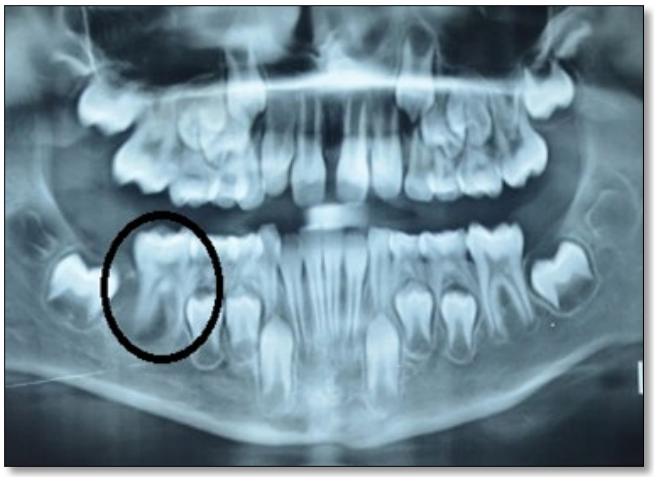


Figure 11. Pre-op OPG pre-op OPG.

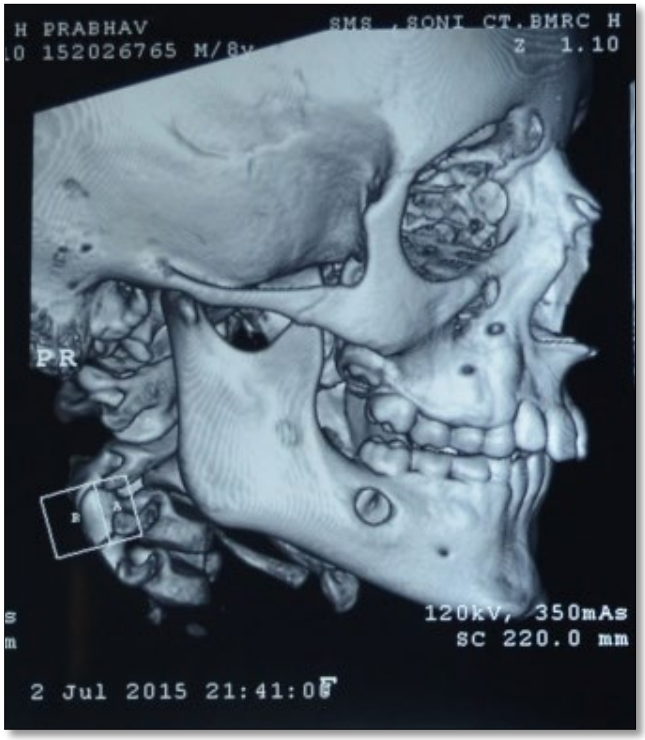


Figure 12. 3D CT-scan.

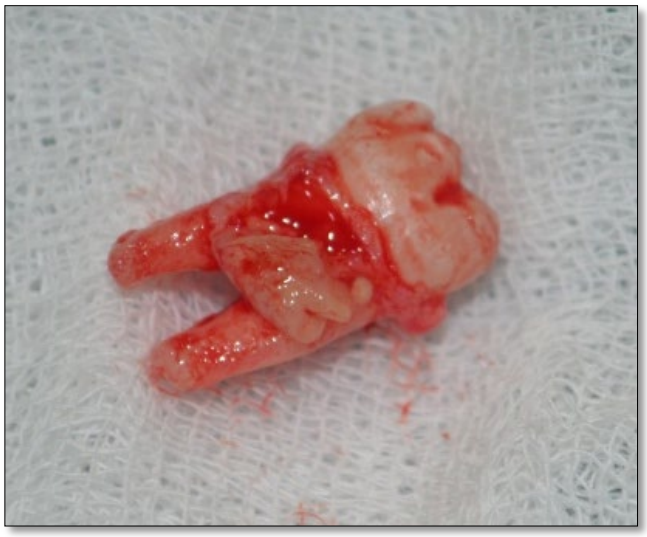


Figure 13. Surgical enucleation and excision of the lesion.

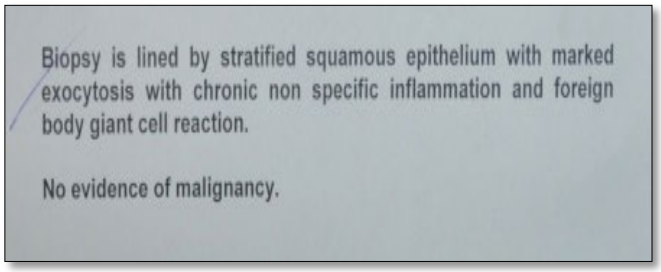


Figure 14. Biopsy report.

CASE REPORT III

Another case with exactly similar clinical presentation, radiographic finding, histopathological reporting and immediate recovery after similar intervention procedure irt to 24,25, but this time the lesion was unilateral and in maxilla **(Figures 15-19)**.

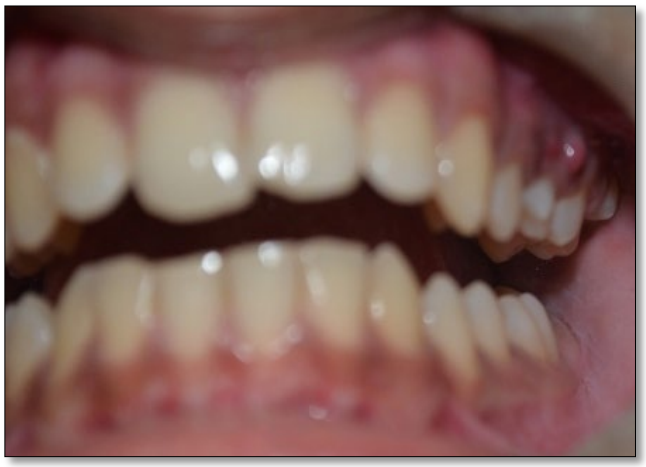


Figure 15. Pre-operative picture of patient with intraoral sinus irt 24 and 25.

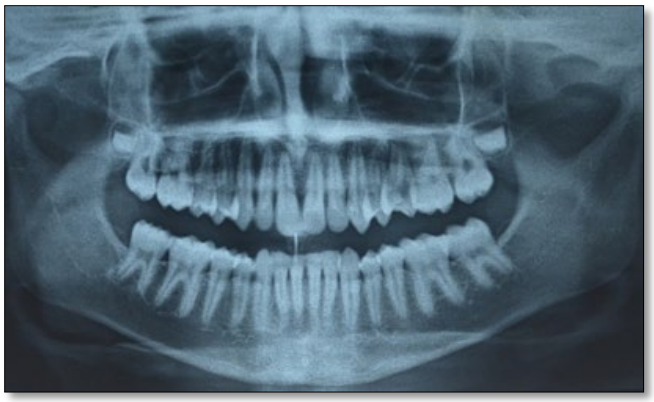


Figure 16. Pre-op Op.

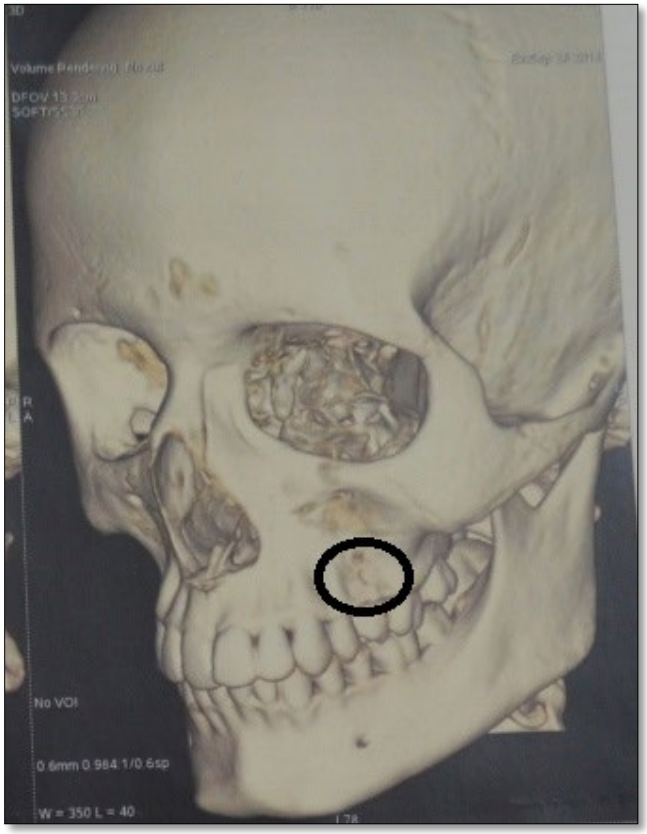


Figure 17. 3D CT-scan.

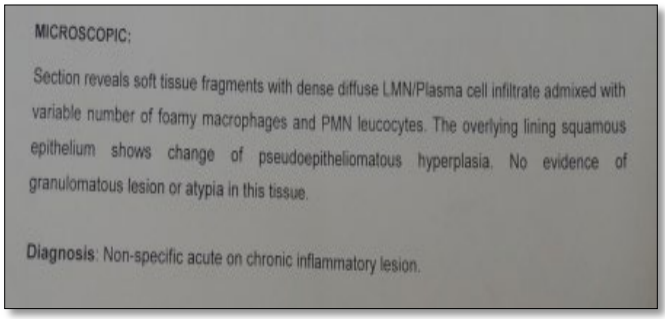


Figure 18. Biopsy report.

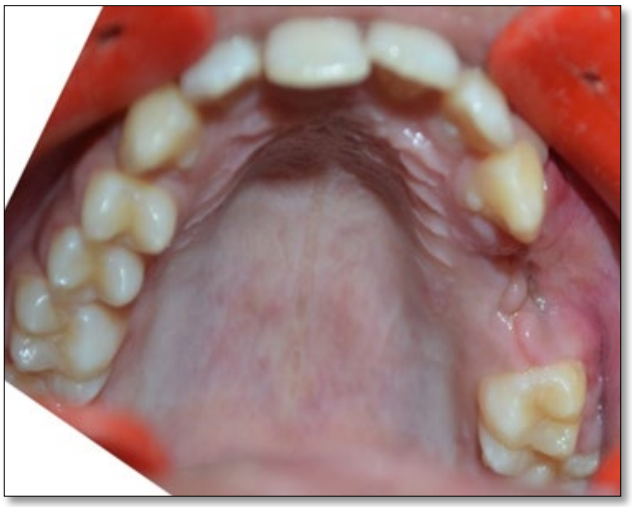


Figure 19. Post-operative healing.

DISCUSSION

In 1953, Jaffe [4] described an apparently reactive intraosseous lesion of the mandible and maxilla following trauma induced intraosseous hemorrhage and containing prominent giant cells. He separated giant cell lesions from long bone giant cell tumor based on differences in their histological pattern and clinical behavior [8]. Due to its reactive in nature, he gave the term reparative giant cell granuloma [9].

It is the disease of the young presenting as a painless expansible mass ranging from a slowly growing asymptomatic swelling to an aggressive lesion causing pain and bone destruction, being more common in the anterior portion of the mandibular body sometimes crossing the midline, the epicentre being anterior to the first molar region. It is an uncommon benign lesion of the jaw which accounts for <7%. Usually in this cortical plates are thinned, with sometimes perforation but gross soft tissue involvement remains limited [10].

In the present case, biopsy report revealed presence of lymphocytes, histiocytes and multinucleated giant cells. No cyst was identified. The result was non-specific granulomatous inflammation. The sign and symptoms were similar to central giant cell granuloma.

But upon enucleation, we can clearly appreciate that the lesion was attached at the cementoenamel junction, just like in dentigerous cyst, making it having mixed presentations.

CT scan showed excessive bony thinning and destruction with resorption of buccal cortical plates, showing signs of its aggressive nature.

In present case, the radiologic features of granuloma was not been clearly defined; the lesion appeared as unilocular radiolucency with ill-defined margins with varying degrees of expansion of the cortical plates. Radiographic appearance

of the lesion is not pathogenomic and may be confused with that of many other lesions of the jaws [6]. Various methods have been described for the treatment of such lesion of jaws. Most often treatment used is Curettage alone or in combination with resection with or without continuity loss [11]. Surgical treatment is usually varied depending upon the anatomic location, size of lesion, clinical behavior, periosteal or nerve involvement. But in this case, perforation of cortical plates prompted us for surgical resection and removal of molars related to the lesion.

CONCLUSION

Granulomatous inflammation may manifest in the oral cavity and usually with an array of non-specific clinical findings. Thus, an extensive clinical, microscopic and laboratory evaluation may be required in order to identify the source of the granulomatous inflammation. However, if the lesions are timely identified and appropriate therapy rendered, prognosis of the condition is significantly improved. This case highlights the difficulty in diagnosing and management of the lesion. Hence, this lesion continues to develop the interest and mystify the clinicians.

DECLARATION OF PATIENT CONSENT

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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