Case Report

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A Rare Case of Central Giant Cell Granuloma Mimicking Solitary Bone Cyst.

Abstract:

Background: Central giant cell granuloma is an uncommon benign tumor of jaws, most prevelant in anterior region of mandible. It has both aggressive and non aggressive nature and occurs in young adults.

Case Presentation : We present a case of a central giant cell granuloma in a 21 year old boy who presented with all the features of solitary bone cyst. Lesion was present in right body of mandible and diagnosis was made histopathologically followed by surgical enucleation.

Conclusion : Our case demonstrates a rare clinical and radiographic appearance of central giant cell granuloma which was also an accidental finding. Prompt diagnosis and treatment plan can prevent further damage and improve outcome.

Key-words: Central Giant Cell Granuloma, Solitary Bone Cyst, Odontogenic keratocyct, Aneurysmal Bone Cyst, Multinucleated Giant Cells

Introduction:

World Health Organization defined Central giant cell granuloma (CGCG) as an intraosseous lesion consisting of cellular fibrous tissue that contains multiple foci of hemorrhage, aggregations of multinucleated giant cells, and a few trabeculae of woven bone [1]. It was first described by Jaffe in 1953 as a reparative granuloma of jaw bones. But the term reparative is bygone, as CGCG causes the destruction of involved bones[2].

The nature of this lesion is contentious with three theories prevailing are - being reactive, a developmental anomaly, or a benign neoplasm[3]. Though there is no known aetiology and pathogenesis, its histology and clinical behaviour has been studied in detail[4].

CGCG accounts for approximately 7% of all benign tumors of the jaws[5] and present with a higher frequency in the mandible than maxilla. There is a slight female predilection, with a peak age of incidence range between 10 to 25 years. Clinically these lesions cause facial swelling, asymmetry, and expansion of cortical plates and radiographically resorption of roots of teeth with cortical perforation[2].

Here we present a case report of central giant cell granuloma of mandible in a young adult which was initially diagnosed as solitary bone cyst.

Case Report:

A 21 year old boy came to the department of Oral Medicine and Radiology, Rishiraj College Of Dental Sciences and

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Research Centre, Bhopal, Madhya Pradesh with the chief complaint of pain in right lower back teeth region since a week. He gave a history of sensitivity on taking hot and cold fluids since 2-3 weeks. Since a week he had pain at midnight after lying down which was insidious in onset, moderate, aching, continuous and non radiating in nature and got relieved on taking analgesics. His medical history was non contributory. On general examination he was of average built and well nourished, alert, responsive, and cooperative. Vital signs were all within the normal range. There was no extraoral swelling or asymmetry of face. On intraoral examination, right mandibular first molar was carious which was not tender on percussion. (Fig.1) Considering dental caries as diagnosis, patient was subjected for routine intra oral periapical radiograph (IOPAR).

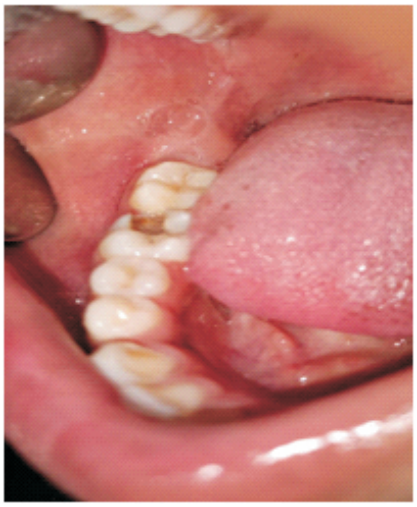

Fig. 1: Clinical presentation with only a carious 46 tooth

Radiographic Features:

The IOPAR showed an ill defined radiolucency involving enamel, dentin and approaching pulp seen disto-occlusally of right mandibular first molar. There was widening of PDL space from crest till apical third portion of the roots. There was loss of lamina dura in apical portion on distal side of root of right mandibular second premolar. A well defined radiolucency was seen extending from right mandibular second premolar till mesial surface of right mandibular third molar with sclerotic border and scalloping around the roots of right mandibular second premolar, first and second molar. (Fig.2.A.) Patient was further subjected for an occlusal radiograph in which radiolucency was present in cancellous bone adjacent to right mandibular first and second molar with no bony expansion. (Fig.2.B.)

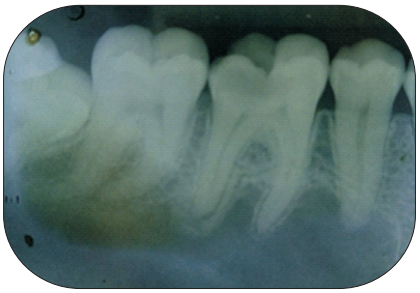

Fig. 2.A. Intraoral periapiacal radiograph showing well defined radiolucency extending from tooth no. 45 till 48 with sclerotic border and scalloping around the roots of 45, 46 and 47.

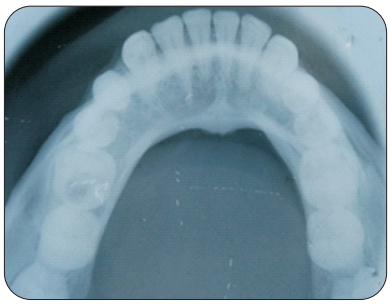

Fig. 2.B. Occlusal radiograph showing radiolucency in cancellous bone adjacent to tooth no. 46 and 47 with no bony expansion

We then sent him for a panoramic radiograph which showed a large, oval shaped, radiolucency extending from right mandibular second premolar till mesial surface of right mandibular third molar of approximately 4cm ×2 cm. The lesion had well defined sclerotic border with scalloping around the roots of right mandibular second premolar, first and second molar. Internally lesion was completely radiolucent. There was widening of periodontal ligament space of right mandibular first and second molar. Lesion involved mental foramen and mandibular canal completely. (Fig.3)

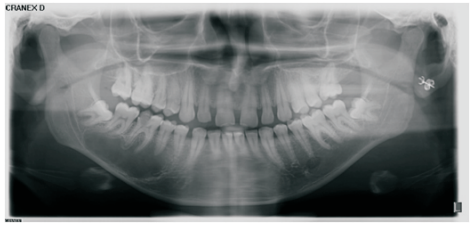

Fig.3: Panoramic radiograph showing large, oval shaped radiolucency measuring 4cm ×2 cm in demensions. There is widening of periodontal ligament space of tooth no. 46 and 47 and lesion is involving mental foramen and mandibular canal completely

A further investigation of Cone Beam Computed Tomography (CBCT) revealed a single, large, expansile periapical radiolucency seen extending from right mandibular second premolar till right mandibular third molar measuring approximately 48mm × 18mm. Internal structure was completely radiolucent with scalloped margins around root surfaces. Thinning of buccal and lingual cortical plates were seen. Discontinuity was seen in lingual cortical plate. Lesion was involving mental foramen and mandibular canal completely (Fig.4-6).

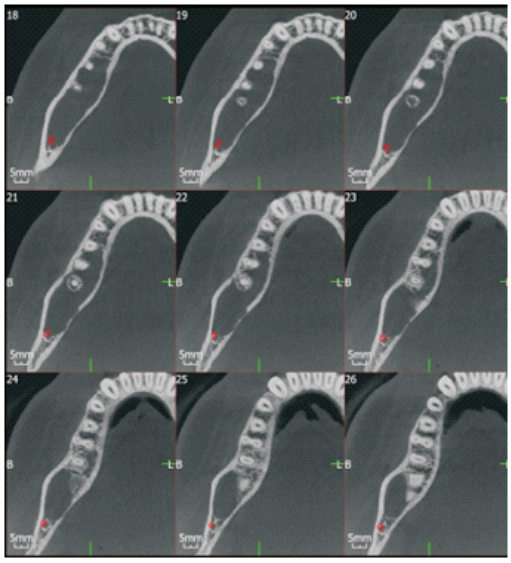

Fig.4: Axial CBCT image of the mandible showing exapansile lesion with thinning of buccal and lingual cortical plates and discontinuity of lingual cortical plate

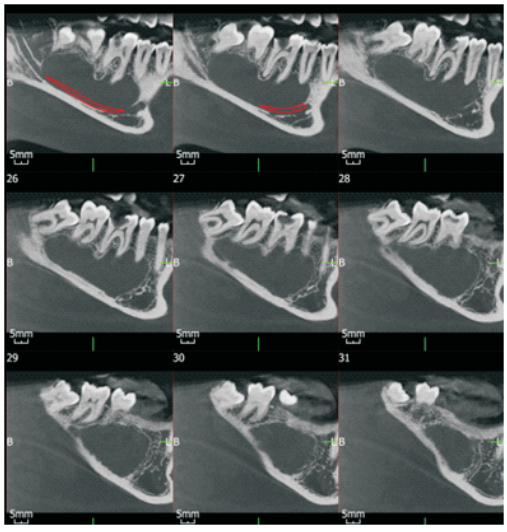

Fig. 5: Sagittal CBCT image of the mandible showing a large well defined radiolucency. Internal structure is completely radiolucent with scalloped margins around root surfaces. Lesion is involving mental foramen and mandibular canal

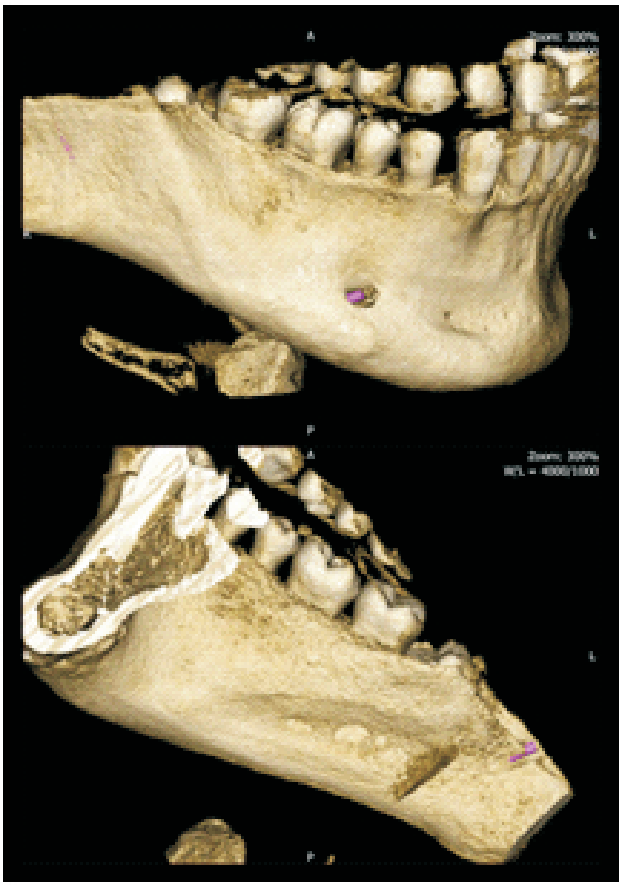

Fig 6 :Three-dimensional CBCT reconstruction of mandible

Diagnosis and Management:

After comparing all the clinical and radiographic features provisional diagnosis of Solitary bone cyst was given. The patient underwent extraction of right mandibular second premolar and first, second and third molars followed by enucleation of cyst. The specimen was sent for histopathological examination which showed connective tissue only. The connective tissue was fibrovascular with many interspersed proliferating fibroblasts and small capillaries. Multinucleated giant cells of varying sizes were present throughout the stroma but not abundant. ln addition, there were numerous foci of extravasated RBC's with associated hemosiderin pigment. Foci of trabeculae of osteoid or bone were also seen with mild infiltration of chronic inflammatory cells consisting predominantly of lymphocytes confirming the diagnosis of Central Giant Cell Granuloma (Fig 7). There was satisfactory healing seen 3 months and 10 months post operatively with no evidence of recurrence (Fig 8,9).

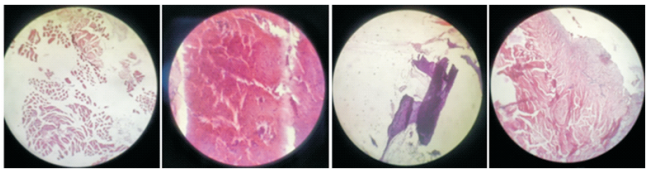

Fig. 7 : Histopathological picture confirming Central Giant Cell Granuloma (40x)

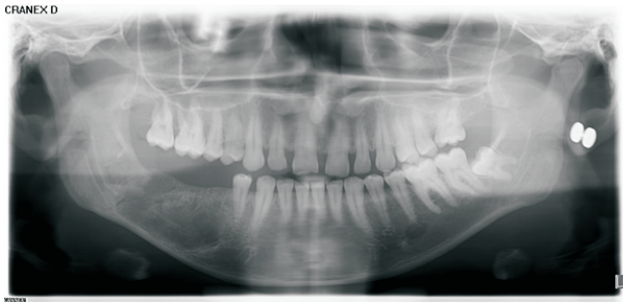

Fig. 8 : Healing lesion 3 months post surgery

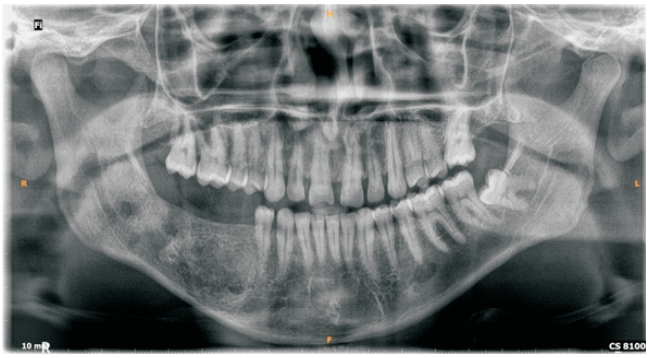

Fig. 9 : Healing lesion 10 months post surgery

Discussion:

The central giant cell lesion of the jaw is a rare benign tumour with an unknown aetiology accounting for up to 7% of tumours in the mandible and the maxilla[2]. The etiopathogenesis of the CGCG of jawbones has not been clearly established but it has been suggested that it is the result of an exacerbated reparative process related to an earlier trauma and intraosseous haemorrhage that triggers the reactive granulomatous process[4]. It has been noted that the development of CGCG may coincides with the onset of gestation or menarche[3]. In our case there was no history of trauma but presence of a carious tooth.

The incidence of CGCG in the general population is estimated to be 0.0001% with 60% of cases occurring before the age of 30. Gender predilection reports are variable, but maximum being females with a female:male ratio 2:1. In this case patient was a male. CGCG is more prevalent in the anterior than the posterior jaws, often crossing the midline (50%), and the mandible is more commonly affected than the maxilla and confined to the tooth-bearing areas of the jaws[3]. In our case lesion was present on right side in body of mandible. It can be aggressive or nonaggressive. According to Chuong *et al*.[2], aggressive giant cell lesions were defined as lesions exhibiting a size equal to or greater than 5 cm as well as showing rapid growth, tooth displacement, root resorption, cortical bone thinning, or perforation or recurrence after curettage.

Aggressive lesions have high recurrence rate as compared to nonaggressive lesions. Nonaggressive lesion is usually slow-growing and asymptomatic, does not show cortical resorption by the lesion or root perforation in teeth affected, and it is significantly less likely to recur than the aggressive type[4].But histologically there is no difference[2]. In this case there was no extraoral or intraoral finding except a carious right mandibular first molar.

Radiographically, the lesion generally presents as a solitary radiolucency with a multilocular appearance or a unilocular appearance which is less common. Lesions develop doubly as frequently in the mandible than maxilla. In young patients the epicenter is anterior to the first molar and after the first two decades of life there is tendency for the epicenter to occur in the posterior aspect of the jaws. The epicenter is more commonly anterior to the canine in maxilla. The periphery may be well-defined or ill-defined and show variable expansion and destruction of the cortical plates. The internal structure may show granular pattern of calcification which is organized into ill-defined, wispy septa which radiate at right angles to the periphery of the lesion. There is apparent displacement and resorption of teeth[3]. However the present case exhibited unilocular radiolucency with well defined, regular sclerotic border scalloping around roots of right mandibular second premolar, first and second molar. Internal structure was completely radiolucent. Thinning of buccal and lingual cortical plates was seen with discontinuity in lingual cortical plate. In our case all the above clinical and radiographic features were in favor of Solitary Bone Cyst as showed in table 1. Only histopathological examination led to the final diagnosis of Central giant cell granuloma.

Table Comparison of features of solitary bone cyst and central giant cell granuloma with our case presentation

FEATURES	SOLITARY BONE CYST	CENTRAL GIANT CELL GRANULOMA	PRESENT CASE PRESENTATION
AGE	1-20 years	Below 30 years	21 years
GENDER	Male > Female	Female> Male	Male
REGION	Mostly body of mandible in premolar and molar region	Mostly anterior region of mandible	Body of mandible
CLINICAL PRESENTATION	Clinically asymptomatic, with no swelling or other functional signs. Often discovered accidentally on routine radiographic examination	Initially asymptomatic but later causes facial swelling and asymmetry	Asymptomatic even at a very late stage. Discovered accidentally on a radiograph
RADIOGRAPHIC FEATURES	Unilocular radiolucent area with well defined or poorly defined borders;with or without asclerotic lining around the lesion The internal structure is totally radiolucent Scalloping around the roots of the associated teeth Larger lesions can cause expansion Root resorption is uncommon;surrounding teeth are vital	Solitary radiolucency with mostly a multilocular appearance with well-defined or poorlydefined periphery, expansion and destruction of the cortical plates The internal structure may show granular pattern of calcificationwith septations Scalloping may be present Expansion may vary with destruction of the cortical plates Displacement and resorption of teeth is evident	Unilocular, well defined radiolucency with sclerotic border. Internal structure completely radiolucent. Scalloping around roots present Cortical thinning and discontnuity in lingual cortical plate No resorption or displacement of teeth present, teeth were vital

The key feature of the central giant cell granuloma is the presence of multinucleated giant cells dispersed unevenly forming clearly recognizable clusters separated by scar-like stromal tissue, markedly seen in areas of hemorrhage. Trabeculae of newly formed reactive bone may be seen on the periphery of the lesion. Sometimes, the bone formation may be extensive and obscure other elements of the lesion[1], as seen in our case also.

Giant cell granulomas are enigmatic lesions. CGCG can be confirmed only histologically as we saw in this case[6]. What made our case veritably intriguing was occurrence of CGCG in mandibular posterior region which is a less common site in association with a carious tooth with no other clinical or radiological finding in favor of CGCG.

Differential Diagnosis:

CGCG should be discerned from odontogenic keratocyst (OKC), unicystic ameloblastoma, aneurismal bone cyst (ABC) and solitary bone cyst (SBC).

OKC can be differentiated from CGCG, as it is asymptomatic, occurs 65% in mandibular third molar region, superior to alveolar canal, presents as a multilocular radiolucency with scalloped borders, and has a high-recurrence rate.

Unicystic ameloblastoma is asymptomatic, occurs in mandibular third molar region, and radiographically appears unilocular with thinning and expansion of the cortical plates.

ABC occurs in mandibular ramus and molar area more than the maxilla, is symptomatic, seen in younger age group and radiographically appears unilocular with thinning, and cortical plate expansion [3]

SBCs may appear similar to central giant cell granuloma; however, the giant cell granuloma has a greater propensity to displace teeth and develop areas of internal mineralization. This is in stark contrast to what was seen in our case presentation[9].

Patients who present with a central giant cell lesion in the maxilla or mandible should be screened for hyperparathyroidism (HPT) to separate it from brown tumour. Although rare, CGCG of facial bones can be the first manifestation of HPT[2].

Treatment:

Management of CGCG is controversial and depends on the aggression, size, and location. Proposed treatment modalities include surgery, radiation, interferon, intralesional steroids and tyrosine kinase inhibitors (Imatinib)[7]. Still the gold standard of treatment of CGCG is surgical enucleation and curettage[8] as was done in this case also.

Conclusion:

Central giant cell granuloma mimicks various lesions as reported here also. All the clinical and radiographic features were in favor of Solitary bone cyst. Solitary bone cyst being an uncommon differential diagnosis for CGCG, it was possible to diagnose it only on histopathological examination. Therefore clinical, radiographic and histopathological examination are all necessary for correct diagnosis and treatment planning. In this case the lesion was treated with surgical enucleation and post surgery panoramic radiograph after 3 and 10 months showed satisfactory healing of the lesion.

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