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Case Report

Maxillary central giant cell granuloma: An unusual presentation with a difficult management

Abhishek Kumar Tiwari[1,*], **Praveen Kumar Pandey**[2]

[1] *Career Post Graduate Institute of Dental Sciences and Hospital, Lucknow, Uttar Pradesh, India*
[2] *Chandra Dental College and Hospital, Dharsania, Uttar Pradesh, India*

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ABSTRACT

Central giant cell granuloma (CGCG) is a rare benign intraosseous aggressive lesion commonly occurring in the mandible with its 2 subtypes, non-aggressive and a relatively rare aggressive type. The present case is a rare aggressive CGCG at an unusual maxillary region associated with swelling, rapid growth, pain and cortical growth perforation. The clinical, radiographic, and histopathological features along with difficult management have been described in detail.

1. Introduction

Central giant cell granuloma (CGCG) is a benign, proliferative intraosseous lesion, first described by Jaffe in 1953 in order to distinguish (CGCG) from the giant cell tumour of long bones. It accounts for almost 7% of all benign lesions of the jaws commonly involving young adults below the age of 30 years showing female predilection[1] CGCG can be divided into two subtypes, aggressive and non-aggressive. The non-aggressive variant is most common, presenting as a slow-growing, painless lesion with expansion of cortical bone. In contrast, aggressive giant cell granulomas tend to present in younger patients with the following possible features: greater than 5 cm in size, rapid growth, tooth displacement leading to malocclusion, cortical bone thinning or perforation, and recurrence after curettage.[2] According to World Health Organization 1992 classification, CGCG is defined as "an intraosseous lesion consisting of more or less fibrous tissue containing multiple foci of hemorrhage, aggregates of multinucleated giant cells, some amount of trabeculae of woven bone forming within the septa of more mature fibrous tissue that may traverse the lesion."[3] We hereby report an uncommon case of an aggressive CGCG in a22-year-old male located in the right maxillary antero-posterior region.

2. Case Report

A 22-year-old male reported to with a complaint of swelling and pain in upper right front and back region of jaw past 2 months developed after 3-4 months of trauma in the same region. Initially the swelling was smaller in size and increased rapidly in last 2 weeks. Patient also complains of difficulty in chewing and heaviness on the side of nose on the affected side. His past dental history was significant with previous examination for the same region without any intervention. Extraoral examination revealed an ovoid, hard swelling on right side of face resulting into an obvious facial asymmetry. The swelling was tender on palpation. Superioinferiorly, it extended from right infraorbital ridge to the vermillion border of upper lip. Medially it extended from lateral wall of nose obliterating the nasolabial fold and extending till the anterior border of zygomatic process. Distinct deviation of face is seen towards right side.

* *Corresponding author*.
E-mail address: dr.abhishek0404@gmail.com (A. K. Tiwari).

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However, the overlying skin is smooth and intact with same color as surrounding tissue. (Figure 1)

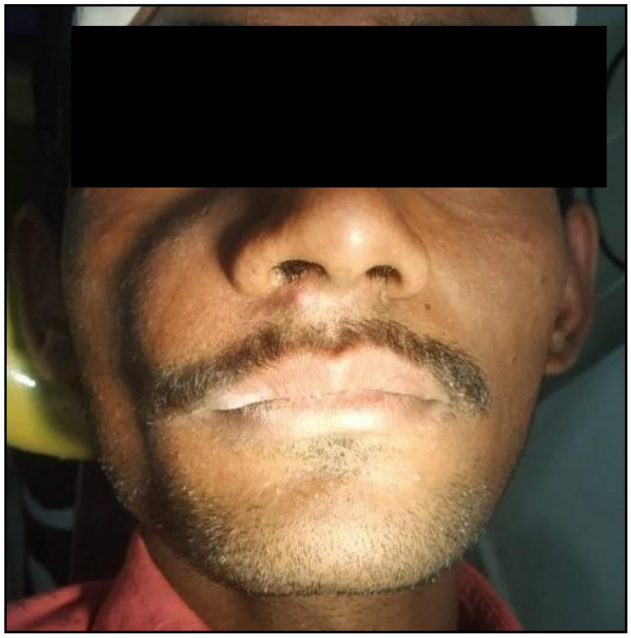

Fig. 1: Extraoral examination

Intraoral examination revealed a diffuse, firm and tender swelling on right side of maxilla extending from distal aspect of 11 to distal aspect of 15 labially. Superior border is ill-defined and inferiorly it extends upto the marginal gingiva of 11-14. There is obliteration of labial vestibule. Palatally the swelling is seen localised within the incisive papilla to the mesial region of 15 not extending beyond the midline. Teeth 13 and 14 shows grade I mobility. The overlying mucosa shows same color as that of adjacent tissues. (Figure 2)

Patient was referred for a Cone-beam CT (CBCT) imaging series. Routine Cone Beam Computed Tomography was performed for QUADRANT MAXILLA SCAN.(Figure 3a,b) Mixed radiopaque-radiolucent expansile lesion seen in the anterior maxilla with significant labial and palatal expansion extending from #11 to #16 region mesio-distally (26.3 mm), from crest of alveolar bone to compressing the antero-lateral wall of right maxillary sinus and lateral nasal wall supero-inferiorly (33.8 mm) and from mid-palatal region to expanding into the labial soft tissue region medio-laterally (31.8 mm) with displacement of teeth wrt 13,14. Periphery of the lesion is well demarcated with thin irregular cortical outline. Internally lesion show thick coarse and straight septa with few patches of radiolucent pockets giving multilocular appearance. Internal margin of the lesion is scalloped with projected septa from the peripheral margin. Loss of cortical wall of lateral nasal wall and antero-medial corner of right maxillary sinus noted. Differential of lesion may include

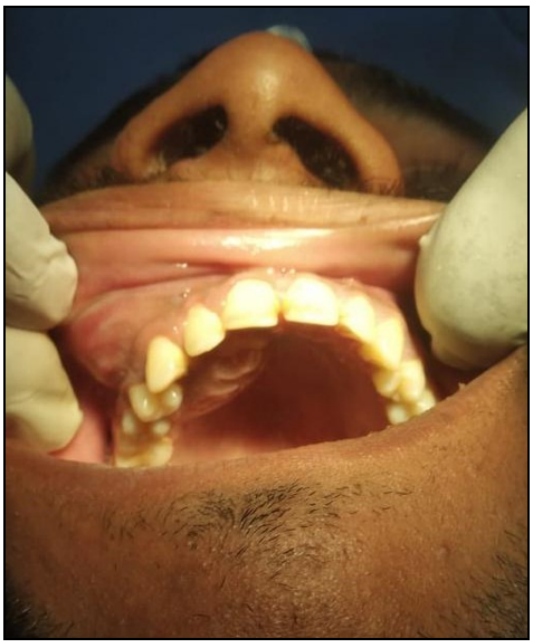

Fig. 2: Intraoral examination

odontogenic lesion of myxomatous origin or a central bony neoplasm**.** A provisional diagnosis of odontogenic myxoma was made initially.

Extraction of 12,13,14,15 was done with surgical curettage and decortication of the lesion was done through an intraoral approach. Tissue from maxillary lesion comprised of multiple bits of tissue with largest one measuring about 3.8×3.0×2.0 cm.(Figure 4 a,b,c)

Histopathological evaluation under higher magnification revealed focal thin superficial parakeratinized stratified squamous epithelium. Larger portion shows dense connective tissue yet hypercellular with numerous bony trabeculae. Connective tissue depicts biphasic cellular population consisting of plump shaped fibroblasts and numerous evenly distributed multinucleated large giant cells with many giant cells containing more than 20 or more nuclei. Areas of hemorrhage are evident. Connective tissue near the areas of hemorrhage are marked with cystic spaces and focal necrosed regions and chronic inflammatory infiltrate. Histopathological features were suggestive of central giant cell lesion and giant cell tumor of bone. To differentiate between Giant Cell Tumor of bone and CGCG, P63 IHC was used which was negative, confirming the lesion as central giant cell granuloma.(Figure 5 a,b,c) On the basis of clinical, radiological, and histopathological examination, a final diagnosis of giant cell granuloma of aggressive type has been achieved.

Patient was under follow up for next 3 months which were uneventful. After a period of about 7 months patient returned to the OPD with a complaint of swelling and pain in the same region with a history of trauma again in the

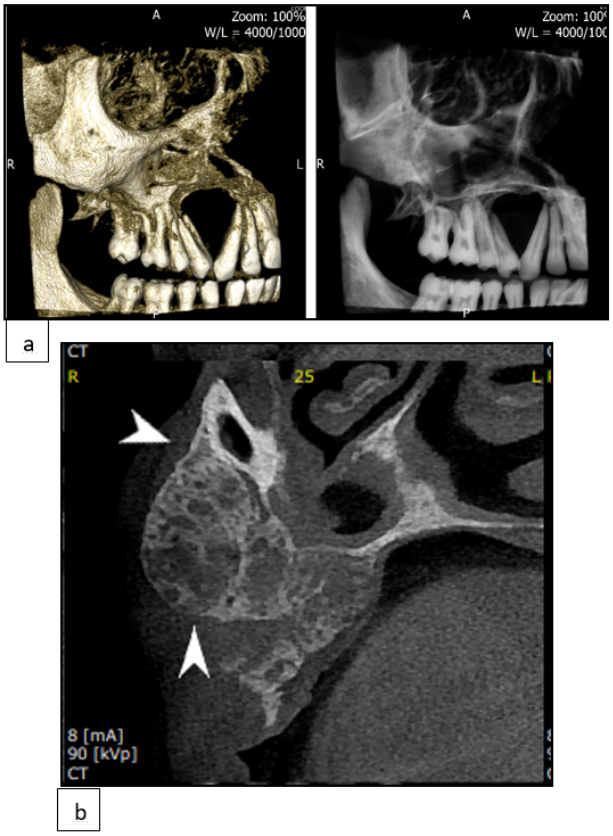

Fig. 3: a): Displacement of teeth wrt 13,14; **b**): Mixed radiopaque-radiolucent expansile lesion seen in the anterior maxilla with significant labial and palatal expansion. Periphery of the lesion is well demarcated with thin irregular cortical outline. Internally lesion show thick coarse and straight septa with few patches of radiolucent pockets giving multilocular appearance

same region. The patient was referred for a Cone-beam CT (CBCT) imaging series for QUADRANT MAXILLA SCAN again.(Figure 6 a,b) Dimensions of the lesion were 30.16 mm x 32.35 mm x 36.98 mm in bucco-palatal, mesio-distal and supero-inferior directions. Periphery of the lesion was well-defined with interrupted borders and shape of the lesion is ovoid in axial sections. Internally lesion show radiolucent vacuoles/sac interspersed in the lesion with coarse wispy septae. Few perpendicular trabeculae septae noted in the periphery of lesion. Superior displacement of cortical floor of maxillary sinus and nasal floor noted with erosions. Findings are indicative of reactive bone lesion. Mild mucosal thickening seen in the left maxillary sinus s/o sinusitis.

Considering the size of the lesion and proximity to sinus biphasic management with 2 ml of intralesional corticosteroid, triamcinolone acetonide at a dose of 10 mg/ml was given twice weekly for 6 weeks. Following the course of the phase I treatment, there was no such significant decrease in size of the lesion clinically and

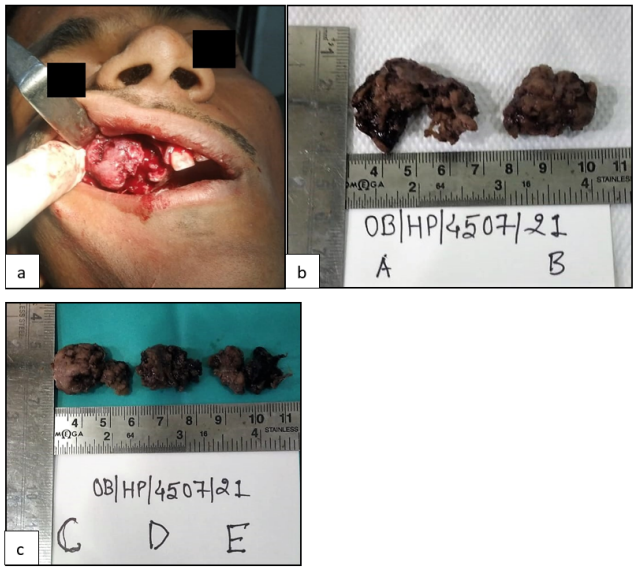

Fig. 4: a): Surgical curettage and decortication of the lesion was done through an intraoral approach; **b):** Tissue from maxillary lesion comprised of multiple bits of hard and soft tissues

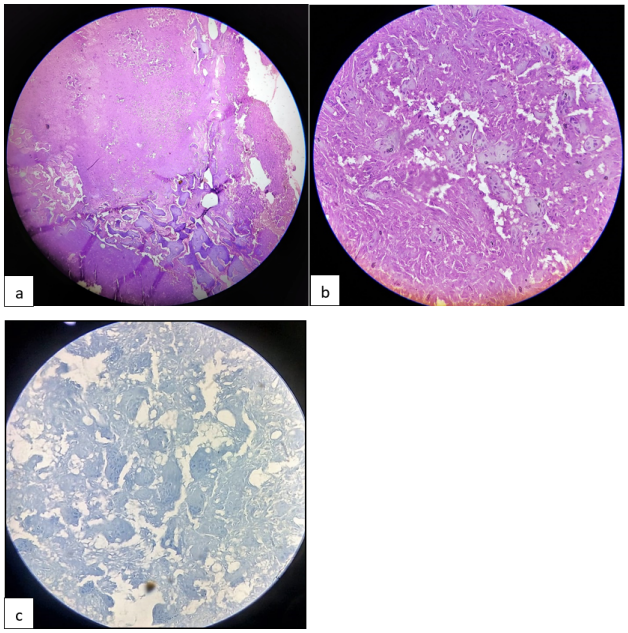

Fig. 5: a) Larger portion shows dense connective tissue yet hypercellular with numerous bony trabeculae; **b**): Connective tissue depicts biphasic cellular population consisting of plump shaped fibroblasts and numerous evenly distributed multinucleated large giant cells with many giant cells containing more than 20 or more nuclei; **c**): To differentiate between Giant Cell Tumor of bone and CGCG, P63 IHC was used which was negative, confirming the lesion as central giant cell granuloma

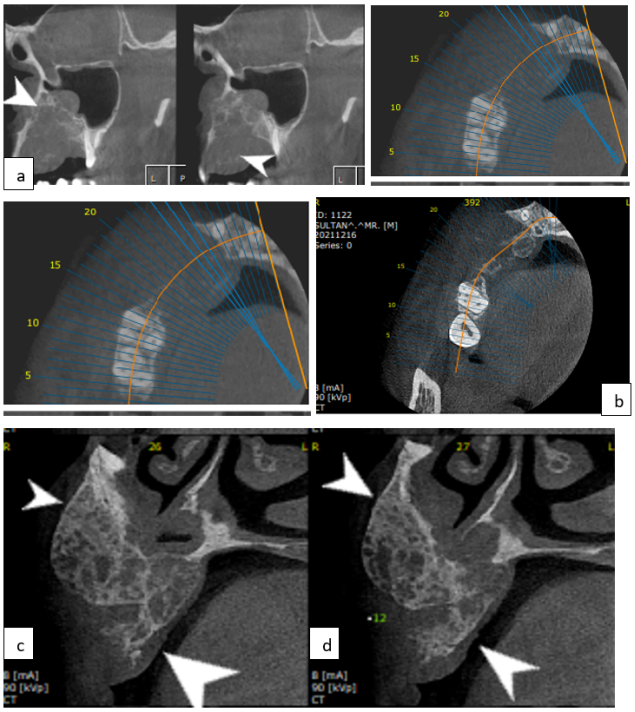

Fig. 6: Recurrence of the lesion after 7 months with subsequent CBCT findings at the interval of 2 months post corticosteroid injection therapy. Although no significant decrease in size was noted yet some significant resistance was developed during intralesional injections

radiographically which was evaluated using CBCT imaging at the interval of 2 months, yet some significant resistance was developed during intralesional injections. (Figure 6c,d) Through second phase management surgical curettage and decortication of the lesion was done through an intraoral approach. Post-operative course was uneventful and patient recovered well. Patient is under regular follow-up and no recurrence has been seen after 2 year of surgery.

3. Discussion

Our case is interesting as the lesion has unusual site at maxillary region with aggressive behavior, post-surgical recurrence, difficult management and showed significant cystic areas. Small rare group of CGCG cases can show cystic areas as in our case which supports the speculation that CGCG is the primary lesion and aneurysmal bone cyst is a secondary change.[2] Though CGCG is a benign reactive osseous lesion, it has been classified into two types based on its clinic-radiologic features into a slow growing asymptomatic, nonaggressive lesion, and an aggressive type encountered in younger patients which is painful grows rapidly into a large size, perforating the cortex, displacement of normal anatomical structures and has a tendency to recur.[3] The treatment of choice is typically surgical curettage of the involved area. The recurrence rate given in various literature reports after conventional surgical curettage ranges from 11 to 49%.[4] Histopathologically, CGCG is composed of proliferating endothelial cells, numerous small capillaries, multinucleated giant cells, and active fibroblasts.[5] Present case have similar histopathological features of biphasic cellular population consisting of plump shaped fibroblasts and numerous evenly distributed multinucleated large giant cells with many giant cells containing more than 20 or more nuclei. Areas of hemorrhage and bony trabeculae are evident. Connective tissue near the areas of hemorrhage are marked with cystic spaces and focal necrosed regions and chronic inflammatory infiltrate. Our case showed normal levels of calcium, parathormone and alkaline phosphatase ruling out Brown's tumor of hyperparathyroidism.

Management of aggressive forms of CGCGs has always been a challenging problem due to their high recurrence rates. Therefore, alternative management modalities using pharmacological agents like corticosteroids, calcitonin and systemic interferon alpha have been developed and reported with encouraging result.[6] There are two treatment modalities for treatment of CGG. i.e. medical and surgical. Surgical part includes curettage and resection. Medical treatment includes intra-lesional injection of corticosteroids.[7] Initial management in our case includes surgical curettage with decortication of bone. Lesion recurred after 7 months and subsequent management included biphasic treatment modalities. Initial intralesional injections of conrticosteroids and surgical intervention thereafter. Literature studies have revealed other innovative methods to restore the lost tissues with autologous matrices. Native extracellular matrix (ECM) scaffold is an emerging tool in tissue engineering for the reconstruction of three-dimensional (3D) tissues and organs, respecting their structural and functional features.[8]

Recent studies using immunohistochemistry (IHC) and molecular methods have demonstrated overexpression of p63 in the stromal cells of most GCTs (Giant cell tumor) of bone and advocate its use as a diagnostic marker as in our case p63 profiling was negative.[9]

4. Conclusion

Multidisciplinary approach is vital for complete management of giant cell lesions. Clinical examination, radiographic interpretation, blood investigation, histopathological examination and complete surgical elimination are the key. As in our reported case despite aggressive and complete removal recurrence of the lesion occurred which was further managed by biphasic treatment modalities including medical and surgical measures.

5. Source of Funding

None.

6. Conflict of Interest

Nil.

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Author biography

Abhishek Kumar Tiwari, Assistant Professor https://orcid.org/0000-0002-8795-0505

Praveen Kumar Pandey, Professor https://orcid.org/0009-0001-7433-7111

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