Original Research Paper

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Radio-Diagnosis

CENTRAL GIANT CELL GRANULOMA OF MAXILLA.

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ABSTRACT **Background:** Central giant cell granuloma are rare non-neoplastic lesions. They are histologically different from giant cell tumour of bone. It contains cellular fibrous tissue with multiple haemorrhagic foci with aggregates of multinucleated giant cells. Sometimes trabeculae of woven bone may also be seen. It was first described by Jaffe in 1953 as an idiopathic non-neoplastic proliferative lesion. It is benign, intraosseous lytic lesion which has debatable aetiology. It doesn't have characteristic clinical and radiographical features which can help it to be distinguished from other lesions in this region. HPE analysis still remains the gold standard for identifying this disorder. **Case presentation:** A 19-year-old male patient presented to the author's centre with a swelling on upper jaw of his face on right side for 6 months along with tooth pain. External examination revealed a diffuse swelling over his right maxilla extending till the infraorbital region causing upliftment of the base of the nose. There is associated obliteration of the right nasolabial fold and causing facial asymmetry. There was missing 32 teeth with bony expansion in right vestibular region extending from first molar till central incisor on right. There was a large expansile soft tissue density lesion seen arising from the maxilla on right side causing cortical thinning and breach measuring approximately 4.2 x 6.5 x 5.1 cm (AP X ML X CC). Multiple bony septations are seen within the lesion with discontinuous peripheral calcifications. Based on the clinical and radiological presentation, a provisional diagnosis of an odontogenic cyst was made. Histopathological examination revealed normal cortical bone with a normal inflamed cellular stroma with patchy distribution of multinucleated giant cells suggestive of giant cell granuloma. **Conclusions:** Surgical management of the patient was done with resection of the lesion and primary reconstruction with bone graft. The result of the surgery was successful.

KEYWORDS : Central giant cell granuloma (CGCG), jaw bones, maxilla, reconstruction

BACKGROUND

Central giant cell granuloma of jaw is a rare and osteolytic disease. It shows aggressive nature in young patients. The disease is idiopathic in occurrence. The lesion presents as proliferative intraosseous lesion which consists of cellular fibrous tissue. The lesion also has multiple foci of haemorrhages, various aggregates of multinucleated giant cells, and also trabeculae of woven bone. It is characterized by rapidly enlarging swelling with bony expansion and associated tooth displacement. Cortical perforation has been reported.[1],[2] World Health Organization in its recent classification of histological classification of odontogenic and maxillofacial bone tumours (4th edition, 2017) has delineated under the giant cell lesions and bone cysts.[3] They account for a small percentage of all head and neck masses. It is most commonly seen in 20 to 40 years of life (1,10). It is seen more commonly in females due to the hormonal factors although they very rarely show presence of estrogen receptors (11,12). No racial predilection is noted. Most of the lesions are seen in the mandibular or maxillary regions. Very rarely they can also be seen in other craniofacial bones and also the small bones of the hands and feet (5) CGCG mostly presents as slow growing lesion and thus delay in diagnosis occurs commonly. The patient most commonly presents with the features of mass effects which is dependent on the site involved. Generally, the most common features of clinical presentation are soft-tissue swelling and pain. Other rare and non-specific features of presentation are proptosis, nasal obstruction, and cranial nerve palsies.

Central giant cell granuloma (CGCG) of jaw is a benign, intraosseous, osteolytic lesion of debatable aetiology. It doesn't have characteristic clinical and radiologic features, so it is difficult to form diagnosis solely on radiological basis. Histopathology still remains the predominant diagnostic modality to identify the disorder. The lesions which are aggressive leads to early damage and requires aggressive therapy.

The aim of this report is to present a case of CGCG in an adult patient and its treatment challenge.

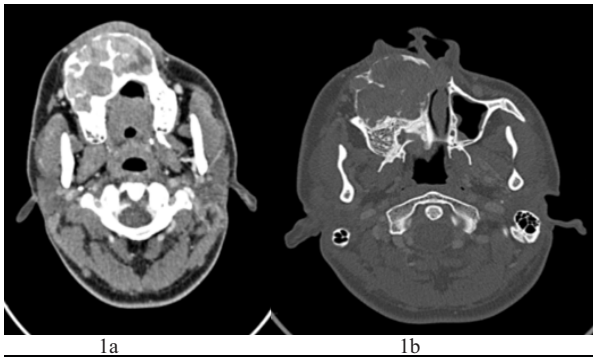
Case Presentation

A 19-year-old male patient presented to the author's centre with a swelling on upper jaw of his face on right side for 6 months along with tooth pain. The swelling as per the patient was insidious in onset, gradually progressive, with progressive loosening of associated teeth and soft tissue mass underneath the upper lips. There was history of difficulty in chewing of food. Patient was a known case of epibulbar dermoid right eye for which he was undergoing treatment. Patient also had wart over his right temporal region.

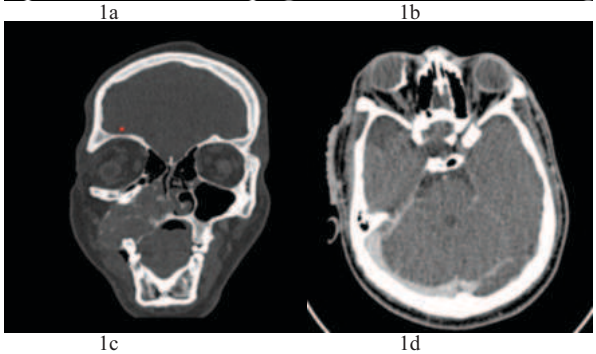
External examination revealed a diffuse swelling over his right maxilla extending till the infraorbital region, lifting the base of the nose. There was associated obliteration of the right nasolabial fold with facial asymmetry. The overlying skin was normal. There was missing 32 teeth with bony expansion in right vestibular region extending from first molar till central incisor on right.

Radiological Examination

The radiograph and computed tomography (CT) of the maxilla were acquired. The coronal and axial sections and three-dimensional reconstructions were performed. There was a large expansile soft tissue density lesion seen arising from the maxilla on right side causing cortical thinning and breach measuring approximately 4.2 x 6.5 x 5.1 cm (AP X ML X CC). Multiple bony septations are seen within the lesion with discontinuous peripheral calcifications. Two teeth are seen within the lesion at its superior and medial aspect respectively with displacement of the maxillary teeth. Anteriorly, the lesion is causing focal contour bulge of overlying skin and subcutaneous tissue with soft tissue component extending into subcutaneous plane. Posteriorly, there is bulge of overlying maxillary cortex into oral and nasal cavity. Superiorly the lesion is extending into and causing widening of the right nasal cavity with extension into the left nasal cavity. The supero-posterior part of the lesion is indenting the anterior wall of right maxillary sinus. The lesion shows heterogeneous post contrast enhancement.



1a 1b

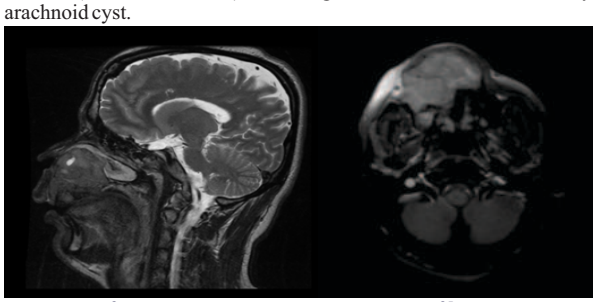


1c 1d

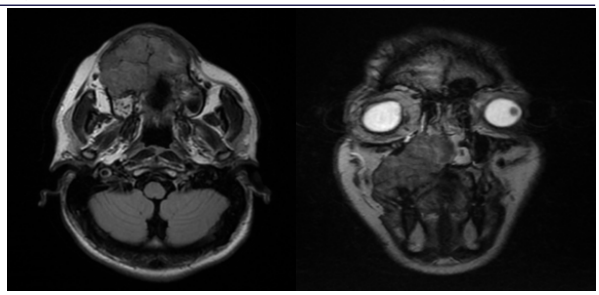
Fig 1a CECT PNS axial view, 1b – Bone window CECT PNS axial view, 1c- Bone window coronal view, 1d- CECT PNS axial view. There is heterogeneously enhancing soft tissue density lesion seen epicentred in the maxilla towards right side extending up to the midline with bony destruction. The lesion is seen extending into the right maxillary sinus and nasal cavity. There is proptosis of right side seen. There is calcification noted at the posterior aspect of the right globe.

On CEMRI- There is a well-defined lobulated altered signal intensity lesion noted in right maxilla measuring 48 x 64 x 55 mm (AP X ML X CC). Anteriorly the lesion is seen to cause focal contour bulge of the skin and subcutaneous tissue. Superiorly the lesion is seen to extend into bilateral nasal cavities but is not seen to reach upto the cribriform plate. Inferiorly the lesion is seen to produce contour bulge of the hard palate.

Incidental note was made of a CSF intensity lesion measuring 17 x 27 x 57 mm (AP X ML X CC) in the right middle cranial fossa likely arachnoid cyst.



2a 2b



2c 2d

Fig 2a Sagittal T2WI, 2b Post contrast T1WI images, 2c FLAIR axial image, 2d T2WI coronal images. There is heterogeneously enhancing lesion seen epicentred in the maxilla towards right showing extension into the right maxillary sinus and left nasal cavity.

No abnormality was detected in biochemical and hematological parameters of the patient.

Based on the clinical and radiological presentation, a provisional diagnosis of an odontogenic cyst was made. Differential diagnosis included radicular cysts, maxillary mucocele, giant cell tumor, radicular cyst, adenomatoid odontogenic tumor, and unicystic ameloblastoma.

Management

An incisional biopsy of the lesion was done. Histopathological analysis showed normal cortical bone with a normal inflamed cellular stroma with patchy distribution of multinucleated giant cells suggestive of giant cell granuloma. Based on the clinical, radiological, and histopathological findings, a diagnosis of CGCG was made. Due to the anatomical location of the lesion and the extent of involvement of the structures surgery was done as the preferred treatment. So, the decision was made to perform surgical resection and primary reconstruction with autogenous bone graft. The result of the surgery was successful.

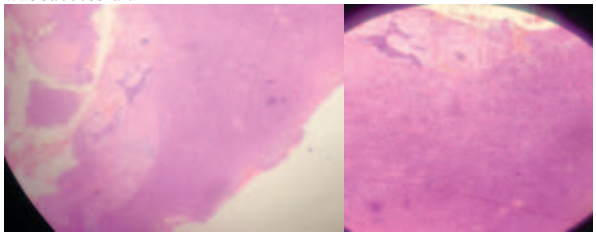


Fig 3(a) Fig 3 (b)

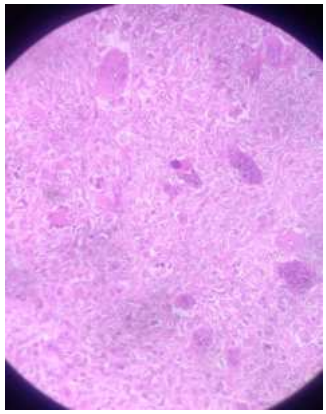


Fig 3(c)

Figure 3: (a) Histopathology of the section (H and E, ×4). Note the cellularity of the connective tissue, the intensity of inflammatory reaction, and the numerous giant cells spread across the section. (b) Histopathology of the section (H and E, ×10). Note the cellularity of the connective tissue, the intensity of inflammatory reaction, and the numerous giant cells spread across the section. (c) Histopathology of the section (H and E, ×40). Note the cellularity of the connective tissue, the intensity of inflammatory reaction, and the numerous giant cells spread across the section. The giant cells have several nucleus and nucleolus.

DISCUSSION:

CGCG as an entity was first described in 1953 as a benign lytic lesion that usually occurs in the jaw bones. Since then, several cases have been reported from India.[1],[2],[4],[5] In pertinent contemporary Indian biomedical literature, CGCG involved children whereas the present case was an adult male. The radiological findings of our case was consistent with the findings from a large series of cases reported till date.[6]

There were several differential diagnoses for this case. The age and the site of involvement were not favouring for a diagnosis of ameloblastoma. Due to absence of dental caries the possibility of periapical and radicular cyst was ruled out. Normal serum picture helped us to rule out Brown tumour (hyperparathyroidism). The lesion didn't appeared like cherubism due to its presentation, size, and radiographic appearance. The incisional biopsy of the lesions helped us reach a definitive diagnosis of CGCG.[1],[2],[4],[5]

Several nonsurgical treatment options have been described for the treatment . These include giving of intralesional steroids, administration of subcutaneous or nasal calcitonin, subcutaneous injections of alpha-interferon, and imatinib drug therapy. [7]

The recurrence rate post surgery is described to be 13%–22% within a short postoperative period. Curettage is the advised treatment for small lesions, but this carries the risk of recurrence. Radical excision is the treatment of choice when the lesion is extensive and involves the cortex, but this would result in loss of multiple teeth as is seen in the present case. Sacrificing teeth instead of performing root canal serve two purposes – (1) the involved teeth exhibit radiological evidence of involvement and resorption and (2) If Root Canal Treatment is performed, the healing of the surgical defect would be impeded. Hence, in unfavourable situation, the removal of teeth may be required. In our present case, surgery was preferred over conservative treatment because of anatomic location that is close association with maxillary sinus and other vital organs, relatively large size of lesion, aggressive clinical course and thinning out of cortical plates. So, the decision was made to perform surgical resection and primary reconstruction with autogenous bone graft. The result of the surgery was successful.

CONCLUSIONS

The successful management of a rare maxillofacial lesion in a young adult is reported. The lesions are generally seen in the mandible with very rare cases in maxilla. There are multiple differential diagnosis noted on imaging ranging from brown tumor, ameloblastoma, aneurysmal bone cyst, odontogenic myxoma and others. Imaging alone cannot distinguish various lesions so the histologic analysis is required for the exact diagnosis. Central giant cell granulomas have similar characteristics with giant cell tumors and differentiation is very important as they have high risk of recurrence, metastasis and malignant transformation.

Declarations

Ethics Approval And Consent To Participate

The inclusion of patient data was done only after acquiring approval on the Institutional Ethical Committee of Base hospital Delhi Cantt.

Consent For Publication

Written and informed consent was obtained from the patient before use of the data pertaining to the patient in the study.

Availability Of Data And Materials

The datasets used and/or analyzed during the current study are available from the corresponding authors on reasonable request.

Competing Interests

The authors declare that they have no competing interests.

Funding:

Not applicable.

Authors' Contribution:

TM and KP reported the radiograph, CECT PNS and CEMRI of the patient. RC performed the biopsy of the patient and gave the HPE diagnosis of the pathology. All the authors were involved in extensive history taking of the patient by their relatives and colleagues. All authors have read and approved the manuscript.

List Of Abbreviations:

CGCG- Central giant cell granuloma.
HPE- Histopathological examination.
AP- Antero-posterior
ML- Medio-lateral
CC- Cranio-caudal
CT- Computed Tomography
MRI- Magnetic Resonance Imaging
CSF- Cerebrospinal fluid.

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