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Recurrent Giant Cell Tumour of the Mandible: A Case Report

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Abstract

Giant Cell Tumour (GCT) is a rare benign, osteolytic, pseudo cystic solitary localised lesion. The aetiology of giant cell tumours remains uncertain. The incidence shows a predilection for the female gender. The curative treatment for these tumours is surgical curettage or resection; undesirable damage of the teeth or tooth germs is unavoidable and surgical removal may lead to rupture of the cyst or iatrogenic fractures. The local recurrence rates are as high as 25%. We present the case of a 28-year-old lady who reported with a swelling in the left mandible and bone erosion.

Keywords: *Giant Cell Tumour, Mandible, Mononuclear Cells, Recurrence, Surgical Approach.*

Introduction

Although Ambroise Pare is credited with the first description of what was probably a giant cell tumour during the sixteenth century, it was not until 1912 that Joseph Bloodgood first coined the term "Giant Cell Tumour"**(**1**)**. Giant cell tumour **(**GCT**)** is an aggressive but benign neoplasm containing spindle-shaped stromal cells, mononuclear round to oval cells resembling histiocytes and abundant evenly distributed osteoclastic giant cells. This lesion constitutes 5% of all primary bone tumours. The frequency of occurrence is 25% in the epiphyses of long bones and 2% in the craniofacial bones**(**2**)**.

GCTs are characterised by profuse multinucleated giant cells scattered with surrounding stroma of mononuclear cells. These giant cells have some similarity with osteoclasts and so these tumours are also known as osteoclastoma.

GCTs share a strict differential diagnosis with giant cell reparative granuloma **(**GCRG**)**, which is an uncommon and benign reactive tumour. Although histologically GCRG are very similar to GCTs, GCTs are distinguished on the basis of their high mitotic activity**(**3**)**.

The traditional treatment of choice for GCT has been surgical excision. The extent of tissue removal ranges from simple curettage to en bloc resection. Recurrence rates following surgery have been reported at 11–72% and were higher in patients with aggressive lesions**(**4**)**.

The aim of this study is to report the rare case of a 28 year old patient with recurrent GCT in the mandible, managed by curative surgery.

Case Report

A 28 year-old lady with known history of solitary left dysplastic kidney, type-I diabetes mellitus and systemic hypertension under regular medication presented with a slowly enlarging, painless swelling in the left body of the mandible for 6 months. Patient had a similar episode in the past with swelling in the same region 4 years before for which biopsy was done. Microscopy of the biopsied specimen revealed a diagnosis of GCT and underwent surgical excision of the lesion from another centre.

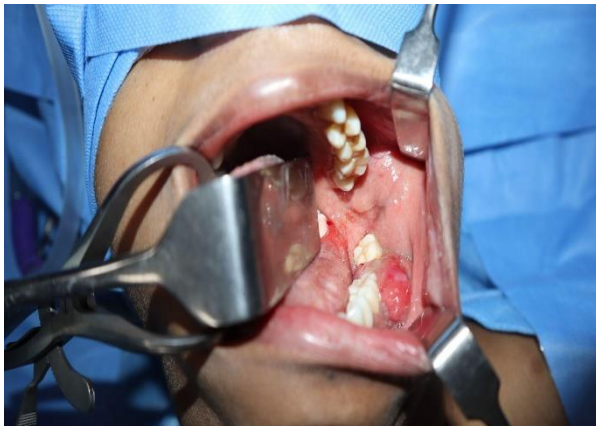
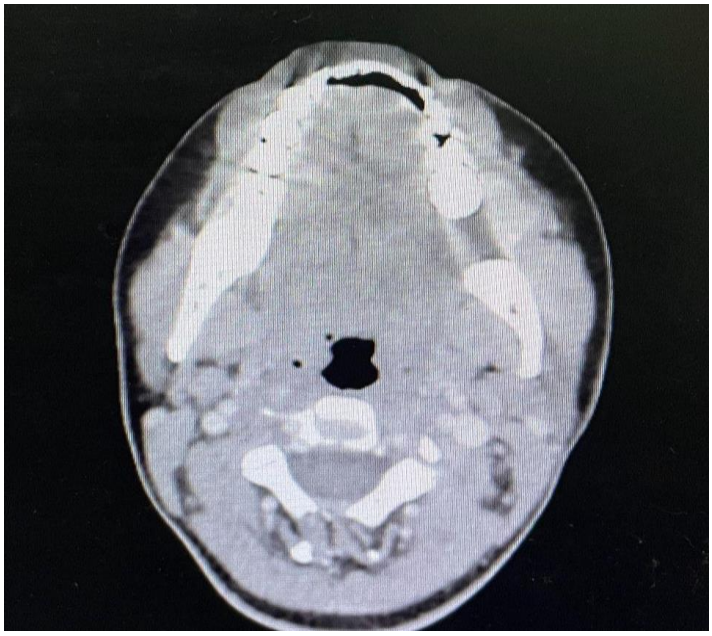


Fig.1 Lesion involving left lower alveolus

On local examination, there was a swelling over the left body of the mandible in relation to the 36, 37 region which was of size 3x3 cm. Mucosa over the swelling was erythematous with a point of discharge. The skin over the cheek was normal. The temporomandibular joint **(**TMJ**)** movements were normal. On palpation, there was a firm, sessile and multilobulated swelling of size 3x3 cm extending supero-inferiorly from occlusal surface of 37 to the buccal vestibule and antero-posteriorly from 35 to 38 region. Swelling was tender on palpation. Neck nodes were not palpable.

CECT scan of the neck revealed an heterogeneously enhancing lesion involving the left lower alveolus measuring 2x2 cm in size. It was associated with erosion of the cortex.



Intraoral biopsy revealed a neoplasm composed of spindle shaped cells with eosinophilic cytoplasm and vesicular oval nuclei. In between, there were multinucleated giant cells of varying sizes. At the edge of the lesion, there was squamous epithelium mucosa with subepithelial connective tissue stroma which was highly cellular with sheets of plump fibroblast and numerous giant cells and admixed with inflammatory infiltrate. There was no evidence of any malignancy.

On the basis of clinical, radiological and histopathological examination, findings were suggestive of a giant cell rich lesion possibly giant cell tumour. The serum values of calcium, phosphorus, and parathyroid hormone were normal, thereby excluding the possibility of hyperparathyroidism.

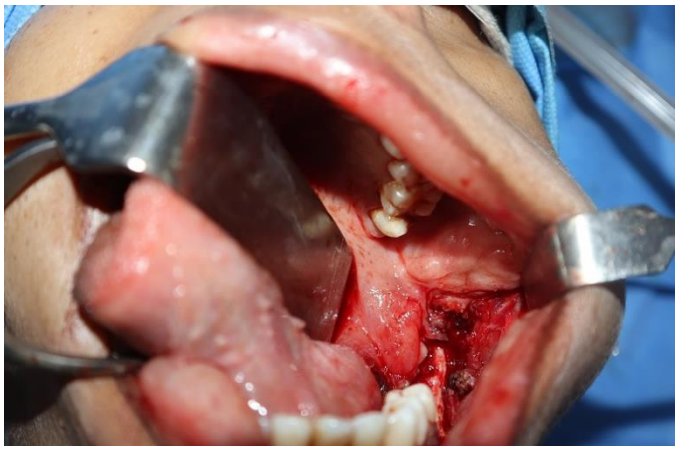


Fig. 2 Post Excision Defect

The patient underwent surgical excision of the tumour via peroral approach with marginal mandibulectomy followed by primary closure of the wound. Post operative histopathological report revealed hyperplastic hyperkeratotic squamous mucosa with subepithelium showing numerous evenly distributed osteoclastic like multinucleated giant cells in a stroma of spindle cells. Multiple foci of haemorrhage and trabecular bone also seen. Mitosis scanty.

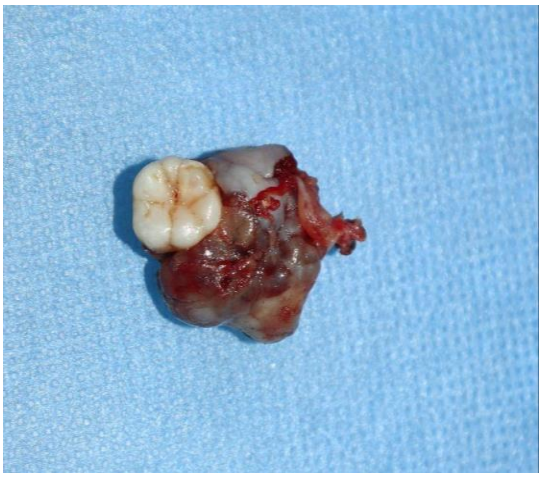


Fig.3 Postoperative Specimen

Discussion

Giant Cell Tumour **(**GCT**)** is an uncommon, osteolytic, benign, pseudo cystic, solitary localised lesion**(**5,6**)**. GCT typically affects the meta-epiphyseal region of long bones, mainly the distal femur and the proximal tibia with a slight predominance in females in the 2nd and 3rd decades of life. This lesion is uncommon in the craniofacial skeleton**(**7**)**.

The incidence of these tumours in facial bone is <1%. The incidence of GCT in the various regions of mandible; 90% of the cases affect the posterior mandible i.e., Body of mandible 40%, ramus 30%, angle 19%, symphysis 9%, condyles 2%**(**5**)**. There are few reports found in the literature describing involvement of the coronoid process by GCT **(**8**)**. This adds to the diversity of clinical and biological behaviour of GCT in maxillofacial region.

The most frequent clinical findings are swelling, local pain, tender mass and pathological fractures**(**5,6**)**. Aggressive lesions are defined by the presence of one or more of the following signs: pain, paresthesia, root resorption, rapid growth, cortical perforation and a high recurrence rate after surgical curettage**(**10,11**)**.

The enlargement may be slow or rapid with blow out distension of part of the affected bone causing bony expansion. There is usually an intact periosteum and very thin shell of bone covering the cyst(12). The teeth may be missing or displaced but root resorption is rarely seen(13).

Histologically, these cysts are described as blood-filled cavities and sinusoidal spaces separated by fibrous connective tissue septa with osteoid trabeculae(13). Variable amounts of hemosiderin and giant cells can be found(12,15). Differential diagnoses include aneurysmal bone cyst, chondroblastoma, giant cell-rich osteosarcoma and brown tumour of hyperparathyroidism(5,16,17).

Despite an improved understanding of the molecular and cellular biology underlying the GCT pathogenesis, the behaviour of this tumour is often heterogeneous and can be difficult to predict on the basis of clinical, radiographic, or histologic features. The most commonly accepted theory of Hillerup and Hjorting-Hansen suggested that GCT, Central Giant Cell Granuloma (CGCG), Traumatic Bone Cyst (TBC) were all related lesions. Minute trauma or the presence of unidentified small aneurysmal enlargements may result in intramedullary bleeding leading to haematoma. If the blood supply is lost TBC may develop. If only small vessels or low pressure is present then capillary and endothelial proliferation occurs resulting in CGCG. If circulation is maintained, creating high pressure, large pools of blood are formed and GCT results. The clinical case did not give any history of trauma and hence this cannot be assigned as the cause of pathogenesis(9).

It is believed that these tumours arise in these areas because the bones of the mandible, sphenoid, ethmoid, and parts of the temporal bone form largely through the process of endochondral ossification. In contrast, the other cranial bones (i.e.,frontal and parietal bones) arise from intramembranous ossification and are less frequently affected by GCTB(18).

Although it is regarded as benign, the giant cell tumour is locally aggressive, and malignant variants can even metastasize to the lung(19,20). Malignant giant cell tumours have been reported, usually resulting from secondary malignant transformation after radiation treatment(22). They present with marked atypia and pleomorphism with mitotic activity and vascular invasion(23).

The treatment of choice of GCT is surgical excision(19,24). Regardless of the site of presentation, partial resection or curettage results in a recurrence rate of up to 70%, whereas recurrence after wide resection is about 7%(25). Radiotherapy is generally avoided and is reserved for GCT's that are considered inoperable, due to the risk of sarcomatous transformation(26).

Other treatment modalities including cryotherapy, chemotherapy, and curettage with adjuvant agents have been tried, but have yielded fewer effective results(12,27). Adjuvant therapy is recommended for cases where complete resection cannot be achieved(28). GCTs generally recur within the first 3 years if recurrence occurs(26). But continuous evaluation upto 5 years is recommended, since late distant metastasis have been reported(12).

In conclusion, GCT arising from the mandible is a rare disease whose diagnosis is difficult. Due to the possibility of local recurrence of soft tissue giant cell tumour of low malignant potential, clinical follow-up is recommended. Wide complete excision is required since incomplete excision results in a high incidence of recurrence.

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