



AGGRESSIVE CENTRAL GIANT CELL GRANULOMA OF MAXILLA: A CASE REPORT

Maxillofacial Surgery

Dr Rangeet Bhadra	Junior Resident, Department of Oral and Maxillofacial Surgery, Faculty of Dental Sciences, King George's Medical University, Lucknow U.P
Dr Hari Ram	Professor, Department of Oral and Maxillofacial Surgery, Faculty of Dental Sciences, King George's Medical University, Lucknow U.P
Dr Heena Yadav	Junior Resident, Department of Oral and Maxillofacial Surgery, Faculty of Dental Sciences, King George's Medical University, Lucknow U.P
Dr Kipa Guma*	Junior Resident, Department of Oral and Maxillofacial Surgery, Faculty of Dental Sciences, King George's Medical University, Lucknow U.P *Corresponding Author

ABSTRACT

Central giant cell granuloma (CGCG) is a non-odontogenic, osteolytic lesion of unknown etiology. It affects the craniofacial region. The commonly affected age group is below 30 years, with female predilection. Histologically it is characterized by an abundance multinucleated giant cells within a sea of spindle-shaped mesenchymal stromal cells, scattered throughout the fibro-vascular connective tissue stroma containing areas of haemorrhage and hemosiderin deposits. A case of a large destructive CGCG in a child involving the entire right maxilla and the lateral wall of nasal cavity causing gross facial asymmetry is presented. It was treated by surgical excision of the entire lesion while preserving the right orbital floor. This was followed by placement of a medicated gauze pack dressing to hasten wound healing and bone formation. Following surgical intervention, the facial aesthetics and oral intake of the child was significantly improved giving a better quality of life.

KEYWORDS

Central Giant Cell Granuloma, Fibro-osseous lesions

INTRODUCTION

The central giant cell granuloma (CGCG) is a non-odontogenic, osteolytic lesion of unknown etiology, which affects the craniofacial region, particularly the anterior mandible. WHO has defined CGCG as "A localized benign but sometimes aggressive, osteolytic proliferation consisting of fibrous tissue with haemorrhage and hemosiderin deposits and presence of osteoclast-like giant cells with reactive bone formation". First described by Jaffe in 1953 and termed "giant cell reparative granuloma". The "reparative" term was later dropped as the tumour did not present always with a consistent reparative process¹. It is histologically benign, although locally proliferative and destructive from osteoclastic activity². It accounts for 7% benign maxillary tumours, with females predominantly affected in 2/3 of cases before age 20 years⁴. Radiographically it presents as a multilocular radiolucency with honeycomb or soap bubble-like appearance having scalloped margins. Aetiology of the lesion include inflammation, intraosseous haemorrhage, local trauma, and gene mutations may be involved⁵. It can be of two types:-non-aggressive and aggressive type. The aggressive type which is seen mostly in younger age groups has been seen to present with pain, numbness, root resorption, rapid spread, bone perforation, and a high rate of recurrence⁶. Depending upon the extent, clinical behaviour, and characteristics treatment option varies from non-surgical to surgical approaches. Surgical approaches includes excision or resection, cryotherapy, enucleation local curettage with or without chemical cauterization⁷. Excision of CGCG by curettage with removal of peripheral bone margins is the gold standard. Early intervention and treatment gives good prognosis. Surgical resection has good prognostic value and low rate of recurrence

CASE REPORT

A 10 year old girl was referred to Department of Oral and Maxillofacial Surgery, King George's Medical University on 15/2/2023 from a private clinic in Lucknow. Patient had developed a large swelling on the right side of the face in the upper jaw over the last 8 months. Initially it started as a diffuse intraoral swelling of the left side of the hard palate which rapidly increased to its present size to cause gross facial asymmetry and loss of multiple teeth.

On examination there was obvious extra oral swelling measuring 7.5cmx7.0cm. Antero-posteriorly it extended from philtrum of the upper lip to 4 cm lateral to the right corner of the mouth and supero-inferiorly extended from mid region of right ala tragus line to 2 cm above the lower border of mandibular body region. This swelling was due to intraoral expansion of the lesion which has caused deviation of the nose. Intraoral swelling measured 4cmx6.5cm, antero-posteriorly extends from region of tooth number 21 till mesial of 55. Medio-

laterally extends from right buccal vestibule to the hard palate and crossing the midline. Swelling is well defined having normal mucosal lining with isolated erythematous areas and smooth surface texture. It was firm on palpation, non-tender having well defined borders, fixed to the underlying bone and bleeding on probing with an explorer point and pus discharge from some areas. Bilateral submandibular and submental lymph nodes were tender and palpable measuring approximately 5mm to 1cm (Refer to Figure 1a and 1b)



Figure 1a:- Pre-operative Clinical Photographs



Figure 1b:- Pre-operative Clinical Photographs

Patient underwent incisional biopsy under general anaesthesia on

21/2/2023. Gross examination showed multiple greyish white soft tissue pieces largest measuring 1.0*0.8*0.5cm. Microscopic examination showed fibro-collagenous tissue lined by stratified squamous epithelium with numerous multinucleated osteoclast like giant cells within cellular, vascular and fibrous stroma with focal red blood cell extravasation. Brown's tumour of Hyperparathyroidism was ruled out by investigating serum PTH, calcium and alkaline phosphatase which were within normal limits

Contrast enhanced computed tomographic (CECT) revealed to be well circumscribed lytic expansile hypodense non enhancing ovoid lesion of size approximately 31x28x30mm seen involving the right superior alveolar arch causing its bony destruction superiorly, abutting right maxillary sinus. Lesion was involving the right nasal cavity, posterior nasal cavity, ethmoid sinus as well (Refer to Figure 2)

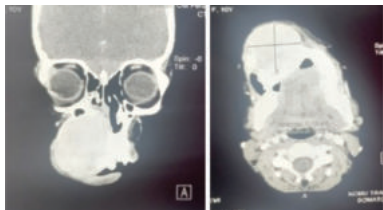


Figure 2:- CECT Of Maxillofacial Region For The Patient

Patient was posted for surgical excision of the lesion (Refer to Figure 3). Intra oral approach was undertaken and lesion was removed in toto (Refer to Figure 4) followed by clearance of the nasal cavity and maxillary sinus. This was followed by placement of an iodoform gauze pack in the region of the surgical defect (Refer to Figure 5) and placement of a surgical obturator. Extra oral tape dressing was placed as well. The specimen was sent for histopathological examination and reporting. There was significant improvement of the facial deformity that had afflicted the patient for the last 8 months in the immediate post-operative period (Refer to Figure 5).

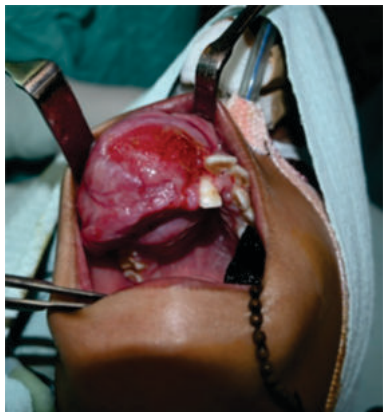


Figure 3:- Intraoperative Photograph Of Lesion

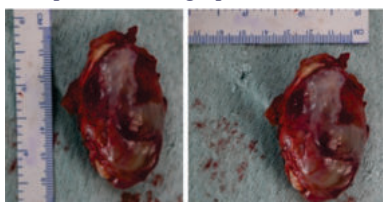


Figure 4:- Excised Tumour



Figure 5:- Iodoform Gauze Pack Placed Intraoperatively Shown In OPG

Post-operative period showed remarkable improvement within a week of surgery. There was significant improvement in the facial symmetry and intake was improved. Patient was also encouraged for improving her intake and maintaining oral hygiene. (Refer to Figure 5 and Figure 6).



Figure 5:- Post Operative Clinical Photograph



Figure 6:- Intraoral Photograph Showing Healed Surgical Site

Gross pathology of the sample revealed its outer surface to be smooth and congested measuring 6.0x3.5x2.5cm. Cut surface showed greyish white firm areas. Section shows multiple tissue bits partially lined by stratified squamous epithelium, proliferation of fibroblastic spindle cells along with many osteoclastic giant cells and proliferating capillary sized blood vessels as well as extravasated red blood cells. (Refer to Figure 6)

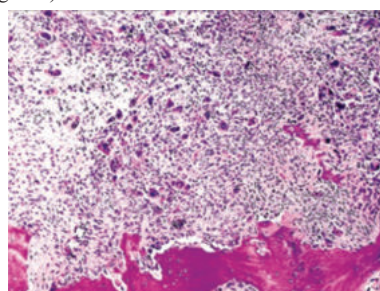


Figure 6:- Histopathological Slide Of Resected Tissue Sample

Discussion

The term Giant Cell Reparative Granuloma was first given by Jaffe to describe lesions which he believed to be caused by traumatic hemorrhage to the jaw⁸. It has been observed to occur mostly in children and young adults usually below the age of 30 years in 75% of cases reported⁹. There appears to be a female to male predilection of 2:1¹⁰⁻¹². Most common site has been observed to be mandible than maxilla. Mandibular CGCG occurs equally in anterior and posterior segments. Maxillary CGCG occurs mostly in anterior segment. Histologically giant cells are a characteristic feature of CGCG, however it has been seen in other tumors as well, hence a complete diagnosis is dependent upon a combination of clinical, radiological and histopathological criteria. In 1988, Jacoway et al. were the first to use corticosteroids for the treatment of CGCG. They used 150mg of Prednisolone per cubic centimeter of tumor over 6 weeks¹³. This is a well-studied role of corticosteroid as per many authors but have shown

to be associated with Cushing's syndrome¹⁴. Injection Calcitonin as well as inhalation method by nasal spray have also been used by some authors. Chien et al. have used bisphosphonates such as alendronate¹⁵. It causes reossification of lesion but often causes BRONJ^{16,17}. Alfa interferon have been used to reduce the aggressive nature of CGCG. It has antiangiogenic properties. Surgery remains the mainstay treatment and better choice for CGCG as per literature. Surgical enucleation and curettage is the treatment of choice in most CGCG as done in the above case. Segmental resection is used in recurrent and aggressive cases. Lowest recurrence has been reported with en bloc resection with a 5 mm margins¹⁸. Our case of CGCG is an aggressive type because it developed in a short period of time. It also occurred in a less common site which is the maxilla. Radiological examination and incisional biopsy was done to reach a confirmatory diagnosis. After ruling out systemic diseases the lesion was treated by surgical enucleation and curettage which resulted in immediate improvement of esthetics and oral intake of the patient.

CONCLUSION

Complete removal of the neoplasm without any recurrence should be the key towards successful treatment of such lesions. It involves understanding of the pathogenesis of the mechanisms that underlie such lesions to formulate a constructive and effective treatment plan. Continuing efforts to minimize recurrence should be advocated.

REFERENCES

1. Kruse-Löslér, B., Diallo, R., Gaertner, C., Mischke, K. L., Joos, U., & Kleinheinz, J. (2006). Central giant cell granuloma of the jaws: a clinical, radiologic, and histopathologic study of 26 cases. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology*, 101(3), 346-354.
2. Motamedi, M. H. K., Eshghyar, N., Jafari, S. M., Lassemi, E., Navi, F., Abbas, F. M., ... & Eshkevari, P. S. (2007). Peripheral and central giant cell granulomas of the jaws: a demographic study. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology*, 103(6), e39-e43.
3. Dimitakopoulos, I., Lazaridis, N., Sakellariou, P., & Asimaki, A. (2006). Giant-cell granuloma in the temporal bone: a case report and review of the literature. *Journal of oral and maxillofacial surgery*, 64(3), 531-536.
4. Stage, D., Pusel, J., Janser, J. C., Rodier, D., & Philippe, E. (1986). Lésions à cellules géantes de la mandibule: problèmes de diagnostic différentiel. A propos de trois cas de granulome central de réparation. In *Annales d'oto-laryngologie et de chirurgie cervico-faciale* (Vol. 103, No. 3, pp. 159-166).
5. Lin, Y. J., Chen, H. S., Chen, H. R., Wang, W. C., Chen, Y. K., & Lin, L. M. (2007). Central giant cell granuloma of the mandible in a 7-year-old boy: a case report. *Quintessence International*, 38(3).
6. Chuong, R., Kaban, L. B., Kozakewich, H., & Perez-Atayde, A. (1986). Central giant cell lesions of the jaws: a clinicopathologic study. *Journal of oral and maxillofacial surgery*, 44(9), 708-713.
7. Rachmiel, A., Emodi, O., Sabo, E., Aizenbud, D., & Peled, M. (2012). Combined treatment of aggressive central giant cell granuloma in the lower jaw. *Journal of Cranio-Maxillofacial Surgery*, 40(3), 292-297.
8. Sentilhes, C., & Michaud, J. (1986). Lésions à cellules géantes du maxillaire. Difficultés diagnostiques. *Revue de stomatologie et de chirurgie maxillo-faciale*, 87(2), 102-107.
9. Whitaker, S. B., & Waldron, C. A. (1993). Central giant cell lesions of the jaws: a clinical, radiologic, and histopathologic study. *Oral surgery, oral medicine, oral pathology*, 75(2), 199-208.
10. Austin Jr, L. T., Dahlin, D. C., & Royer, R. Q. (1959). Giant-cell reparative granuloma and related conditions affecting the jawbones. *Oral Surgery, Oral Medicine, Oral Pathology*, 12(11), 1285-1295.
11. Waldron, C. A., & Shafer, W. G. (1966). The central giant cell reparative granuloma of the jaws. An analysis of 38 cases. *American Journal of Clinical Pathology*, 45(4), 437-447.
12. Chuong, R., Kaban, L. B., Kozakewich, H., & Perez-Atayde, A. (1986). Central giant cell lesions of the jaws: a clinicopathologic study. *Journal of oral and maxillofacial surgery*, 44(9), 708-713.
13. Jacoway, J. R. (1988). Central giant cell granuloma-an alternative to surgical therapy. *Oral Surg Oral Med Oral Pathol*, 66, 572.
14. El Hadidi, Y. N., Ghanem, A. A., & Helmy, I. (2015). Injection of steroids intralesional in central giant cell granuloma cases (giant cell tumor): Is it free of systemic complications or not? A case report. *International journal of surgery case reports*, 8, 166-170.
15. Chien, M. C., Mascarenhas, L., Hammoudeh, J. A., & Venkatramani, R. (2015). Zoledronic acid for the treatment of children with refractory central giant cell granuloma. *Journal of pediatric hematology/oncology*, 37(6), e399.
16. da Silva, N. G., Carreira, A. S. D., Pedreira, E. N., Tuji, F. M., Ortega, K. L., & de Jesus Viana Pinheiro, J. (2012). Treatment of central giant cell lesions using bisphosphonates with intralesional corticosteroid injections. *Head & Face Medicine*, 8(1), 1-6.
17. Naidu, A., Malmquist, M. P., Denham, C. A., & Schow, S. R. (2014). Management of central giant cell granuloma with subcutaneous denosumab therapy. *Journal of Oral and Maxillofacial Surgery*, 72(12), 2469-2484.
18. Inchingolo, F., Tatullo, M., Abenavoli, F. M., Marrelli, M., Inchingolo, A. D., Inchingolo, A. M., & Dipalma, G. (2011). Non-Hodgkin lymphoma affecting the tongue: unusual intra-oral location. *Head & neck oncology*, 3(1), 1-5.