

Reparative giant cell granuloma of the maxilla

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K. A. Jeevan Kumar, S. Humayun¹, B. Pavan Kumar, J. Brahmaji Rao

Department of Oral and Maxillofacial Surgery, Kamineni Institute of Dental Sciences, Narketpally, Nalgonda, ¹Department of Dental and Maxillofacial Surgery, Kamineni Hospitals, King Koti, Hyderabad, Andhra Pradesh, India

Address for correspondence:

Dr. S. Humayun, H.No.10-2-283/A/2, 1st Floor, Near Shantinagar Police Quarters, Vijayanagar Colony, Hyderabad – 500 057, Andhra Pradesh, India. E-mail: drshumayun@yahoo.com

ABSTRACT

Giant cell reparative granuloma accounts for 1–7% of all benign lesions of the jaw. It often arises in the maxilla followed by mandible and affects children and young adults. It is usually a slow-growing lesion. The fast growing lesions are rare and despite the innocent histological appearance, has an aggressive behavior mimicking a malignant lesion. In the present report, the clinical features, diagnosis, and surgical treatment of an unusually large aggressive variety of reparative giant cell granuloma found in the cheek with extensions into maxilla, antrum, and infratemporal region in a 23-year-old female is described. The impact of delay in correct diagnosis on massive enlargement of the lesion, the importance of computed tomography-guided biopsy in the diagnosis of such inaccessible lesions, and the role of a general dentist in the early detection are also emphasized.

Keywords: Benign tumor, brown tumor, giant cell granuloma, giant cell lesion, maxilla, partial maxillectomy, Weber-Fergusson incision

INTRODUCTION

Reparative giant cell granuloma (RGCG) is not a true neoplasm but rather a reactive process; its origin can be triggered by trauma or inflammation.^[1,2] It is a rare bony lesion in the head and neck region. It is a nonodontogenic lesion never seen in any other bone of the skeleton.^[3] It most commonly affects maxilla followed by the mandible and often seen in children and young adults, predominantly females, in the second and third decades of life.^[4] RGCGs are classified, according to location, as central (bone) and peripheral (gingival tissues).^[5]

Fast-growing lesions have rarely been reported. In these cases, RGCGs are characterized by an aggressive behavior against an innocent histological appearance, pain, and rapid facial swelling and high recurrence rate (as early as 3 and as late as 22 years).^[6,7] The clinical importance of these benign tumors is that they clinically mimic a malignant lesion.^[8] The present report illustrates a rare aggressive variety of RGCG, with an atypical clinical presentation in a 23-year-old female; attention has been focused in particular on computed tomography (CT)-guided biopsy and surgical treatment.

CASE REPORT

A 23-year-old female reported with complaints of a slowly expanding swelling of right cheek and upper jaw, pain in right upper teeth, and painful watering of the right eye of 2-3 months duration.

Her dental history revealed that she had pain in 16 about 1½ year back and got root canal treatment done for it by a local general dentist and was apparently asymptomatic for the next 6 months. There was no history of trauma. Then she had intermittent pain in the same tooth and antibiotics were given. Later she developed a swelling on right cheek and felt severe pain so the tooth 16 was extracted by the same dentist about 3 months back. As the pain and swelling did not subside even after extraction, incisional biopsy was performed twice from the region of 16 both from the socket and vestibule by oral and maxillofacial surgeons. It was histologically diagnosed as nonspecific inflammatory lesion.

On examination, extraorally a swelling measuring about 4 × 5 cm with diffuse borders was seen on right cheek extending from the corner of the mouth, nose, to lower eyelid. On palpation,

the swelling was firm to hard in consistency with moderate tenderness [Figure 1]. Skin over the swelling was reddened and compromised facial nerve function on right side on smiling was also observed. Obliteration of right nasolabial fold and mild edema over right lower eyelid was noticed. Right eye showed no change in visual acuity and eyeball movements.

The intraoral examination revealed a purplish discoloration with expansile mass in the right upper vestibular region extending from 13 to 17 with complete obliteration of the vestibule [Figure 2]. Oroantral fistula resulted after the earlier biopsies in the regions of 14, 15, and 16. There was no caries in the associated teeth but were periodontally compromised: 14, 15, and 17 showed grade II mobility and 13 and 18 exhibited grade I mobility. Significant palatal expansion in the molar region was also observed. Aspiration was negative and the lesion was found to be bleeding actively on slightest provocation.

OPG showed a radiolucent lesion with ill-defined borders in the regions of 15 to 17. CT scan revealed a soft tissue mass completely obliterating the right vestibule, cheek, and extending on to maxilla and maxillary antrum with thinning and destruction of parts of

the antral walls [Figure 3]. The mass extended inferiorly into the body of the maxilla up to alveolus, involving the teeth. Medially, it almost obliterated the posterior and inferior third of the right maxillary antrum [Figure 4]. Superiorly, it extended up to the floor of the orbit and laterally into the body of the zygoma. Posteriorly, it reached the lateral and medial pterygoid plates, infratemporal fossa, and lower portion of the temporal space [Figure 5]. CT also showed evidence of weakness [Figure 6]. 3D reconstruction CT shows perforation of anterolateral surface of the maxilla and uninvolved right orbital floor [Figure 7].

Keeping in mind the clinical and radiological features, it was thought that the lesion could not be a nonspecific inflammatory one but it could be a benign tumor. Therefore, a biopsy was done to obtain more deeper tissue and to have an idea about the exact nature of the lesion, and it was histologically revealed as a giant cell granuloma [Figure 8].

With this provisional diagnosis, an incisional biopsy was performed under general anesthesia, and the histopathological examination confirmed it as RGCC [Figure 9].



Figure 1: Facial photograph showing swelling of right cheek and obliteration of right nasolabial fold



Figure 2: Intraoral view of the lesion which shows obliteration of the vestibule

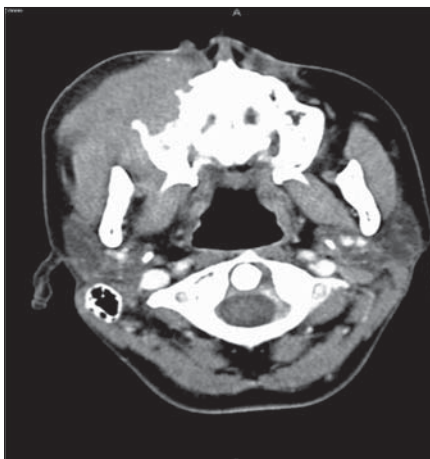


Figure 3: Axial section of CT scan. The lesion involves cheek and extends into infratemporal region



Figure 4: Coronal section shows extension of tumor into the antrum

The patient was advised routine and special blood investigations to rule out hyperparathyroidism. Complete excision of the tumor mass along with partial maxillectomy to gain access to the infratemporal region was planned. Partial maxillectomy was carried out through a Weber-Fergusson incision under general anesthesia [Figures 10 and 11]. Palatal mucosa was separated from the bony segment and greater palatine artery ligated. Entire tumor mass was removed along with portions of invaded bone and corresponding teeth. Pressure resorption of the alveolus with exposure of the roots of the teeth was observed intraoperatively.

A careful and thorough curettage of the residual bony cavity was performed [Figure 12]. The defect was reconstructed with full-thickness skin graft harvested from right thigh region [Figures 13 and 14]. The graft was secured by interrupted sutures. Postoperatively nasogastric feeding was maintained for 10 days. The postoperative course was uncomplicated and the patient was discharged on postoperative day 10. The patient was regularly observed at follow-up visits and the wound healing was uneventful [Figure 15]. Patient recovered well and was followed up for 1 year. She is asymptomatic and no recurrence is evident till date.

DISCUSSION

RGCG is a rare disease.^[9] It can occur at any age but most frequently in the second and third decades and involves the maxilla more than mandible.^[10] It is twice as frequent in females than males.^[11] World Health Organization defines it as an intraosseous lesion consisting of cellular fibrous tissue and contains many foci of hemorrhage, aggregations of multinucleated giant cells, and occasionally trabeculae of woven bone.^[12] In our present case, the lesion might have started in the soft tissues of the cheek and vestibule and involved the maxilla secondarily by pressure erosion, showing the rarity of the case.

Although lesion is expansive and invasive, it does not usually involve perineural sheaths, for this reason paresthesia is usually not observed in these patients.^[13] But there was weakness of right facial nerve function in the present case; it may be due to invasion into or pressure on nerve fibres by the tumor mass. Despite the fact that the course of the disease is considered benign, there still exist some reports in literature where metastasis has been observed.^[14] Furthermore, malignant transformations to osteosarcoma or fibrosarcoma have also been reported.^[15]

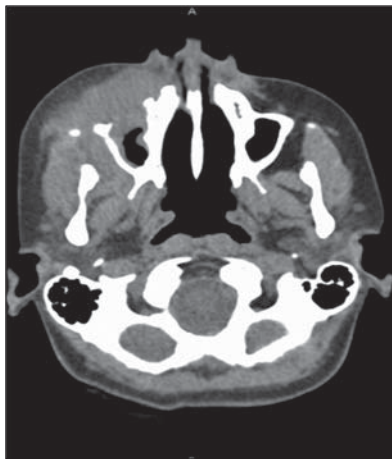


Figure 5: Extension of tumor mass over the pterygoid plates and infratemporal fossa



Figure 6: CT scan showing evidence of weakness the facial nerve function



Figure 7: 3D Volume rendering image shows perforation of anterolateral surface of maxilla



Figure 8: CT-guided biopsy of the lesion

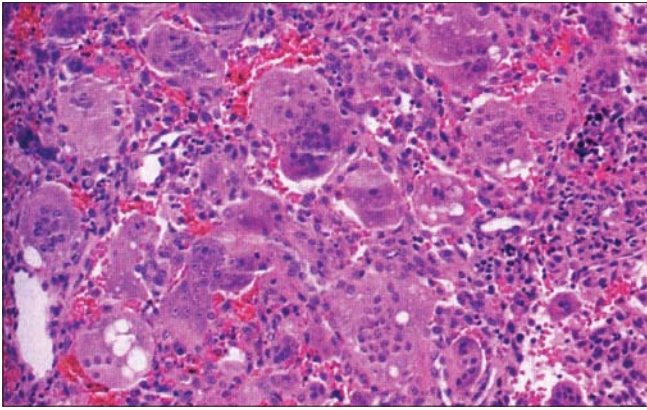


Figure 9: Histopathological section of the lesion showing giant cells. (H and E, 20x)



Figure 10: Exposure of the lesion after Weber-Fergusson incision and reflection of the cheek flap



Figure 11: Partial maxillectomy osteotomizing the portion of the maxilla to gain access to the infratemporal region



Figure 12: Surgical defect following excision of the tumor showing portions of the maxilla and orbital floor through antrum



Figure 13: Harvesting of full-thickness skin graft from right thigh region



Figure 14: Reconstruction of the defect: skin graft is secured by interrupted sutures

Histologically, it is indistinguishable from other giant cell lesions such as cherubism and aneurysmal bone cyst. Giant cell granuloma forms a lobulated mass of proliferative vascular connective tissue packed with giant cells. These giant cells are seen lying in vascular stroma.^[10] These cells have a patchy distribution, and signs of bleeding into the mass and deposits of

hemosiderin are frequently seen.^[11] Brown tumors are identical to RGCG, both histologically and radiographically, but they were ruled out on the basis of normal serum levels of calcium,



Figure 15: Immediate postoperative view showing final closure

phosphorus, alkaline phosphatase, and good renal function.^[8]

Imaging plays an essential role in the detection, characterization, presurgical evaluation, and in postoperative follow-up of these lesions.^[16] Radiological appearance of RGCG is nonspecific, and conflicting descriptions have appeared in various articles.^[17] CT scan allows an optimal view of the bone and soft tissue and provides essential data for differentiating benign from malignant lesions, for diagnosing difficult inaccessible lesions, and for planning correct surgical procedures.^[18]

Surgery is the most accepted and traditional form of treatment. However, tissue removal ranges from simple curettage to bloc resection.^[19] Radiation therapy in such a case is contraindicated.^[20] There have been cases reported in which radiation-treated lesions have undergone malignant transformation.^[21] Incidence of recurrence after surgery is 4–20%, whereas locally aggressive giant cell lesions have a higher recurrence rate and it usually occurs due to incomplete removal of the tumor.^[10,22] Several surgical techniques have been proposed for removal of more aggressive giant cell granuloma and for an aggressive lesion that shows rapid growth and facial swelling, bloc resection, and suitable reconstruction of the affected area is considered to be the most appropriate approach.^[3]

The lesion in the present case might have started in the soft tissues and involved the maxilla and antrum secondarily as described earlier. As the patient first reported with tooth pain in 16, probably the dental surgeon concentrated more on pulpal pathology and forgot about adjacent structures. And the previous oral surgeons also might have taken the biopsy from the extracted socket of 16 and this area was not yet invaded by the tumor mass, hence, the diagnosis was totally misleading.

Due to the delay in correct diagnosis, patient's valuable time was lost and the tumor mass increased to a huge size and involved the infratemporal and temporal spaces which were surgically difficult to access. If the lesion was diagnosed at an early stage, these large extensions into infratemporal regions and extensive resection and reconstructive surgery could have been avoided and facial esthetics might not have been compromised.

Nonsurgical approaches to avoid disfigurement have also been used, including daily systemic doses of calcitonin and intralesional injection with corticosteroids.^[19] Some giant cell granulomas can be sterilized thermally using laser or cryoprobe.^[22] Weekly intralesional injections with corticosteroids have reported successful results in literature.^[23] Corticosteroids, however, are contraindicated in certain conditions such as diabetes mellitus, peptic ulcer, and immunocompromised state.^[11] Nonsurgical treatment is good for slow-growing lesions; however, successful treatment of large, rapidly growing lesions is more likely to be achieved surgically.^[8,11]

CONCLUSION

RGCG is an uncommon benign tumor, but in some cases it is aggressive and locally destructive. Giant cell granuloma is one of those lesions which have obscure etiopathogenesis with differing clinical presentations and treatment modalities. Diagnosis of these lesions at an early stage will make the treatment simple and more conservative; it can also avoid spread of lesions into inaccessible regions, radical surgeries, and facial disfigurement. The present case report emphasizes the need for a thorough assessment of any case in clinical practice to prevent morbidity due to late and/or false diagnosis.

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