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Multifocal Central Giant Cell Granuloma - A Case Report

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ABSTRACT

Central giant cell granuloma is a benign, aggressive neoplasm composed of multinucleated giant cells that almost exclusively occurs in the jaws though extra-gnathic incidence is rare.Multifocal CGCGs of the jaws are very rare and suggestive of systemic diseases such as hyperparathyroidism,an inherited syndrome such as Noonan-like multiple giant cell lesion syndrome or other disorders.Very few cases of multifocal CGCGs in the jaws without any concomitant systemic disease have been reported. This paper describes an unusual case reported to the Oral Surgery Department of Dr. D.Y.Patil Dental College & Hospital, Nerul, Navi-Mumbai in 2014 in a 45-year-old male with multifocal central giant cell granuloma involving maxilla and mandible. The serum alkaline phosphatase, calcium and phosphorus levels were within the normal limits. After complete clinical examination hyperparathyroidism and clinical characteristic of any syndromes such as Noonan-like syndrome and neurofibromatosis were ruled out. Thus this paper reports a non-syndromic multifocal central giant cell granuloma.

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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 (1,2).The etiopathogenesis of this lesion is unclear with genetic and reparative response being hypothesised as positive factors (3,4).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 (3). The clinical behaviour of CGCG is variable being aggressive and associated with pain, osseous destruction, cortical perforation, root resorption, and recurrence (1,5). Multifocal central giant cell granulomas in the jaws are rare and is suggestive of hyperparathyroidism, Noonan-like multiple giant cell lesion syndrome, Giant cell tumour, Cherubism or Paget's disease. Multifocal, CGCGs without any concomitant systemic disease are extremely rare (4). To the best of our knowledge, only 10-15 such cases have been reported in English literature (4).

All of them have been seen in females. This paper describes an unusual case of non-syndromic multifocal Central Giant Cell Granuloma, in a male patient.

Case Report

A 47 year old male patient reported to the Oral Surgery Department of Dr. D.Y.Patil Dental College & Hospital, Nerul, Navi-Mumbai in 2014 with the chief complaint of growth in the lower jaw (Figure 1a &1b) since 5 months which was initially small and gradually attended the present size and was associated with intermittent pain.

On examination, a diffuse growth of 5×3×2 cm in dimension was seen involving the anterior region of mandible extending in the region of 35 to 45 (Figure 1c).The surface of the growth was smooth. It was tender on palpation and firm in consistency.

Ethical approval of this case was taken by the Ethical Committee of Dr. D.Y.Patil Dental College, Nerul, Navi Mumbai and patient consent received.

Orthopantomogram and Computed Tomography were performed. Orthopantomogram revealed an extensive osteolytic lesion extending in the region of 33 to 45 crossing the midline and measured 3x2 cm in dimension (Figure 2).

The three dimensional reformatted image of the CBCT scan showed displacement of the maxillary lateral incisor, canine and perforation of the labial and lingual cortices in mandible. There was no evidence of pathologic fracture (Figure 3a). The axial scan of CBCT of the maxillary lesion at the level of maxillary sinus revealed a well- defined osteolytic lesion in the anterior region of the maxilla measuring 13×16 mm in its maximum dimension, crossing the midline. This section showed intact interior wall of the sinus and increased thickness of the mucosal lining (Figure 3b).The coronal section passing through two molars revealed thinning of the lining of the nasal cavity on the lateral aspect and was discontinuous and eroded towards the midline (Figure 3c).The axial section of the mandible at the level of the roots of the mandibular teeth showed a soft tissue growth on the labial aspect of the mandible which measured 35.77 x 27.30 mm in its maximum dimension extending from 35 to 45 region crossing the midline. This section revealed break in the continuity of the labial and lingual cortices of the mandible (Figure 3d). Routine haematological and urine investigations were normal. The serum calcium and alkaline phosphatase levels were within normal limits excluding the possibility of endocrinal disturbances.

On incisional biopsy, the microscopic examination of the tissue showed numerous giant cells with proliferating fibroblasts and delicate collagen fibres, along with budding capillaries and dense inflammatory cell infiltration. Deeper section showed new bone formation. The features were suggestive of Central Giant Cell Granuloma.

Two months later, the patient reported to the department again with the chief complaint of swelling in the upper jaw. On examination, an oval swelling of 3×2×2 cm in dimension was seen on the anterior palatal region which was hard in consistency. The overlying mucosa was normal.

An excisional biopsy was performed for both lesions involving the maxilla and mandible and tissue was sent for histopathology. Tissue from maxillary lesion comprised of multiple bits of soft tissue measuring 1×1.5×0.5 cm to 0.2×0.2×0.2 cm in dimension whereas tissues from mandible consisted of section of mandible along with associated teeth and measured 3.2×4.5×2 cm in dimensions (Figure 4a&b). All the tissues were irregular in shape, brownish in colour and firm in consistency. Microscopic examination revealed highly cellular connective tissue stroma comprised of bi-phasic cell population with numerous proliferating fibroblasts and multinucleated giant cells. Foci of extravasated RBC's were evident (Figure 5a&b). Special stains such as Prussian blue and Van Gieson were performed

(Figure 5c&d). Prussian blue stain demonstrated hemosiderin pigment. Final diagnosis of Central Giant Cell Granuloma was given.

Discussion

The WHO has defined (CGCG) as "an intraosseous lesion consisting of cellular fibrous tissue that contains multiple foci of haemorrhage, aggregations of multinucleated giant cells and occasionally trabeculae of woven bone"(2).

The etiopathogenesis of the CGCG of jawbones is controversial and has been a reactive, an inflammatory, an infective, or a neoplastic process occurring secondary to previous trauma and involving intraosseous haemorrhage by various authors (2, 6). Donoff and Rosenberg suggested the local changes in the blood flow throughout the bone and local bone dysplasia as the etiologic factors (7). Cossio P. et al. have suggested traumatic etiology (6).

CGCG occurs in patients younger than 30 year old with female preponderance (1). However in our case, it involved a 45 year male patient. It commonly involves the anterior region and more frequently mandible, usually crossing the midline. Clinically, the lesion may vary from an asymptomatic lesion that grows slowly without expansion, seen in this case,to an aggressive, painful process followed by root resorption, cortical bone destruction, and extension into the soft tissue (4).

Some authors classified CGCG into aggressive and nonaggressive types based on their clinical and radiographic characteristics (8). This case presented with intermittent pain. CGCG of the jaw is usually unifocal, though multifocal lesions have been reported in literature (4). The radiographic examinations of CGCG are not specific. CGCG are expansile, radiolucent lesions, multilocular and with uncorticated borders (1).

Of 20 cases studied, 55% of the lesions were radiolucent, 45% showed a mixed feature, and 65% presented well-defined limits (9). Kaffe et al. observed in their study on 80 cases as 51% of the lesions multilocular, 44% unilocular, 5% non-loculated, and 68% of the multilocular lesions in the mandible (2). 7.7%, 12% and 37% of radicular resorption respectivelywas reported (9).The present case demonstrated root resorption. CGCG affecting the maxillary sinus may expand or erode bone walls (9).

Computed tomography is the best modality for determining the extent of the lesion (8). The CT scan of the present case demonstrated an osteolytic lesion with well-defined borders in the anterior region of the maxilla. The axial section of the mandible revealed break in the labial and lingual cortices.

The histopathology of CGCG is characteristic with numerous multinucleated giant cells embedded in a fibro-cellular stroma with blood vessel walls. CGCG consist of spindle-shaped stromal cells (fibroblasts or myofibroblasts) loosely arranged in a fibrous stroma. Foci of haemorrhage with hemosiderin pigment along with newly formed osteoid or bone is observed in the stroma (2). Morphologically, the giant cells resemble foreign body type or osteoclast-like. The osteoclast-like multinucleate giant cells arise from the fusion of the mononuclear component, and the mononuclear cells may be the osteoclast precursors (2).

Miloro M, et al reviewed all the cases of Central Giant Cell Granuloma with multifocal involvement reported in literature till 1999 of which maximum were syndromic and very few were non-syndromic (4). Edwards P, et al reported a case of central giant cell granuloma with bilateral mandibular involvement and associated with Noonan syndrome (10).

Soundarya N et al reported a case of multifocal central giant cell granuloma associated with

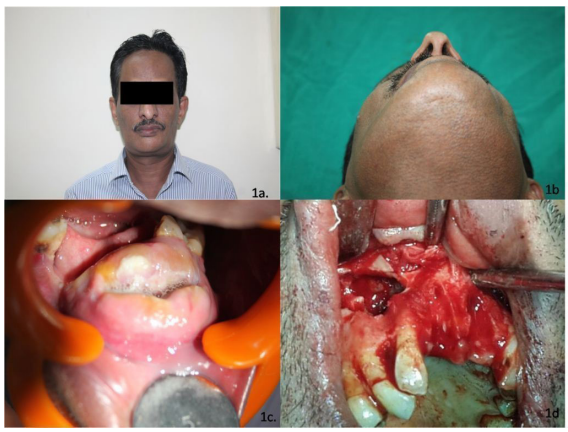

Fig. 1
a&b: Extraoral examination. c: Diffuse growth in the anterior region of mandible extending from 35 to 45.d-Intraoperative (Maxillary lesion).

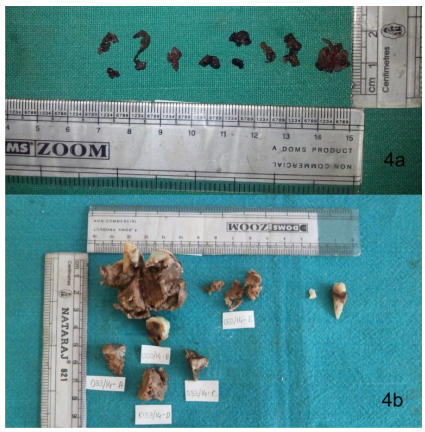

Fig. 4
Macroscopic examination of excisional biopsy from maxilla (a) mandible (b).

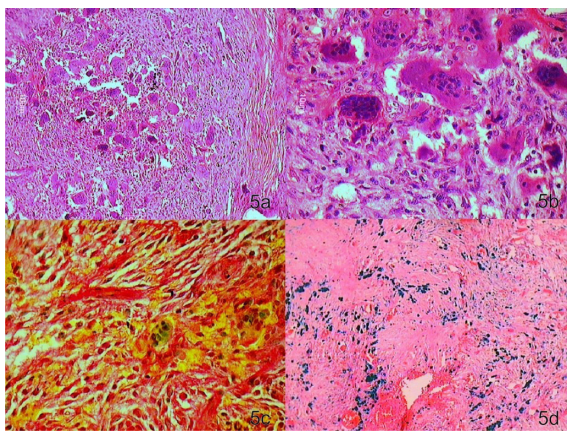

Fig. 5
a&b: H and E stained section (10X&40X) showed numerous giant cells with proliferating fibroblasts and delicate collagen fibres, along with budding capillaries and dense inflammatory cell infiltration. c:VanGieson stain showing red colour imparted to collagen. d:Prussian blue stain showing blue coloured hemosiderin pigment.

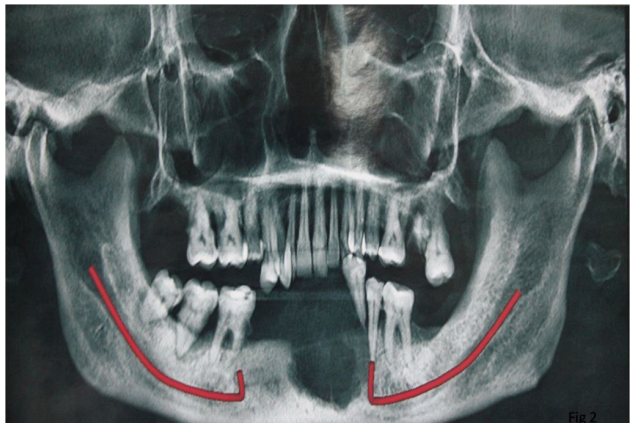

Fig. 2
Orthopantomogram showing an extensive osteolytic lesion extending from 33 to 45.

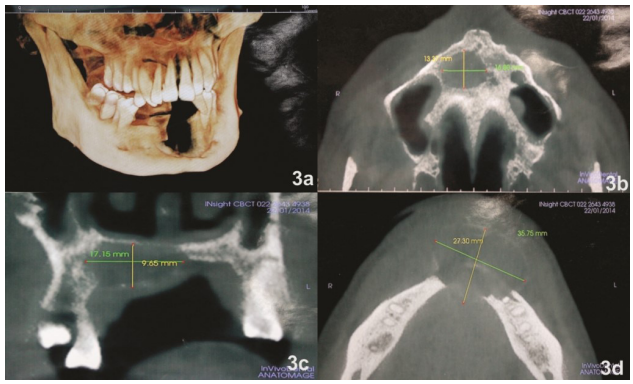

Fig. 3
a: The three dimensional reformatted image of the CBCT scan showing perforation of the labial and lingual cortices in mandible. b:The axial scan of maxillary sinus showing osteolytic lesion in the anterior region of the maxilla. c: The coronal section passing through two molars revealed thinning of the lining of the nasal cavity. d:The axial section of mandible showing break in the continuity of the labial and lingual cortices of the mandible.

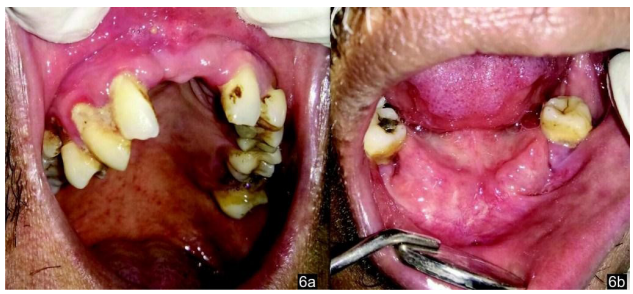

Fig. 6
a&b: Post-operative healing of maxillary and mandibular lesions was satisfactory after two months

Browns tumour (11). Thus it is evident from the literature that most of the reported cases of multifocal central giant cell granuloma are syndromic and very few are non-syndromic .However in the present case reported the enzyme levels were within the normal limits excluding the possibility of hyperparathyroidism. Thus the report presented a clinically aggressive, multifocal and non-syndromic case of CGCG. Conventional management of CGCG involves curettage or resection whereas Non-surgical treatment includes systemic calcitonin therapy and intralesional injections with corticosteroids (2). In this case, surgical management was done by peripheral ostectomy, curettage and enucleation and the patient was recalled for follow-up.Post-operative healing of maxillary and mandibular lesions (Fig 6a&b) was satisfactory after two months. CGCG though controversial in its aetiology, is aggressive in nature and can show multifocal involvement.

Conclusion

With the help of radiographic examination the aggressive nature of CGCG should be determined and treated accordingly. In case of multifocal involvement the possibilities of abnorm alities such hyperparathyroidism should be ruled out by laboratory investigation. Any specific syndromes and systemic conditions should also be suspected.

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