Case Report

Appearances can be deceptive – Innocuous swelling on the gingiva masking an aggressive lesion within the maxilla

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Abstract

The central giant cell granuloma displays a varied biologic behaviour ranging from simple reactive lesions to aggressive neoplasms. The pathogenicity still remains enigmatic and needs to be differentiated from other giant cell containing lesions. Both maxilla and mandible are affected and 80% involve the region anterior to the first premolar region. CGCL arises centrally within bone, whereas PGCG is a gingival soft tissue lesion. Clinical and radiographic correlation is required to rule out a peripheral giant cell granuloma. The case described here was a rare presentation of a large epulis clinically with involvement of maxilla radiographically and was histologically diagnosed as a central giant cell lesion.

Keywords: Central giant cell granuloma, gingiva, maxilla

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INTRODUCTION

Jaffe in 1953 described central giant cell granuloma (CGCG) as an idiopathic non-neoplastic proliferative lesion,[1] and the World Health Organization defines it as an intraosseous lesion composed of cellular and dense connective tissues that contain multiple haemorrhagic foci, an aggregation of multinucleated giant cells, and occasional bone tissue trabeculae.[2] Although giant cell granuloma and its related lesions in the jaw are grouped under a single umbrella, they exhibit varied clinical behaviour ranging from simple reactive lesions to neoplasms sometimes even manifesting as aggressive malignant neoplasms.[3]

Although benign, they can be locally destructive and surgery is the most accepted method of treating the condition. The case reported herewith is a giant cell lesion that involved the left maxilla. This pedunculated swelling on the gingiva gave a deceptive appearance of an epulis, however proved to be a more aggressive lesion on exploration.

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CASE REPORT

A 53-year-old female patient reported to the Department of Oral and Maxillofacial Surgery with a painless swelling in upper right front region of jaw since 6 months, which was initially small in size and had gradually increased. The swelling caused difficulty while eating and speaking. No history of trauma, fever or any systemic illness was reported.

Extraoral examination revealed a firm swelling on the right side of the face with no palpable lymph nodes. Intraorally,

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a solitary, well-defined, firm, nontender pedunculated, roughly oval swelling measuring roughly 4 × 5 cm was seen on the labial gingiva of 12, 13, 14 and 15, which was reddish purple with dilated capillaries. The swelling extended from the maxillary vestibule to 2 mm below the occlusal surface and its lobulated surface showed indentations from lower teeth [Figure 1]. Grade 1 mobility was observed in 14, 15 and Grade 2 mobility and bleeding on probing was present in 12 and 13 regions.

Based on clinical findings, a provisional diagnosis of peripheral ossifying fibroma was made and pyogenic granuloma, peripheral giant cell granuloma and peripheral odontogenic tumour were considered in the differential diagnosis.

OPG and CBCT revealed an ill-defined mixed radiopaque-radiolucent lesion in the periapical area of 12, 13 and 14 extending from distal aspect of 12 to the distal aspect of 14. The internal structure was mixed radiolucent radiopaque with thick straight septa and thin delicate amorphous radiopacities at places intermixed with peripheral radiolucency [Figure 2]. Loss of lamina dura and spacing in 13 and 14 regions was noted. Mixed and amorphous extensions on buccal aspect of 14, 15 were noted. The buccal cortical plate was expanded in 13, 14 and 15 regions; however, at places, it was not perceptible. Thinning of palatal cortical plate was observed between 13 and 14 regions [Figure 3].

Considering the mixed radiopaque–radiolucent nature of the lesion, along with thick straight septa and thin delicate amorphous radiopacities causing displacement of 13, 14 and loss of lamina dura of involved teeth, a radiographic diagnosis of central-ossifying fibroma was given.

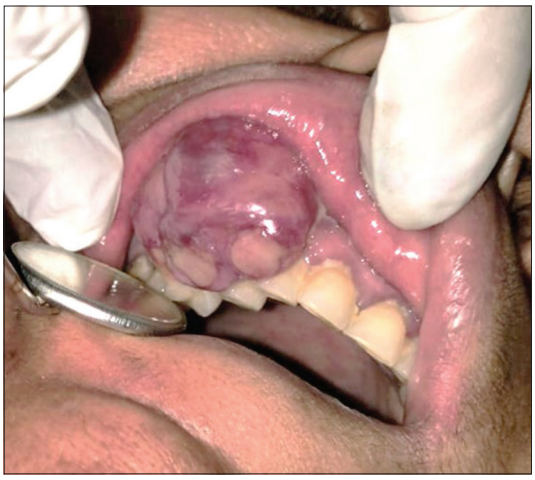

Figure 1: Well-defined reddish purple swelling seen on the labial gingiva of 12, 13, 14 and 15

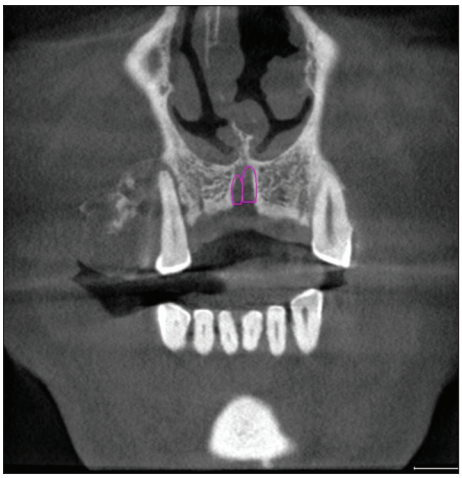

Figure 2: Anterior maxilla in the region of 12, 13 and 14 shows a mixed radiopaque–radiolucent lesion with thick straight septa and thin delicate amorphous radiopacities at places

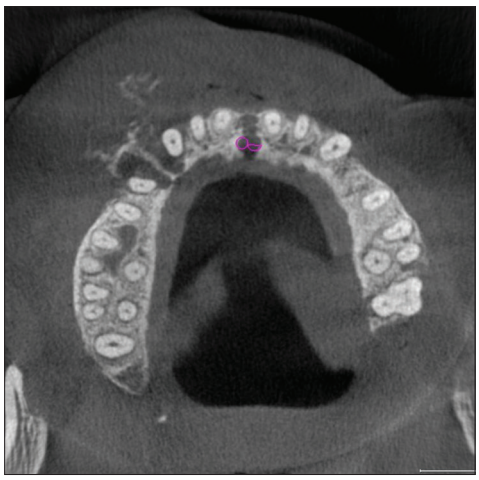

Figure 3: Expansion and loss of buccal cortical plate seen in 13, 14 and 15 regions with mixed and amorphous extensions on buccal aspect of 14 and 15 and thinning of palatal cortical plate between 13 and 14

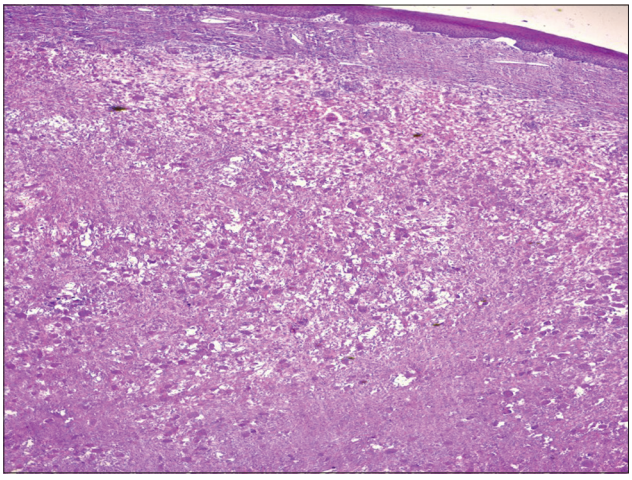

Figure 4: Peripheral lesion showing epithelium and underlying connective tissue, which is separated by a zone of dense bundles of collagen fibres. Evenly distributed giant cells are noted in the tissue. (H&E, 4X)

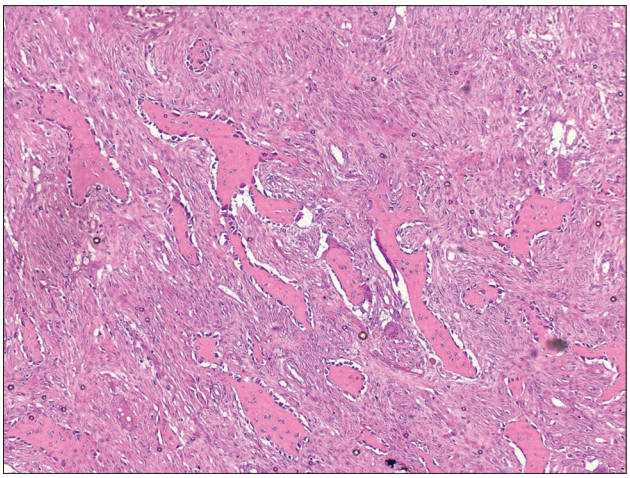

Figure 5: Central lesion decalcified bone shows bony trabeculae of varying sizes, which shows interspersed giant cells and osteoblastic rimming and osteocytes in lacunae. (H&E, 10X)

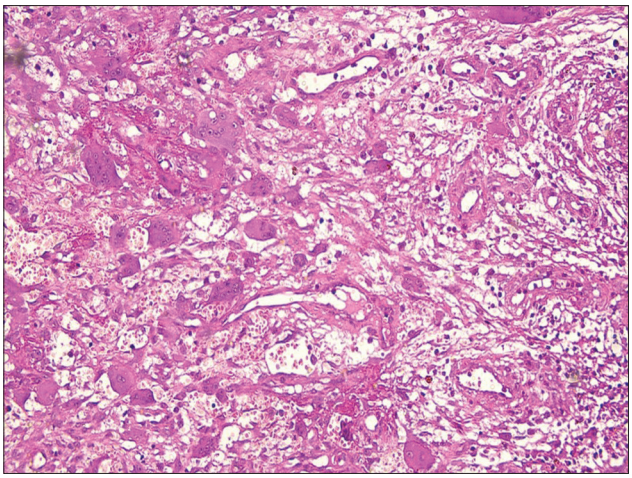

Figure 6: Multinucleated giant cells diffusely distributed within the background of plump and ovoid mononuclear cells. The giant cells contain 6-20 nuclei. Extravasated RBCs also noted. (H&E, 40X)

Differential diagnosis was given as central giant cell granuloma.

Since the patient was noncompliant and not willing to make multiple visits, an excisional biopsy was planned. Haematological investigations and blood chemistry revealed values within normal limits thereby ruling out hyperparathyroidism.

Under local anaesthesia, all aseptic precautions were followed. Wide local excision was followed by aggressive marginal treatment. Aggressive marginal treatment of bony margin was done with a surgical handpiece and round bur, after which hand instrumentation was done for removal of remaining soft tissue and hard tissue. Hard and soft tissue margins were cauterized, haemostasis was achieved, and buccal flap advancement and closure was performed. The excised specimen was submitted for histopathologic examination.

Histopathology of the peripheral lesion showed lesional tissue separated from superficial epithelium by a grenz zone [Figure 4]. Multinucleated giant cells containing 6-20 nuclei diffusely distributed within the background of plump and ovoid mononuclear cells was noted [Figure 6]. The connective tissue also showed blood vessels, extravasated RBCs and a mild inflammatory cell infiltrate. H&E decalcified stained section of the intraosseous lesion showed bony trabeculae of varying sizes showing osteoblastic rimming and osteocytes in lacunae and a similar histology as the peripheral tissue [Figure 5]. Thus, a diagnosis of giant cell lesion was made for both the central and peripheral excised tissue.

DISCUSSION

The WHO in 2017 defined CGCL (Central giant cell lesion) as an unencapsulated proliferation of mononuclear spindle-shaped and polygonal cells with osteoclast-type multinucleated giant cells in a vascular background.[4] The age range of CGCG varies from 2 to 81 years of age with a mean age of 23 years and 64% of lesions appear before the age of 30 years. A distinct female predilection has been noted as 63% of all patients were female.[5] In an extensive review by Chrcanovic, it was found that lesions were more prevalent in the mandible in comparison with the maxilla, but there was no clear prevalence concerning the different regions of the jaws.[6]

Although CGCL is frequently only a painless swelling, growth in some cases is so rapid and the mass can also rarely erode through bone particularly of the alveolar ridge to produce a soft tissue swelling. Extra-gnathic incidence is rare.[7] The uniqueness of this case was that it presented as a pedunculated mass masquerading as a peripheral lesion having perforated the buccal cortical plate, which raises queries as to could this be a collision of PGCG and CGCG. A rare case of PGCG reoccurring as CGCG has been reported with both the distinct lesions sharing the same anatomic location.[7] CGCL and PGCG are both composed of mononuclear stromal cells that mimic osteoclast precursors and multinucleated giant cells that mimic differentiated osteoclasts.[8] The expression of NFATcI expression is also increased in both of these lesions. However, CGCL arises centrally within bone, whereas PGCG is a gingival soft tissue lesion.[9]

Generally, smaller lesions of CGCL are totally radiolucent. These lesions may appear similar to a cyst. However, in a majority of the times CGCL appears as a mixed lesion. If the internal structure is less striated, it can be confused with cemento-ossifying fibroma. The lesion bony defects size and nature varies according to the aggressiveness of

the lesion. Chuong R *et al.* and Ficarra G *et al.* classified CGCG as aggressive or nonaggressive based on six criteria including pain, growth rate, swelling, tooth root resorption, cortical perforation and recurrences.[10,11] Aggressive lesions are characterised by pain, rapid growth and swelling, as well as cortical bone perforation, tooth displacement and root resorption and are prone to recurrence. Nonaggressive lesions, on the other hand, grow slowly, have few symptoms and may be devoid of associated features.

Shrestha S *et al.* presented a case series of seven cases of CGCL.[12] They thoroughly discussed clinical features, radiologic features of all the selected cases. Radiographically, most of the cases showed unevenly dense expansile mass causing bone destruction and cortical thinning. These findings are similar to that of our case. All their cases showed significant damage to the involved bone. Also, these cases were well-defined and multilocular. These findings are contrasting to that of our case. The lesion in our case was poorly defined and lacked the multilocular appearance. This disparity could be explained by the fact that in our case, maxilla was involved, and the radiographic appearance of CGCL in the maxilla is always diverse.

Lesions surgically treated with resection or curettage and additional treatments have practically no chance of recurrence.[13] However, in large and aggressive lesions when curettage is relatively mutilating, medical treatment with calcitonin and intralesional injection of steroids have been attempted.[14] Interferon-alpha-2A has been suggested as additional treatment of CGCG on the basis of its anti-angiogenic action.[15]

CONCLUSION

This report describes a rare case of giant cell lesion, which presented as an innocuous pedunculated swelling on the gingiva which masked an aggressive lesion intraosseously. However, radiographic examination and intrasurgical findings revealed the presence of an intraosseous lesion, which had a similar histopathology consistent with that of a central giant cell lesion. To conclude, the present case is an unusual presentation and requires multidisciplinary approach for proper management of the lesion.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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