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Case Report

Central Giant Cell Granuloma Associated with Cement- Ossifying Fibroma: the Histopathological Spectrum of a Hybrid Lesion- A Rare Case Report

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ABSTRACT

Swellings of the maxillo-mandibular region are a common occurrence in the oral cavity. Even though a majority of these manifest as a single pathology arising out of a single etiology, there are some instances, when patient present with a single lesion showing features of different pathologies, referred to as hybrid lesions. Hybrid lesions comprising central giant cell granuloma with fibro- osseous components are rare. We report a case of a hybrid lesion in a 56-year-old male, who presented with a swelling in the left side of mandible extending from incisor to first molar. Orthopantamogram reveals a well-defined multilocular radiolucent lesion of the body of the mandible with scattered radio-opacity that pointed to the diagnosis of odontogenic tumor. Incisional biopsy was performed, and histopathological examination showed features of both central giant cell granuloma and cement-ossifying fibroma, a hybrid lesion.

Key words: Hybrid lesions, Central giant cell granuloma (CGCG), Cemento-ossifying fibroma (COF)

INTRODUCTION

Lesions consisting of an association of characteristics from different pathologies are considered as hybrid lesions. Hybrid involving central giant lesions granulomas (CGCG) and fibro-osseous components are very rare in the jaws, with only seven cases reported in the literature till date. [1] CGCG is defined by the World Health Organization (WHO) intraosseous lesion consisting of cellular fibrous tissue with multiple foci of haemorrhage, aggregation of multinucleated giant cells and, occasionally, immature bone trabeculae. [2] Radiographically, it presents as multilocular radiolucent areas, usually in

the mandible. [3] Females under 40 years old are more commonly affected. Fibro-osseous lesions of the jaws are a heterogeneous group of lesions characterized by the replacement normal of bone bv fibrovascular tissue containing formed mineralized material. They present cellular variability and the amount and content of the mineralized material is also variable. We report a case of CGCG associated with cemento- ossifying fibroma and discuss the clinical, radiological and histological features of this hybrid lesion. [1-

CASE REPORT

A 56 year old male presented at the department of oral medicine at our tertiary institute with chief complains of swelling on the left side of mandible since last 6 months. The swelling was small, slow-growing and non-tender.

Extra-oral examination revealed presence of swelling on the left side of mandible extending from angle of mandible to angle of mouth. The swelling was nontender, hard on palpation and was approximately 3cms in diameter. The overlying skin was normal and submandibular lymph nodes of the left side were palpable.

Intra-orally, the swelling was seen on the floor of the mouth extending from incisors to first molar. (Fig 1) The swelling was firm in consistency and the overlying mucosa was normal. The radiograph revealed a well defined multilocular radiolucent lesion of the body of the mandible with scattered radio-opacity, extending from incisors to first molar. (Fig 2) A provisional diagnosis of odontogenic tumor was made.



Fig 1. Shows swelling (intraorally) on the floor of the mouth, extending from left mandibular central incisor to first molar tooth.



Fig 2. OPG revealing a well- defined multilocular radiolucent lesion of the mandible with scattered radio- opacity, extending from left mandibular central incisor to second molar area, involving the roots of left mandibular canine to first molar tooth of .

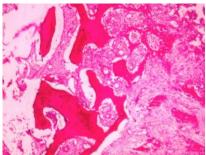


Fig 3. Showing curvilinear bony trabeculae in the central giant cell granuloma (CGCG) area of the lesion (H & E, 40X).

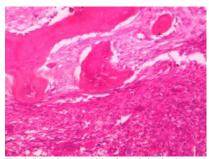


Fig 4. Showing osteoblastic activity in the cement-ossifying fibroma (COF) area of the lesion. (H & E. 40X).

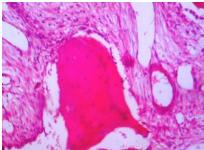


Fig 5. Showing areas of metaplastic bone formation in CGCG part of the lesion (H & E.40X)

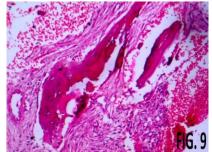


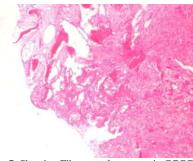
Fig 6. Showing areas of woven bone formation in COF part of the lesion (H & E, 40X)

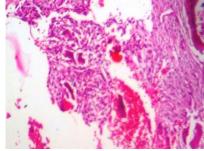
The lesion was surgically incised and examined. Macroscopically, the specimen consisted of two soft tissue bits, brownish yellow in color, irregular surface and soft to firm in consistency. The dimensions include $3x \ 1x \ 1 \ mm^3$ for specimen A and $5x \ 2x \ 1mm^3$ for other. Microscopically, sections

showed decalcified tissue with interconnecting bony trabeculae displaying a curvilinear pattern (Fig 3), metaplastic/woven bone formation (Fig 5,6), osteoblastic activity (Fig 4), fibrous and loose well-vascularized connective tissue stroma (Fig 7), fibroblasts with plump

nuclei, multinucleated giant cells (with 4- 10 nuclei/ giant cells), inflammatory cell infiltrate are seen. Other areas presented haemorrhagic foci (Fig 8). The transition zone that separates areas of CGCG from

COF was also seen in our case (Fig 9). A final diagnosis of cement- ossifying fibroma with central giant cell granuloma - Hybrid tumor was made on histopathological examination (HPE).





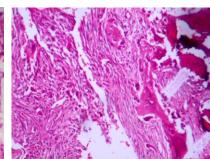


Fig 7. Showing Fibrovascular stroma in CGCG Fig 8. Showing hemorrhagic areas in the area of lesion (H & E, 40X)

connective tissue stroma of COF part of lesion (H & E, 40X)

Fig 9. Transition zone showing hybrid of CGCG and COF (H & E, 40X)

DISCUSSION

CGCG is a relatively uncommon lesion. The association of a CGCG with a fibro-osseous lesion affecting maxillamandibular bones is a very rare condition, with only a few cases described in the literature. Hybrid lesions of CGCG and COF have been reported in the 3-84 years range, with a mean age of 31.9 years and with a higher female predilection. The lesion has predilection for the mandibular posterior aspect in the premolar and molar area, presenting as slow- growing, hard, and nontender swelling. [4] Radiologically, this hybrid lesion presents as a well-defined multi or unilocular radiolucency except in one case in which complete radio opacity was seen. ^[5] The location and radiological appearance of our case correlates with the existing literature.

Histologic features such as curvilinear trabeculae, metaplastic or woven formation, bone osteoblastic activity. fibrovascular connective tissue, fibroblasts with plump nuclei, multinucleated giant cells, hemorrhagic areas and inflammatory cells seen in our case are in accordance with Crusoe- Rebello et al [6] who reported a case of hybrid lesion in 38 year old female

patient who presented with a swelling in the left mandibular parasymphysial region. Radiological features suggested a mixed type of lesion. Histological finding revealed interconnected bony trabeculae displaying a pattern. Some areas showed curvy activity, osteoblastic and also frequently, multinucleated giant cells are seen. Fibrous and loose well-vascularized connective tissue was also observed. Other areas presented haemorrhagic foci and accumulation of multinucleated giant cells of various shapes and sizes. [7,8]

Similar histological findings were also observed by Farzaneh Ah et al, [9] Shetty K et al, [10] Kaplan I et al [8] and Penfold CN et al, [7] except the feature of osteoblastic rimming which was only observed in the case report of Crusoe-Rebello et al and presence of a characteristic transitional zone that separates areas of CGCG from COF, was observed in our case.

Farzaneh et al [9] and Penfold et al [7] argue that the occurrence of giant cells in association with fibroosseous conditions may represent a reaction that stimulates modifications in the stroma of the original tumour. Osteoblasts may activate osteoclasttype giant cells through paracrine mechanisms. ^[7,9] The present case may also be related to such a phenomenon. Histologically, COF may present giant cells, which represent osteoclasts. In hybrid lesions, however, giant cells appear scattered in fibrovascular tissue. These features can be observed in our case.

CONCLUSION

The paucity of information on hybrid lesions makes it tough for the surgeons to reach a definitive diagnosis and plan appropriate treatment. Further, hybrid lesions have a variable biological behavior. Our case thus adds relevant information on the clinic- radiologic- pathologic spectrum of hybrid lesions

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