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Case report

Combined lesion of central giant cell granuloma and ossifying fibroma: A case report of a rare event in oral cavity

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ABSTRACT

Introduction and importance: Giant cell granulomas (GCG) and ossifying fibroma (OF) of the jaw are benign reactive lesions. GCG characterized by the presence of abundant multinucleated giant cells in a cellular stroma. On the other hand, the characteristic feature of OF is benign connective tissue replaces the normal bone. Combination of these two lesions, GCG with OF, in the jaw is extremely rare.

Case presentation: A 35-year-old woman presented with complains of right jaw swelling with no history of previous disease or lesion and no family history of such lesions. By physical examination, a painful swelling in the right jaw was observed. A computed tomography scan of the facial bone showed a large, expansible, lytic lesion with narrow zone of transition and internal septations, involving the right side of the maxilla and floor of the right maxillary sinus. Histopathologic examination revealed a lesion with combined features of OF and GCG.

Clinical discussion: GCG and OF are two of the most frequent oral lesions. Presence of both tumors in one patient as a combined lesion is highly unusual, with only a few reported cases in the literature. On clinical and radiologic examination, diagnosing such a combined lesion is not possible, however, such lesions can be easily diagnosed by microscopic examination, indicating the importance of pathologic examination.

Conclusion: Occurrence of combined OF and GCG is a rare event. Proper histopathologic evaluation can contribute to accurate diagnosis and better management of such lesions. Confirmative diagnosis of such lesions by radiology alone is not possible.

1. Introduction

Giant cell granuloma (GCG) of the jaws is a benign reactive lesion with no exact cause and pathogenesis [1]. Two forms of GCTs of jaws are known: central giant cell granuloma (CGCG) and peripheral giant cell granuloma (PGCG). Clinically and radiologically their difference is based on their location, as CGCG located within bone and PGCG occurs in the gingiva or edentulous alveolar process [2].

Ossifying fibroma (OF) is another benign fibro-osseous lesion of jaw, which is non-odontogenic lesion with unknown etiology. In this lesion the benign connective tissue replaces the normal bone, so, it characterized by connective tissue matrix having varying amounts of mineralized material that can be bone and/or cementum [3].

CGCG is a non-neoplastic, slow-growing lesion which grows inside a bone with locally aggressive and variable clinical behavior. It commonly occurs in female and constitutes less than 7% of all benign jaw lesions [4]. The characteristic morphological feature of both these lesions is the presence of abundant multinucleated giant cells in cellular stromal cells with a fibrous connective tissue stroma. Although most CGCGs are asymptomatic, aggressive form of CGCG can also exist, characterized by pain, cortical perforation, root resorption and tendency to recur after treatment [1].

CGCG and OF are somehow common lesions of the jaw and oral cavity but combination of these two lesions is extremely rare to occur [5]. Here we present a rare case of combined lesion of CGCG with OF involving the right jaw. Our work has been reported in concordance with the SCARE criteria [6].

Abbreviations: CGCG, central giant cell granuloma; PGCG, peripheral giant cell granuloma; OF, ossifying fibroma; H&E, hematoxylin & eosin.

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2. Case presentation

A 35-year-old married, rural resident woman and mother of five children presented with complaints of right jaw swelling of one-year duration. The patient did not narrate history of previous disease or lesion, family history of such lesion or tumor and drug history. By physical examination, a swelling in the right lower jaw was observed which had pain with touching. No other abnormality was noted. Computed tomography (CT) scan revealed a large, expansible, lytic lesion with narrow zone of transition and internal septations involving the right side of the maxilla and floor of the right maxillary sinus. There was no chondroid matrix within the lesion. No soft tissue component was noted. Similarly, no erosion of the adjacent teeth or pathological fracture was noted. The features were in keeping with benign lesion with the differential diagnosis of aneurysmal bone cyst, brown tumor, fibrous dysplasia, ossifying fibroma while the CGCG was suggested as a less likely possible lesion (Fig. 1).

Patient underwent excisional biopsy procedure under general anesthesia and by a maxillofacial surgeon with two decades experience of surgery in a governmental hospital. Post operatively the recovery was uneventful and the patient discharged after four days of stay in the recovery ward. The biopsy specimen was received at the Department of Pathology and Clinical Laboratory, French Medical Institute for Mothers and Children (FMIC). Gross examination revealed a gray-dark irregular tissue, measuring 6 × 4 cm with firm texture on cut section. The tissue was submitted in multiple paraffin embedded blocks and processed. Microscopic examination revealed a lesion consisting of fibro-osseous tissue having anastomosing and irregular trabeculae of bone with osteoblastic rimming, intervening fibro-collagenous stroma and multinucleated giant cells (Fig. 2A–D). Based on above histologic description the diagnosis of combined lesion of CGCG with OF was confirmed. Patient was followed up for a period of six months with no complications/recurrence, whatsoever during this time.

3. Discussion

Fibro-osseous lesion is a broad term that refers to a variety of different bone related lesions in which bone is replaced by fibrous connective tissue. This entity comprised of OF, fibrous dysplasia, and cemento-osseous dysplasia [7]. CGCG occur more commonly in female than males and the mostly affecting individuals aged between 10 and 25 years old. This lesion is exclusive to the bones of jaw and oral cavity. Mandibular bone is more frequently affected than maxillary bone. The first case of CGCG was published in 1953 [8]. In 1987, Waldron and El-Mofty, for the first time, described and introduced the term "hybrid lesions" when working on cases of ameloblastoma [9]. By definition, hybrid lesions are those lesions which comprises of main histopathologic components of two or more different lesions [10]. The occurrence of CGCG associated with fibro-osseous lesions is very rare. The so far reported hybrid lesions associated with CGCG were OF, cemento-osseous dysplasia and fibrous dysplasia [11].

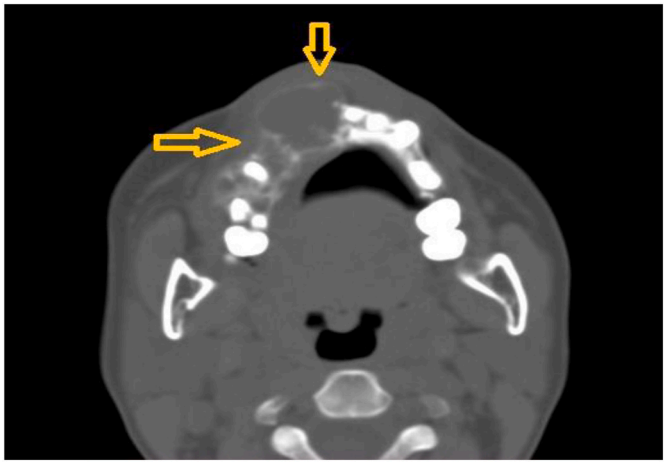

Fig. 1. CT scan shows a large, expansible, lytic lesion with narrow zone of transition and internal septations in maxilla bone and floor of the right maxillary sinus.

The histologic definition of CGCG by WHO is; an intraosseous lesion comprising of cellular fibrous tissue which includes multiple hemorrhagic foci, numerous multinucleated giant cells, and very few fragments of woven bony trabeculae [12] Previously, due to its locally destructive nature, it was called reparative GCG. However, now the lesion is believed to be initially destructive, so the term "reparative giant cell granuloma" is now avoided [12]. CGCG is further divided into aggressive and non-aggressive groups. Pain, paresthesia, resorption of the tooth root, rapid growth, cortical perforation, and high recurrence rate after curettage are the characteristics which constitute the aggressiveness of the lesion [13]. OF histologically, shows replacement of normal bones with fibrous tissue, and morphologically shows dense cellular stroma of fibrous tissue with intervening irregular bony trabeculae. The bony trabeculae have osteoblastic rimming at the periphery and this histologic characteristic can distinguish the OF from fibrous dysplasia, which is a close mimicker of it [14].

Presence of both of CGCG and OF components in single tumor is a very rare occurrence. Hybrid or combined lesion is defined as presence of large areas of both lesions microscopically in a single lesion, which is separated from each other [15]. For ethiopathogenesis of CGCG and OF, it has been hypothesized that the primary lesion in CGCG-OF is OF. The cytokine released from mesenchymal spindle cells of the tumor by unknown cause or triggering factors, which causes differentiation toward osteoclast/giant cells [16].

4. Conclusion

Combined lesion of CGCG and OF is a rarely reported lesion in the jaw and oral cavity. As it is composed of two different lesions, therefore the accurate diagnosis is made through histopathologic evaluations by correlation of radiological findings.

Patient perspective

The husband of the patient stated that it would be useful to publish and share this case with other healthcare workers, to enable better understanding and correct diagnosis of such cases (combined lesion), that would cause a specific treatment for two lesions instead of one lesion.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

Not commissioned; externally peer-reviewed.

Ethical approval

Since this was a retrospective observational study and did not involve actual patients or patient's images, ethical approval was not sought for this study.

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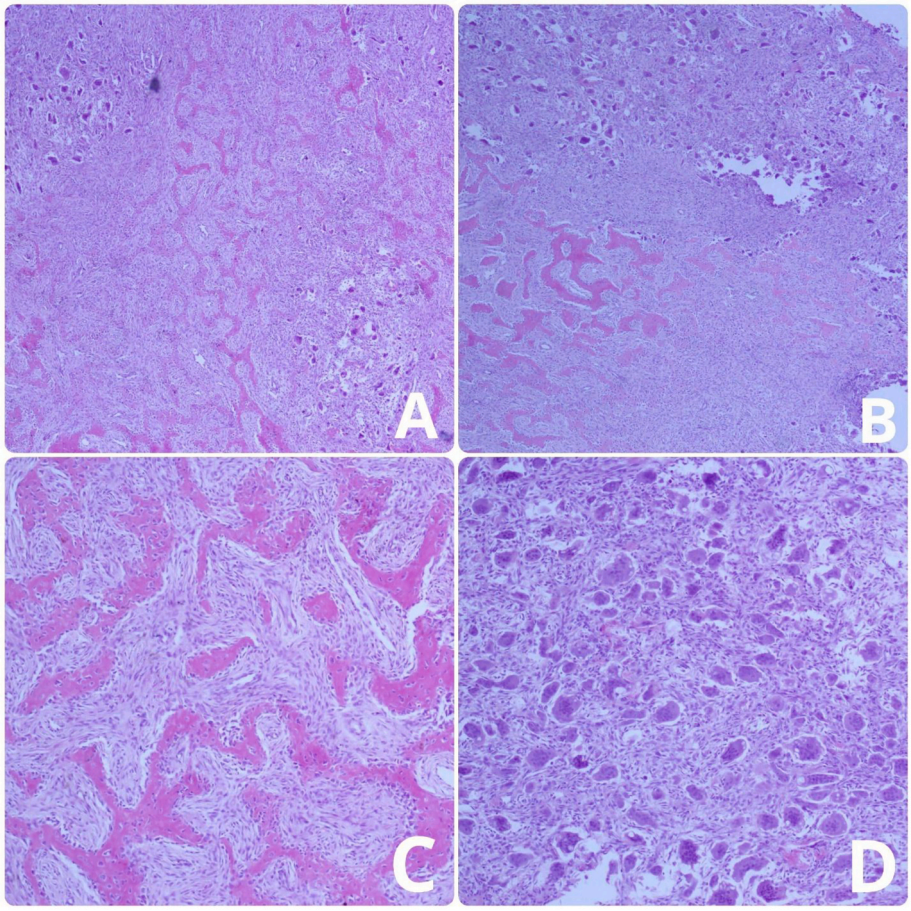

Fig. 2. Microscopic low power view shows areas of fibro-osseous lesion in center and giant cell granuloma in periphery (A, B). High power view of fibro-osseous components of the tumor shows fibrous tissue with interspersing bone trabeculae with osteoblasts rimming (C). High power view of giant cell granuloma comprising multinucleated giant cells in a fibrous stroma.

Guarantor

Jamshid Abdul-Ghafar, MD, PhD.

Research registration number

None.

CRediT authorship contribution statement

RS convinced the idea. JAG, and RS diagnosis the case. RS was involved in literature review and drafted the manuscript. RS and MAA helped to collect clinical and follow-up data of the cases. FRF provided radiological findings and report. AMH and JAG participated in reviewing the drafted manuscript. JAG participated with the corresponding, editing the drafted manuscript as per journal policy, and submission of the article. All authors read and approved the final manuscript.

Declaration of competing interest

It is declared that all authors have no conflict of interest.

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