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Case report

Hybrid peripheral giant cell granuloma and peripheral ossifying fibroma lesion: A rare case report and review of the literature

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ABSTRACT

Hybrid lesions containing a similar component to the giant cell granuloma (GCG) associated with the central odontogenic fibroma (COF) and fibro-osseous lesions have been reported in the literature since 1992; in contrast, peripheral cases are extremely rare. Thus, the purpose of this manuscript is to report a case of a hybrid lesion constituted by peripheral giant cell granuloma (PGCG) and peripheral ossifying fibroma (POsF), emphasizing the clinical, radiographic, histopathological, therapeutic and prognostic aspects of such lesion. In that way, our case presentation comprised a 31-year-old feodermic female patient that presented a large asymptomatic exophytic lesion in the left mandibular alveolar mucosa, radiographically showing an irregular radiopaque area within the soft tissue lesion. An excisional biopsy was performed under local anesthesia. Histopathological analysis displayed a fragment of oral mucosa lined by parakeratinized and atrophic stratified squamous epithelium with ulcerated areas; the lamina propria exhibited dense connective tissue containing numerous inflammatory multinucleated giant cells (MGCs) permeated by fusiform cells, blood vessels, hemorrhage and hemosiderosis; in addition, deposition of trabecular bone matrix intermixed by MGCs, proliferation of fibroblasts and collagen fibers was observed. When this association between PGCG and POsF occurs, the possibility of a PGCG exhibiting bone formation should be rule out. Therefore, the hybrid PGCG and POsF may represent a diagnostic challenge to pathologists, and more cases with histopathologic features similar to our case should be reported in order to elucidate the questionable histogenesis of this lesion.

1. Introduction

A hybrid lesion consists of the association of characteristic features from different pathologies into one single lesion [1]. Since 1992, hybrid lesions containing a similar component to the giant cell granuloma (GCG) [2] mainly associated with the central odontogenic fibroma (COF) [2–7], and more rarely with the central ossifying fibroma (COsF) [1,8,9], trabecular variant of the juvenile ossifying fibroma (TJOsF) [10,11], fibrous dysplasia (FD) [12–14] and cemento-osseous dysplasia (COD) [15] have been reported. In constrast, hybrid lesions allocated peripherally and involving the above mentioned pathologies have not yet been addressed, except for a recent report in which predominates the doubtful diagnosis between a true hybrid lesion formed by the combination of peripheral giant cell granuloma (PGCG) and peripheral ossifying fibroma (POsF), and a lesion consisting of PGCG with an extensive bone formation [16].

PGCG and central giant cell granuloma (CGCG) are benign disorders, which arise either peripherally in the periodontal ligament and alveolar mucoperiosteum, or centrally inside the bone [17]. Both are histopathologically similar, being characterized basically by the presence of numerous inflammatory multinucleated giant cells (MGCs) within a stroma containing ovoid or fusiform mesenchymal cells [18] However, PGCG and CGCG have distinct

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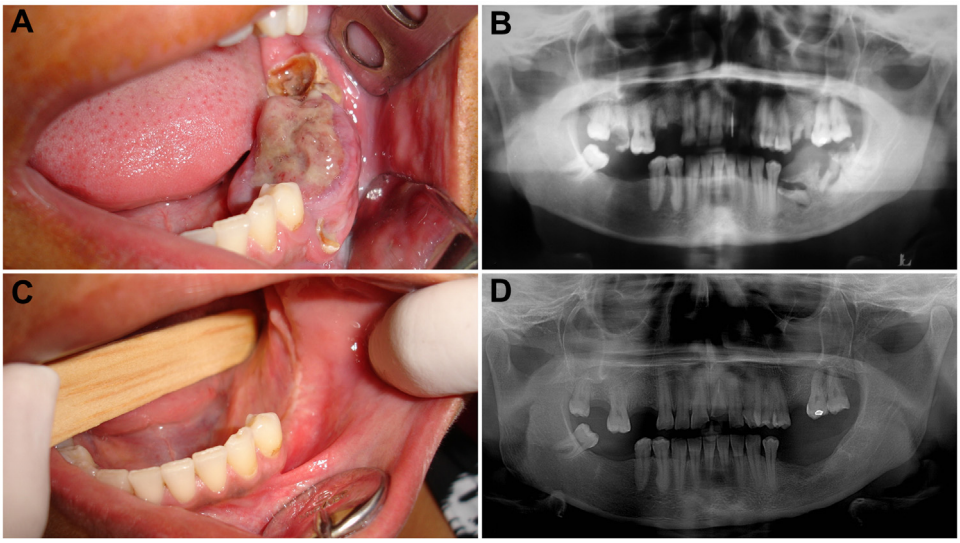

Fig. 1. Clinical and radiographic aspects, and follow-up. (A) Sessile nodule of firm consistency, purplish, presenting superficial ulceration covered by pseudomembrane, extending from the distal face of the lower left first premolar to the mesial face of the lower left third molar, and measuring approximately 2.5 cm in its greatest extension; (B) Initial radiograph: irregular radiopaque area within the soft tissue lesion and residual roots of the lower left second premolar and lower left second molar; (C) Complete healing of the operated area; (D) Final radiograph: 9 months of follow-up, observing no signs of recurrence.

prognoses; PGCG is a reactive lesion that may arise in response to a local irritating factor, and demonstrates low recurrence after eliminating the etiological factor, while CGCG has an uncertain pathogenesis and may be divided into non-aggressive lesions (asymptomatic with slow growth and low recurrence) and aggressive lesions (painful with fast growth and tendency to recurrence, resembling a benign tumor) [9,17,19].

POsF, as well as PGCG, is also considered a reactive lesion that originates from the pluripotent cells of the periodontal ligaments [16]; this pathology is characterized microscopically by a proliferation of fibroblasts associated with the formation of mineralized material [20]. On the other hand, COsF is a benign neoplasm that consists of fibrous tissue with different degrees of cellularity, also containing variable amounts of mineralized material [9]. Focal clusters of MGCs can be found within POsF and COsF, and represent osteoclasts associated with eventual mineralization [1,9]. In hybrid lesions, however, MGCs appear scattered within the fibrovascular tissue [1,9].

In that way, the purpose of this manuscript is to report a rare case of a hybrid lesion constituted by PGCG and POsF in the mandibular alveolar mucosa, emphasizing the clinical, radiographic, histopathological, therapeutic and prognostic aspects of such lesion. In addition, a review of the related literature in order to identify and characterize all reported cases of oral hybrid lesions containing a similar component to the GCG will be presented and analyzed.

2. Case report

A 31-year-old feodermic female patient presented an asymptomatic exophytic lesion in the left mandibular alveolar mucosa that was reported to have 8 years of duration and a significant growth meanwhile. The extraoral physical examination did not reveal any significant change, and intraorally there was a sessile nodule of firm consistency, purplish, presenting superficial ulceration covered by pseudomembrane, extending from the distal face of the lower left first premolar to the mesial face of the lower left third molar, and measuring approximately 2.5 cm in its greatest extension (Fig. 1A). A panoramic radiography showed an irregular radiopaque area within the soft tissue lesion, and also the presence of residual roots of the lower left second premolar and lower left second molar underlying the lesion (Fig. 1B). Based on these clinical and radiographic aspects, the diagnostic hypothesis was of POsF. An excisional biopsy and extraction of the residual roots and lower left third molar were then performed under local anesthesia. During the surgical excision the lesion was easily detached from the alveolar bone crest at a superficial level; the lesion did not show an intrusive behavior towards mandible's inner trabecular bone whatsoever. So, we can affirm that the lesion presented a peripheral location. The specimen was sent for histopathological analysis. Microscopically, the sections stained with hematoxylin and eosin revealed a fragment of oral mucosa lined by parakeratinized and atrophic stratified squamous epithelium with ulcerated areas covered by a pseudomembrane (fibrin and polymorphonuclear inflamm atory cells); the lamina propria showed dense connective tissue containing numerous MGCs permeated by fusiform cells randomly distributed, blood vessels, hemorrhage and hemosiderosis; in addition, deposition of trabecular bone matrix was observed (Fig. 2A-B), some already completely mineralized and having osteocytes inside lacunae, osteoblasts (Fig. 2C) and intermixed by MGCs, proliferation of fibroblasts and collagen fibers (Fig. 2D–F). Thus, the diagnosis of hybrid PGCG and POsF lesion was established. One month after surgery, there was complete healing of the operated area, and 9 months of follow-up indicated no signs of recurrence (Fig. 1C and D).

3. Discussion

A literature review of all cases of hybrid lesions containing a similar component to the GCG was performed using PubMed/Medline database. The keywords used in the search were: hybrid lesion AND oral; giant cell granuloma; central odontogenic fibroma; and fibro-osseous lesions. Moreover, the following inclusion criteria were adopted: (1) English language; (2) full-text available; (3) histopathological diagnosis; and (4) oral location. In that way, the initial literature search identified 205 potential articles. After full-text review, 16 articles were qualified for further analysis, meeting then a total of 37 cases of hybrid lesions containing a similar component to the GCG in addition to the present case report. However, considering the peripheral location, as seen in our case, only 1

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