CASE REPORT
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Peripheral Dentinogenic Ghost Cell Tumor of the Alveolar Mucosa: A Rare Case Report

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Abstract

Objectives: Peripheral (extra-osseous) dentinogenic ghost cell tumors are an extremely rare type of odontogenic lesions. They show a slight male predilection and most commonly occur in the anterior gingiva of the mandible. The peak incidence is above 50 years of age, and unlike the intraosseous type, they are generally nonaggressive. Diagnosing intra-oral lesions can be challenging, and less common lesions are often overlooked. **Case:** This report aimed to describe a peripheral dentinogenic ghost cell tumor as a painless, pedunculated mass in a 62-year-old male. **Conclusion:** Understanding this unusual entity can facilitate a proper diagnosis and enhance our comprehension of its biological behavior.

Keywords: Mandible; Jaw; Odontogenic Tumor; Dentinogenic Ghost Cell Tumor

Introduction

Dentinogenic ghost cell tumor (DGCT) represents an uncommon neoplastic variant of the calcifying odontogenic cyst (COC).[1] It is a benign odontogenic tumor identified microscopically by abnormal keratinization of the epithelium, the presence of ghost cells, and dentinoid material.[2] DGCT is classified into peripheral and central types.[1] The formation of ghost cells results from an abnormal process of terminal differentiation and keratinization of neoplastic cells, which involves apoptosis.[2] The peripheral or extraosseous type is much less common than the central type and typically presents as a painless swelling or nodule on the gingiva.[1] Peripheral DGCT (PDGCT) arises from remnants of the dental lamina or the surface epithelium of the alveolar mucosa, with several studies highlighting the continuity between the basal layer and the neoplastic parenchyma. Evidence suggests that the activation of the Wnt/β-catenin/TCF-Lef signaling pathway, through β-catenin accumulation caused by mutations in the CTNNB1 gene, is linked to the development of ghost cell lesions.[3] The recommended treatment is conservative surgical excision with underlying bone curettage.[1, 3] This case report details a 62-year-old male with PDGCT of the anterior mandibular alveolar mucosa. Gaining a deeper understanding of this rare condition can aid in accurate diagnosis and enhance knowledge of its biological behavior.

Case Report

A 62-year-old male patient was sent for consultation to an oral and maxillofacial pathology center in Tehran, Iran, for assessment of a painless, pedunculated, pink, exophytic mass with an ulcerated surface in the anterior mandibular alveolar mucosa of uncertain duration. The patient was edentulous and had worn a complete denture for 22 years. He had no other medical conditions, and the extra-oral examination was normal, with no cervical lymphadenopathy. The mass was firm, measuring 2.5×1 cm, and a panoramic radiograph revealed no signs of an intraosseous lesion, except for minor erosion of the underlying bone. The provisional differential diagnoses included reactive lesions such as peripheral giant cell granuloma (PGCG) and healed pyogenic granuloma (PG). An excisional biopsy was conducted with the use of local anesthesia, and the specimen exhibited a solid, white, homogeneous cut surface. It was processed for histopathological study using Hematoxylin and Eosin staining. Low-power microscopic examination showed a peripheral odontogenic tumor covered by oral mucosa, with islands of ameloblastic epithelium arranged in a follicular pattern within the fibrous connective tissue (Figure 1). Examination at higher magnification showed neoplastic islands made up of a single layer of tall columnar ameloblast-like cells encircling a core of loosely arranged angular cells (stellate reticulum), which contained numerous “ghost cells.” Calcification within the ghost cells, several giant cells, and dysplastic dentin (dentinoid) were

observed (Figures 2, 3). These histopathologic findings confirmed the diagnosis of PDGCT. However, the patient failed to attend the follow-up appointments.

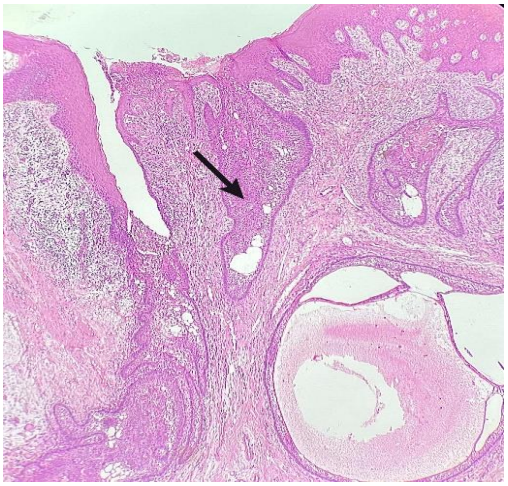

Figure 1: Histopathologic section shows islands of ameloblastic epithelium containing ghost cells filling the lamina propria. Connection of the islands with the basal layer of the surface epithelium is also seen (black arrow) 100X; H&E stain.

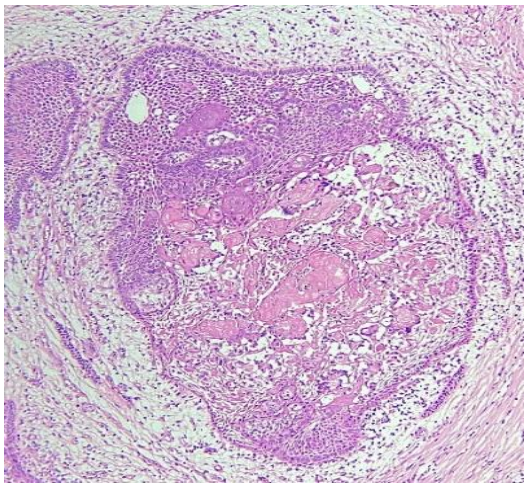

Figure 2: A large tumoral island displays palisaded peripheral cells surrounded by a core of loosely arranged angular cells and many eosinophilic ghost cells 400X; H&E stain.

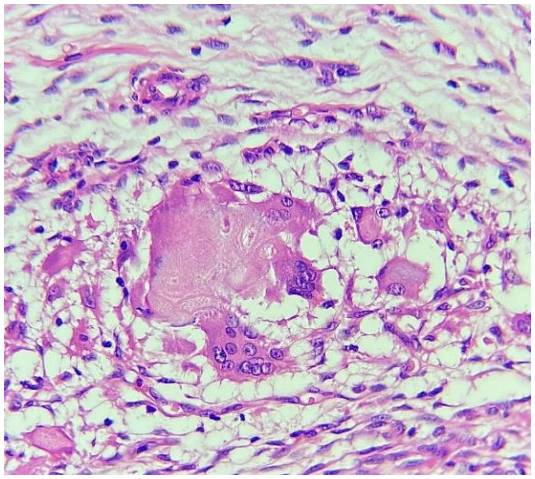

Figure 3: Histopathologic section shows a foreign body reaction by forming multinucleated giant cells around the ghost cells in the connective tissue 400X; H&E stain.

Discussion

DGCT is an uncommon benign tumor that involves both epithelial and mesenchymal tissues in odontogenesis. [2] The peripheral variant, PDGCT, is rarer and tends to exhibit less aggressive behavior compared to the central form.[1, 3] This tumor typically occurs in the anterior mandible of older males.[1, 3] However, de Arruda et al. [4] suggested that the peripheral variant is more frequently found in the anterior regions of both jaws. In this case, the patient, who was in his 7th decade of life, also had involvement of the anterior mandibular alveolar mucosa.

PDGCT typically appears as a firm, painless nodular mass, usually measuring up to 3.0 cm [3], and can mimic common oral reactive or inflammatory lesions, such as PGCG, PG, irritation fibroma, epulis, or parulis. [1, 3] In the present case, the lesion was excised with a provisional diagnosis of PGCG and PG. The most common radiographic finding is a mild "cup-shaped" erosion of the underlying cortical bone [3], and histopathological examination is required for a definitive diagnosis. [1]

PDGCT exhibits small and large islands of odontogenic epithelium within fibrous connective tissue. The epithelial islands are characterized by peripheral palisaded columnar cells and central stellate reticulum, resembling ameloblastoma. Ghost cell nests are present within the epithelium, and juxtaepithelial dentinoid material is typically observed. These microscopic features help distinguish PDGCT from peripheral ameloblastoma. [1-3, 5] It is important to note that other lesions, such as pilomatrixoma and craniopharyngioma, may also contain numerous ghost cells on microscopic examination. Additionally, scattered ghost cells can be found in other odontogenic lesions like ameloblastoma, ameloblastic fibro-odontoma and odontoma. Dentinoid material has been noted in, complex odontoma, adenomatoid odontogenic tumor, ameloblastic fibrodentinoma/fibroodontoma, odontogenic carcinomas, and sarcomas. The combination of these histopathological features contributes to the diagnosis of PDGCT [3], and this patient exhibited all the related microscopic characteristics. Ghost cells in contact with the connective tissue can provoke a foreign body reaction, leading to the formation of multinucleated foreign body-type giant cells, often accompanied by calcifications. [1] This feature was also observed in this case, though PDGCT typically follows a more indolent course. [3] Therefore, simple surgical excision along with underlying bone curettage is considered the preferred treatment, with a very low recurrence rate. [1, 3]

Conclusion

Given the rarity of PDGCT, this case report provides valuable clinical and histopathological information to aid in its diagnosis and treatment. The lesion exhibits unique microscopic features, highlighting the importance of a biopsy for an accurate diagnosis.

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All authors have read and agreed to the published version of the manuscript.

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Conflict of Interest: No conflicts of interest to declare.

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