



CASE REPORT

Central granular cell odontogenic tumor: The first reported case in Oriental people and literature review



Chien-Ta Chiang ^a, Kwang-Yu Hu ^b, Chien-Chen Tsai ^{a,*}

^a Department of Anatomical Pathology, Far Eastern Memorial Hospital, Taipei, Taiwan

^b Department of Oral Maxillofacial Surgery, Far Eastern Memorial Hospital, Taipei, Taiwan

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KEYWORDS

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The central granular cell odontogenic tumor (CGCOT) is a rare odontogenic neoplasm, usually occurring in the mandible of middle-aged women. Previous studies have reported only 34 cases, all of whom were white or black individuals. The present study reports an additional case of CGCOT, occurring in the posterior mandible of a 69-year-old Taiwanese man. To the authors' knowledge, this is the first reported case in Oriental people. The odontogenic epithelium exhibited strong positive immunoreactivity for pan-cytokeratin, and focal weak staining for bcl-2. The granular cells showed strong positivity for vimentin and α 1-antichymotrypsin, and focal weak staining for carcinoembryonic antigen (CEA), neuron-specific enolase (NSE), and CD68. These features indicated a mesenchymal origin and possible histiocytic lineage for the granular cells. This study also presents a literature review and describes immunohistochemical features of the tumor.

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Introduction

Central granular cell odontogenic tumor (CGCOT) is a rare odontogenic benign neoplasm comprising varying amounts of large eosinophilic granular cells and an apparently

inactive odontogenic epithelium. There have been 34 cases of CGCOT reported, using varied terminology.^{1–21} The recent classification of odontogenic tumors of the World Health Organization does not include CGCOT; however, many authors regard this type of tumor as a distinct entity, recently naming it CGCOT.^{13–18,20}

CGCOT tends to surface in the mandible, occurring especially in women older than 50 years. Previously reported cases all regarded white or black people. The present study reports the first case of CGCOT occurring in an Oriental person.

Conflicts of interest: The authors have no conflicts of interest relevant to this article.

* Corresponding author. No. 21, Section 2, Nanya South Road, Banciao District, New Taipei City 220, Taiwan.

E-mail address: zerosedi@hotmail.com (C.-C. Tsai).

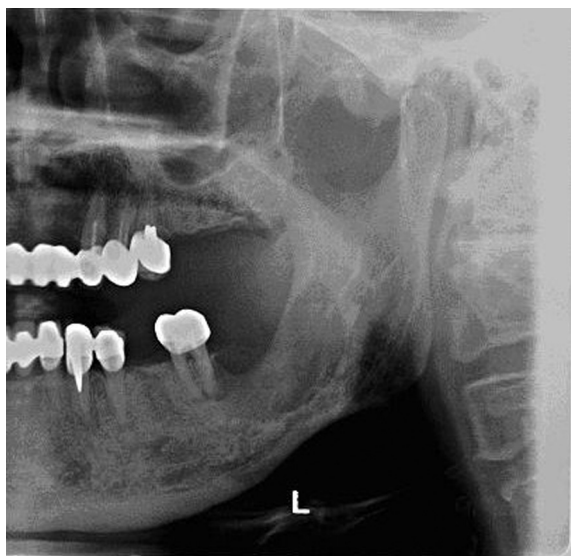


Figure 1 A well-demarcated radiolucent lesion noted in the left mandibular bone.

Case report

Clinical findings

A 69-year-old Taiwanese man attended the Oral Maxillofacial Surgery Clinic of the Far Eastern Memorial Hospital, New Taipei City, Taiwan, complaining of swelling of the left lower posterior gingiva for one month. He had a medical history of L-spine spondylosis and adenomatous polyp of the colon with high-grade dysplasia. Intraoral examination revealed a firm, asymptomatic swelling on the left side of the mandible. The overlying mucosa showed focal erythematous change with a smooth surface. The panoramic radiograph showed a well-demarcated radiolucent lesion measuring 4.0 cm in greatest dimension, centrally located over the ramus area of the left mandibular bone, with involvement of the apex of the second molar tooth and bulging into the retromolar pad (Fig. 1). After a clinical diagnosis of bone tumor, a surgeon performed tumor excision with extraction of the second molar tooth. During the

Table 1 Immunohistochemical findings.

Antibody	Cell immunoreactivity	
	Granular	Epithelial
Cytokeratin	–	+
Vimentin	+	–
Carcinoembryonic antigen	+/-	–
Neuron specific enolase	+/-	–
Bcl-2	–	+/-
α 1-antitrypsin	–	–
α 1-antichymotrypsin	+	–
CD68	+/-	–

+/- = focal weak staining.

operation, the surgeon noted lingual plate destruction. A rubber-like capsule surrounding the tumor was also found. The postoperative clinical diagnosis was ameloblastoma. No tumor recurrence had occurred 2 months after the operation.

Pathological findings

Gross pathology: The tumor arrived for pathological examination as 6 tissue fragments measuring up to $2.8 \times 0.9 \times 0.8$ cm, and fixed in 10% neutral buffered formalin. The tumor fragments were white to slightly yellow, and the cut surface was smooth, homogenous, and firm.

Histological and immunohistochemical analysis: Microscopic examination revealed a submucosal tumor comprising sheets or lobules of large polygonal granular cells (Fig. 2A). The nuclei were round to ovoid and slightly eccentric. The cytoplasm was uniformly finely granular and eosinophilic. The stroma contained foci of thin-walled vessels and fibrous-connective tissue. Focally myxoid and clear cell-like appearances were noted, suggesting a degenerative change. Inactive odontogenic epithelium comprising cords and nests of cuboidal to low columnar basal cells were dispersed between the granular cells. Epithelial cells containing a clear cytoplasm were

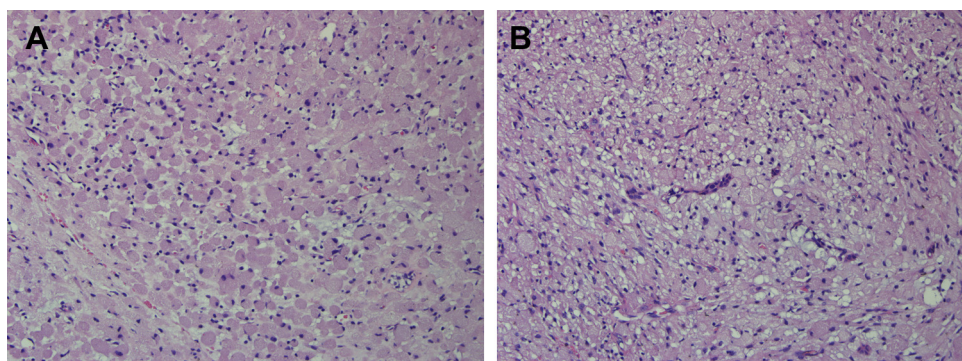


Figure 2 (A) Granular cells revealed eccentric nuclei and eosinophilic granular cytoplasm (hematoxylin and eosin stain, original magnification $200\times$). (B) Odontogenic epithelium with vacuolated changes (hematoxylin and eosin stain, original magnification $200\times$).

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