

**Case Report**

## Granular Cell Odontogenic Tumor, an Extremely Rare Case Report

Saede Atarbashi-Moghadam, DDS, Msc<sup>1</sup> - Homeira Saebnoori, DDS<sup>2</sup> - Nafiseh Shamloo, DDS, Msc<sup>3</sup>, Mehrdad Dehghanpour Barouj, DDS, Msc<sup>4</sup>, Sara Saedi, DDS<sup>5</sup>

<sup>1</sup> Dept. of Oral and Maxillofacial Pathology, School of Dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

<sup>2</sup> Postgraduate Student, Dept. of Oral Pathology, School of Dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

<sup>3</sup> Dept. of Oral and Maxillofacial Pathology, School of Dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

<sup>4</sup> Dept. of Oral and Maxillofacial Surgery, School of Dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

<sup>5</sup> Postgraduate Student, Dept. of Endodontic, School of Dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

### KEY WORDS

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### ABSTRACT

The granular cell odontogenic tumor is an extremely rare odontogenic neoplasm which about 38 cases has been reported with sufficient documentation in the literature. It has a prominent predilection to occur in the posterior of mandible of middle-aged women. Here, we report a case of mandibular granular cell odontogenic tumor in a 57-year-old female with chief complaint of swelling and tooth mobility. As rarity of these lesions, their clinical behavior and prognosis are not clear; hence, reporting more such cases may be beneficial to correct diagnosis and prevent unnecessary treatment.

**Corresponding Author:** Saebnoori H, Dept. of Oral and Maxillofacial Pathology, School of dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran. Tel: +98-2122403080 Email: saebnoori@yahoo.com

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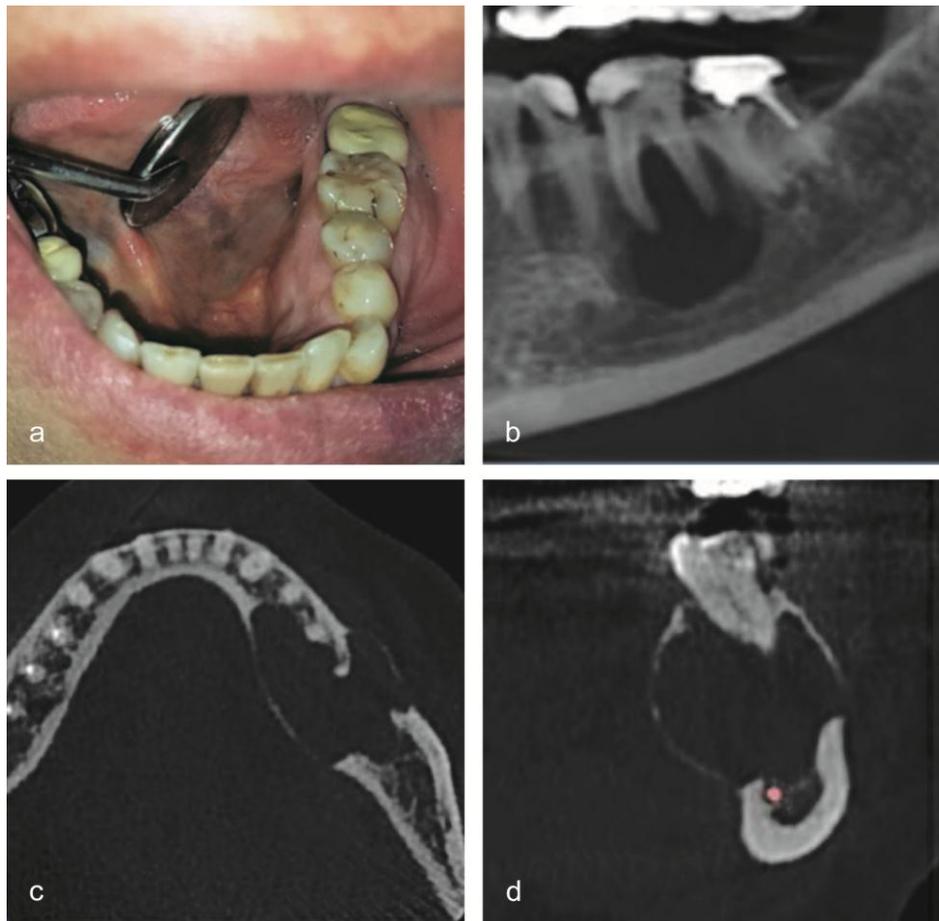
### Introduction

Granular cell odontogenic tumor (GCOT) is one of the most rare odontogenic neoplasms of jaws. This tumor had been nominated with so many different terms. Granular cell ameloblastic fibroma and granular cell odontogenic fibroma were other terms for this lesion [1]. However, this entity has not been included in the 2017 World Health Organization (WHO) classification of odontogenic tumors [2]. It has a striking tendency to occur in the posterior of mandible and middle-aged women [1]. GCOTs only arise in tooth bearing regions of the jaws and usually show a painless swelling. Radiographic features include a well-defined radiolucency or mixed radiolucent-radiopaque lesion [3]. Extrasosseous variant and granular cell odontogenic sarcoma have also been described [4-5]. Histopathologic examination shows sheets and lobules of large eosinophilic granular cells. Narrow cords or small islands of odontogenic epithelium are scattered among the granular cells. Cementum-like materials associated with the granular cells have also been reported. It seems that GCOT is completely benign and responds well to curettage [1]. The purpose of this case report is to describe a rare case of

mandibular GCOT affecting a 57-year-old female individual.

### Case Report

A 57-year-old woman was referred to the Department of Oral and Maxillofacial Surgery, Shahid Beheshti University of Medical Sciences (Iran, Tehran) for evaluation of painless swelling of the left posterior mandible and tooth mobility in that region for two weeks. The patient had no history of previous trauma or medical problems. Clinically, the general head and neck examination were normal. Intraoral examination showed a firm swelling extending from teeth no 34 to 36 with intact mucosa. The involved teeth were vital. The patient's oral hygiene was good. In the panoramic radiograph, a well-defined radiolucency in apical area of tooth no 34 -36 were present. Cone beam computed tomography (CBCT) demonstrated expansion of buccal and lingual cortical plates, which cause eggshell appearance and perforation in some areas (Figure 1). It appeared that mandibular canal was intact. Aspiration of the lesion was negative. Due to clinical, radiographic and aspiration findings, odontogenic tumor and central



**Figure 1a:** Photograph shows buccal and lingual expansion in left side of mandible with normal colored intact mucosa. **b:** CBCT reveals, a well- defined radiolucency in apical area of tooth no 34 -36. **c** and **d:** Axial and frontal view of the lesion shows expansion of buccal and lingual cortical plates which cause egg shell appearance and perforation in some areas.

giant cell granuloma were considered in the differential diagnosis. The patient underwent root canal therapy for 35, 36 and then the lesion completely excised under local anesthesia. On gross examination, the lesion consists of one piece of solid, creamy-white soft tissue with elastic consistency measuring 1.9×1×0.5 cm.

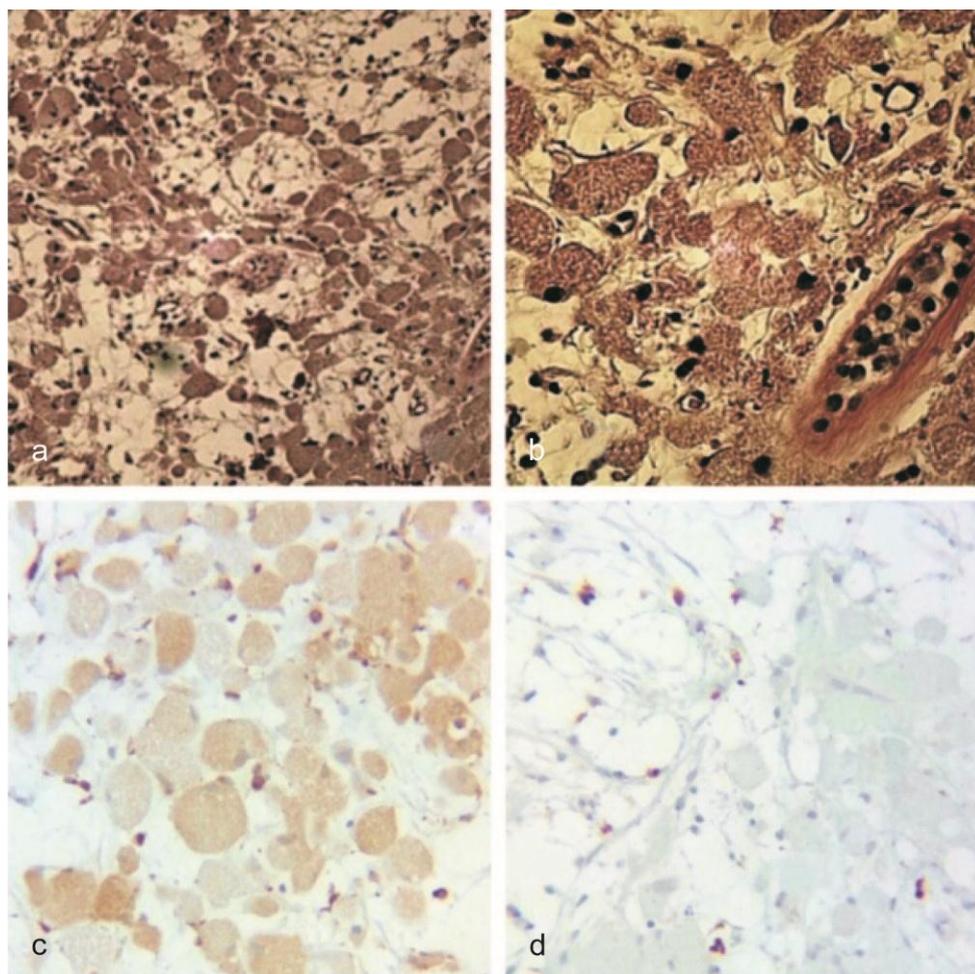
Histopathologic sections revealed a mesenchymal odontogenic tumor composed of sheets and lobules of large eosinophilic granular cells with scant fibrous to myxomatous stroma. Small odontogenic epithelial islands with juxtaepithelial hyalinization were also seen. The nature of the granular cells was determined through immunohistochemistry (IHC). The granular cells were diffusely positive for CD68 and negative for S-100 protein (Figure 2). According to aforementioned histopathologic, IHC and radiographic features, the diagnosis of central granular cell odontogenic tumor (CGCOT) was performed. Because it appears that CGCOT shows non-aggressive behavior, no additional treatment was done. About one month after surgery, the patient stated that

the teeth mobility was improved. There was no recurrence during a 12-month follow-up period. (Informed consent was obtained from the patient for publishing her clinical photography and radiography).

### Discussion

CGCOT is a rare neoplasm which had been designated with so many diverse terms [1]. Yin *et al.* [6] stated that lack of dental papilla-like stroma and inactive odontogenic epithelium with no proliferation or histodifferentiation is inconsistent with the name of “Granular cell ameloblastic fibroma”. In addition, the average age of patient’s with ameloblastic fibroma is 14.6 years compare to CGCOT with average age of 45.2 years [1]. Furthermore, Gardner [7] declared that this tumor never shows dense fibrous tissue of odontogenic fibroma. Although the WHO has eliminated this entity from the classification of odontogenic tumors, it has proposed the name of CGCOT for this lesion [1].

CGCOT occurs predominantly in the premolar- mol-



**Figure 2:** Histopathologic sections show, **a:** Sheets and lobules of eosinophilic granular cells in a myxomatous stroma (H&E×100). **b:** Large granular cells and one odontogenic island with juxtaepithelial hyalinization (H&E ×400). **c:** granular cells was positive for CD68 diffusely (IHC×400) and **d:** negative for S-100 protein (IHC×400).

lar region of mandible with a strong predilection for middle-aged women [1, 3, 6, 8]. More than half of the cases happened during the sixth to eighth decades of life [9]. Current case was a 57-year-old female with posterior mandibular involvement which is similar to most previous studies. The majority of reported cases showed painless swelling [1, 3, 9-11]. Nevertheless, cortical perforation, pain, ulceration, tooth displacement, and root resorption and extension to the adjacent soft tissue was described [1, 9, 12-13].

In the current case, however, tooth mobility (grade II) was also existed. The radiographic features were not pathognomonic and the majority of cases show unilocular radiolucency with sclerotic borders [1]. Focal opacity may be seen in few cases [14]. Anbiaee *et al.* [14] mentioned that the mixed appearance of their case report was due to the multi locular pattern and very coarse septa of the lesion resembling calcifications in the panoramic view. Therefore, they suggested using three-

dimensional techniques such as CBCT for better diagnosis and prediction of lesion behaviors.

In microscopy, this tumor is frequently well-demarcated and often shows thin pseudocapsule [15]. It demonstrates sheets and lobules of round to polygonal granular cells with eccentric nuclei, which intermixed with inactive cords and nests of odontogenic epithelium. The lobules are separated with fibrous connective tissue stroma. Cementum-like material, dystrophic calcification, Juxtaepithelial hyalinization and occasional palisading or polarization of the peripheral epithelial cells of odontogenic islands are also reported [1]. In present case, myxoid connective tissue was seen intermixed with granular cells that were not reported before. In immunohistochemistry, granular cells of CGCOT are reacted with CD68, lysozyme,  $\alpha$ -1 antichymotripsin and vimentin that support the histiocytic origin [3, 8]. Furthermore, these cells positive for vimentin and bcl-2 [1, 3, 11]. They are negative for neuron specific enolase

(NSE), cytokeratin (CK) and S-100 protein. Abundant lysosome-like particles has been reported in electron microscopy which were similar to those previously described for granular cell tumor of soft tissue [14].

The histopathologic differential diagnosis consists of granular cell tumor (GCT) of soft tissue, granular cell variant of ameloblastoma and congenital epulis. Granular cells of GCT demonstrate similarities to those of CGCOT. Although, GCTs doesn't show odontogenic islands, cementum-like material or dystrophic calcification and are strongly positive for S-100 protein [1]. Granular cell ameloblastoma reveals changes within the ameloblastic islands and replacing the stellate reticulum-like cells. These cells are positive for cytokeratin and negative for S-100 protein [1]. In the case of congenital epulis, the patient's age and location of the lesion (alveolar ridge) are helpful in diagnosis. In addition, it is positive for NSE and negative for S-100 [1]. It seems that CGCOT is completely benign and the treatment of choice is conservative surgery. Though, close follow-up is essential to determine the long-term result [1, 15].

### Conclusion

In conclusion, as CGCOTs are rare in the literature, their clinical behavior and prognosis are not clear; hence, reporting more such cases may be beneficial. The current case had some interesting points comprising tooth mobility and myxoid areas that was not reported before in the literature.

### Conflict of Interest

The authors declare that there is no conflict of interests regarding the publication of this paper

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