

Central Granular Cell Odontogenic Tumor: Report of a Case with CBCT Features

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Abstract

Central granular cell odontogenic tumor (CGCOT) of the jaw is an exceedingly rare benign odontogenic neoplasm with 35 reported cases in the literature. Among these, very few studies have focused on the cone-beam CT features of CGCOT. Here, we report a case of an asymptomatic CGCOT in a 16-year-old girl and focus on the cone-beam CT features. Only 36 cases of this lesion, including this one, have been reported so far. The case presented is of special importance due to the young age of the patient, the posterior location of the lesion and the multilocular pattern in the cone beam CT images

Key Words: Granular cell tumor; Odontogenic tumors; Cone beam computed tomography

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INTRODUCTION

Central granular cell odontogenic tumor (CGCOT) of the jaw, which was previously known as central granular cell odontogenic fibroma or granular cell ameloblastic fibroma, is an exceedingly rare benign odontogenic neoplasm [1, 2] with 35 reported cases in the literature [3]. Some of these lesions have very few clinical features so they are often found accidentally in the routine radiographs. The extension of some types of this lesion and the absence of capsule around them has made resection the necessary treatment for these CGCOTs [1]; therefore, the precise and early diagnosis of these lesions could refrain their extreme extension. CGCOT is usually seen in the posterior areas of the female mandible in

the fifth decade of life [3]. In most reported cases, they have a well-defined unilocular radiolucent pattern. Focal areas of opacity may be evident in few cases. A sclerotic rim may also be present [4]. As very few studies have focused on the cone-beam CT features of CGCOT, here we report a case of an asymptomatic CGCOT in a 16-year-old girl with descriptions on CBCT findings.

CASE REPORT

In October 2010, a 16-year-old girl was referred to our maxillofacial radiology clinic for radiographic examination for orthodontic treatment. Accidentally, in the panoramic radiograph, a unilocular mixed lesion was detected in the left mandibular angle.



Fig 1. Cropped panoramic radiograph shows a unilocular mixed lesion in the left mandibular angle.



Fig 2. Lateral cephalogram shows a mild expansion in the posterior and inferior borders of the mandibular angle.

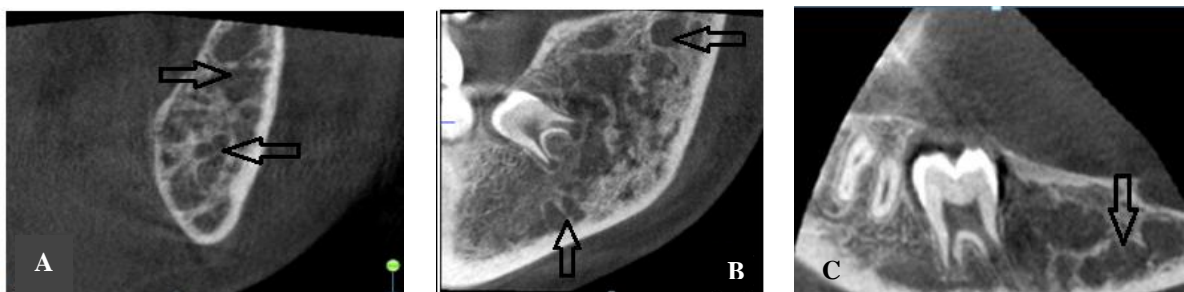


Fig 3. CBCT images: A: coronal, B: sagittal and C: oblique sagittal views show multilocularity and coarse septa of the lesion (arrows). The oblique sagittal section (C) shows the intact cortical borders and follicular space of the unerupted left third molar.

The posterior borders of the lesion were poorly-defined; whereas, the anterior borders were ill-defined. In the panoramic view, intra-lesion calcifications were evident (Figure 1). In the panoramic radiography and lateral cephalography, a mild expansion was observed in the posterior and inferior borders of the mandibular angle (Figure 2). The mandibular cortical borders appeared completely intact. Root resorption was not present on any of the teeth adjacent to the lesion. The approximate dimensions of the lesion were 3×5 cm. The patient had no noteworthy medical history. She did not experience pain in the region, and the lesion was non-tender on palpation. On clinical examination, a slight swelling was observed in the left mandibular angle. No enlarged lymph nodes were detected on palpation.

On intra-oral examination, the overlying mucosa of the region was smooth and of normal color.

To localize the intraosseous lesion and perform further investigations on the effects of the lesion on the surrounding structures, we performed cone beam computed tomography. On CBCT examination, a multilocular lesion with coarse septa, without sclerotic borders was observed.

The cortical borders of the mandible and follicular space of the unerupted left third molar were normal. No displacement and root resorption was found on the teeth adjacent to the lesion (Figure 3).

Moreover, no displacement in the inferior alveolar nerve canal position was noted, but the cortical borders of the canal were thinned (Figure 4).



Fig 4. CBCT images show the effect of the lesion on the mandibular canal. A: sagittal view shows disruption of the superior and inferior borders of the mandibular canal in some areas (arrows). B: cross sectional views show thinning of the mandibular canal cortical borders.

Based on age, clinical behavior, and radiographic findings, the differential diagnoses of fibrous dysplasia and desmoplastic ameloblastoma were made. Incisional biopsy of the lesion was performed.

Histologic examination of the lesion revealed sheets and islands of large eosinophilic cells with abundant granular cytoplasm with small islands of odontogenic epithelium (Figure 5A-C). Foci of dystrophic and cementum-like calcifications were detectable in the lesion (Figure 5D). The fibrous stroma contained fibroblasts, and in the decalcified sections, bone trabecules were evident (Figure 5A and B). Histologic examination confirmed the diagnosis of central granular cell odontogenic tumor (CGCOT). Resection of the lesion was carried out to treat the patient. For mandibular reconstruction, allograft from the fibula was applied (Figure 6). Histopathological examination of the excised specimen confirmed the diagnosis of CGCOT.

The patient is on follow-up periodic check and there has not been any radiographic evidence of recurrence on the follow-up radiograph after 2 years of operation (Figure 7).

DISCUSSION

CGCOT of the jaw is an exceedingly rare, benign odontogenic neoplasm with 35 reported cases in the literature until 2012 [3].

A historical controversy regarding its nomenclature and histogenesis is evident in previous literature [1, 4]. Werthemann reported the first case of CGCOT in 1950 as “spongiotic adamantoma” [1, 5-8]. This lesion was later termed as “granular cell ameloblastic fibroma” by Couch et al. [9]. Most authors, including Gardner [10] and Brannon [1, 11] introduced CGCOT as a separate entity from central odontogenic fibroma (COF). CGCOT was not considered as an entity in the recent WHO classification of head and neck tumors published in 2005[4].

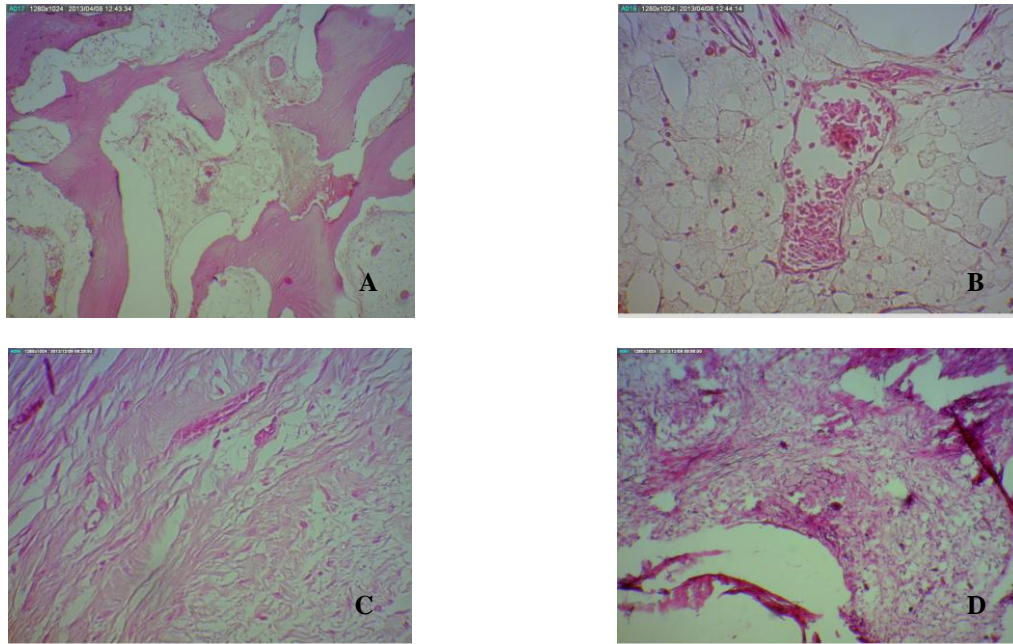


Fig 5. Photomicrograph shows granular cells with eosinophilic and granular cytoplasm in the fibrous stroma. (A: H&E staining, ×100 magnification and B:×400 magnification) C: Island of odontogenic epithelium (arrow). D: Foci of dystrophic and cementum-like calcifications (arrows)



Fig 6. Post-operative cropped panoramic shows the mandibular reconstruction.



Fig 7. Follow-up panoramic radiograph after 2 years of operation does not show any evidence of recurrence.

This evolution in nomenclature and the concurrent debate on the histogenesis of this lesion was due to the uncertain histogenesis of the granular cells, i.e., it is not clear whether these cells are neoplastic, reactive, or metabolic in nature [2].

On electron microscopy, White et al. [12] found numerous lysosome-like particles that were similar to those previously described for “granular cell tumor.”

According to de Sousa et al. [8], the granules found in this lesion are not specific to a particular cell line.

Clinically, more than 70% of CGCOT cases occur in females [13]. Although CGCOT is more often reported in patients older than 40 years at the time of diagnosis [13], there has only been one case of CGCOT reported by Regezi (1978) in a 16-year-old boy [1] similar to the age range of our case.

CGCOT is usually seen in the mandible and most often in the premolar and molar regions in previously reported cases [1, 3, 7, 13, 14].

In the current case, however, the lesion was located posteriorly in the mandibular angle region rather than in the premolar-molar areas. CGCOT is usually presented as a painless swelling according to the report of Gesek et al. [14]. However, cortical expansion, teeth displacement, and locally aggressive behavior have been reported in few cases [1]. In the present case, mild expansion of the mandibular angle was not obvious due to coverage of the masseter muscle in the region, which led to accidental detection of the lesion. However, the mild expansion was detectable on radiographic examination.

From a radiographic point of view, in previous cases, most lesions were reported as radiolucent with sclerotic borders [4]. The cases reported by Gesek et al. [14], Machado de Sousa et al. [8], Reichart et al. [1] and Brannon et al. [1] were all multilocular with sclerotic borders. Furthermore, in the review of literature by Ardekian et al. [15], 10% of the reported cases of CGCOT were multilocular. The current case also showed a multilocular pattern without sclerotic borders. The multilocularity of this lesion might be related to the young age of the patient or the posterior location of the lesion, although this hypothesis warrants further investigation. According to the report by Ardekian et al. [15], 90% of CGCOTs were radiolucent, whereas 10% showed a mixed pattern. They reported that the intralesional calcifications were microscopically observed in 50% of the cases, but they were not detectable in the radiographic view because of the scarceness of these calcifications. In the present case, the mixed pattern and calcifications were evident in the panoramic radiograph, but in the further evaluations by means of CBCT, otherwise specified.

Clinically, CBCT has a wide range of applications in dentistry. It may be used to determine the extent and condition of the internal struc-

ture of odontogenic lesions. CBCT can allow the three-dimensional evaluation and determination of the amount of expansion and extension of the odontogenic lesions [16]. Compared with CT-scan, the relatively low dose and high speed image acquisition of this technique results in the widespread application in dentistry. Despite the mentioned benefits, low contrast resolution prevents appropriate detection of soft tissue, which can be detected in CT scan images as well [17]. In the present case, The CBCT evaluation of the lesion revealed that this mixed pattern is actually due to the multilocular pattern and very coarse septa of the lesion resembling calcifications in the panoramic view. This concept highlights the importance of using new techniques such as CBCT or CT-scan, which are superior in better description of the lesions.

CONCLUSION

CGCOT can also affect adolescents. Different behaviors of this lesion could be related to the age of the patients. Various radiographic appearances have been reported for CGCOT. This variation could be due to the application of different radiographic techniques. It seems that use of three-dimensional techniques could be useful in better perception of the radiographic views and behavior of the lesion.

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