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Multilocular radiolucent jaw lesion: A diagnostic pitfall

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Abstract

Dentinogenic Ghost Cell Tumour (DGCT) is a rare, locally invasive, solid neoplastic variant of the Calcifying Odontogenic Cyst (COC). It accounts for 2% to 14% of all COCs. In this present report, we discuss a case of a 30-year-old female patient with the chief complaint of pain and swelling in the lower right back region of the jaw. CECT maxillomandibular region reveals a large well-defined expansile multilocular hypodense lesion involving the posterior part of the mandibular body, angle, and ramus of the right mandible with diffuse thinning and erosion of cortical margins. The histopathological analysis revealed the presence of odontogenic epithelium with calcifications, ghost cells and dextrinoid material. The clinical features, imaging and histopathological findings confirmed the diagnosis of Dentinogenic Ghost Cell Tumor.

Keywords: Dentinogenic; Ghost cell; Ghost cell tumour; Leisgang rings; DGCT.

Introduction

Dentinogenic Ghost Cell Tumor (DGCT) is a rare benign odontogenic tumor, characterized by ameloblastomatous epithelium, ghost cells and extensive dextrinoid deposition [1]. In 1972, Fejerskov and Krogh suggested the term 'calcifying ghost cell odontogenic tumor' [2]. The term "dentinogenic ghost cell tumor" (DGCT) was proposed for the solid neoplastic type of Calcifying Odontogenic cyst (COC) [3]. According to the WHO 2005 classification, the spectrum of odontogenic ghost cell tumours comprises Calcifying cystic odontogenic tumor (CCOT), DGCT and ghost cell odontogenic carcinoma. The World Health Organization (WHO) recently recognized DGCT as the solid counterpart of the Calcifying Odontogenic cyst [1]. Dentinogenic ghost cell tumours (DGCT) are classified into two types: extraosseous (peripheral) and intraosseous (central). Intraosseous DGCTs are more aggressive, exhibit an infiltrative growth pattern, and have a high recurrence rate after resection. Therefore, intraosseous DGCTs require extensive surgical resection with an adequate safety margin, unlike the extraosseous variant, which is less aggressive [3,4]. Here, we present a case report of 30-year-old female diagnosed as Dentinogenic Ghost Cell tumor with its clinical, radiographic, histopathological features, along with management and follow up.

Case presentation

A 30-year-old female patient reported to the OPD of tertiary care dental hospital with chief complaint of pain and swelling on right side of face for past 5 months. The patient was hypothyroid and was under medication for the past 6 months. On inspection, Extra oral examination revealed diffuse swelling of

right side of face extending super inferiorly from a point 5 cm below right infraorbital rim till the lower border of mandible, and anteroposterior 4 cm from right side ala of nose till 1 cm in front of the right tragus of ear (Figure 1). Overlying skin was non-stretched and normal in colour with no evidence of secondary changes like surface ulceration, sinus opening or pus discharge. On palpation, there was pain during wide mouth opening as well as during protrusive and lateral mandibular movements. No evidence of right-side submandibular lymphadenopathy was noted. On intra oral examination, no buccolingual expansion was present, however tenderness was noted with respect to anterior border of ramus of mandible. Overlying mucosa was normal in colour as that of surrounding mucosa with no evidence of sinus opening/ pus discharge. Missing 46 and 36 with medially tilted 47 was present. Investigations including routine blood investigations such as complete blood count, differential leucocyte count, ESR, random blood sugar, tricot and Orthopantomogram (OPG) were advised. Blood investigations were within normal limits. OPG revealed well defined multilocular radiolucent lesion in right hemimandible extending, anteroposterior from distal aspect of 44 till posteriorly border of ramus of the mandible and super inferiorly involving condyle and coronoid process till lower border of the mandible with few internal bony septa dividing the lesions into multiple compartments approximately 7 cm x 4 cm in size (Figure 2). Cortical outline w.r.t right condyle was intact with gross thinning of buccal and lingual cortical plates; sigmoid notch is eroded with break in continuity seen at posterior border of mandible. Lesion caused resorption of roots of right mandibular second and third molar with loss of lamina dura noted. The inferior alveolar canal was displaced inferiorly on the right side. Missing 46, 36 with grossly decayed 28 was present. Further, a Contrast Enhanced Computed Tomography (CECT) scan of maxilla and mandible was advised. CECT revealed large well defined expansile multilocular hypodense lesion measuring approximately 3.9 x 3.2 x 7 cm in size, involving posterior part of mandibular alveolar arch (body), angle and ramus of right mandible (Figure 3). There is diffuse thinning and erosion of cortical margins, mild displacement of molars with erosions of roots of the same was noted. No evidence of any intralesional tissue component/calcific foci was seen. Based on the clinical and radiographic features, Odontogenic Keratocyst, Ameloblastoma, Odontogenic Myxoma, Calcifying Epithelial Odontogenic Tumor, Central Giant Cell Granuloma and Giant Cell Tumor of Hyperparathyroidism were considered in the differential diagnosis. Incisional biopsy was carried out under local anaesthesia and the tissue specimen was sent for histopathological analysis. Macroscopic examination revealed that the specimen was irregular in shape and brown in colour. Soft tissue bits were firm and hard tissue bit was bony hard in consistency. Microscopic examination revealed lesional tissue composed of fragmented bits of odontogenic epithelium with cuboidal hyperchromatic cells. Many follicles and cords of the epithelium were present in lesional tissue showing peripheral layer of columnar to cuboidal cells with hyperchromatic palisaded nuclei and subnuclear vacuolization while central cells resemble stellate reticulum with areas of acanthomatous changes. Abundant eosinophilic masses of anucleated ghost cells staining yellow in van Gieson stain were present throughout the lesional tissue associated with numerous clusters of multinucleated foreign body giant cells. Abundant eosinophilic to basophilic calcifications varying from osteodentin with cellular inclusions, globules of cementum like material, dentinoid and bony trabeculae with entrapped osteocytes were present staining bright red in van Gieson stain (Figure 4). The surrounding connective tissue is moderately collagenous with haphazardly arranged collagen fibres, ovoid to spindle shaped fibroblasts, variably sized blood vessels and dense chronic inflammatory cell infiltrate chiefly composed of lymphocytes and plasma cells. CK 19 showed negative staining for ghost cells. Features were suggestive of Dentinogenic Ghost Cell Tumor. The patient was planned for segmental resection under general anaesthesia from right side condyle to 44 tooth regions. A reconstruction plate was placed along with costochondral graft which was taken from 5th rib (Figure 5). The gross specimen (Figure 6) was submitted for histopathological evaluation which confirmed the diagnosis of Dentinogenic Ghost Cell Tumor. Patient was thereafter kept under regular follow up for last 2 years and no recurrence is noted (Figure 7).

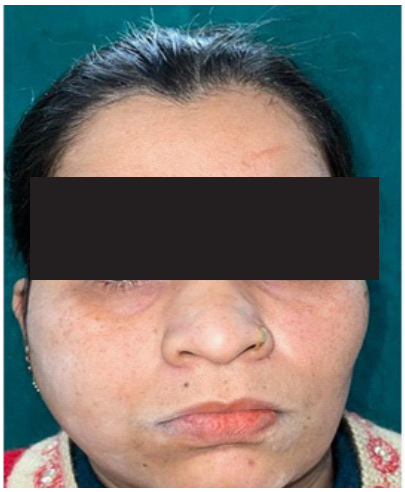

Figure 1: Extraoral photograph showing facial asymmetry with diffuse swelling on middle third of face on right side.

Discussion

The Dentinogenic Ghost Cell Tumour (DGCT) is an odontogenic neoplasm, which is uncommon. DGCT may develop at any age from the second to the eighth decade of life, mean age of occurrence being 50 years with no gender predilection [5]. However, Shah et al [6] found that this lesion occurs more commonly in males than in females. Intraosseous DGCT has been reported to occur predominantly in canine to first molar region [7]. It may be seen in the edentulous region of the jaws as well. De Arruda et al [8] who reviewed 55 cases of DGCTs, observed that it mostly occurs in the fourth decade of life and mandible is the most common site of involvement. Majority of cases were seen in males. The present case was noted in mandible involving right mandibular body, ramus and coronoid process in a 30-year-old female patient, which is at a comparatively younger age than the average age as reported by Candido who found that the age ranges from 41 to 83 years for DGCTs with an average age of 62 years. Clinically intra-osseous DGCT manifests as painless visible swelling causing obvious facial asymmetry due to expansion of the jaw, however occasionally pain and numbness is associated with the lesion. Swelling can be accompanied by pus discharge, tooth displacement or mobility [9]. The reported case presented as a diffuse extraoral soft tissue swelling without causing expansion of buccal or lingual cortex of man-

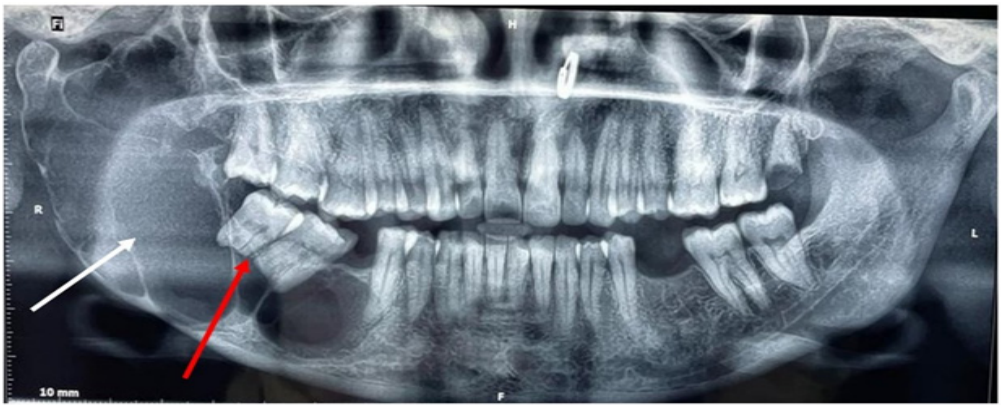

Figure 2: Orthopantomograph (OPG) showing well defined multilocular radiolucent lesion (White arrow) in right side of mandible extending from region of 44 till the sigmoid notch with thinned cortical margins and there is resorption of apical ends of roots of 47, 48 (Red arrow).

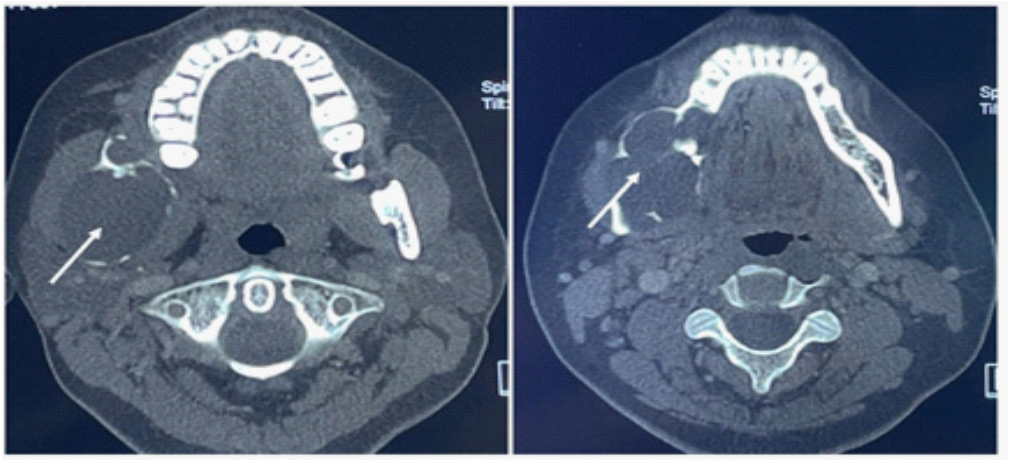

Figure 3: Axial section of CT showing well defined expansile multilocular hypodense lesion (White arrow) involving posterior part of mandibular alveolar arch (body), angle and ramus of right mandible with diffuse thinning and erosion of cortical margins.

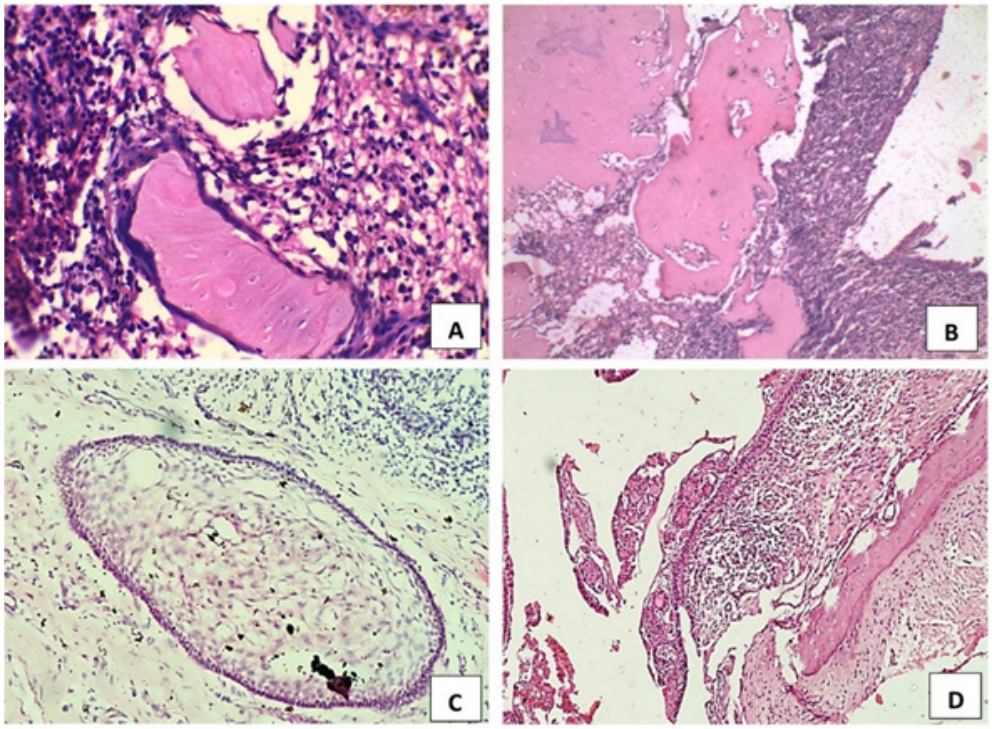

Figure 4: **(A)** Microphotograph (40x) showing clusters of ghost cells with nuclear remnants **(B)** Microphotograph (10x) showing large deposits of dentinoid material surrounded by inflamed connective tissue stroma **(C)** Microphotograph (10x) showing follicle of odontogenic epithelium with low columnar peripheral cells and central stellate reticulum-like cells in a loose connective tissue matrix. **(D)** Microphotograph (10x) showing cystic cavity lined by ameloblastomatous epithelium containing ghost cells.

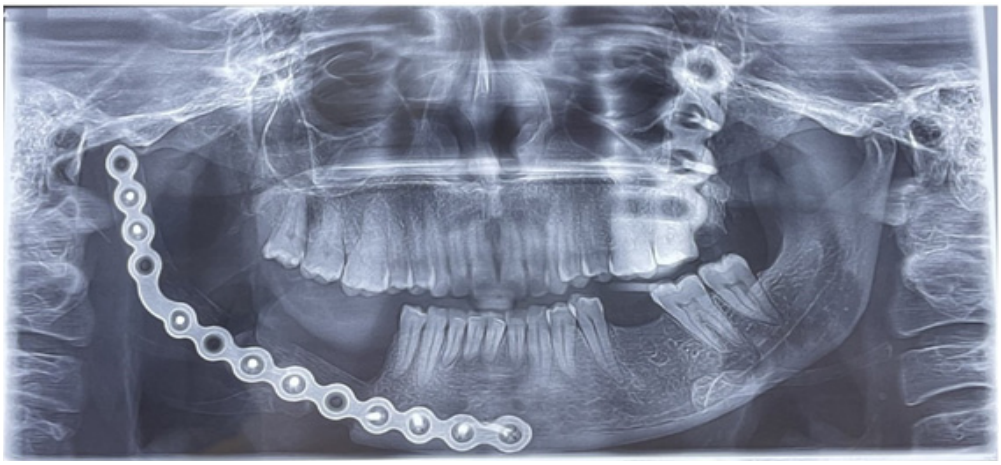

Figure 5: Postoperative radiograph (OPG) showing segmental mandibular defect on right side with reconstruction plates with screws extending from symphysis till condylar region on right side.

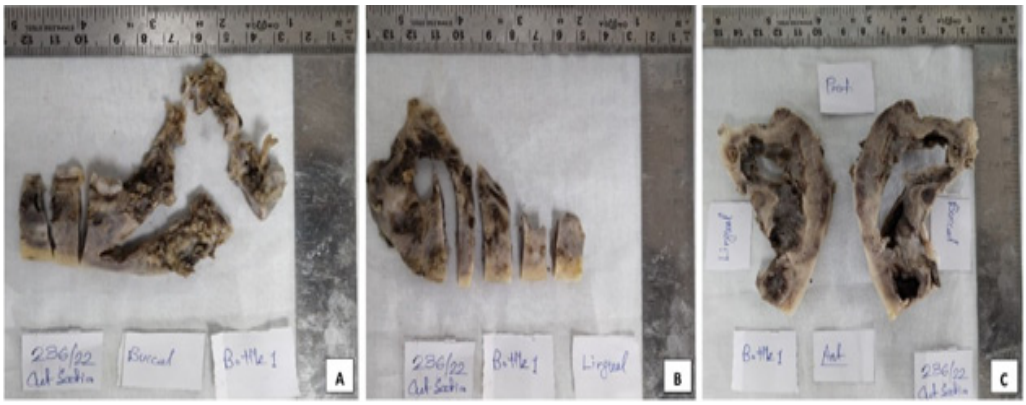

Figure 6: Photograph of the gross specimen after surgical resection from **(A)** Buccal aspect, **(B)** Lingual aspect, and (C) Antero-posterior aspect.

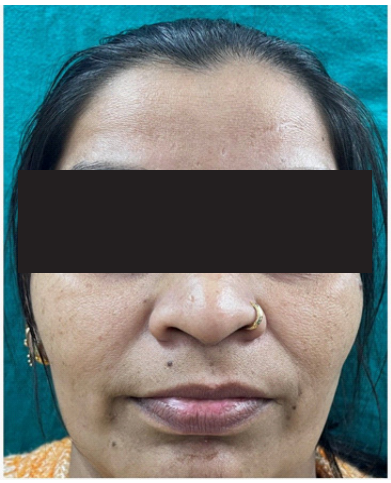

Figure 7: Postoperative follow up facial photograph of the patient.

dible. Amounting to the presence and extent of calcification, DGCT may appear radiographically as radiolucent, radiopaque or mixed lesion. Lesions can be unilocular or multilocular with either well defined or ill-demarcated margins [10]. These findings were in accordance with the literature review which was carried out by Constantias et al [9] in 2013 and by De Arruda et al in 2018, majority of cases present as mixed radiolucent radiopaque lesion. Other features like presence of impacted teeth, displacement and/or root resorption of adjacent teeth have also been reported [11]. This case presented with large well defined expansile multilocular hypodense lesion involving posterior part of the mandibular alveolar arch, angle and ramus of right mandible with diffuse thinning and erosion of cortical margins, mild displacement of 2nd and 3rd molar tooth with erosion of roots of the same. Final diagnosis of DGCT is usually made only after the histopathologic correlation. Microscopic features of DGCT consists of variable amounts of ameloblastomatous epithelial proliferation, showing pseudo-ductal structures. Large deposits of dentinoid material containing entrapped epithelial cells will be readily found, associated with clusters of ghost cells surrounded by whorl-like epithelial arrays and ameloblastic epithelium. The stroma will be fibrous and rich in small blood vessels. Similar histopathological features were seen in the current case. However, in most of DGCT cases ghost cells will show positive staining with cytokeratin 14 and 19, which is negative in the present case. Similarities are seen in epithelium of ameloblastoma and DGCT. However, recognition of ghost cells, dysplastic dentin, and calcifications will help to distinguish DGCT from ameloblastoma [12]. DGCT needs to be distinguished from its malignant counterpart odontogenic ghost cell carcinoma. Both lesions show ghost cells and infiltrative growth. However, characteristic microscopic features like hypercellular proliferation of small cells with scanty cytoplasm, hyperchromatic nuclei, brisk mitotic activity are usually seen in odontogenic ghost cell carcinoma, thus helping in differentiation. Thorough microscopic examination of DGCTs should be done as malignant transformation into an odontogenic ghost cell carcinoma is possible [13]. Early diagnosis of DGCT is essential for better prognosis of the patient. Intraosseous lesions are usually treated by block excision or segmental resection with adequate safety margins to prevent recurrence, which is frequent with conservative management of the lesion. Recurrence potential for intraosseous DGCT appears similar to that of conventional ameloblastoma i.e

5-8 years following initial treatment. Kasahara et al studied 11 patients with intraosseous DGCT and found that the recurrence rate was 36% [13]. In another study by Sun et al [14], 7 patients with intraosseous DGCT were reviewed and 5 patients treated with conservative treatment showed recurrence of the lesion. Aggressive treatment was done for the present case, showed no recurrence till date.

Conclusion

In conclusion, we reported a case of giant DGCT of the mandible, highlighting its clinical, radiological, and microscopic features. Hence, we need amalgamation of clinical examination, advanced radiographic imaging, and detailed histopathological examination to reach the final diagnosis of DGCT.

Declarations

Funding: None to declare.

Conflicts of interest/competing interests: Authors have no conflict of Interest.

Ethics approval: Not applicable.

Informed consent: Informed consent has been obtained from subject for inclusion of their details & images in manuscript.

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