

Case Report

A rare case of calcifying cystic odontogenic tumor

Vivek Narayan*, James Antony Bhagat**, Muthukrishnan Arvind***, M R Muthusekar****

*Senior Lecturer, ***Prof and Head (Academics), Deptt. of Oral Medicine and Radiology, Saveetha Dental College, Chennai

**Senior Lecturer, Department of Oral and Maxillofacial Surgery, Ragas Dental College, Chennai

****Prof and Head, Department of Oral and Maxillofacial Surgery, Saveetha Dental College, Chennai, India

DOI: 10.5455/jrmds.20153214

ABSTRACT

Calcifying odontogenic cyst is a rare odontogenic cyst. It was described by Gorlin in 1962 and is also called as Gorlin's cyst. Ghost cells in the epithelium are the hallmark of Gorlin's cyst. A 25 year old male patient complained of a painless swelling in his lower anterior teeth region which was present for the past 3 years. It was slow growing with absence of infiltration, soft in its center and bony hard in its periphery, radiograph showing multilocular radiolucency, with associated root resorption, tooth displacement. Histopathological examination revealed a cystic lesion, lined by odontogenic epithelium and amorphous eosinophilic round or oval structures suggestive of ghost cells were seen within the epithelium. A long standing multilocular radiolucent lesion in the mandible crossing the midline with clinical features imitating ameloblastoma and odontogenic keratocyst was later diagnosed as a Gorlin's cyst with histological evidence.

Keywords: Calcifying cystic odontogenic tumor, Ghost cells, Multilocular radiolucency.

INTRODUCTION

Calcifying odontogenic cyst is a unique and rare odontogenic cyst [1]. It was described by Gorlin in 1962 and is also called as Gorlin's cyst. It was classified as a benign odontogenic tumor and renamed as calcifying cystic odontogenic tumor in 2005 [2-3]. This unusual odontogenic lesion possesses characteristics of both cyst and neoplasm. A case is presented here in which the cyst occurred in the mandible crossing the midline, with multilocular radiolucency and associated with tooth displacement and root resorption resembling a benign odontogenic tumor. But on histological evaluation it was confirmed as calcifying cystic odontogenic tumor by the presence of ghost cells.

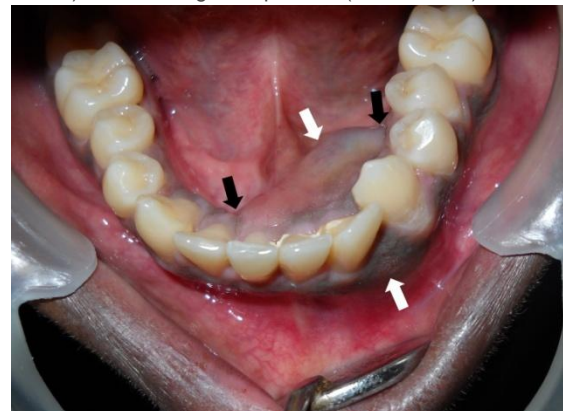
CASE REPORT

A 25 year old male patient visited the Department of Oral Medicine and Radiology, Saveetha Dental College, Chennai, India. The patient's chief complaint was a painless swelling in his lower anterior teeth region, present for the past 3 years. History revealed that the swelling started before 3 years and had an insidious onset. It was initially small and gradually increased in size. The patient developed such a swelling for the first time. Medical, surgical, dental and family histories were not noteworthy.

Intraoral examination revealed a single, localized swelling present in the lower anterior alveolar

mucosa in relation to teeth 31, 32, 33, 34, 35, 36, 41, and 42. The swelling was 3 cm × 4 cm faciolingually and mediolaterally respectively. The swelling was ovoid in shape. It had well-defined margins. Surface was lobulated. Swelling extended from teeth 34 to 42 with faciolingual expansion [Figure- 1]. Mucosa over the swelling was normal with no secondary changes.

Figure – 1: Clinical picture showing an intraoral swelling with lobulated surface extending from teeth 34 to 42 (black arrows) with faciolingual expansion (white arrows)



On palpation, swelling was non tender. The center of the swelling was soft and the periphery was bony hard in consistency. The surface was lobulated with well-defined edges. The surrounding structures were normal and the swelling was attached to the bone. Mucosa over the swelling was intact and tooth 33 was tilted lingually. None of the involved teeth had caries or periodontal lesion. Regional

lymphadenopathy was absent. The differential diagnosis of this swelling which was chronic in nature, non-tender, slow growing with absence of infiltration, soft in its center and bony hard in its periphery, and no loss of sensation in the lower lip are,

1. A benign neoplastic swelling (Ameloblastoma).
2. Developmental odontogenic cyst (keratocystic odontogenic tumor).

Panoramic radiograph revealed a multilocular radiolucency from teeth 36 to 43 with root resorption in 34, 35, and 36 and displacement of teeth 31, 32 and 33 [Figure-2]. Mandibular occlusal radiograph revealed expansion of the anterior mandible faciolingually and mediolaterally [Figure-3]. CT scan revealed expanding cystic lesion in the left body and symphysis of mandible with displacement of roots of adjacent teeth [Figure-4]. Incisional biopsy was done for the lesion, which revealed the underlying connective tissue to be myxomatous and fibrocellular rather than a typical fibrous wall encountered in a cystic lesion which suggested the lesion to be unicystic ameloblastoma. Under GA, the lesion was enucleated and curettage was done. Histopathological examination revealed features consistent with calcifying cystic odontogenic tumor [Figure-5].

Figure – 2: OPG showing a multilocular radiolucency from teeth 36 to 43 (white arrows) with root resorption in 34, 35, and 36 with displacement of teeth 31, 32 and 33

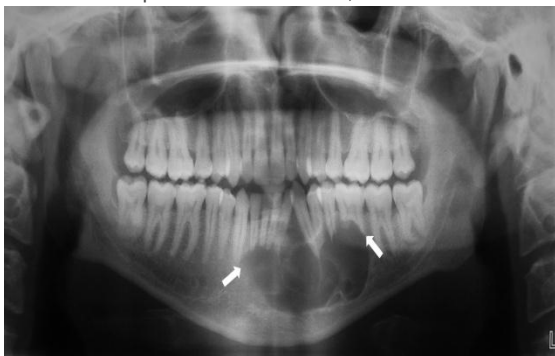
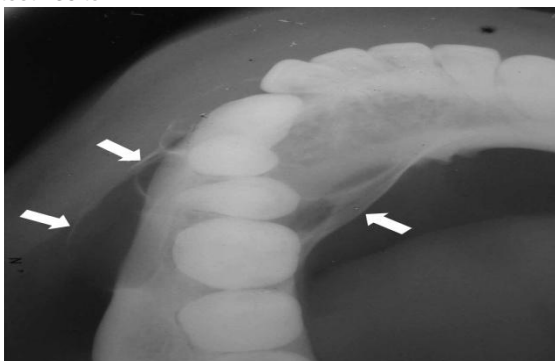


Figure – 3: Mandibular occlusal radiograph showing faciolingual expansion of the lesion (white arrows) from teeth 36 to 42



Correlating the history, clinical findings, radiographic and histopathologic findings a final diagnosis of calcifying cystic odontogenic tumor was made. No clinical and radiographic evidence of recurrence was noted after 3 years of follow up.

Figure – 4: Coronal section CT scan revealing expanding cystic lesion in the left body and symphysis of mandible (yellow arrows)

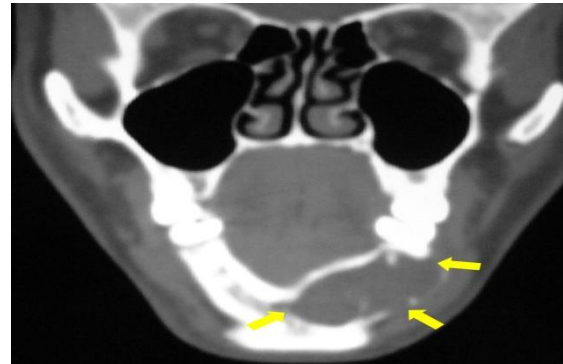
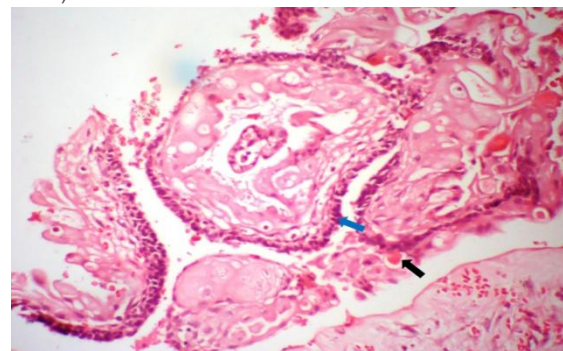


Figure – 5: Haematoxylin and Eosin stained section under 10X magnification of the specimen showing ameloblast like cells (blue arrow) and characteristic ghost cells (black arrow)



DISCUSSION

Calcifying cystic odontogenic tumor is an uncommon odontogenic cyst contributing to 1% of all odontogenic cysts [2]. Clinically the lesion might present as a slow growing painless swelling unless secondarily infected. Thinning of the cortex might be seen as a result of expansion and with center of the swelling being soft and yielding on pressure and the periphery was hard on palpation. On aspiration the lesion usually yields a viscous, yellow granular fluid, but in our case it was serous and blood tinged. Many classification systems were proposed for calcifying cystic odontogenic tumor considering it as 2 entities, a cyst and a neoplasm [4-5]. Calcifying epithelial odontogenic cyst is a rare developmental, unusual odontogenic cyst arising from dental lamina rests [1]. A prominent microscopic feature of COC is ghost cells, which may induce the calcification that occurs in this lesion. This cyst can occur over a wide age group but peak incidence is noted in second and third decades of life, with close to equal

distribution among males and females. The maxilla and mandible are also about equally affected, and there may or may not be an association with an unerupted tooth. Presenting symptoms include a firm, painless swelling. The cyst is predominantly an intraosseous lesion. The lesion can occur both in maxilla and mandible and mostly affects the incisors and canines. The cyst can appear as a unilocular or a multilocular radiolucency. Radiopaque structures within the lesion, either irregular calcifications or tooth like densities can be seen [1]. Root resorption of the adjacent teeth is also sometimes present. The most characteristic histologic feature is presence of variable number of ghost cells. These are eosinophilic altered epithelial cells characterized by loss of nuclei with preservation of basic cell outline.

In majority of COC cases, features such as impacted tooth most often a canine and presence of calcifications were common and the lesion usually appears unilocular in a radiograph [5-6]. But our case had no impacted tooth, had a multilocular radiolucency which is only seen occasionally and the lesion was crossing the midline [7]. Root resorption and tooth displacement indicated long term existence of pathology.

CONCLUSION

Our case was unique in that, the clinical features were not favoring calcifying cystic odontogenic tumor especially the lesion did not have any calcifications or impacted tooth, arriving at a provisional diagnosis for this case was challenging. Hence, we always must consider Gorlin's cyst as a differential diagnosis when encountering such cases. As it does not have any characteristic, clinical and radiological features, the histopathological evaluation is mandatory in order to obtain a definitive diagnosis. Thus thorough and careful examination can give an accurate diagnosis.

ACKNOWLEDGEMENT

Authors are thankful to Dr. Sivapathasundaram, Head of Department of Oral and Maxillofacial

Pathology, Meenakshi Ammal Dental College, for his expert opinion of the biopsy specimen.

REFERENCES

1. Robinson HBG: Classification of cysts of Jaw. Am J Orthod Rel Surg 1947;31:370 – 7.
2. Praetorius F, Ledesma – Montes C (2005) Calcifying cystic odontogenic tumor. In WHO classification of tumors. Pathology and genetics of head and neck tumors, Barnes L, Eveson JW, Reichart P, Sidransky D eds, IARC press, Lyon, 313.
3. Kramer, I.R. H, J. J. Pindborg, M. Shear: Histological typing of odontogenic tumors. (Springer Verlag, Berlin 1992).
4. Praetorius F, Hjorting-Hansen E, Gorlin RJ, Vickers RA. Calcifying odontogenic cyst: Range, variations and neoplastic potential. Acta Odontol Scand 1981; 39:227-40.
5. Hong SP, Ellis GL, Hartman KS Calcifying Odontogenic cyst: A review of 92 cases with reevaluation of their nature as cysts or neoplasms, the nature of ghost cells and sub classifications. Oral Surg Oral Med Oral Pathol 1991; 72 : 56 – 64.
6. Buchner A: The central (intraosseous) calcifying odontogenic cysts: An analysis of 215 cases. J Oral Surg 1991; 49:330–9.
7. Charles A Waldron. Odontogenic cysts and tumors. Oral and Maxillofacial Pathology (3rd ed) Noida: Elsevier 2009:695-6.

Corresponding Author:

Dr. Vivek Narayan
Senior Lecturer,
Department of Oral Medicine and Radiology,
Saveetha Dental College,
Chennai, India
E-mail: viveknarayan8985@gmail.com

Date of Submission: 25/01/2015

Date of Acceptance: 09/02/2015

How to cite this article : Narayan V, Bhagat JA, Arvind M, Muthusekar MR. A rare case of calcifying cystic odontogenic tumor. J Res Med Den Sci 2015;3(2):155-7.

Source of Support : None

Conflict of Interest : None declared