(International Print/Online Journal)

SJIF IMPACT FACTOR: 4.617
PUBMED-National Library of Medicine ID-101739732

ISSN (Print): 2209-2870
ISSN (Online): 2209-2862

International Journal of Medical Science and Current Research (IJMSCR)
Available online at: www.ijmscr.com
Volume2, Issue 6, Page No: 153-156
November-December 2019

Rare presentation of calcifying epithelial odontogenic Cyst (Type 1A) of maxilla– A diagnostic challenge

Dimla Denny Cheruvathoor[1], Bhupender Singh Negi[2], Sruthy Davis[3]
[1, 2, 3] MDS, Oral Medicine and Radiology Department,of Oral Medicine and Radiology
Government Dental College Calicut (Kozhikode) Kerala

***Corresponding Author:**

Dimla Denny Cheruvathoor
MDS, Oral Medicine and Radiology Department,of Oral Medicine and Radiology
Government Dental College Calicut (Kozhikode) Kerala

Type of Publication: Case Report
Conflicts of Interest: Nil

ABSTRACT
Odontogenic neoplasms are relatively uncommon in routine clinical practise. Most are diagnosed easily based on typical clinical and radiological features. Calcifying epithelial odontogenic cyst (CEOC) are peculiar due to its duality in existence, having both neoplastic and cystic features. This case report discusses a rare presentation of CEOC, which was extensively involving maxilla.

Keywords: NIL

INTRODUCTION

Odontogenic tissue is exclusive to jaw bones and neoplasms arising from these structure are also restricted to this area of human body. There are numerous pathologies arising from odontogenic apparatus, one among them being odontogenic tumors. Clinical, radiological, histopathological and prognostic features are quiet typical for each of these tumors. One must have a thorough knowledge for diagnosing such neoplasms. When they deviate from classic presentation, it becomes a difficult task for a clinician to diagnose.

Calcifying epithelial odontogenic cyst (CEOC), also called 'Gorlin cyst' is an unusual pathology, which is only 1% of all odontogenic tumors [1]. This is an interesting lesion having duel features of a cyst as well as a proliferating tumor. Accordingly different classifications and terminologies exist based on its histological picture.

Most common location of CEOC is anterior mandible. This case report discusses a rare presentation of CEOC in maxilla, having atypical features and mimicking adenomatoid odontogenic tumor.

Case report

A 20 year old male patient complained of painless swelling of right cheek for 2 months, with frequent watery discharge from nose since last 1 month. There was no associated pain, parasthesia, pus discharge or bleeding. He was not having relevant medical history and had habit of chewing tobacco.

On clinical examination, there was a swelling in the right cheek of size 5.5×4 cm, hard, non tender and with well- defined margins. The skin over the swelling appeared dry and darker in colour compared to adjacent area with a scar towards the anterior aspect. Intraorally, a swelling was present in the right maxilla of size 6×4 cm , both bucally and palatally. It was extending anteriorly from distal half of 21 and posteriorly upto distal aspect of 17. Superior margin was not visible intraorally (Figure 1). Inferiorly it

extends upto the marginal gingiva of 21,11,12,13,14,15,16 &17. Margins were well defined except the superior margin, which was not visible. Surface was appearing irregular with prominent vessels. The swelling was hard and non-tender. There were no ulcerations, bleeding or any other discharges, sinus openings or visible/palpable pulsations. 53 was retained with mobility and clinically missing 13. Palatally displaced 11 and 12 was in infraocclusion. No mobile teeth were present.

Figure 1: Clinical Images

Clinical examination features were suggesting possibility of Odontogenic tumor due to the following reasons - Hard consistency of swelling with bosselated surface, swelling on both buccal and lingual aspect and prominent blood vessels in the buccal vestibular surface.

Routine blood examination detected no abnormal values. Vitality of associated teeth was assessed using electric pulp testing. No response for 53, and delayed response of 11,12,14,15, 16 & 17 was obtained. 21 showed normal response.

Panoramic radiograph showed a radiolucent area in the right maxilla, of size 5.5×4 cm, which is extending medially from distal aspect of 11 and right lateral wall of nasal cavity upto 18 regions laterally. Superiorly it was extending from right infraorbital margin and inferiorly upto apical 3rd of roots of 17,16,15,14,12 &11. The margins were well defined and corticated in the medial, antero inferior, superior and distal aspects. posteroinferior margin was ill-defined. The internal aspect of the lesion is completely radiolucent except for the radiopacity in the superomedial aspect, suggestive of impacted 13. Root resorption present on 11,12,53,14,15,16 & 17. Floor of maxillary sinus was not visible on right side.

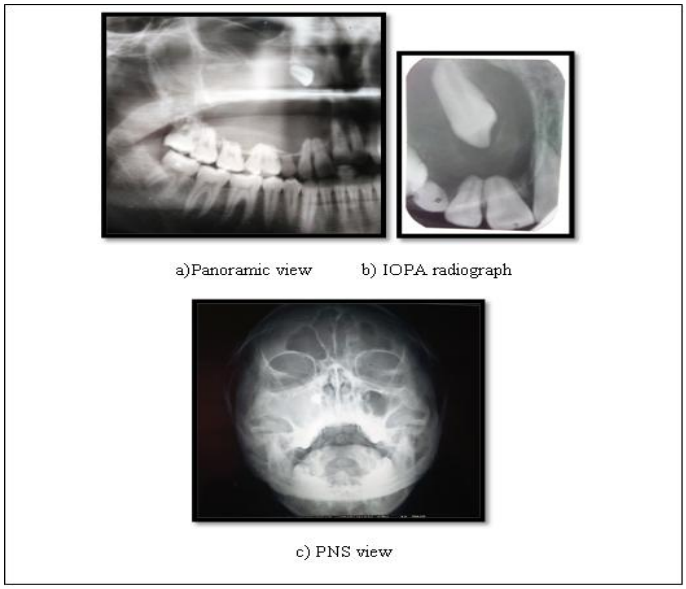

Figure 2: Radiographs

Volume 2, Issue 6; November-December 2019; Page No.153-156

Intra oral periapical (IOPA) radiograph of 12 region showed a radiolucent lesion was not within the confines of the IOPA radiograph. Impacted 13 was present within the lesion with increased radiolucency noticed surrounding the crown of 13. Medial and inferior walls of the lesion were corticated. No evidence of internal calcifications (Figure 2).

Paranasal sinus (PNS) view showed haziness in whole of right maxillary sinus with impacted tooth within the sinus, in its superomedial aspect. CT of PNS showed a well-defined soft tissue density of size 4.6 ×3.8 ×2.6 cm, involving right upper alveolar process extending into right maxillary sinus and osteomeatal complex. Adjacent bony defects noted. Impacted tooth noted within. Thinning of vault of palate noted. Nasal septum was deviate to left (Figure 3).

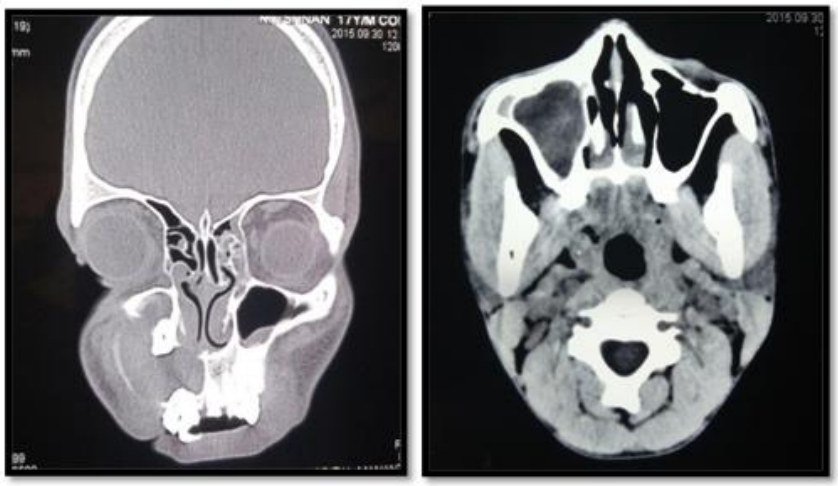

Figure 3: Computed Tomographic views

After the assessment of radiographic findings, we came to the provisional diagnosis of Adenomatoid odontogenic tumor(AOT) of maxilla due to the following reasons- Lesion in the anterior maxilla, which is related to impacted canine, patient of younger age (average age 10-21), slow and continuous growth pattern causing expansion of cortical plates.

The patient underwent incisional biopsy, and the report was contradictory to the clinical diagnosis. Histopathologic findings were suggestive of Calcifying epithelial odontogenic cyst type IA.

Discussion

The uniqueness of Calcifying epithelial odontogenic cyst (CEOC) lies in its dual nature, since it is showing characters of both as cyst and tumor. Controversies were there regarding its classification sysytem, and many types were postulated by different authors (Table1).

CEOC can present in maxilla and mandible, with equal probability and is more common in second decades of life. [5]. Its duality was making its terminologies more complicated, the most accepted one is “Calcifying epithelial odontogenic cyst”. Many times it is co existing with odontomas. It has peripheral variants too. Radiographically, most appear as radiolucent lesions, with radiopacities of internal calcifications.

Volume 2, Issue 6; November-December 2019; Page No.153-156

Table 1: Classification systems of Calcifying Epithelial Odontogenic Cyst

Sl No	Author	Classification
1.	Praetorius[2]	Type I (cystic type) (a) Simple unicystic type, (b) Odontome-producing type, and (c) Ameloblastomatous proliferating type Type II (neoplastic type) [dentinogenic ghost cell tumor]
2.	Toida[3]	Cyst: Calcifying ghost cell odontogenic cyst (CGCOC) Neoplasm: A. Benign - Calcifying ghost cell odontogenic tumor (CGCOT) Cystic variant - Cystic CGCOT Solid variant - Solid CGCOT B. Malignant - Malignant CGCOT Combined lesion: Each of the categories above associated with the following lesions: Odontoma, Ameloblastoma, Other odontogenic lesions
3.	WHO, 2005[4]	Cystic type - CCOT Solid type - DGCT
4.	WHO,2017[4]	Cystic forms - under developmental odontogenic cysts Solid forms – under mixed odontogenic neoplasms

Management of the disease is surgical enucleation, and cystic variant has better prognosis with least chance of recurrence compared to neoplastic type. Recurrence is rare and depends on extent of removal of pathologic tissue.

Conclusion

Odontogenic tumors usually have typical features that help in diagnosis. But this case has atypical clinical and radiological findings, mimicking AOT due to its location and relationship with impacted canine. Diagnosis is definitely challenging here, which makes the present case interesting.

References

1. Bhat S, Shetty SR, Babu SG, Shetty P, Ka F. A rare variant of calcifying odontogenic cyst with ameloblastoma presentation. Stomatologija 2015; 17:131-4.
2. Shear M. Calcifying odontogenic cyst. In: Shear M, editor. Cysts of the Oral Regions. 3 rd ed. Oxford: Wright Publishers; 1992. p. 102-10.
3. Sonawane K, Singaraju M, Gupta I, Singaraju S. Histopathologic diversity of Gorlin's cyst: A study of four cases and review of literature. J Contemp Dent Pract 2011; 12:392-7.
4. J. M. Wright and M. Vered, "Update from the 4th Edition of the World Health Organization Classification of Head and Neck Tumours: Odontogenic and Maxillofacial Bone Tumors," Head & Neck Pathology, vol. 11, no. 1, pp. 68–77, 2017.
5. Gorlin RJ, Pindborg JJ, Odont, Clausen FP, Vickers RA. The calcifying odontogenic cyst -A possible analogue of the cutaneous calcifying epithelioma of Malherbe. An analysis of fifteen cases. Oral Surg Oral Med Oral Pathol 1962; 15:1235-43.

Volume 2, Issue 6; November-December 2019; Page No.153-156