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BIZARRE CELLS IN CEOT- DOES THE COUNT MATTER?

Dental Science

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ABSTRACT

Calcifying epithelial odontogenic tumor (CEOT) is a rare entity comprising about 1% of all odontogenic tumors. It usually occurs in the fourth to sixth decades of life and commonly seen in mandibular posterior regions with or without an impacted teeth. The diagnosis of this entity is dependant mostly on histopathology. We report an unusual case of a patient having CEOT in a less common location showing presence of diffuse bizarre cells, raising a suspicion of malignancy. The objective of this report is to highlight the rare presentation and its histopathologic importance in the diagnosis of CEOT.

KEYWORDS:

Bizarre, CEOT, Malignancy

INTRODUCTION

Bizarre cells are generally referred to cells which have a morphology of irregular-shape and large nuclei with or without hyperchromatism. These cells are usually seen in malignant neoplasms inclusive of calcifying epithelial odontogenic tumour which is a benign entity. CEOT which was first described by Pindborg, is a rare benign odontogenic tumour as it accounts for 0.4%- 3% of odontogenic tumours[1-4]. The objective of this report is to highlight the rarity in clinical appearance and histopathologic findings showing large amount of bizarre cells which aroused a suspicion of malignant lesion.

CASE REPORT

A 21yr old female patient presented with an asymptomatic, slow growing swelling on the left side of the maxilla for a duration of 3 months. On extraoral examination, mild swelling was seen in the maxillary region. Intraorally the swelling was seen in the region of first permanent molar where the tooth was missing clinically. Swelling was well defined, firm on palpation and pale pink in color. The margins were well-defined, extending antero-posteriorly from the distal margin of the tooth 25 to the mesial margin of 27. Provisionally it was considered as Dentigerous cyst as 26 was missing clinically. Differential diagnosis clinically was thought to be any odontogenic cysts or tumors.

Radiographic findings showed radiolucency in the left maxillary molar region which was associated with an impacted tooth with the radiolucency encircling it. The impacted tooth was seen above the root level of the adjacent erupted teeth 27. Radiolucency was seen extending from the occlusal level and encircling the impacted 26 (fig.1). Radiographic differential diagnosis considered was Dentigerous cyst, Unicystic ameloblastoma and Adenomatoid odontogenic tumour.

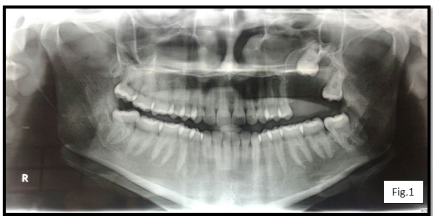

Figure:1- Preoperative panaromic view shows a radiolucency encircling the impacted left first permanent molar.

Considering the differential diagnoses an excisional biopsy along with extraction of the involved tooth was done.

On gross examination, the submitted specimen showed both hard tissue and soft tissue bits. It had the impacted 26 along with soft tissue attachment at the cervical area and multiple bits of soft tissue were submitted.

Histopathological examination of H&E stained specimens showed epithelium in sheets and strands made of squamous cells. It was interspersed with eosinophilic material suggestive of amyloid and few calcifications.

Sheets of squamous cells showed hyperchromatic and pleomorphic nucleus giving a bizarre appearance with prominent intercellular bridges.

Bizarre cells or pleomorphic cells were mononucleated and multinucleated, giving a bizarre appearance that had a diffuse distribution. The density and distribution of the bizarre nuclei seemed to be more in this case giving a suspicion of malignancy of the lesion. Hence the number of bizarre cells were counted per field to understand the distribution of it in the lesion which averaged about 10-15 per field in 400 magnification in 18 fields(Fig 2).

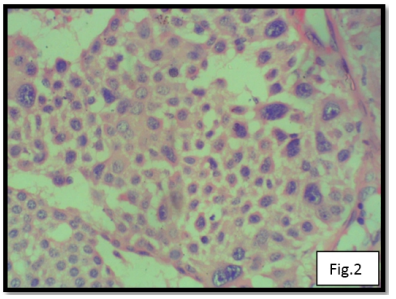

Figure:2- Large number of cells showing bizarre nuclei appearance (H&E stain, 40x)

Few areas of calcification were also seen. No abnormal mitosis was seen. Congo – red staining was done to confirm the presence of amyloid – like material.

Even though it showed diffuse distribution of bizarre of cells, due to the absence of mitotic figures it was diagnosed as Calcifying Epithelial Odontogenic Tumour.

DISCUSSION

CEOT is a benign, slow-growing lesion showing aggressiveness occasionally[3]. The tumour had been reported prior to 1955 under different names, such as ameloblastoma of unusual type with calcification, calcifying ameloblastoma, malignant odontoma, adenoid adamantoblastoma, cystic complex odontoma and as a variant of the simple ameloblastoma[3].

It has two variants, intraosseous with 94% and extraosseous with 6% occurrence[5]. Patients between 4th and 6th decade with mean age of 40 are usually affected with no gender predilection. It commonly occurs in the posterior mandible in the premolar-molar region, twice as much as maxilla[6]. It is also seen to involve premolar region predominantly[4]

In our case, the patient had lesion involving upper alveolus molar region.

Radiologically, it appears either as a diffuse or well-circumscribed unilocular radiolucent lesion or shows combined pattern of radiolucency and radiopacity[7].

Histopathologically the classic form of CEOT shows sheets of polyhedral epithelial cells with well defined borders, eosinophilic and homogenous deposits within sheets of tumor cells and few calcified structures[8,9]. Though presence of cellular and nuclear pleomorphism is not rare in CEOT, the amount and appearance of these bizarre cells seen in this case aroused a suspicion of malignancy.

In our case, even though there was a diffuse distribution of bizarre cells there was no evidence of mitotic figure and invasion into adjacent structures which are the features required for considering malignant CEOT as reported in the literature[9-12].

Bizarre cells being a hallmark of malignancy and also a pathognomonic feature in CEOT, its distribution and its role in benign entity has not been discussed in the literature to the best of our knowledge.

To our knowledge, literature shows no evidence on the amount of bizarre cells that can be seen in CEOT which can be still considered as benign and the substantiation of appearance of pleomorphic cells in a benign tumour. This case report is to arouse a suspicion on the benign nature of the tumor even in case of large amount of bizarre cells and to put forward the thought amongst pathologists.

CONCLUSION

CEOT is a rare odontogenic tumour with distinct histopathological feature reported with 1-3% occurrence among all odontogenic tumours. Though our case showed an atypical occurrence associated with an impacted maxillary first molar, raised a suspicion of malignancy due to diffuse appearance of bizarre cells, it was ruled out considering various factors. Hence for the pathologists to understand the science of appearance of bizarre cells and to evaluate the degree of pleomorphism which can still be considered as a benign entity, further literature evidence is warranted.

REFERENCES

1. Franklin CD, Pindborg JJ. The calcifying epithelial odontogenic tumor: a review and analysis of 113 cases. Oral Surg Oral Med Oral Pathol 1976;42:753-65.
2. Pindborg JJ, Vedtofte P, Reibel J, Praetorius F. The calcifying epithelial odontogenic tumor: a review of recent literature and report of a case. APMIS Suppl 1991;23:152-7.
3. Philipsen HP, Reichart PA. Calcifying epithelial odontogenic tumour: biological profile based on 181 cases from the literature.Oral Oncol 2000;36:17-26.
4. Hada MS, Sable M, Kane SV, Pai PS, Juvekar SL: Calcifying epithelial odontogenic tumor: A clinico-radio-pathological dilemma. J Can Res Ther 2014;10:194-6.
5. Cheng Y, Wright J, Walstad W, Finn D: Calcifying epithelial odontogenic tumor showing microscopic features of potential malignant behavior. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2002;93:287-95
6. Shetty D, Jayade B V, Jayade G, Gopalkrishnan K :Peripheral calcifying epithelial odontogenic tumor - Case repor. Jjournal of oral biology and craniofacial research 4 (2014) 147 -150
7. Neville, B.; Damm, D.; Allen, C.; Bouquot, J. Odontogenic Cysts and Tumors. Oral & Maxillofacial Pathology. St. Lousi Missouri: SAUNDERS/ELSEVIER; 2009.
8. Ai-Ru L, Zhen L, Jian S. Calcifying epithelial odontogenic tumors: a clinicopathologic study of nine cases. J Oral Pathol. 1982;11:399-406.
9. Kawano K, Ono K, Yada N, Takahashi Y, Kashima K, Yokoyama S, Yanagisawa S. Malignant calcifying epithelial odontogenic tumor of the mandible: report of a case with pulmonary metastasis showing remarkable response to platinum derivatives. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2007;104:76-81
10. Franklin CD, Pindborg JJ. The calcifying epithelial odontogenic tumor. A review and analysis of 113 cases. Oral Surg 1976;42:753-65.
11. Cheng Y-SL, Wright JM, Walstad WR, Finn MD. Calcifying epithelial odontogenic tumor showing microscopic features of potential malignant behavior. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2002;93:287-95.
12. Goldenberg D, Sciubba J, Koch W, Tufano RP. Malignant odontogenic tumors: A 22-year experience. Laryngoscope 2004;114:1770-4.