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Research Article

DIAGNOSTIC PITFALL OF CLEAR CELL ODONTOGENIC CARCINOMA: IS IT AN IGNORANCE OR AN ENIGMA

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ABSTRACT

Clear cell odontogenic carcinoma is a rare malignant odontogenic tumour which primarily affects the older age group and mandibular posterior region commonly. Surgical excision is considered as the treatment of choice. Here we present a case report of 63- year old male patient diagnosed with clear cell odontogenic carcinoma of right mandibular posterior region

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INTRODUCTION

Clear cell odontogenic carcinoma is a very rare epithelial malignancy reported by Hansen et al (1985). Initially, it was considered as a benign neoplasm having local invasive behaviour. Some others considered it as an odontogenic malignancy due to its local invasiveness, local aggressiveness and occasional metastasis. [1] Waldren et al described two similar cases; He referred the neoplasm as odontogenic carcinoma. [2] The WHO classification of 1992 divided clear cell tumours of odontogenic origin into four distinct varieties.(1) calcifying epithelial odontogenic origin (2) clear cell odontogenic origin (3) clear cell Ameloblastoma(4) clear cell odontogenic carcinoma. Some pathologists believed that clear cell ameloblastoma and clear cell odontogenic carcinoma are a similar neoplasm, but this is controversial. [2] The WHO classification of 2005 reclassified CCOT as a malignant carcinoma of odontogenic origin. Diagnosis of clear cell tumour has put the surgical pathologists into a diagnostic dilemma. The biologic behaviour and prognostic behaviour of CCOC still remain confused. [3]

Case Report

A 63 -year old male patient visited the outpatient department of Yenepoya dental college, Mangalore with a history of painless

swelling in the lower right mandibular posterior region. On examination, smooth surfaced, mucosal swelling of about 2c.mx 2c.m was observed. On palpation, the swelling was non-tender, firm in consistency with grade II mobility of regional tooth 37 & 38. Bilateral cervical lymph nodes were not palpable. Intraoral peri-apical radiograph revealed unilocular radiolucent lesion with bone loss from the region of 36 to 38. Incisional biopsy was performed under local anaesthesia. Along with incisional biopsy, 37 &38 were also removed. On gross examination, the specimen was un-encapsulated, firm in consistency and creamy white in colour.

Microscopically, sections stained with H&E shows biphasic nature of cells.(Figure1) Connective tissue stroma shows two cell population, mainly clear cells with hyperchromatic nuclei and polygonal cells (figure2&3). Under higher magnification, connective tissue stroma is composed of clear cells having eosinophilic, granular cytoplasm. The nuclei is centrally placed showing pleomorphism. The cells are arranged in nests and separated by thin fibrovascular connective tissue septa (figure4). The polygonal shaped cells show hyperchromatic nuclei with ill -defined cellular borders, arranged in small nests and cords. The abundant cytoplasm of the clear cells showed PAS- positive granules, indicating intra-cytoplasmic glycogen deposition (figure 5).

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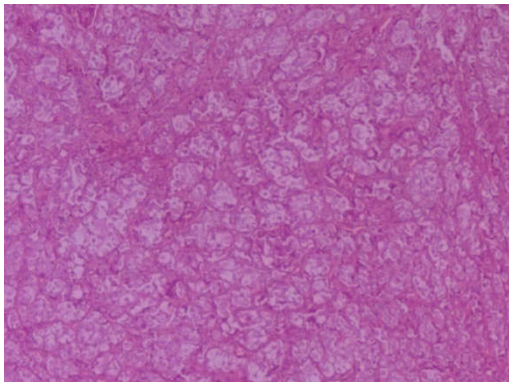


Figure 1 Section showing biphasic nature of the tumor.

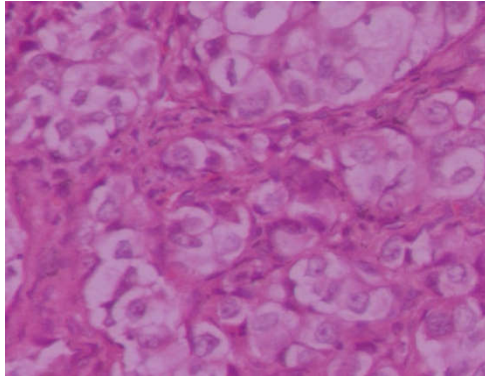


Figure 2 Section shows clear cells with hyperchromatic nuclei

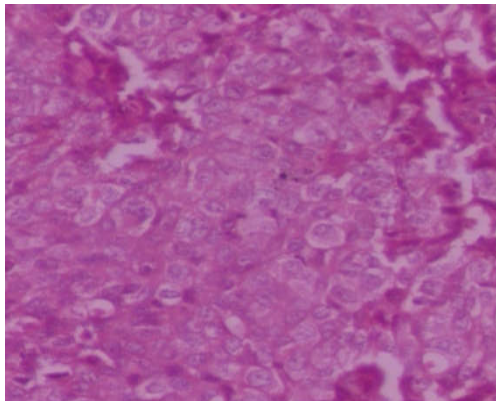


Figure 3 Section shows polygonal shaped cells.

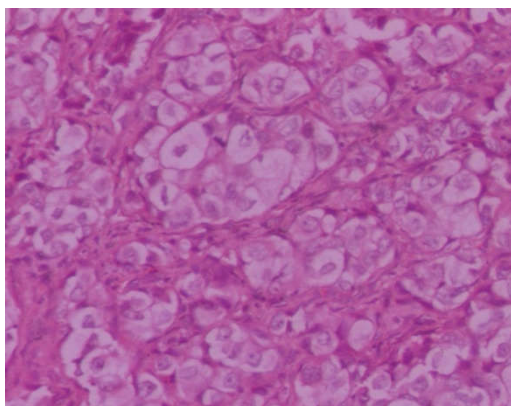


Figure 4 Cells arranged in nests, separated by thin connective tissue core

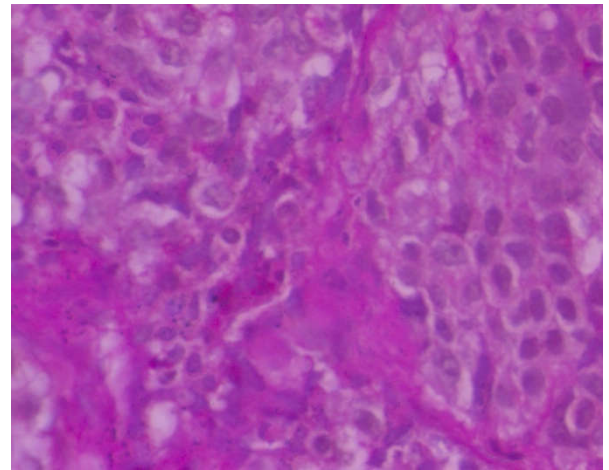


Figure 5 Section shows PAS positive granules

DISCUSSION

Clear cell variant of odontogenic tumours is considered as rare entities of the jaw. [3] A clear cell is a physiologic/ neoplastic cell which is finely vacuolated with central hyperchromatic nuclei, which contains abundant glycogen or other material which does not take up hematoxylin /eosin during staining that makes the cytoplasm appear clear. [4] CCOC has got its clinical presentation at a mean age of 55 years with a slight female predilection. The ratio between female and male ratio is 2:1. Mandibular arches are more commonly affected than maxillary. Tumour is commonly associated with clinical symptoms of tenderness, localized swelling along with the mobility of the teeth. But most of the patients represent with painless, slow-growing enlargement for a few months to years. [5]

Radiographically, most of the CCOC reported cases showed radiolucency. In literature, there are only very few reported cases showing mixed radiolucent and radio -opacity. Histologically CCOC shows three different patterns. The biphasic pattern, monophasic pattern and the ameloblastomatous pattern. The biphasic pattern of CCOC shows two different groups of cells, which are arranged in nests. The two morphological cell types seen in this pattern are the clear cells along with polygonal cells with hyperchromatic nuclei having eosinophilic cytoplasm. The monomorphic pattern shows predominantly clear cells. The last and rarest among the three is the ameloblastomatous pattern which comprises of clear cells arranged in nests within a follicular network. Certain authors consider CCOC and clear cell ameloblastoma as same pathological process, but this didn't gain much recognition. Also, the tumour shows other histopathological features like nuclear hyperchromatism and pleomorphism and a varying number of mitotic figures. These features vary according to each case. Rarely the tumour shows encapsulation, it has the potentiality to metastasize and invade into the medullary bone, muscle and neural tissue. [5, 6]

The diagnosis of clear cell odontogenic carcinoma is very much important due to its close resemblance to other pathological entities like calcifying epithelial odontogenic tumour, mucoepidermoid carcinoma, myoepithelial carcinoma, epithelial-myoepithelial carcinoma, clear cell ameloblastoma, hyalinizing clear cell carcinoma, amelanotic melanoma, metastatic renal cell carcinoma. The brief features like histopathology, special stain and immunohistochemical

markers to distinguish between clear cell odontogenic carcinoma and other pathological entities are enumerated in table 1. [7]

Differential Diagnosis	Histological features	Special stains & IHC
Calcifying epithelial odontogenic tumour	<ul style="list-style-type: none"> Cells are arranged in cords, nests or sheets having nuclear pleomorphism and prominent intercellular ridges. Amyloid deposits. Psammoma bodies. 	<ul style="list-style-type: none"> Amyloid deposits will be positive for Congo red stain. Clear cells can be PAS positive or negative.
Mucoepidermoid Carcinoma	<ul style="list-style-type: none"> Three groups of cells- epitheloid like cells, intermediate cells, mucous cells. Cystic spaces containing mucin. 	<ul style="list-style-type: none"> Mucous cell will be PAS positive, diastase resistant. Mucous cells show mucicarmine, alcian blue positive. IHC-CK 7, 8 & 13- positive for epithelial cells. CK 14 positive for epithelial cells & intermediate cells CK 19 positive for epithelial cells and mucous cells.
Myoepithelial Carcinoma	<ul style="list-style-type: none"> Cells are arranged in sheets. Presence of spindle cells, plasmacytoid cells, clear /epidermoid like cells. Cells show pleomorphism and mitotic activity. 	<ul style="list-style-type: none"> Clear cells are PAS positive Clear cells are – S100, Vimentin, SMA, Calponin, Caldespin positive
Clear cell Ameloblastoma	<ul style="list-style-type: none"> Presence of peripheral tall columnar cells with palisading nuclei and reversal of polarity. Clear cells are arranged in cords, nests, trabeculae or islands. 	<p>Calretinin. CK8, 13, 19 & AE1/3- +VE.</p>
Hyalinising Clear Cell Carcinoma	<ul style="list-style-type: none"> Connective tissue shows hyalinization. Cells show a biphasic pattern. 	<p>PAN CK positive.</p>
Epithelial-Myoepithelial Carcinoma	<ul style="list-style-type: none"> Clear cells have centrally placed nuclei, admixed with pleomorphic polygonal epithelial cells. Cells are arranged in nests. 	<p>Cells show positive for Calponin, Caldesmin, CK, EMA, PAN CK, S-100, vimentin, SMA.</p>
Amelanotic Melanoma	<ul style="list-style-type: none"> Cells will be polygonal with clear to weakly eosinophilic cytoplasm. 	<p>Cell shows positive for S-100, Melan A, and HMB-45.</p>
Metastatic Renal cell Carcinoma	<ul style="list-style-type: none"> Cells are arranged in solid, organoid pattern. High vascularity. 	<ul style="list-style-type: none"> Mucicarmine positive. Clear cells are positive for vimentin, renal cell carcinoma antigen.

The treatment modality for clear cell odontogenic carcinoma is surgical resection with a wide margin with /without lymph node dissection. Adjuvant therapy like chemotherapy and radiotherapy is indicated in certain cases. Recurrence rate is more in cases having lymph node metastasis. Keeping the above facts patients diagnosed with clear cell odontogenic carcinoma should have a long term follow-up in order to avoid recurrence and distant metastasis. [8]

References

1. Kwon IJ, Kim SM, Amponsah EK, Myoung H, Lee JH, Lee SK. Mandibular clear cell odontogenic carcinoma. *World journal of surgical oncology*. 2015 Dec; 13(1):284.
2. Ferreira S, Faverani LP, SANTOS GM, Martins EP, Garcia Junior IR. Clear cell odontogenic carcinoma of the mandible: a treatment strategy. *Journal of Applied Oral Science*. 2018; 26.
3. Jayapalan CS, George A, Noufal A, Pynadath MK, Mangalath U. Clear Cell Odontogenic Carcinoma (CCOC): Mini-Review of Literature and Case Report of Mandibular Radiolucency in 17-year Girl. *Diagnostic Pathology Open*. 2016; 1(120):2.
4. Walia C, Chatterjee RP, Kundu S, Roy S. Clinical enigma: A rare case of clear cell odontogenic carcinoma. *Contemporary clinical dentistry*. 2015; 6(4):559.
5. Hideshi Yamamoto, Madoka Inui, Atsusi Mori, Toshiro Tagawa. Clear cell odontogenic carcinoma: A case report and literature review of odontogenic tumors with clear cells. *Oral Surgery Oral Medicine Oral Pathology*. 1998; 86(1).
6. Chera BS, Villaret DB, Orlando CA, Mendenhall WM. Clear cell odontogenic carcinoma of the maxilla: a case report and literature review. *American journal of otolaryngology*. 2008 Jul. 1; 29(4):284-90.
7. Swain N, Dhariwal R, Ray JG. Clear cell odontogenic carcinoma of maxilla: A case report and mini review. *Journal of oral and maxillofacial pathology*. 2013 Jan; 17(1):89.
8. Harbhajanka A, Lamzabi I, Jain R, Gattuso P, Kluskens L. Cytomorphology and immunohistochemistry of a recurrent clear cell odontogenic carcinoma with molecular analysis: A case report with review of literature. *Diagnostic cytopathology*. 2015 Sep; 43(9):743-6.

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