

# CASE REPORT

## A RARE CASE REPORT: CLEAR CELL ODONTOGENIC CARCINOMA INVOLVING MAXILLA

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**ABSTRACT:** Clear cell odontogenic carcinoma (CCOC) is a rare tumor occurring predominantly in posterior mandible during 5-7 decade with a female preponderance, potentially aggressive tumor capable of frequent recurrence and loco regional spread and distant metastasis. Till date around 75 cases have been reported in literature. In this paper, we report case of a 60-year-old male with CCOC in maxilla extending from distal surface of the right maxillary canine to retromolar area and involving maxillary sinus with no association with impacted teeth. The diagnosis was confirmed by aspiration cytology and histologically, the tumor was composed of sheets of epithelial cells, with areas of clear cell changes. The presence of clear cells in the histological sections, accounts for the aggressive nature of the tumor simulating the clinical appearance. Prevention of recurrence can be achieved by radical resection.

**KEYWORDS:** Biphasic pattern, clear cell odontogenic carcinoma, clear cells.

**INTRODUCTION:** CCOC is first described by two separate group of researcher hensen at all and Waldron at all in 1985.<sup>(1),(2)</sup>

In 1992 it was included in benign tumor with a capacity of locally invasive growth and was more aggressive than ameloblastoma.

Reichart and Philipsen proposed a revision of the classification of odontogenic tumors in 2003, clearly considering the clear cell odontogenic tumor as a carcinoma.<sup>(2)</sup>

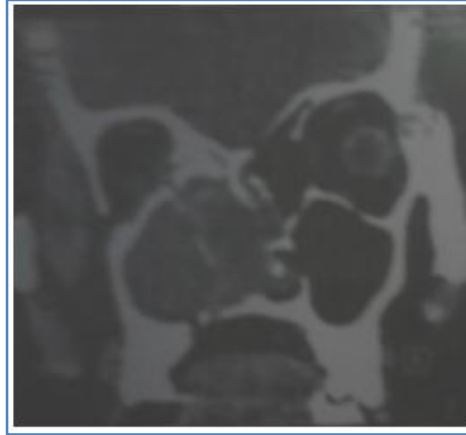
Now WHO classification of 2005 ccoc denoted as a malignant tumor of odontogenic origin.<sup>(3)</sup> In the past, the terms "clear cell ameloblastoma" and "clear cell odontogenic tumor" were synonymous for CCOC.<sup>(3),(4)</sup>

**CASE REPORT:** A 65 year old male reported to our department of otorhinolaryngology of Govt. Medical College Kota with a one year h/o of painless swelling in upper jaw region with blocked nose and mass coming out through right nostril. On inspection a smooth mucosal coloured swelling of around 4 cm wide with loosening of premolar teeth in jaw.

On palpation swelling was non tender, non-fluctuant and firm in consistency. Cervical Lymph node are nor palpable.

Medical history of patient is known case of asthma taking t/t for it. Clinical examination revealed a generally fit man, with a blood pressure of 150/96 mm Hg, with a massive swelling of the face over the right maxilla. The overlying skin was freely movable. NO ANY OTHER significant h/o present. Routine blood INV is hb-9.3 blood sugar fasting is 65mg/dl and blood urea is 22mg/dl. Radiological investigation CT shows a radiolucent mass of size ap 8.5, H 4.6, & W 3.8 cm, seen in RT nasal cavity and RT maxillary sinus. Mass is adherent to nasal wall and involving septum, caudally mass extended to RT side of hard palate and cranially mass involving the RT ethmoidal, sphenoidal and RT frontal sinus, medially up to the orbital wall erosion, superolat destruction on in orbital wall.

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**Figure 1: CT Image of PT**

Now with provisional diagnosis intra osseous odontogenic tumour punch biopsy was performed under local anesthesia. Punch biopsy HPE shows sheets and islands of large clear cells separated by a delicate fibrous connective tissue stroma. Under higher magnification, biphasic population of cells characterized by polygonal, clear cells and hyperchromatic, basaloid cells with eosinophilic cytoplasm were seen. Occasional islands showed peripheral palisading. Nuclear pleomorphism was minimal, and mitotic figures were rare.

PT is treated with maxillectomy and post-operative period is uneventful and stitch removed after 12<sup>th</sup> day. After that radiotherapy is given and after one year of follow up there is no recurrence.

**DISCUSSION:** CHOC is now considered as a rare malignant neoplasm with peak incidence in 5<sup>th</sup> -7<sup>th</sup> decade with female: male: 17: 10 and more in mandible than maxilla. Post mandible involve more than anterior mandible. Classical presentation is painless swelling of mandible and maxilla.

Histologically show 3 type of pattern:<sup>(5)</sup> biphasic, monophasic and ameloblastomatous, biphasic pattern of tumor growth comprise of nests of cell containing eosinophilic cytoplasm. Monophasic pattern comprises only of clear cell while ameloblastomatous pattern,<sup>(6)</sup> resembles the growth pattern of ameloblast with nest of cell showing cystic changes and squamous differentiation and peripheral nuclear palisading with reverse polarity. The degree of nuclear pleomorphism, hyperchromatism, and number of mitoses in CCOC are quite variable. In general, encapsulation is seldom seen and it frequently invades the medullary bone, muscle Glycogen storage is quite common in these tumors displaying PAS positivity which is diastase sensitive.<sup>(7),(8),(9),(10)</sup>

Differential diagnosis can be categorized in odontogenic and nonodontogenic. Odontogenic included odontogenic cyst, clear cell variant of calcifying epithelial odontogenic tumor, clear cell ameloblastoma and CCOC. Non odontogenic tumor with clear cell feature include Acinic cell carcinoma, squamous cell ca with clear cell, clear cell oncocytoma, glycogen rich salivary tumor, hyalinising clear cell carcinoma, sebaceous cell tumor, epithelial myoepithelial ca, clear cell variant of adenocarcinoma of salivary gland tumor.

Intraosseous salivary gland tumor with prominent cytoplasm clearing include epithelial-myoeplithelial carcinoma myoeplithelial cell are immunoreactive for s-100 protein, vimentin., the clear amyloid deposition and/or calcifications in the tumor cells or the intercellular space must be absent

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to exclude cell variant of calcifying epithelial odontogenic tumor on Congo red-stained slides. Finally, a metastatic lesion can be excluded on clinical and radiological grounds. Immunohistochemistry may be useful in the differential diagnosis of CCOC, as clear cell salivary gland tumors tend to express positive results for S-100 protein, CK, vimentin, and muscle actin, whereas odontogenic tumors with clear cell differentiation react negatively for vimentin and muscle actin. Treatment for CCOC is primarily resection with a wide margin. Other treatment modality which is given in literature is enucleation, curettage, surgical resection with radiotherapy, chemotherapy. Adjuvant radiation therapy may be beneficial in patients with extensive soft tissue or perineural invasion, in cases in which tumor-free margins are not possible or in patients with positive nodes and/or extra capsular spread.

**CONCLUSION:** CCOC is a rare malignant odontogenic neoplasm with benign looking histology. The acknowledgment of this rare tumor and its distinction from other clear cell neoplasms is crucial in establishing the appropriate therapeutic plan and there are very less number of cases are reported so that ideal treatment modality is still to be decide.

Metastatic lesion to bone must be considered they usually come from breast (33%) LUNG (18%) KIDNEY (16%) THYROID (6%) prostate and colon. clear cell ca of kidney closely resemble CHOC. salivary gland tumors (Primary or Secondary) and metastatic renal carcinoma. The latter two were eliminated on morphological, immunohisto chemical and clinical grounds.

CHOC can be distinguished from the clear cell variant of calcifying epithelial odontogenic tumor lacks calcification and amyloid deposit.

Lymph node metastasis is infrequent but it's frequent in the recurrence cases. Surgical control of CCOC with en block resection of bone and soft tissue decrease the risk of recurrence. Regional lymph node dissection can be performed for staging and t/t of regional ds. Adjuvant radiotherapy may contribute to control local ds.

**CONCLUSION:** At the end we conclude that the CHOC is not a very common tumor. Fewer cases are reported involving maxilla, to know the actual origin of tumor are very difficult whether it's from odontogenic epithelium, or from salivary gland, or from secondary metastasis from kidney. To rule out distant metastasis bone scientigraphy is best modality.

Best radiography technique is CT scanning with 3D reconstruction.

Best treatment modality is wide free margin surgical resection with radiotherapy.

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