Case Report

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CLEAR CELL ODONTOGENIC CARCINOMA OF MANDIBLE: A RARE CASE REPORT

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ABSTRACT:

Malignant odontogenic tumors (MOT) are rare entities that represent up to 6.1% of all odontogenic tumors. Here we report a case of clear cell odontogenic carcinoma in a 63 year old female patient and describing it's clinical, radiographic, histological and immunohistochemical features. The patient was referred to the nearest tertiary cancer centre. The patient underwent wide surgical excision. Regular follow-up was done for 1 year and no recurrence was reported.

Keywords: Odontogenic, clear cell odontogenic carcinoma

INTRODUCTION:

Malignant odontogenic tumors (MOT) are rare entities that represent up to 6.1% of all odontogenic tumors.[1] Clear cell odontogenic carcinoma (CCOC) is a rare neoplasm of the jaws and was first described by Hansen and Waldron et al in 1985 and they termed it as clear cell odontogenic tumor considering its local destructive nature.[2]. CCOC was initially known as clear cell odontogenic tumor or clear cell ameloblastoma. In 1992, CCOC was classified as odontogenic tumor by the WHO.[3] Most of the cases occur during the 5th to 7th decades of life with marked female predilection. Radiographically, a poorly delineated, unilocular or multilocular radiolucent lesion with prominent bone destruction is observed.[4] Owing to its behavior as an infiltrative neoplasm with a marked tendency for local recurrence, regional lymph node metastasis and possible distant pulmonary metastasis, in the WHO classification of 2005, CCOC was denoted as a malignant tumor of odontogenic origin.[5] Here we report a case of clear cell odontogenic carcinoma in a 63 year old female patient and describing it's clinical, radiographic ,histological and immunohistochemical features.

CASE-REPORT:

A 63-year-old female patient reported to the Department of Oral Medicine and Radiology, with a complaint of swelling on the left lower gums of 1 year duration. Swelling was associated with pain and mobility. The patient had extracted mobile teeth 1 year back which

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relieved her symptoms hence did not attend furthur follow-ups. 1 month back patient developed paresthesia over the lower lip towards left side for which she consulted a doctor and was referred here. She had underwent multiple extractions which was uneventful. No relevant medical, family and personal history. General examination was non-contributory.

No obvious swelling present on extraoral examination. On intraoral examination a swelling was noted in the left lower edentulous alveolar ridge in relation to 33 to 38 region of size 5x4 cm extending anteroposteriorly from mesial of 33 to the mesial of 38 in buccal aspect and mesial of 35 to mesial of 38 in lingual aspect and superoinferiorly from the alveolar ridge to inferiorly obliterating the vestibule. Inferior extent was not visualized in both buccal and lingual aspect. Borders were ill-defined. Surface mucosa appeared similar to the adjacent mucosa. No bleeding, discharge, ulcerations, sinus openings, visible pulsations noted. On palpation the swelling was slightly larger in size 7x4 cm and was extending from mesial of 32 to distal of 38. The swelling was non tender and hard. Surface was bossellated and margin except on the inferior aspect was well defined. No discharge, bleeding and palpable pulsations. Grade I mobilily of 34 and 38 were noted.

Radiographic examination was performed to assess the nature and extent of the lesion. A panoramic radiograph revealed a multilocular radiolucency in the left body of mandible of size 7x5cm extending anteroposteriorly from the distal aspect of apex of 32 to the distal aspect of apex of 38 and superoinferiorly from the alveolar crest in relation to 36, 37 and 38 region to the lower border of mandible. The borders were well-defined and corticated except in the anterior and superior aspect. Margins were irregular. Multiple curved septae of variable thickness was seen within the lesion. No evidence of calcification was noted. Resorption of the apical part of root of 34 was noted. Destruction of alveolar crest in relation to 35, 35 and 37 region seen. Destruction of the lower border of mandible seen. Inferior alveolar canal cannot be traced anteriorly. A true occlusal radiograph of left side body of mandible showed a multilocular radiolucency with expansion of the lingual cortical plate. CECT sections showed lytic lesion involving the left side of mandible with enhancing soft tissue component, causing destruction and cortical break. Laterally the lesion was extending into left gingivobuccal sulcus with lost fat plane. Medially abutting left half of tongue and anteriorly into the subcutaneous tissue of face. Electric pulp testing of the teeth adjacent to the lesion was also carried out and 34, 38 showed delayed response. Based on the clinical and radiographic findings a provisional diagnosis of ameloblastoma was made.

Blood investigations and urine analysis were within normal limits. Incisional biopsy was performed and H&E stained sections showed sheets and islands of oval or polyhedral cells with dark staining eccentric nuclei with clear cytoplasm(clear cells) and round to ovoid cells with basophilic nucleus and eosinophilic cytoplasm in a moderately collagenous stroma. Cellular pleomorphism and mitosis were not observed. Mucicarmine was negative. Immunohistochemistry was performed for CK, S100 and p63.CK was strongly positive in the tumor islands.p63 nuclear positivity and S100 cytoplasmic staining were also observed.Based on the radiographic, histopathological and immunohistochemical findings, confirmatory diagnosis of clear cell odontogenic carcinoma was made. The patient was referred to the nearest tertiary cancer centre. The patient underwent wide surgical excision. Regular follow-up was done for 1 year and no recurrence was reported.

DISCUSSION:

Clear cell odontogenic carcinoma (CCOC) is a low-grade odontogenic carcinoma showing cells with clear cytoplasm arranged in sheets and islands[6]. The histogenesis of CCOC is unclear literature suggests that clear cells in jaw lesions originate from dental lamina or cell rests of Malassez.[7] Literature review reveals that CCOC commonly occurs in the sixth decade of life (range 17–89 years).[8] CCOC characteristically has female preponderance with male:female ratio 1:1.8 and affects mandible more frequently than maxilla.[4] Clinically, it presents as asymptomatic, slow growing swelling of long duration with a mean size of 4 cm in diameter; however, cases with pain, tooth mobility, paresthesia, and nonhealing ulcerations have been reported[1]. Radiographically, CCOC manifests as either unilocular or multilocular radiolucency which may be well or poorly demarcated.[9] In the present case, multilocular radiolucent lesion on left posterior mandible was observed.

The light microscopic features of CCOC reveal three histological patterns: Biphasic, monophasic, and ameloblastomatous. The most common biphasic pattern shows two sets of cellular population arranged in sheets and islands. Center cells are clear, round to polygonal in shape mixed with another population of cuboidal to columnar cells with eosinophilic cytoplasm at the periphery. The present case also showed Biphasic pattern. The monophasic pattern has islands of clear cells wholly. The ameloblastomatous pattern is least common, characterized by the presence of clear cells inside the follicular network.[10,11] The degree of nuclear pleomorphism, hyperchromatism, and mitotic figures are variable. Encapsulation is rarely present. It has a higher tendency for the invasion to the medullary bone, muscle, and the neural tissue.[11]

Differential diagnoses include other clear cell lesions of head and neck region like calcifying epithelial odontogenic tumor(CCCEOT), clear cell mucoepidermoid carcinoma(CCMEC), myoepithelial carcinoma, hyalinising clear cell carcinoma(CCC), epithelial-myoepithelial carcinoma, amelanotic melanoma, and metastatic renal cell carcinoma. Absence of amyloid and Liesegang's ring calcification in the present case negated the possibility of CCCEOT. Lack of mucin and negative staining for alpha SMA, Vimentin and Calponin, ruled out CCMEC.[12] Intraosseous location with central bone destruction, presence of palisaded peripheral cells, differentiated the present case from CCC.[8] Absence of sinusoidal vascularity, intramural hemorrhage, noncontributory findings of USG abdomen and chest radiographs ruled out the possibility of metastatic renal cell carcinoma.[13]Clear cells appear to be clear due to the presence of an intracellular accumulation of nonstaining compounds, such as glycogen, mucopolysaccharides, lipids, and mucin. It is believed that during late bell stage, cells of inner enamel epithelium undergo histodifferentiation to presecretory ameloblast. Electron microscope shows the presence of lysosomes, mitochondria, tonofilaments, and desmosomes. It is now known that bone morphogenetic protein (BMP-2) plays a crucial role in regulating Ms × 1 and Ms × 2 along with Dl × 2. The combined effect of Ms × 1, Ms × 2, and Dl × 2 influences Dl × 3, which normally help ameloblast cell to differentiate. Absence of these two homobox genes may be due to dysregulation of BMP-2 expression affecting the terminal differentiation of ameloblast cell. In addition to this, the genomic analysis also revealed a polypoid population of cells with DNA index of 1.93 and overall S-phase of 10.2% suggesting chromosomal alterations in cases of CCOC.[13]

The overall recurrence rate is as high as 55%. The treatment should aim at wide surgical resection with tumor-free margins adjunct to radiotherapy. A long-term follow-up is necessary to look for any loco-regional recurrence and distant metastasis.[7]

CONCLUSION:

Till date only 97 cases of CCOC have been reported in English literature excluding the present case. Differentiating CCOC from other clear cell tumors of head and neck is very difficult. Clinical, radiographic presentation and histopathological pattern primarily aided in the diagnosis of CCOC in our case. Correlation of histologic type of CCOC with prognosis has not studied yet. CCOC warrants a close and long-term follow-up since it has a low survival rate and recurrence and metastasis were reported as late as 20 years after first intervention.

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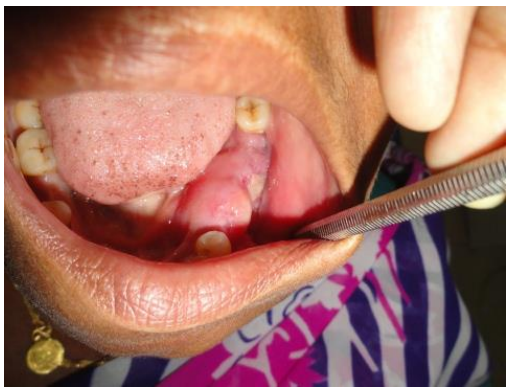
FIGURES:



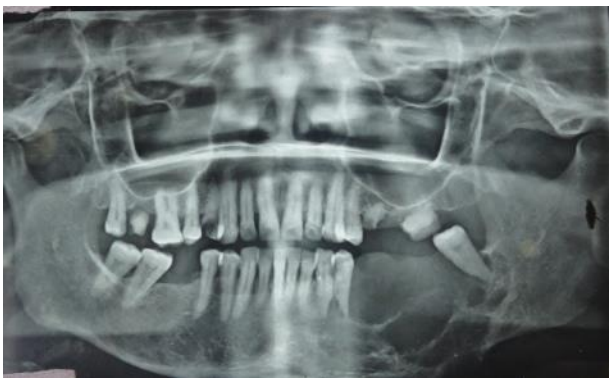
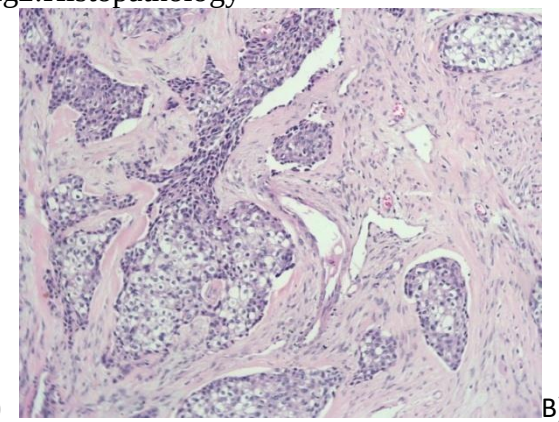
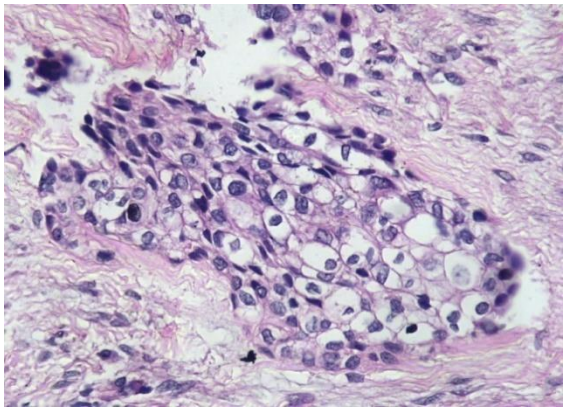


Fig1:A) Intraoral photograph B) Orthopantomogram showing multilocular radiolucency on left posterior mandible

Fig2:Histopathology

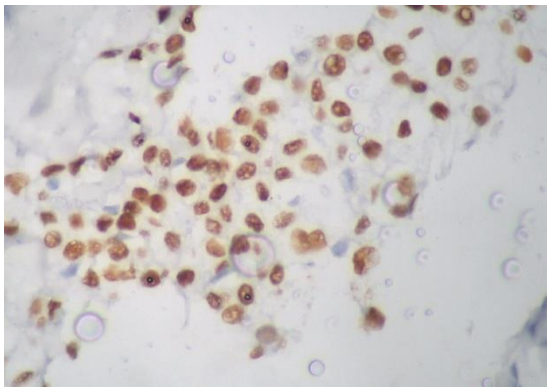
A) 

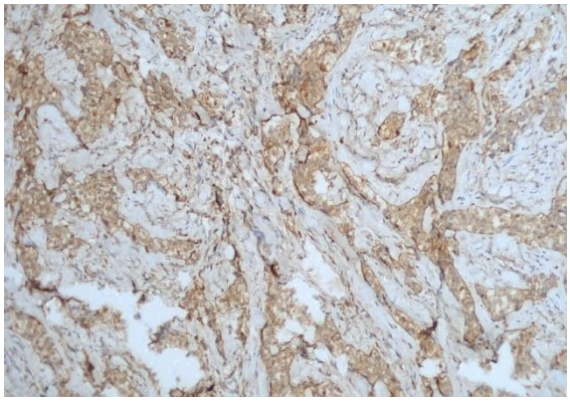
 B)

A) Tumor islands showing biphasic pattern with centrally placed clear cells and peripheral basaloid cells (H and E, 10x)

B) Central clear cells with centric small basophilic nucleus, distinct cell membrane and clear cytoplasm (H and E ,40x)

Fig3: Immunohistochemical staining

A) 

 B)

A)P63 nuclear positivity B) S100 cytoplasmic staining.

Fig 4: CK positivity in the tumor islands.