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## Case Report

## Clear cell odontogenic carcinoma: A case report and literature review

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## ABSTRACT

Clear cell odontogenic carcinoma (CCOC) is a rare odontogenic tumor of the jaw with a particular histology and a deceptive behavior. The clinical presentation may be misleading, that is why the contribution of incisional biopsy, molecular biology, immunohistochemistry and cytogenetics is essential in the diagnostic process.

This review article aims to discuss its various aspects, summarizing the currently published studies of CCOC to enhance the diagnosis and to present a case report of a 41-year-old man that presented to the Department of Maxillofacial Surgery at Gemelli University Hospital in Rome, Italy.

## 1. Introduction

Clear cell odontogenic carcinoma (CCOC) is a rare intraosseous tumor of the jaw.

Clear cell changes have been identified in various benign and malignant tumors of epithelial, melanocytic, mesenchymal and hematopoietic origin but are perceived as a rare occurrence in the head and neck region. It was formerly defined as Clear cell odontogenic tumor (CCOT) or Clear cell ameloblastoma (CCA).

Hanson et al. were the first to describe this tumor in 1985 and they classified it as a benign odontogenic tumor. According to the World Health Organization (WHO) classification of odontogenic tumors in 1992, CCOC was classified as a benign neoplasm with the capacity of undergoing local invasion. In 2003, a revision of the classification of odontogenic tumors was advanced by Reichart and Philipsen, in which the clear cell odontogenic tumor was identified as a carcinoma. In 2005, CCOC was reclassified by the WHO as a tumor of low grade malignancy, characterized by locally destructive and aggressive behavior with local recurrence, regional lymph node metastasis, and rare distant metastasis [1,2].

Although a long time has passed since its first description, there is still much to discover about this type of tumor. Epidemiology,

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differentiation aspects, predictive factors and treatment outcomes are yet a topic of debate.

Here, we report a case of a maxillary clear cell odontogenic carcinoma affecting a 41-year-old male.

## 2. Case report

A 41-year-old man presented to the Department of Oral and Maxillofacial Surgery at Gemelli University Hospital with a 12-month history of a painless and gradually growing lesion of the attached gingiva of the dental element 5. The patient had been previously assessed by a general dentist who referred the patient to a specialist, for a second opinion. A complete history was taken and the examination was performed.

There was no significant past medical, surgical or dental history.

The extra-oral examination showed no anomalies. The intraoral examination revealed an alteration of the attached gingiva of the region of the elements 5 and 6 with pseudo-nodular swelling. The overlying alveolar mucosa was normal. The involved teeth were sound, positive at cold sensitivity test and without mobility. No swollen cervical lymph nodes were found, and the remainder of the physical examination was within normal limits.

A computed tomography scan without administration of contrast revealed: "presence of an erosive area of the cortical and trabecular bone located in correspondence of the upper right hemiarch, in the interdental space between dental elements 5 and 6. The aforementioned area, 14 mm in size CC x 6.6 RL, x 4 AP, causes evident erosion of the maxillary superficial cortical profile and disappearance of the trabecular design in the interdental space. There are no changes in the deep cortical profile of the maxillary bone. There is no involvement of the roots of the aforementioned dental elements, which however are in continuity with the erosive area" (Fig. 1).

Based on the clinical and radiographic findings a preventive histological diagnosis was considered for treatment planning.

An incisional biopsy was performed under local anesthesia and revealed the presence of a CCOC.

A surgical resection followed by reconstruction with local flaps was planned.

The patient was admitted to the hospital for intra-oral excision of the tumor under general anesthesia.

An oscillating saw was used to perform a partial maxillectomy involving the antral floor, the alveolar bone, and teeth 4 to 6, allowing a complete en bloc resection of the tumor.

The buccal fat pad was harvested and moved to repair the oroantral communication.

The excised tumor was sent for histopathological examination.

On macroscopic examination, the tumor mass was of 1.6 cm in maximum diameter, with infiltration the maxillary bone.



**Fig. 1.** Head CT without contrast performed before surgery showed the presence of an erosive area of the cortical and trabecular bone located in correspondence of the upper right hemiarch, in the interdental space between 1.3 and 1.4.

The neoplasm was arranged in small nests, trabeculae and strands connected to the superficial mucosal epithelium and composed of epithelial cells with pale eosinophilic to clear cytoplasm, well demarcated cell membranes and oval to round dark nuclei, with mild nuclear pleomorphism and scarce mitotic figures. Clear cell cytoplasm contained diastase-sensitive PAS-positive material indicative of glycogen. Peripheral cells showed sometimes a partial palisading with reversed nuclear polarity, giving to the neoplasm a biphasic pattern (Fig. 2).

Immunohistochemical analyses revealed positive staining for CK 5/6, p63, CK14 and negative for BER-EP4, SMA, calretinin and S100. The index of proliferation (ki67) was about 10–12%. Based on these findings, the lesion was diagnosed as a CCOC (Fig. 3).

Healing was uneventful without any complications and follow-up was performed at 3, 6, and 12 months.

12 months after the surgery, clinical examination and radiographs showed good healing of both of the bone and the soft tissues with no local recurrence (Fig. 4).

### 3. Discussion

To the best of our knowledge, 107 cases of CCOC have been reported (excluding the present one) in the English literature to date. CCOC occurs most often in the fifth decade of life, with a range that goes from 14 to 89 years. Middle-aged women seem to be involved more often than the male counterparts. There seems to be no ethnicity predilection. In the current literature, no cases of Italian patients affected by CCOC are reported. The posterior mandible is the most frequently involved site, followed by the anterior mandible, maxilla and palate [2,3].

CCOC often presents itself as an asymptomatic, painless and slow-growing lesion, according to Loyola et al. [2] and the average dimensions at diagnosis is 4 cm in diameter.

The onset of pain and tooth mobility is common and occurs approximately in one-third of patients, whereas bleeding, paresthesia, and non-healing ulcerations are less commonly reported.

The diagnosis is often late due to these deceptive and misleading signs, the duration of symptoms prior to diagnosis is reported to be of 47.62–91.9 months with a range of 1–504 months, which is uncommonly long for a malignancy [2].

In our case, the patient was a 41-year old male presenting a 12-month history of a painless and gradually increasing lesion of the attached gingiva of the 5 region.

Radiological aspects are unspecific and poorly defined. Radiographically, CCOC is similar to other osteolytic jaw lesions and there are no typical radiographic features, X-ray images are almost always radiolucent with irregular margins; other common features are multiple cortical perforations, root resorption and in some cases, soft-tissue invasion is also noted. A minority of cases exhibit a mixed radiolucent–radiopaque lesion [3–5].

The differential diagnosis of CCOC is a challenge, it comprises odontogenic and non-odontogenic tumors with clear cell changes [6], like acinic cell carcinoma, myoepithelial carcinoma, clear cell variant of calcifying epithelial odontogenic tumor (CCCEOT), ameloblastic carcinoma, intraosseous variant of clear cell mucoepidermoid carcinoma (CCMEC), hyalinizing clear cell carcinoma (CCC), intraosseous variant of melanoma and amelanotic melanoma, metastatic tumors from kidney, thyroid, and prostate. (Table 1) [7].

Therefore, when doing the diagnosis of a patient with CCOC, a thorough investigation is important to understand if we are encountering a primary or a secondary lesion.

Clear cells are a hallmark, but they are not considered to be pathognomonic for CCOC [8].

A clear cell is a cell characterized by a clear cytoplasm when stained with Haematoxylin and Eosin. Normally, clear cells are secretory cells in the epithelium, and are one of the components of eccrine sweat glands. A clear cell's plasma membrane is highly folded, more so on the apical and lateral surfaces. The cytoplasm of clear cells contains large amounts of glycogen and many

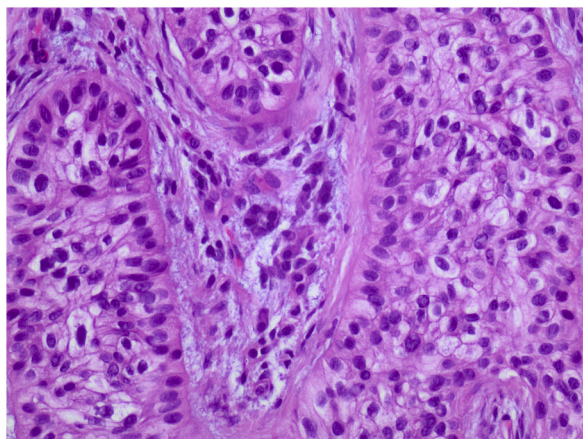
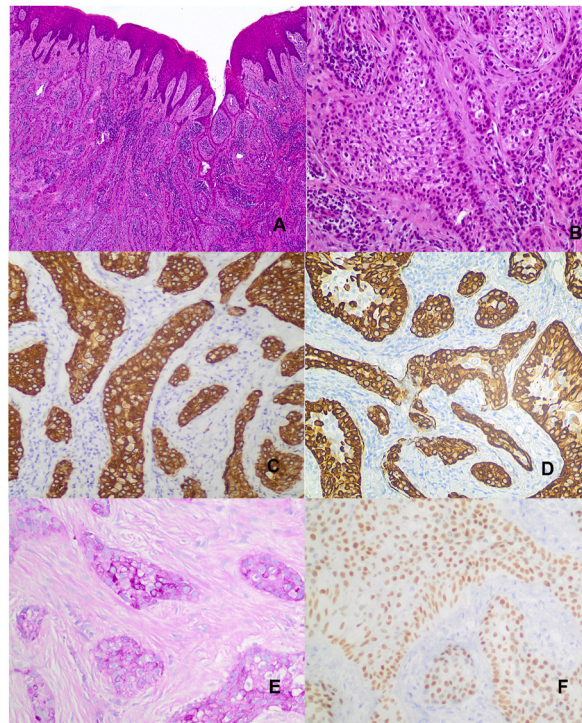


Fig. 2. Clear cell odontogenic carcinoma (Haematoxylin and eosin, original magnification 200×).



**Fig. 3.** Clear cell odontogenic carcinoma (Haematoxylin and eosin; **A**, original magnification 20 $\times$ , **B**, original magnification 100 $\times$ ). **C**. Cytoplasm of neoplastic cells was positive for PAS. (Pas stain, original magnification 100 $\times$ ). Neoplastic cells showed immunoreactivity for CK5/6 (**D**), for CK14 (**E**) and for p63 (**F**). (Haematoxylin counterstaining, original magnification 100 $\times$ ).

mitochondria [9,10].

Clear cells in a lesion may also result from fixation artifacts, cytoplasmic accumulation of water, glycogen, lipids, mucins, hydropic degeneration of organelles, etc [11].

CCOC is identified by the proliferation of neoplastic epithelial cells with clear cytoplasm arranged in islands and strands. Three kinds of cells can be found in CCOC: basaloid to polygonal clear cells, basaloid to polygonal pale eosinophilic cells, and columnar cells with ameloblast-like differentiation.

Depending on the proportion of these cells in the tumor, three different subtypes can be distinguished: (1) monophasic: formed almost entirely of clear cells with well-defined borders and centrally located nuclei;

- (2) biphasic: is characterized by oval and linear nests of large cells intermixed with smaller islands of smaller polygonal cells with eosinophilic cytoplasm
- (3) ameloblastic: characterized by columnar cells with ameloblastic differentiation at the periphery of islands [3,12,13].

The majority of tumors reported have a biphasic pattern, while the ameloblastic pattern is the least common type.

When present, the biphasic pattern with peripheric ameloblastoma-like cells aids in the recognition of an odontogenic neoplasm and not a salivary clear cell carcinoma, narrowing the differential diagnosis considerably.

The monophasic variant, instead, is more arduous to be distinguished from central mucoepidermoid carcinoma, metastatic renal cell carcinoma, or salivary clear cell carcinoma [12,14].

The immunohistochemical profile of CCOCs, is a helpful tool for the differential diagnosis from other tumors of the jaws displaying a prominent clear cell component.

Many immunohistochemical markers can be useful in the differential diagnosis: commonly expressed markers are CK14 and CK19; absent or mildly expressed markers are: CK7, CK8, CK18, vimentin, desmin, enolase, smooth muscle actin, calponin, S-100 protein, a (1)-chymotrypsin, CD10, CD31, CD45, glial fibrillary acidic protein, and chromogranin [15–17].

However, immunohistochemical analysis can be non-decisive. Findings indicate that both CCOC and Hyalinizing clear cell carcinoma (HCCC) of the salivary glands show a high degree of morphologic and immunophenotypic overlap, thereby making it impractical and challenging to distinguish them. Facing this, tumor location (in the jaws versus in oral mucosa/salivary glands) becomes a crucial differentiating criterion. Other aspects suggestive of CCOC are the presence of a more lobulated growth pattern, a myxoid to fibro-cellular stroma, identification of peripheral palisading, and osteodentin deposition [18,19].

Moreover, the positivity of epithelial membrane antigen p63 and the connection with the epithelial lining in CCOC may justify the mucosal origin rather than a glandular one.



**Fig. 4.** Clinical aspect before (a) surgical resection and after (b).

CCOC, like the hyalinizing clear cell carcinoma of the salivary gland, is one of the epithelial neoplasms known to harbor an EWSR1-ATF1 fusion. Therefore, a link between these tumors seems plausible [20].

Another demanding issue is the differential diagnosis of CCOC from ameloblastoma with clear cells and ameloblastic carcinoma. According to some authors, ameloblastic carcinoma and CCOC may represent different clinicopathologic manifestations of a malignant ameloblastic neoplasia associated with clear cell metaplasia [21,22].

Ameloblastic carcinomas at histological examination reveal nests of ameloblastic epithelium with surrounding myxoid stroma epithelium showing multi layering at places. The cells have increased nucleocytoplasmic ratio, hyperchromatic nuclei and were present in sheets. Focal areas of keratinization are also evident, as well as few mitotic figures [23].

While EWSR1 rearrangements is identified in clear cell odontogenic carcinoma, BRAF V600E point mutation is instead identified in ameloblastic carcinoma [24,25].

Due to the rarity of this tumor, the ideal treatment approach has not been yet conclusively determined. In general, a protocol comparable to that used for oral squamous cell carcinoma is adopted.

Due to the high rates of recurrence, the literature agrees in suggesting an early and aggressive surgical treatment with clear margins [2-4].

Recurrences are common when enucleation and local excision are performed, hence they should not be practiced anymore.

In our case, we performed surgical resection with margins of healthy bone tissue.

There was no clinical or radiographic evidence of metastatic spread to the neck and therefore ganglion removal was not performed.

To date, one year after surgery, our patient's clinical exam and radiological exam showed no evidence of local recurrence.

Due to the limited number of data available it is difficult to outline risk factors for tumor recurrence and metastases [26].

The degree of nuclear pleomorphism and hyperchromatism are variable and they seem to be associated with a different metastatic potential of the tumor [27].

Ganglion removal is debated, some of the indications are evidence of extensive soft-tissue invasion, perineural invasion, or cases where resection of the tumor with an adequate margin cannot be guaranteed [28-30].

**Table 1**  
Differential diagnosis of clear cell odontogenic carcinoma.

Type	Tumor	Site	Distinguishing histologic features	Immunoprofile of clear cells
Primary odontogenic	CCOC	Mandible > maxilla	Monophasic, biphasic and ameloblastomatous type of patterns with odontogenic epithelial elements, fibrous stroma, PAS positive, mucin negative	EMA + CK14 + CK19 + Calretin ± EWSR1-ATF1 fusion P63 + CK 5/6 + CK 19 + P63 + BRAF V600E
	Calcifying epithelial odontogenic tumor (CEOT)	Mandible > maxilla	Amyloid and hyalinizing stroma, polygonal epithelial cells, calcification	EWSR1-ATF1 fusion 34βE12 + p63 + EMA +/- P63 + HMWK + MAML2 rearrangements EWSR1-ATF1 fusion Caponin CK EMA S100 Pan CK Vimentin Trasferrin CEA GFAP CD10 S100 CD10 PAX 8 + S100 Melan A HMB45
Primary salivary	Ameloblastoma with clear cell changes	Mandible	Ameloblastic epithelium with myxoid stroma,	
	Hyalinizing clear cell carcinoma	Palate and tongue base	Hyalinizing and fibrocellular stroma, nests of monomorphic clear cells	
	Mucoepidermoid carcinoma	Parotid and palate	Absent or rarely hyalinizing stroma, goblet cells, mucin positive	
Other	Salivary clear cell carcinoma	Minor salivary glands mainly	Polygonal cells with clear cytoplasm, PAS positive, mucin negative	
	Epithelial myoepithelial carcinoma	Most common in parotid, rare in jaws	Polygonal cells with clear cytoplasm, distinct outline, centrally placed nucleus	
Other	Acinic cell carcinoma	Most common in parotid, rare in jaws	Peripherally located nuclei, sparse basophilic granules in some areas	
	Metastatic renal cell carcinoma	Isolated bone metastasis are possible	Epithelial cells with small round hyperchromatic nucleus, clear cytoplasm, PAS positive, mucin negative	
	Amelanotic melanoma	Hard palate, soft palate and gingiva most commonly	Clear to finely vacuolated cytoplasm	

The same conclusions are found in the literature concerning the indication for radiotherapy [31].

Further investigations are needed to better define the indications of radiotherapy and/or chemotherapy in these patients.

Long-term follow-up (including head, neck, and chest imaging examinations) is obligatory due to the evolution of this tumor. The review of Loyola et al. reported a local recurrences rate of 41.0% and distant metastases in 31.0% of the cases [2].

#### 4. Conclusions

CCOC is a rare malignant neoplasm with particular histology and deceptive behavior.

Clinical presentation may be misleading and incisional biopsy guides the diagnosis.

The contribution of molecular biology, immunohistochemistry and cytogenetic are significant in the diagnostic process.

Surgical resection with wide margins is the gold-standard treatment and cervical lymphadenectomy should be considered in certain cases.

The high potential of recurrence of CCOC makes the follow-up mandatory over many years after surgery.

#### Consent

Not applicable.

#### Declaration of competing interest

The authors state that there is no conflict of interest to disclose.

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