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Case Report

Clear cell odontogenic carcinoma, diagnostic difficulties. A case report

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ABSTRACT

Introduction: Clear cell odontogenic carcinoma (COCC) is a rare tumor described by Hansen et al. in 1985. The clinical and radiological manifestations are multiple and the diagnosis is histological.
Observation: A 64-year-old patient consulted us for a right mandibular osteolytic lesion associated to a homolateral labial hypoesthesia. A biopsy was performed under local anesthesia. Histology was consistent with a metastatic lesion of clear kidney cell carcinoma, COCC, or odontogenic squamous tumor. Additional tests eliminated a metastatic lesion. A wide excision of the lesion by hemi-mandibulectomy associated with lymph node dissection and reconstruction by a fibula osteosepto-cutaneous flap was performed. Presence of a fission of the EWSR1 gene on the histological examination of the surgical specimen made the diagnosis of COCC.
Discussion: Our observation illustrates the difficulty of diagnosing COCC. The new contribution of the cytogenetic techniques such as FISH-type techniques makes possible the improvement of the diagnosis.
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1. Introduction

Clear cell odontogenic carcinoma (COCC) is a rare tumor described by Hansen et al. in 1985 [1]. First referred as a clear cell odontogenic tumor, it was considered as a benign, locally aggressive lesion in the 1992 WHO tumor classification. COCC term appears in 2005 in the revision of this classification where it is recognized, because of its aggressiveness, as a malignant tumor [2].

The clinical and radiological aspects are not specific making histology unavoidable for the diagnosis. There are several histological forms of COCC which share morphological characteristics with other clear cell tumors. The diagnosis is complex, and despite the few differential diagnoses, due to the rarity of this type of tumor, it is often a source of misdiagnosis. The contribution of molecular biology, immunohistochemical and cytogenetic examinations are major in the diagnostic procedure.

The objective of this article is to present a clinical case that illustrates this diagnostic difficulty.

2. Observation

A 64-year-old patient with no general history was referred to the Department of Oral Surgery of Timone University Hospital in Marseilles for the management of an intraosseous lesion of the right body of the mandible, discovered following the onset of a hypoesthesia. The right lip and an oral examination were unusual.

CT-scan of this lesion showed an osteolytic image with blurred edges, with bicortical bone destruction giving an aggressive and malignant appearance (Fig. 1).

The MRI found a soft tissue invasion, an osteolysis of the mental foramen and the infra alveolar canal, with a contrast enhancement of the nerve, as far as the lingula region (Fig. 2).

A biopsy was performed. Histology revealed an aggressive epithelial tumor proliferation of odontogenic nature with clear cells, whose diagnosis was difficult. The morphological aspects led to suspect a malignant transformation of a clear cell odontogenic tumor. Immunodetections showed expression of anti-AE1/AE3 and anti-CK8/18 EMA antibodies, minimal expression for CK5/6 and negativity for CK7, CK20, CD10, PS100, PSA, TTF1 and PAX8. The proliferation index measured with the anti-Ki67 antibody was less than 2%.

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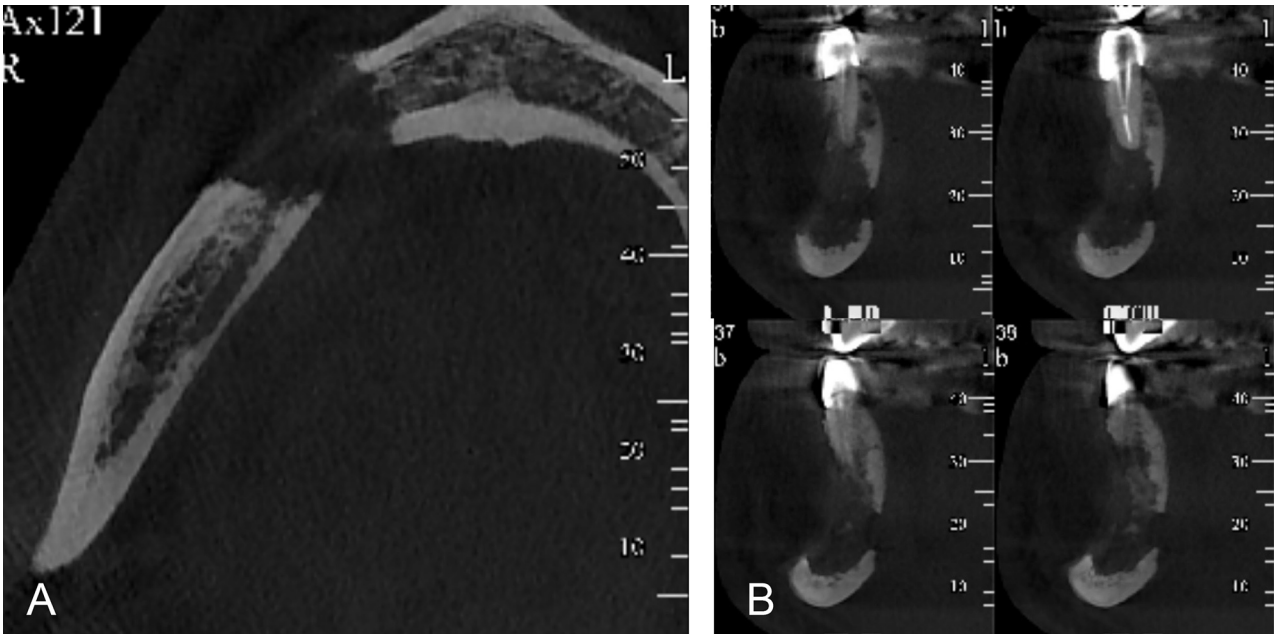

Fig. 1. CT-scan of CCOC showing a bicortical invasion of the tumor.

Diagnostic assumptions were initially: a metastatic lesion of clear kidney cell carcinoma, COCC or odontogenic squamous tumor. Additional tests and the absence of anti-PAX8 immunodetection made it possible to eliminate a primary renal lesion. The blades were addressed to a specialized institution (Toulouse University Institute of Cancer). Due to the absence of mitosis, the diagnosis envisaged as a priority was an odontogenic squamous tumor. After pluridisciplinary discussion and considering the clinical and radiological aspects of this aggressive lesion, treatment consisted in a wide excision of the lesion by hemi-mandibulectomy associated with lymph node dissection and reconstruction by a fibula osteoseptocutaneous flap. Extemporaneous biopsy of the margins and the infra alveolar nerve was negative. Histology examination of the surgical specimen concluded with the presence of marked atypia, moderate mitotic activity (Ki67 to 30%) and especially a fission of the EWSRI gene to the diagnosis of COCC. The patient subsequently benefited from soft tissue infiltrations and concomitant radio chemotherapy (50 Gy on the tumor, 16 Gy on the nodes combined with Cisplatin). There was no sign of recurrence after a one year follow-up.

3. Discussion

COCC is a rare tumor with major diagnostic difficulties.

Clinical and radiological signs are polymorphic. When symptomatic, COCC manifests itself with swelling and dental mobility.

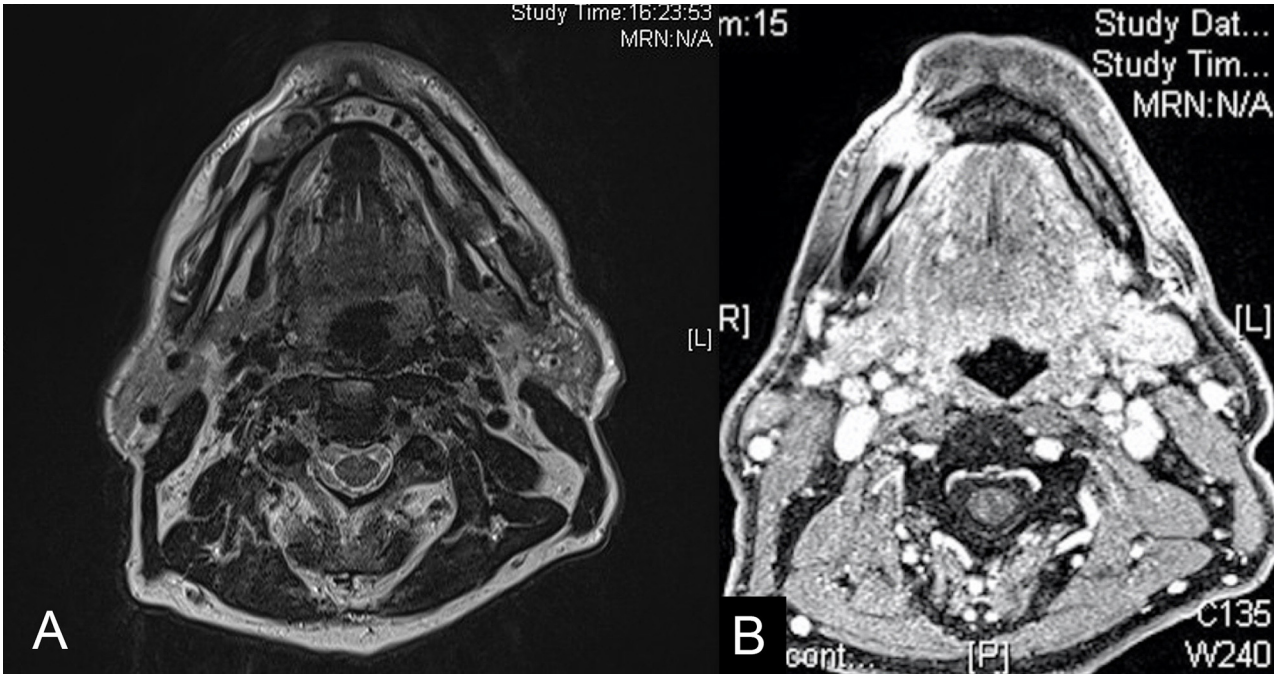

Fig. 2. MRI of CCOC (axial view). A. T1 weighted image showing soft tissues invasion by the tumor. B. T2 weighted image showing mandibular nerve invasion by the tumor.

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