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Clear cell odontogenic carcinoma. A review

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ABSTRACT

Introduction: Clear cell odontogenic carcinoma (CCOC) is described as an exceptional and hard to diagnose malignant tumor which was first reported by Hansen in 1985. The purpose of this review article is to show that CCOC is a not that rare entity and to discuss its various aspects in order to enhance the diagnosis.

Material and methods: A search in the English language literature was performed using the Scopus, ScienceDirect, PubMed and Medline databases between 1985 and 2016. Data were collected on epidemiologic, clinical, radiographic, histological, immunohistochemistrical, cytogenetic, management, follow-up and prognosis features of CCOC.

Results: Sixty-five studies from which a total of 95 case reports were included in the review. CCOC was generally seen in the fifth decade and the most common site was mandibular. The most frequently found symptoms were swelling, tooth mobility and pain. Radiologically, the image was radiolucent and could look like a cyst or a periodontal lesion. In situ hybridization techniques frequently expressed a gene fission of EWSR1. The treatment was mostly a radical surgical excision of the tumor with or without adjuvant radiotherapy or chemotherapy. CCOC showed high rates of recurrence and mortality related with the presence of distance metastasis.

Discussion: Fission of EWSR1 gene could be the main element it the diagnosis of CCOC. A multidisciplinary approach, including a radiologist, pathologist and an oral & maxillofacial surgeon may be helpful in the evaluation and management of these lesions. With 95 reports found in English literature, we cannot say that CCOC is extremely rare anymore.

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1. Introduction

Clear cell odontogenic carcinoma (CCOC) is often considered as a rare tumor, first described by Hansen in 1985 [1]. Formerly known as clear cell odontogenic tumor, a locally aggressive benign tumor in the WHO classification of 1992 [2], it was renamed CCOC and classified among malignant tumors after the revision of this classification in 2005. It is still considered a malignant tumor in the 2017 classification [3].

Literature on CCOC is rare, that makes it hard to diagnose. Clinical and radiological features are not specific, but its aggressive nature can guide the diagnosis.

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http://dx.doi.org/10.1016/j.jormas.2017.08.005 2468-7855/© 2017 Elsevier Masson SAS. All rights reserved. When symptomatic, COCC manifests itself by swelling and tooth mobility. Pain is inconstant. Its aggressive nature is radiologically found with osteolytic lesions with ill-defined limits which may cause dental root resorption.

Histopathology shows three types of cells: basaloid or polygonal cells with clear cytoplasm, basaloid or polygonal cells with eosinophilic cytoplasm and cylindrical cells with ameloblastic differentiation on the periphery of the lobules. Consequently, three types of COCC can be found: monophasic, biphasic and ameloblastic [3].

Morphologically, COCC shares similar characteristics with other clear cell tumors.

The diagnosis is complex, despite of the lack of differential diagnoses. The rarity of this tumor is often a cause of misdiagnosis. The contribution of molecular biology, immunohistochemistry and cytogenetic studies are significant in the diagnostic process.

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The recommended treatment is radical surgery because partial or incomplete resection results in recurrence.

This study is a comprehensive review on its epidemiological, clinical, radiological, histopathological, immunohistochemical, cytogenetical, therapeutic and prognostic aspects, showing that this clinical entity seems to be not as rare as described by previous studies.

2. Material and methods

A literature review was conducted using the Scopus, Science-Direct, PubMed and Medline databases in July 2016. The keywords used were "(clear cell odontogenic tumors, or clear cell odontogenic carcinoma, or clear cells) and (immunology, or immuno, or immunological, or immunohistochemistry, or cytogenetics, or EWSR1, or PAS, or CK) and (oral, or mouth, or buccal)" along with MESH terms.

Reports published in the English language from January 1985 to July 2016 were eligible. No exclusion criteria has been established. Reference lists of included articles were scanned.

For each case report, data on epidemiology, clinical, radiographic, histological, immuno-histochemistry and cytogenetic features of CCOC were collected. Management, follow-up and prognosis data were also noted. Data from previous reviews were checked and corrected.

3. Results

The search resulted in 65 studies from which a total of 95 case reports (the current case and 94 others) were included in the present review.

Clinical features (epidemiology, tumor location, presence of metastasis, management, follow-up and outcome) are described in Table 1.

Table 1

Case no	Author	Age (years)	Sex	Tumor location	Metastasis	Management	Follow-up (months)	Recurrence	Outcom
1	Hansen et al., 1985	64	F	Mandible	_	Resection, ND	_	_	_
2	Hansen et al., 1985	74	F	Maxilla	_	Resection	17	Yes	NED
3	Hansen et al., 1985	60	F	Maxilla	-	Resection	72	_	NED
4	Waldron et al., 1985	50	M	Mandible	_	Curettage	78	_	NED
5	Waldron et al., 1985	66	F	Maxilla	LN	Resection	174	Yes	DOD
3	Muller et al., 1986	14	M	Mandible	-	Local excision	180	_	NED
7	Bang et al., 1989	67	M	Mandible	_	Resection	6	_	NED
8	Bang et al., 1989	74	F	Mandible	LN, lungs	Enucleation	144	Yes	DOD
9	Bang et al., 1989	50	F	Mandible	LN LN	Resection	_	-	_
10	Ng et al., 1990	48	M	Mandible	=	Local excision	60	_	NED
11	Guilbert et al., 1991	66	M	Mandible	LN	Local excision, ND	-	_	_
12	Fan et al., 1992	79	F	Mandible	LN	CTX	24	_	AWD
13	Odukova et al., 1992	15	M	Mandible		Resection	60	_	NED
14	Milles et al., 1993	65	F	Maxilla	LN	Resection, RTX	6	_	NED
15	Nikal et al., 1993	56	F	Mandible	LIN _	Resection	5	_	NED
16		74	г М				60	_	DOD
	Piattelli et al., 1994		F	Mandible	LN, lungs	Local excision, ND	180		
17	Eversole et al., 1995	41		Mandible	LN, lungs	Curettage		Yes	DOD
18	Eversole et al., 1995	48	F	Mandible	/- /	Local excision	18	-	NED
19	Eversole et al., 1995	44	F	Mandible	-	Local excision, RTX	18	_	NED
20	Eversole et al., 1995	43	M	Mandible	-	_	-	_	-
21	Eversole et al., 1995	53	F	Mandible	_	-	_	-	-
22	Mari et al., 1995	71	M	Maxilla	_	Resection	36	Yes	DWD
23	Sadeghi et al., 1995	89	F	Mandible	-	Resection	12	Yes	AWD
24	De Aguiar et al., 1996	30	F	Mandible	⁷ –	Curettage	156	Yes	-
25	Muramatsu et al., 1996	69	M	Mandible	-	Resection	6	-	NED
26	Kumamoto et al., 1998	61	M	Mandible	-	Resection	11	_	NED
27	Miyauchi et al., 1998	56	F	Mandible	_	Resection	_	_	-
28	Yamamoto et al., 1998	67	M	Mandible	_	Enucleation	36	-	NED
29	Behro et al., 1999	66	M	Mandible	_	Resection	_	_	_
30	Behro et al., 1999	53	M	Maxilla	_	Resection	_	_	_
31	Kumamoto et al., 2000	35	M	Mandible	_	Resection	_	Yes	_
32	Nair et al., 2000	41	M	Mandible	_	Resection, RTX	10	_	NED
34	Benton et al., 2001	85	F	Mandible	_	Abstention	3	_	DOD
35	Brinck et al., 2001	39	F	Mandible	Lungs	Local excision	36	Yes	_
36	Li et al., 2001	31	F	Mandible	LN	Local excision	48	Yes	DOD
37	Li et al., 2001	42	F	Mandible	_	Enucleation	84	_	NED
38	Li et al., 2001	58	F	Mandible	_	Enucleation	36	_	NED
39	Li et al., 2001 Li et al., 2001	32	M	Maxilla	_	Resection	24	_	NED
40	Li et al., 2001	49	F	Maxilla	_	Resection	24	_	NED
41	Maiorano et al., 2001	81	F	Mandible	_	Resection	60		NED
42	Maiorano et al., 2001	84	F	Mandible	_	Resection	36	_	NED
42 43	Adamo et al., 2002	49	r M	Mandible	_	Resection, ND	40	_	NED
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14 15	Ariyoshi et al., 2002	60	F	Mandible	_	Resection	36		NED
45 46	Brandwein et al., 2002	81	F	Mandible	_	Local excision	54	Yes	NED
46 47	Dahiya et al., 2002	26	M	Maxilla	_	Resection, RTX	72	Yes	-
47	lezzi et al., 2002	62	F	Maxilla	-	Resection	48	-	NED
48	Mosqueda-Taylor et al., 2002	55	F	Mandible	-	Resection, ND	6	_	NED
49	August et al., 2003	72	F	Mandible	-	Resection	60	Yes	AWD
50	August et al., 2003	73	F	Mandible	-	Resection	24	-	-
51	August et al., 2003	77	F	Maxilla	_	Resection, RTX	24	_	_
52	August et al., 2003	40	F	Mandible	=	_	6	-	NED
53	Braunshtein et al., 2003	72	F	Mandible	_	Local excision	24	_	NED

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