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# Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: [www.elsevier.com/locate/jomsm](http://www.elsevier.com/locate/jomsm)

## Case Report

# Low-grade mucoepidermoid carcinoma with regional lymph node metastasis: A case report and genetic review of criteria for grading



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## ARTICLE INFO

### Article history:

Received 8 June 2015

Received in revised form 15 August 2015

Accepted 26 August 2015

Available online 10 October 2015

### Keywords:

Mucoepidermoid carcinoma

Salivary gland

CRTC1-MAML2 gene fusion

## ABSTRACT

Mucoepidermoid carcinoma (MEC) is the most common salivary gland malignancy. It accounts for 5% of all salivary gland tumors and 20% of salivary malignancies and has various clinical outcomes. Its prognosis is related to the tumor grade in the WHO classification of head and neck tumors. MEC is divided into high, intermediate and low grades. In general, low-grade MEC with cervical lymph node metastasis is rare, and primary MEC that occurs in a minor salivary gland with cervical lymph node metastasis is very rare. Low-grade lesions have quite different behavior from high-grade ones, leading to differences in the treatment course required for disease control.

MEC is associated with recurring chromosomal translocation t(11;19)(q21;p13). CRTC1-MAML2 gene fusion was identified from the recurring translocation, which is often the sole cytogenetic alteration in this disease. This fusion transcript is detected frequently in MEC and is involved in the transformation of epithelial cells. Detection of the CRTC1-MAML2 fusion transcript is associated with a less advanced clinical stage and low-grade tumor histology. The presence of the transcript is associated with longer disease-free and overall survival periods.

Here, we report a rare case of low-grade MEC with cervical lymph node metastasis. The findings for this patient show that the CRTC1-MAML2 fusion transcript may be specific to MEC, and associated with a distinct MEC subset that exhibits favorable clinicopathologic features.

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## 1. Introduction

Mucoepidermoid carcinoma (MEC) is one of the most common malignant salivary neoplasms. In a series of 108 MECs in salivary glands, 55% of the tumors occurred in major salivary glands and 45% in minor salivary glands [1]. Conventional clinical parameters such as age and stage are among the most significant prognostic factors, together with histopathological grading criteria [2–4]. Although high-grade MEC requires adjuvant radiation and neck dissection [5], complete local excision of the tumor is often sufficient for low-grade MECs of minor salivary glands. More widespread disease, however, may require extensive surgical resection [6]. MEC classified as low grade usually does not metastasize and is often cured by appropriate surgery [7]. However, based on 2400 MECs, Chen et al. reported that the incidence of nodal involvement of the neck was 3.3% for low-grade MECs [8]. According to Muzzo and colleagues, for MEC the stage of disease strongly influences survival, suggesting

**Abbreviations:** MEC, Mucoepidermoid carcinoma; FDG, fluorodeoxyglucose; PET, positron emission tomography; CT, computed tomography; CRTC1, CREB-regulated transcription coactivator 1; MAML2, mastermind-like gene 2.

☆ Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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<http://dx.doi.org/10.1016/j.jomsm.2015.08.006>

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the crucial influence of nodal status [1]. Some patients with MEC classified as grade 1 by the prevailing criteria of Auclair, including classification based on the classical non-point based grading system, develop metastases or disease-related mortality [2,3,6,9]. However, clinicopathological analysis of head and neck MEC indicates that advanced histological grade and positive nodal disease continue to be the strongest independent predictors of overall and disease-specific survival, respectively [6]. Here, we report a case of MEC originating in a minor intraoral salivary gland with cervical lymph node metastasis at the time of presentation for which one of the predictive factors for regional lymph node metastasis was discovered after surgery.

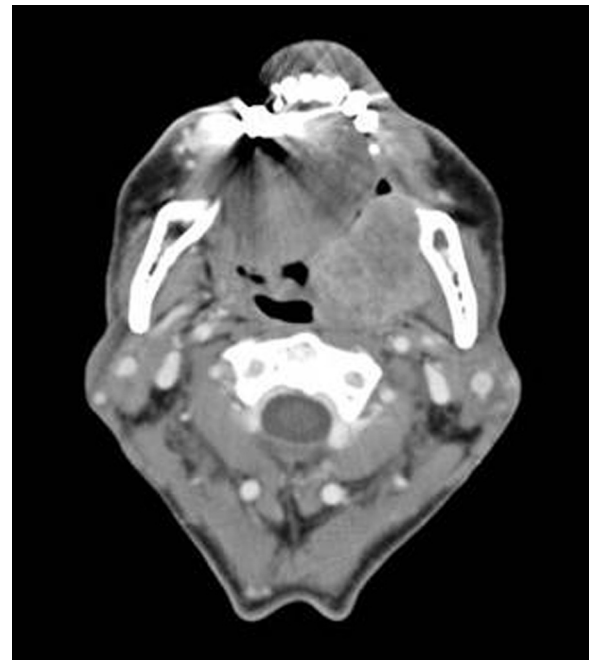
## 2. Case report

A 56-year-old man was referred to our outpatient clinic with a tumor that he had noted on his left retromolar triangle for 2 months. He had been treated for 2 years for disorganized schizophrenia and intraoral bleeding. His tumor was discovered by a nurse who took care of him. An incisional biopsy was performed and low-grade adenocarcinoma was diagnosed by histological staining. There was no significant past medical history. Intraoral examination revealed a 38 mm × 27 mm tumor that was irregular and partly ulcerated on the surface of the left retromolar triangle (Fig. 1). The lesion was painless upon palpation. Extraoral examination revealed ipsilateral 20 mm × 20 mm subjugulodigastric lymphadenopathy.



**Fig. 1.** Macroscopic observation of the intraoral tumor showed an approximately 38 mm × 27 mm irregularly raised submucosal hard mass in the left retromolar triangle. The border of the tumor was well circumscribed but not movable.

A computed tomography (CT) frontal view showed that the inside of the left mandible was suppressed by the tumor and absorbed (Fig. 2). Axial  $^{18}\text{F}$ FDG-PET/CT images revealed a mass with intense  $^{18}\text{F}$ FDG uptake in the left retromolar area, and ipsilateral subdigastric and contralateral submandibular regions [5]. The patient underwent bilateral supraomohyoid neck dissection after frozen section diagnosis of the bilateral level III lymph nodes and marginal resection of the mandible with resection of the retromolar triangle tumor. There was no perineural or perilymphovascular invasion. No necrosis, cellular anaplasia, aggressive tumor invasion or bone invasion was present. There was an under 20% cystic component, and fewer than 4 mitoses were observed per 10 high-power fields (Fig. 3). The tumor was well circumscribed, and composed of multicystic and small columnar structural tumor cells that were positive for Alcian blue-periodic acid-Schiff stain (Al-PAS). Al-PAS showed predominantly intraluminal mucin-positive material, with scattered cells showing intracytoplasmic mucin-positive material (Fig. 4A). Epithelial mucin (MUC1) expression was restricted to the apical cell membranes of normal salivary glands (Fig. 4B). The pathological diagnosis was low-grade mucoepidermoid carcinoma

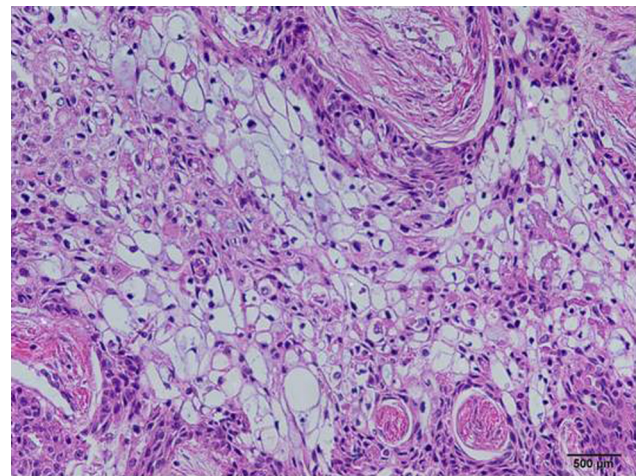


**Fig. 2.** Enhanced computerized coronal tomography shows a mass on the left buccal mucosa.

on the left retromolar pad and metastasis to the left subjugulodigastric lymph node with no extracapsular extension, pT2N1M0. The pathologist had also suggested in previous diagnosis that it was polymorphous low-grade adenocarcinoma. The postoperative course was uneventful and the patient remains tumor free at 32 months after definitive surgery.

## 3. Data collection and review of the literature: relations between the prevalence of MECT1/MAML2 translocation and clinicopathologic variables

Formalin-fixed paraffin-embedded tissue samples from six cases of intraoral MEC, including the present case, were available



**Fig. 3.** The tumor shows solid eosinophilic tumor cells and a cystic area with an admixture of mucus cells, intermediate cells, and epidermoid cells. It has a prominent cystic component (>20%), but lacks neural invasion, necrosis or cellular anaplasia, and is only rarely mitotic. No necrosis, cellular anaplasia, aggressive tumor invasion or bone invasion was present. There was an under 20% cystic component and fewer than 4 mitoses per 10 high-power fields were observed (hematoxylin and eosin stain, original magnification: 200×).

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