



Mucoepidermoid Carcinoma of the Endojugal Accessory Salivary Glands: A Case Report

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How to cite this paper: Bakhil, A., El Hafidi, N., Benbachir, A., Koualla, S., Ribag, Y., Sabani, H., Khalfi, L., Hamama, J. and El Khatib, K. (2022) Mucoepidermoid Carcinoma of the Endojugal Accessory Salivary Glands: A Case Report. *Open Access Library Journal*, 9: e9416.
<https://doi.org/10.4236/oalib.1109416>

Received: October 2, 2022

Accepted: November 5, 2022

Published: November 8, 2022

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Abstract

Mucoepidermoid carcinoma is a malignant tumor that affects the salivary glands, particularly the parotid gland. Involvement of the accessory salivary glands is rarer; some cases have been described in the literature. The clinical symptoms are atypical with a slow and silent evolution which delays the diagnosis. The diagnosis is based on clinical, radiological and anatomopathological examination of diagnostic and prognostic interest. The management is essentially surgical with carcinological margins of exeresis, radiotherapy can be indicated in case of high grade tumor. In our case, it was a mucoepidermoid carcinoma of the endojugal salivary glands which is a rare localization, and no similar case has been found in the literature. The management was surgical with healthy margins of exeresis.

Subject Areas

Dentistry

Keywords

Mucoepidermoid Carcinoma, Accessory Salivary Glands, Endojugal

1. Introduction

Mucoepidermoid carcinoma (MEC) is a malignant tumor of the upper aerodigestive tract, the term mucoepidermoid tumor was first used by Stewart and Foote in 1945, to describe a salivary tumor containing squamous cells and mucin-secreting cells, it mainly affects the salivary glands, especially the parotid gland and represents 15% to 30% of malignant tumors of the salivary glands. Involvement of the accessory salivary glands remains rare. MEC affects the pop-

ulation between the fourth and sixth decade, but it can occur at earlier ages. Its etiopathogenesis is not well elucidated and its clinical manifestations are not specific and are not very evocative, which sometimes makes the diagnosis difficult. Diagnosis is based essentially on anatomopathological examination which allows a staging, of prognostic and therapeutic interest, in 3 grades: low grade, intermediate grade and high grade.

The management of MEC is essentially surgical; radiotherapy is indicated in high-grade MEC as a complement to surgery. The long-term prognosis is still poorly established due to the rarity of the cases studied.

In this article, we will describe the clinical manifestations of mucoepidermoid carcinoma of the endojugal accessory salivary glands and its management through a case report and a review of the literature.

2. Clinical Observation

We report the case of a 61-year-old female patient, with a history of arterial hypertension on dual therapy, poorly controlled diabetes on insulin and oral antidiabetics, the patient has never been operated.

The history of her disease goes back to 6 months before her admission to our department, with the appearance of a left endojugal nodule facing the 1st mandibular molar 36. This nodule was painless, it progressively increased in volume, becoming functionally troublesome, especially during mastication, and morphologically troublesome with a jugal swelling, all evolving in a context of preservation of the general state and apyrexia.

The clinical examination on admission found a patient in a good general condition, hemodynamically, respiratory and neurologically stable. Facial examination revealed a facial asymmetry due to the presence of a centro-jugal swelling filling the left labio-genial fold and covered by normal skin tissue, palpation was painless, and the mass was mobile, well limited with a major axis of about 4 cm (**Figure 1**), adherent to the deep plane. Endobuccal examination found a raised swelling on the inner side of the cheek, below and in front of the stenson orifice, this swelling was partially filling the left upper vestibule, and the mucosa was purplish in some places (**Figure 1**). No palpable cervical adenopathy was found.



Figure 1. Clinical aspect of the left endojugal tumor.

In view of this atypical clinical picture, the patient underwent a surgical biopsy under local anesthesia, which came back in favor of a mucoepidermoid carcinoma of the accessory salivary glands.

An MRI of the facial mass was requested as a local extension assessment, which showed a tissular lesion infiltrating the buccinator muscle and the subcutaneous tissue (**Figure 2**). A cervico-thoraco-abdominal CT scan was performed as part of the general extension assessment, which did not reveal any secondary or distant lesion.

The patient's case was discussed in a multidisciplinary consultation meeting in our hospital. That led us to perform a carcinological excision and to discuss the additional support after the anatomopathological results of the surgical specimen.

The patient was admitted to the operating room after a pre-anesthesia consultation; a transfixing excision of the tumor, with safety margins (**Figure 3**), was performed, the limits were verified by an extemporaneous examination, and catheterization of Stenon's canal was performed. The loss of substance was self-closing. The cervical lymph nodes were then removed by a cervicotomy including the fatty cellulose tissues of lymph node groups I, II and III without any macroscopic adenopathy being detected (**Figure 4**).



Figure 2. MRI images of the lesions.



Figure 3. Monobloc excision piece.

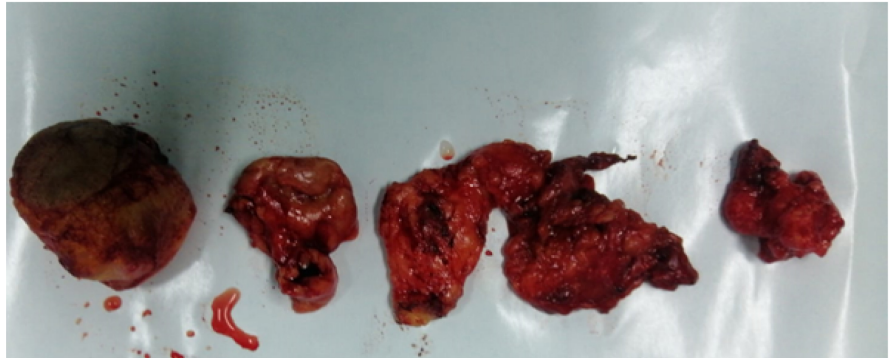


Figure 4. Excisional specimen and obvious lymph node.

To allow rapid healing of the mucosa in the mouth, a nasogastric tube was placed with, liquid and lukewarm food were indicated, and mouthwash for multiday use. The patient was put on parenteral antibiotics and corticosteroids for 48 hours and then relai by mouth on the third day. The postoperative course was simple.

Anatomopathological examination of the surgical specimen and lymph node sampling showed a low-grade mucoepidermoid carcinoma with healthy resection limits and no lymph node invasion.

The patient's case was discussed again in a multidisciplinary consultation meeting and no indication for adjuvant treatment was retained.

3. Discussion

Mucoepidermoid carcinoma is a malignant salivary tumor containing squamous cells and mucin-secreting cells. The heterogeneity of the histological types and their low incidence, makes the search for risk factors and etiopathogeny difficult, most studies are retrospective with a small number of cases.

The most known risk factor is exposure to ionizing radiation; we also count exposure to tobacco and alcohol, and the presence of a previous history of cancer in the patient [1] [2]. Exposure to ionizing radiation, considered for the diagnosis of medical or dental pathologies (radiography of the cephalic extremity) has been implicated in the occurrence of salivary gland cancers, particularly before the 1960s, due to the use of higher doses than nowadays. A study of atomic bomb survivors in Japan in 1945 reported a higher risk of salivary gland cancer, mucoepidermoid carcinoma was the most decribed histological type [3].

MEC is a non-encapsulated tumor with local invasion; it can spread remotely through the lymphatic system in its most malignant forms. Its evolution is slow and not very symptomatic; it is generally limited to a hard, painless mass that has been present for months or even years. A certain number of elements of the clinical examination point to the malignant nature of the tumor, such as its painful nature, the rapid increase in its volume, and the presence of palpable cervical lymph nodes, mostly faced in the presence of a malignant tumor.

The most frequent localization is the parotid gland, followed by the subman-

dibular gland and then the accessory salivary glands [4] [5] [6]. Other locations other than the salivary glands have been reported in the literature such as the conjunctiva, the lung and the breast [7] [8] [9].

Usually, the diagnosis of MEC is relatively easy on microscopic examination. It contains two types of cells: squamous cells and mucin-producing cells [4]. Often a third type is present: intermediate cells. The behavior of MECs is uncertain, local recurrence and the discovery of distant metastases are possible. For these reasons, it was necessary to determine sub-classifications of MEC. This sub-classification is based on the histological aspect: proportion of mucus-producing cells, cell differentiation, anaplasia, cell mitoses, cell growth aspect.

Generally, three grades have been proposed according to the authors [5] [6]:

- Low-grade or well-differentiated tumor: tissue in which mucus-producing cells account for more than 50% of the tumor with macrocysts. The squamous cells are well differentiated and the intermediate cells are limited in number.
- High-grade or poorly differentiated tumor: tissue in which mucus-producing cells are rare (less than 10), with no macrocysts. Tumors tend to form clusters of intermediate or epidermoid cells. Pleomorphism is present with numerous mitoses and prominent nucleoli.
- Intermediate or moderately differentiated tumor. In this group, there are elements present in the two previous ones.

In front of an increase in volume in a progressive way of the tumor, an ultrasound is the paraclinical test of choice, given its simplicity and low cost. It is a non-invasive and rapid examination but it remains operator dependent. It is performed bilaterally and comparatively in case of a parotid or sub-mandibular localization and confirms the diagnosis of intra-parotid tumor in 100% of cases. The criteria in favor of a benign lesion are the homogeneous and well limited character; the malignant character is evoked in the absence of one of these two criteria. The CT scan allows evaluating the intra-glandular tumor involvement and the presence of a bone extension [10]. It can also provide information on the loco-regional extension of the tumor to the mandibular bone and masticatory muscles, and explore the lymph node areas. MRI allows a better exploration of the soft tissues and searches for extension to the muscles and subcutaneous areas. MRI is more efficient than CT for the detection of tumors and the study of their extension, but bone involvement is better visualized in CT [11] [12].

Ultrasound, CT and MRI, do not allow a diagnosis of nature, hence the obligation of an anatomopathological examination on a good quality biopsy allowing a positive diagnosis and a histological staging of the tumor. The most frequently requested workup is a thoracic-abdominal-pelvic CT scan as part of the extension assessment. In this regard, the most frequently affected site is the lung parenchyma, which should be systematically explored in case of a high-grade tumor. The rest of the work-up should be performed according to the specific needs and clinical aspects of each patient (bone scan, brain CT, etc.).

The decision of a therapeutic plan must be taken in multidisciplinary consul-

tation meeting including surgeons oncologists and radiotherapists, the management is generally surgical, additional radiotherapy is indicated in the case of a high grade mucoepidermoid carcinoma.

The prognosis depends on the histological criteria, basically the degree of cellular differentiation, the number of mucus-secreting cells, the extent of mitosis, the presence of necrosis, nerve and vascular invasion, infiltration and invasion of neighbouring structures [13].

4. Conclusions

MEC is a malignant tumor of the salivary glands involving frequently the parotid gland. It affects mostly elderly subjects, but can occur in young people as well. The evolution is often slow, the diagnosis is based on a meticulous clinical examination, medical imaging and anatomopathology which allow both a positive diagnosis and prognosis.

Management is usually surgical with adjuvant radiotherapy in the case of high grade MEC.

Conflicts of Interest

The authors declare no conflicts of interest.

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