

CASE REPORT

Spontaneous partial regression of low-grade mucoepidermoid carcinoma of the maxilla

M.F. Vargas Gamarra*, O. Natsuki, M. Flores and M. Armengot Carceller

ENT Department, Hospital Universitario y Politécnico La Fe, Consonrci Hospital Universitario General de Valencia, Universidad de Valencia, Valencia, España

*Correspondence address. Tel: +34 961244000; E-mail: mariafernandavargasg@gmail.com

KEY CLINICAL MESSAGE

Spontaneous regression has been described in all types of cancers. We report the first case of spontaneous regression of a low-grade maxillary mucoepidermoid carcinoma. Study of the mechanisms underlying spontaneous regression of malignant neoplasms opens the door for future treatments.

INTRODUCTION

Malignant neoplasms of the nasal cavity and nasal sinuses are uncommon accounting for 3–5% of all head and neck tumors [1, 2]. The squamous carcinoma is the most frequent type, followed by the adenocystic carcinoma and the mucoepidermoid carcinoma (MEC).

MEC is a malignant epithelial tumor composed of mucosecretory cells and epidermoid cells that originates in the minor salivary mucosal glands. It accounts for 13% of all malignant tumors of the maxillary sinuses, and is more common in females [1]. Histologically, we differentiate three types: those with low, high and intermediate degrees of malignancy according to the cellular proportions, with mucosal cells predominating in the low-grade type [3].

Spontaneous regression has been described in all types of cancers, although it is more frequent in some than others. We present a rare case of spontaneous partial regression of a low-grade maxillary MEC, not previously described in this tumor, in a 50-year-old woman.

CASE DESCRIPTION

A 50-year-old woman was referred for a progressive facial deformity at the left nasogenian sulcus and bilateral gingivolabial sulcus.

This painless cystic formation was associated with complete bilateral nasal obstruction.

Exploration of the nasal cavity revealed bilateral purulent rhinorrhea and polypoid formations. The nostrils were completely occluded at the height of the middle third by a tumor of bone consistency. There was also a deformity of the bone palate with a prominence of the same consistency.

Maxillofacial computed tomography (CT) showed a voluminous expansive mass, occupying and destroying the maxilla and nasal fossae with pseudocystic formations and an intralaminar calcified matrix. It also affected the ethmoid cells and bulged in the cavum (Fig. 1). Puncture of the gingivolabial tumor was done to obtain 4 ml of serohematic fluid, the analysis of which did not lead to a diagnosis.

Subsequently, abundant biopsies were obtained using a gingivolabial approach, and a definitive diagnosis of low-grade MEC was made.

Since complete excision required a potentially mutilating intervention, and radio- and chemotherapy were not indicated for this tumor, the patient decided to remain under observation without treatment. After 10 years, the patient's symptoms have improved, the nostrils are permeable, and the facial morphology has not changed. In imaging tests, this improvement can be seen more clearly (Fig. 1B).

DISCUSSION

MEC of the Maxilla is a rare entity [4]. In our patient, the tumor had grown into the maxillofacial region and the effect on the anatomical structures was so profound that curative surgical treatment would have generated a severe iatrogeny, without ensuring complete excision. In fact, one of the reasons for the

Received: January 16, 2018. Accepted: April 15, 2018

© The Author(s) 2018. Published by Oxford University Press.

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/4.0/>), which permits non-commercial re-use, distribution, and reproduction in any medium, provided the original work is properly cited. For commercial re-use, please contact journals.permissions@oup.com

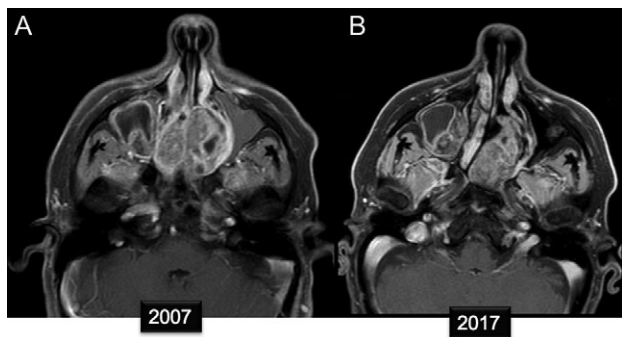


Figure 1: Axial magnetic resonance MR slices and T1-weighted sequences after administration of gadolinium with fat saturation. (A) Soft tissue mass in the posterior region of the nostrils that crosses the midline and shows heterogeneous enhancement after contrast administration. There is obliteration of the nostrils, invasion of the right maxillary sinus and remodeling of the left maxillary sinus; both sinuses present with mucosal retention. (B) In the control study, a reduction in mass size was observed, with permeability of the right nasal cavity and an absence of mucosal retention in the left maxillary sinus (in relation to the permeability of the meatus)

poor prognosis of this kind of tumors is the close anatomic proximity to vital structures as the skull base, orbit, brain and carotid artery that would render an incomplete exeresis of the tumor. This kind of tumor tend to be asymptomatic in early stages causing symptoms only when extensive local extension has been produced. Five-year OS is 35% according to some authors. Early stage diagnosis of this kind of tumors is of paramount importance in order to achieve good survival rates. This kind of tumors should always be considered in as differential diagnosis in the study of maxilla mases.

Although the majority of patients present with early stage MEC (I and II) the ongoing growth is a common feature among all stages. Recurrence appear in the first 2 years after treatment and can be as high as one third of the patients. Surgery is the treatment of choice for maxillary sinus carcinomas and prognosis is better for patients managed by surgery associated with radiotherapy (RT) than for patients submitted to RT or surgery alone [5]. RT as single treatment seems to be indicated only for advanced unresectable tumors.

De Cruz et al. in his 2006 paper describes a series of five cases of MEC in retrospective review from a single institution. His findings in accordance with the literature describe a higher prevalence in woman, a prolonged period of time for the onset of symptoms and a slight trend of early clinical stages at time of consult. Three (42%) were treated with surgery and post-operative RT, the remaining two cases were treated either with surgery or RT alone. Mean follow up time was 57 months; recurrence rate was 28.6% and 5-year OS was 70%. They noted

a clear trend towards better result with the addition of RT to the treatment of these kind of tumors. Early diagnosis, proper surgical approach and associated Rt seems to be the key points for the improvement of overall survival.

According to our literature search, no cases of spontaneous regression of this tumor type have been reported previously.

The mechanisms involved in spontaneous tumor regression may vary, as follows: (i) immune mediation, (ii) inhibition of growth factors, angiogenesis, carcinogens and cytokines, (iii) elimination of or decrease in oncogene expression, (iv) hormonal mediation, (v) psychological factors and (vi) mechanisms that induce apoptosis [6, 7].

Spontaneous regression is possible in MECs of a low maxillary grade. This should be taken into consideration when surgical excision is not possible or would lead to severe iatrogeny. Study of the mechanisms underlying spontaneous regression of malignant neoplasms opens the door for future treatments.

CONFLICT OF INTEREST STATEMENT

None declared.

REFERENCES

1. Myers LL, Nussenbaum B, Bradford CR, Teknos TN, Esclamado RM, Wolf GT. Paranasal sinus malignancies: an 18-year single institution experience. *Laryngoscope* 2002;**112**: 1964–9.
2. Bhattacharyya N. Survival and staging characteristics for non-squamous cell malignancies of the maxillary sinus. *Arch Otolaryngol Head Neck Surg* 2003;**129**:334–7.
3. Triantafyllidou K, Dimitrakopoulos J, Iordanidis F, Koufogiannis D. Mucoepidermoid carcinoma of minor salivary glands: a clinical study of 16 cases and review of the literature. *Oral Dis* 2006;**12**:364–70.
4. Wolfish EB, Nelson BL, Thompson LD. Sinonasal tract mucoepidermoid carcinoma: a clinicopathologic and immunophenotypic study of 19 cases combined with a comprehensive review of the literature. *Head Neck Pathol* 2012;**6**: 191–207.
5. Dulguerov P, Jacobsen MS, Allal AS, Lehmann W, Calcaterr T. Nasal and paranasal carcinoma: are we making progress?: A series of 220 patients and a systematic review. *Cancer* 2001;**92**:3012–29.
6. Bodey B. Spontaneous regression of neoplasms: new possibilities for immunotherapy. *Expert Opin Biol Ther* 2002;**2**: 459–76.
7. Park YH, Park BM, Park SY, Choi JW, Kim SY, Kim JO, et al. Spontaneous regression in advanced squamous cell lung carcinoma. *J Thorac Dis* 2016;**8**:E235.