Oral Oncology 93 (2019) 116–119

Contents lists available at ScienceDirect

Oral Oncology

journal homepage: www.elsevier.com/locate/oraloncology

Letter to the editor

Myoepithelial carcinoma with rhabdoid features in the maxillary sinus: Immunohistochemical and in situ hybridization analysis of a rare case

ARTICLE INFO

Keywords:
Myoepithelial carcinoma
Sinonasal carcinoma
Rhabdoid phenotype
Maxillary sinus
Immunohistochemistry
In situ hybridization

ABSTRACT

Myoepithelial carcinomas of the head and neck are often located in the major salivary glands, notably in the parotid glands, being less frequent in the minor salivary glands. Noteworthy, myoepithelial carcinoma in the maxillary sinus is extremely rare. In fact, only five cases have been previously published to date. Here, we present, for the first time, a detailed immunohistochemical and in situ hybridization analysis of a SMARCB1 (INI-1)-intact myoepithelial carcinoma with rhabdoid features, expanding the histopathological spectrum of high-grade sinonasal carcinomas.

Introduction

Myoepithelial tumors, either benign or malignant, account for about 1% of all primary salivary gland tumors. Approximately 70% and 20% of them arise in the parotid and submandibular glands, respectively, whereas 10% in the minor salivary glands [1,2]. Approximately 10% of all myoepithelial tumors are myoepithelial carcinomas. Notably, myoepithelial carcinomas arising in unusual head and neck locations have been reported, such as nasal cavity [3], nasopharynx [4] and maxillary sinus [5–8]. In the maxillary sinus, to date, only 5 cases of myoepithelial carcinoma have been previously published. Recently, it was observed that 66 (0.009%) out of 7190 salivary gland tumors showed maxillary sinus involvement, being none of them diagnosed as myoepithelial carcinoma, confirming its rarity in this anatomical location [9].

Here, we present, for the first time, a detailed immunohistochemical (IHC) and in situ hybridization (ISH) analysis of a myoepithelial carcinoma with rhabdoid features arising from maxillary sinus, expanding the histopathological spectrum of high-grade sinonasal carcinomas.

Case report

A 44-year-old man was referred presenting painful facial asymmetry evidenced by increase of volume in the left maxilla with several months of evolution. Computerized tomography scan examination showed an extensive osteolytic lesion on the left maxillary sinus. Clinical examination and computerized tomography of the neck, thorax and abdomen showed no alterations. The histopathological analysis showed extensive sheets and foci of isolated cells, which presented polygonal morphology with large hyaline cytoplasmic inclusions and eccentric nuclei with single prominent nucleoli. Moreover, a smaller component (< 15% of all tumor cells), exhibited plasmacytoid or epithelioid cell features. Neither glandular structures nor benign component were visualized. Multinucleated tumor cells were absent (Fig. 1). Immunohistochemistry revealed positivity for pan-cytokeratin (CK) AE1/AE3, EMA, vimentin, CD10, CD138, α-SMA, focally for p53 and CD68 (KP1), and scarce cells for CAM5.2, GFAP and Cyclin D1. SMARCB1 (INI-1) expression was intact. Moreover, CK5/6, CK7, CK20, S100, p16, p40, p63, D2-40, calponin, desmin, CD34, CD56, CD57, CD99, CD163, HLA-DR, HMB-45, chromogranin and synaptophysin were negative. ISH for human papillomavirus (HPV) wide-spectrum and types 6/11 and 16/18, as well as for Epstein-Barr virus (EBV) (EBER1/2), were negative. Ki-67 was > 40% (Fig. 2). A diagnosis of myoepithelial carcinoma with rhabdoid features was made. The anatomopathological examination of the surgical specimen confirmed the diagnosis. The patient was referred to an oncology center, which reported progressive deterioration of the patient's health. After this, he was lost to follow-up.

Discussion

The cytological features in myoepithelial tumors includes epithelioid, spindle, hyaline (plasmacytoid), clear and mixed cell types [1,6], with myoepithelioma occasionally presenting basaloid and oncocytic cytoplasmic features [1]. Moreover, a mucinous variant of myoepithelial tumors has also been recognized [1]. Approximately 10% of all myoepithelial tumors arising from salivary glands are myoepithelial carcinomas. Remarkably, some myoepithelial carcinomas arise from unusual head and neck locations, such as nasal cavity [3], nasopharynx [4] and maxillary sinus [5–8]. Recently, a study assessed 7190 salivary gland tumors, of them, only 66 cases (5 benign [pleomorphic adenoma

https://doi.org/10.1016/j.oraloncology.2019.04.015
Received 17 April 2019; Accepted 22 April 2019
Available online 30 April 2019
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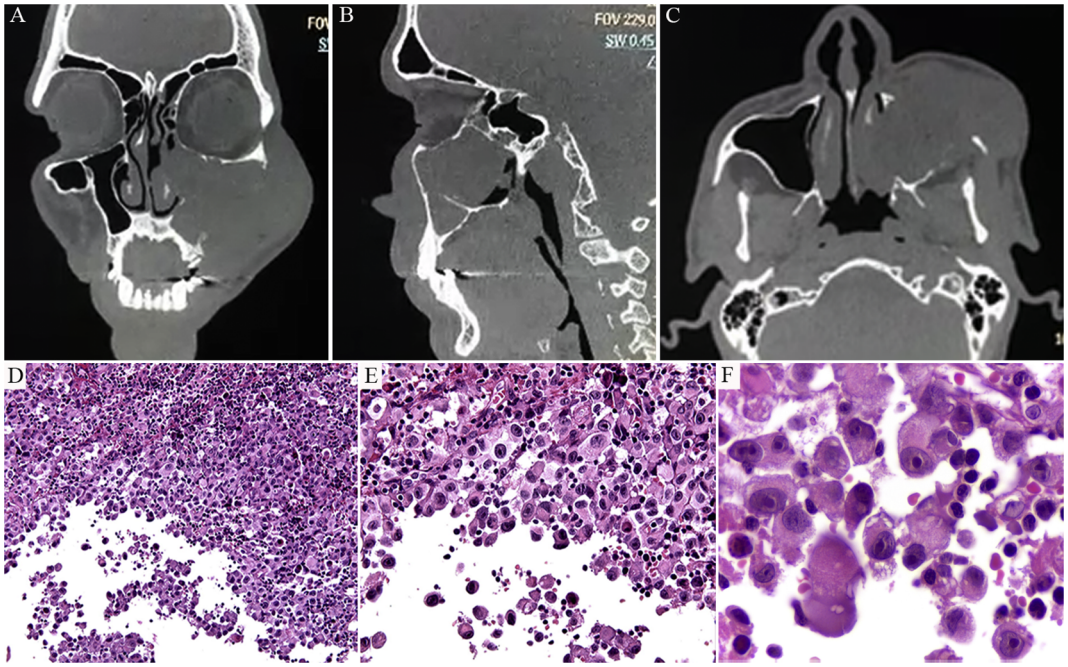

Fig. 1. Computerized tomography scan showing extensive osteolytic lesion involving the left maxillary sinus in frontal (A), sagittal (B) and axial (C) views. Histophatological features on H&E stain, exhibiting sheets and discohesive rhabdoid cells with large nuclei, prominent nucleoli and abundant eosinophilic cytoplasmic inclusions (D, ×20; E, ×40; F, ×100).

and myoepithelioma] and 61 malignant cases [adenoid cystic carcinoma, adenocarcinoma NOS, mucoepidermoid carcinoma, among others]) showed maxillary sinus involvement, with none case diagnosed as myoepithelial carcinoma [9]. To the best of our knowledge, only 5 cases of myoepithelial carcinoma in the maxillary sinus have been reported to date [5–8]. Of them, 3 were male and 2 females. The age range varied from 38 to 67 years (mean, 50 years). The treatment of choice (3 cases) was maxillectomy [5–7].

Although rhabdoid features have been rarely described in salivary duct carcinoma [10] and carcinoma ex-pleomorphic adenoma [11], this distinctive microscopic finding, with apparent prognostic impact, has not been demonstrated in detail assessing sinonasal myoepithelial carcinoma. In fact, there is two myoepithelial carcinoma with rhabdoid features affecting the breast [12] and vulva [13], and one soft tissue myoepithelial carcinoma of the neck with rhabdoid morphology, which showed loss of nuclear SMARCB1 (INI-1) immunoexpression and EWSR1 rearrangement by fluorescence ISH [14], confirming the rarity of such findings. Noteworthy, EWSR1-rearranged myoepithelial carcinomas of salivary gland origin can also be observed [15].

The first morphological description of rhabdoid features in myoepithelial carcinoma seems to correspond to Savera et al. [6] study, which observed this phenotype assessing 3 (predominantly plasmacytoid) out of 25 cases of salivary myoepithelial carcinoma. Similarly, in the current case, a plasmacytoid/epithelioid cell component was also observed. Moreover, in this study [6], only one case (low-grade, clear cell predominant) with maxillary sinus involvement was described, affecting a 38-year-old man, which despite maxillectomy and chemotherapy, presented recurrence and metastasis on the scalp, dying of disease after 72 months of follow-up.

Squamous cell carcinoma (SCC) with a rhabdoid phenotype is extremely rare in the maxillofacial region. To date, only 3 cases in the oral cavity have been reported, often associated with a worse prognosis [16–18]. Two were female and one male, with mean age of 55 years. The sites involved were gingiva, buccal mucosa and vestibular fornix.

Based on the large epithelioid and rhabdoid cell morphology in the sinonasal region, the differential diagnosis includes sinonasal undifferentiated carcinoma (SNUC), SMARCB1-deficient carcinoma, HPV-related multiphenotypic sinonasal carcinoma, malignant melanoma, anaplastic large cell lymphoma, adamantinoma-like Ewing sarcoma, large cell neuroendocrine carcinoma, NUT carcinoma, PEComa, and epithelioid variants of fibrosarcoma, myxofibrosarcoma, leiomyosarcoma, rhabdomyosarcoma, angiosarcoma, liposarcoma, malignant peripheral nerve sheath tumor, among others [19]. Interestingly, the SMARCB1-deficient carcinoma (in the last WHO classification, belonging to the SNUC spectrum) is notably by variable presence of plasmacytoid/epithelioid and rhabdoid cells [19]. All these tumors can be differentiated from the current case, through strict and detailed morphological, IHC and ISH analysis.

In summary, although rare, myoepithelial carcinoma with rhabdoid features should be considered in the differential diagnosis when assessing maxillary sinus malignant neoplasms.

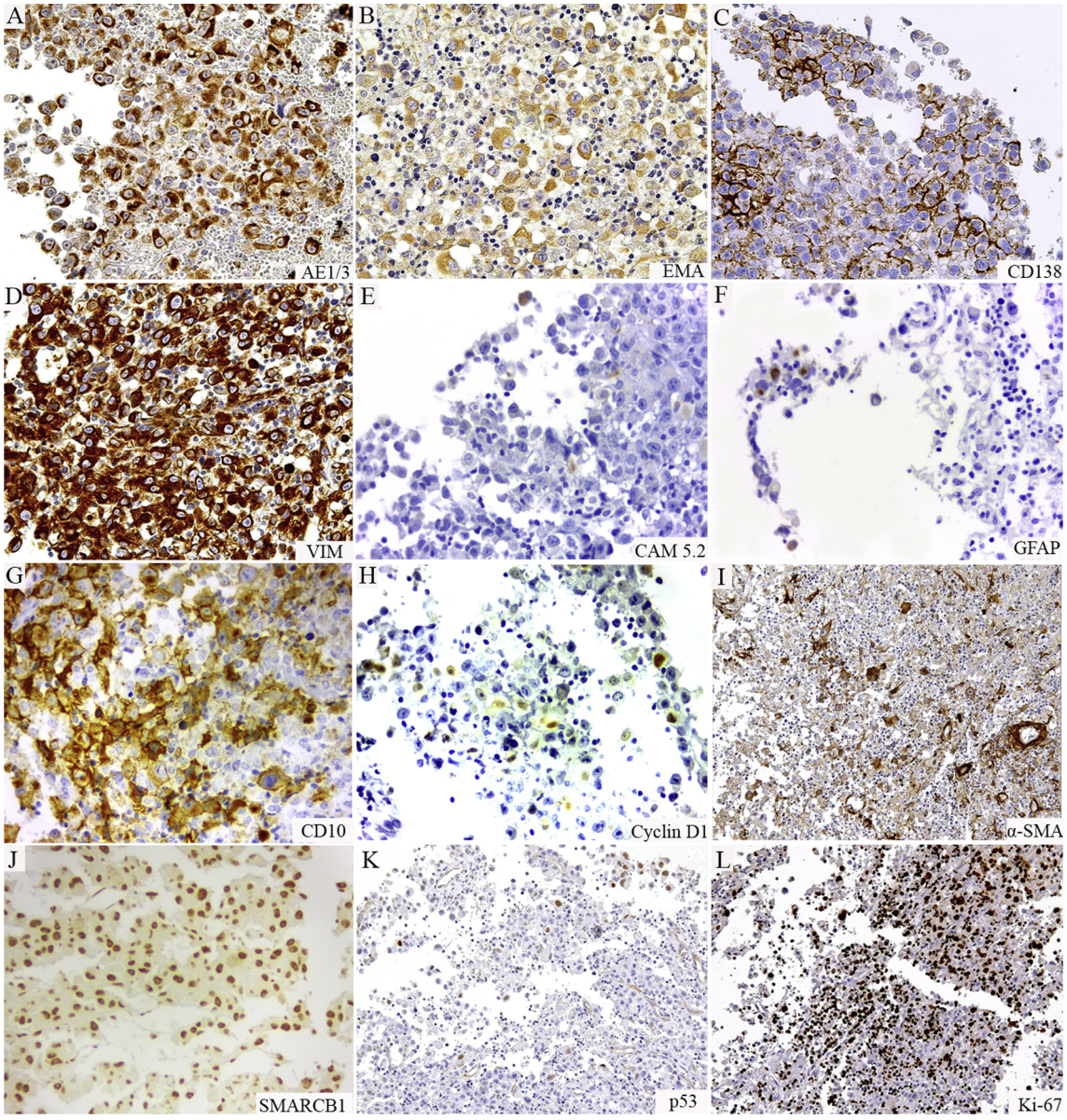

Fig. 2. Immunohistochemical analysis showing strong paranuclear cytoplasmic positivity for CK AE1/AE3 (A, ×40). Similarly, EMA exhibited a cytoplasmic rather than membranous pattern positivity (B, ×40). Strong CD138 membranous pattern positivity (C, ×40). Vimentin showing strong paranuclear cytoplasmic pattern positivity (D, ×40). Scarce cells weakly positive for CAM5.2 (E, ×40) and GFAP (F, ×40). The most cells were CD10 positive in dot-like cytoplasmic and membranous staining patterns (G, ×40). Cyclin D1 was expressed by few cells (H, ×40). Notably, α-SMA cytoplasmic positivity was evident (I, ×20), and SMARCB1 (INI-1) expression was intact (J, ×40). Focal expression of p53 was visualized (K, ×20), and Ki-67 labeling index was > 40% (L, ×20).

Funding

Luciana Yamamoto Almeida (2016/02713-2), Heitor Albergoni Silveira (2018/12734-2) and Jorge Esquiche León (2016/11419-0) have received research Grants from State of São Paulo Research Foundation (FAPESP).

Conflict of interest

The authors have no conflict of interest.

Ethical approval

This article does not contain any studies with human participants or animals performed by any of the authors.

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