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Case Report

A Rare Case of a Minor Salivary Gland Carcinoma with a Voluminous Cardiac Metastasis

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Abstract

Minor salivary gland carcinomas are rare and usually they have not tendencies to metastatize. Metastases to the heart are extremely rare too. We report the case of a 78 year old woman with minor salivary gland carcinoma and a voluminous heart metastasis, virtually unknown in this type of carcinomas.

Keywords: Cardiac magnetic resonance; Metastasis; Salivary gland carcinoma

Introduction

Minor salivary gland carcinomas of the head and neck are a group of rare cancers with significant heterogeneity in histological types and ultimate clinical behavior. Literature on minor salivary tumors is limited due to the rarity and diversity of these tumors [1]. Few centers have enough volume of cases to report on meaningful outcomes. The WHO classification of Head and Neck tumors was used for the histological classification and for definitions of grade within pathological groups. Tumors were categorized into low, intermediate and high histology risk groups to allow for analysis because of the large number of histological types. Tumors categorized as high risk were salivary duct carcinoma, high grade Mucoepidermoid Carcinoma (MEC), high grade Carcinoma Ex-Pleomorphic Adenoma (CEPA), high grade adenocarcinoma, high grade myoepithelial, high grade Adenoid Cystic Carcinoma (ACC), and high grade acinic cell carcinoma. Intermediate risk tumors included ACC. Tumors classified as low risk included low grade acinic cell carcinoma, low and intermediate grade MEC, low grade adenocarcinoma, low grade CEPA, low grade myoepithelial carcinoma and all Polymorphous Adenocarcinoma (PLGA) and epithelial-myoepithelial carcinomas. Recently PLGA has been reclassified as Polymorphous Adenocarcinoma (PAC).

Metastases are the most common tumors of the heart followed by primary benign cardiac tumors while primary heart carcinomas come last [2]. Metastatic spread to the heart has been identified in approximately one-fifth of all patients who have metastatic cancer, with lung carcinoma being the most common primary tumor and with an overall incidence of 1.23% [3]. Symptoms of cardiac metastases are extremely various, largely depending on the site and extent of the lesions, ranging from the complete absence of symptoms to signs and symptom of acute heart failure or embolic events. Treatment also varies depending on the etiology of the primary tumor and in some cases heart surgery may be indicated, mainly with the final aim of symptomatic relief. The advent of advanced cardiovascular imaging modalities has allowed the precise non-invasive diagnosis and characterization of such formations [2]. We present a case of a metastatic PAC with a voluminous cardiac metastasis which is virtually unseen in this type of carcinoma and where advanced cardiac imaging modalities played a central role in its characterization.

Clinical Case

A 70 years old woman with no significant past medical history attended the hospital because of palpable nodule on her tongue. She underwent a right hemiglossectomy and lymphnodes sampling. The histopathologic diagnosis was polymorpus adenocarcinoma (PAC) of the minor salivary glands (stage I). One year later the patient had a laterocervical lymphnode recurrence and she had lymphnodes dissection followed by radiotherapy. After further 8 years, recurrence in right retroauricular lymphnode was documented. Histology confirmed the initial diagnosis. Total-body CT documented bilateral lung metastases, multiple subcutaneously metastases, and a voluminous round-shaped mass (maximum dimensions 45 x 40 mm on the axial view) expanding

within the right ventricular cavity (Figure 1). In order to evaluate the hemodynamic impact of the mass and to perform a precise tissue characterization, we performed a contrast-based CMR. CMR cine imaging confirmed the presence of the mass occupying most of the right ventricular cavity but surprisingly, with no significant hemodynamic impact (Figure 2, panel A). The short Tau inversion recovery imaging (STIR), a T2-weighted imaging protocol designed to visualize increased water content/edema and suppress fat signal, showed diffuse enhancement, indicating high water (vascular) content within the mass (Figure 2, panel B). On CMR first pass contrast perfusion there was heterogeneous enhancement within the formation, which was also evident on delayed gadolinium enhancement imaging (LGE), confirming the vascular nature of the mass (metastasis), and with no evidence of extracardiac involvement (Figures 2, panel C, D). Overall, clinical conditions remained stable but in consideration of the extent of the cardiac mass, we decided to start a chemotherapic treatment with weekly paclitaxel. The CT performed after 3 cycles showed partial response of disease and stability of the mass in the heart. She continued until 6 cycles of chemotherapy with stable disease and good clinical conditions.

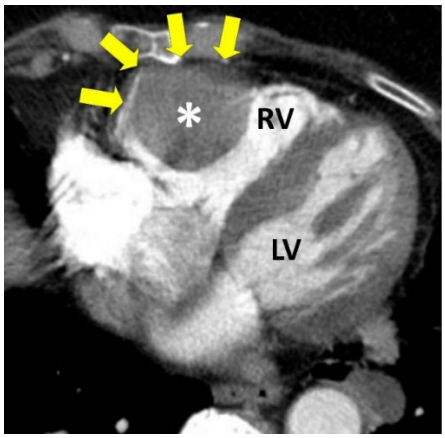

Figure 1: Contrast-based Chest CT. Axial view showing a voluminous round-shaped mass (white asterisk) expanding within the right ventricular cavity (arrows). RV: right ventricle; LV: left ventricle.

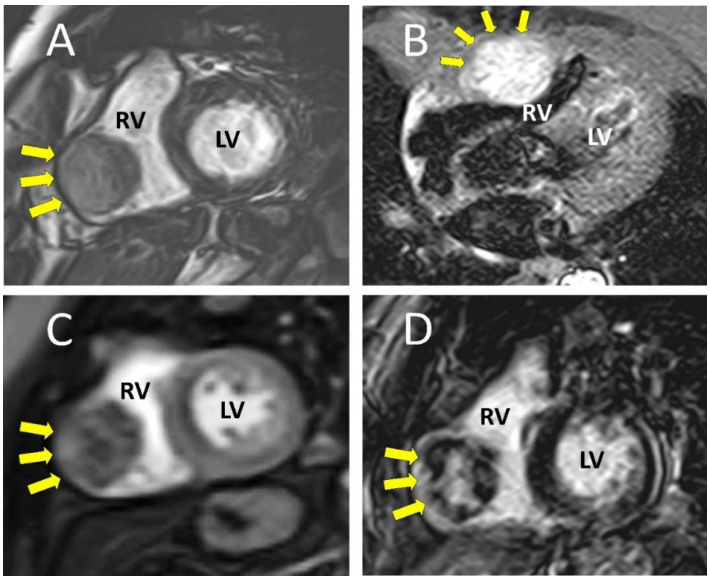

Figure 2: Contrast-based Cardiac Magnetic Resonance. Panel **A:** CMR-Cine imaging in the short axis view showing a round-shaped mass at the level of the basal lateral wall of the right ventricle with well-demarcated borders (arrows). Panel **B:** STIR-T2-weighted imaging in the 4-chamber long axis view, showing hyperintense signal of the entire cardiac mass (arrows) indicating high water content (vascularized tissue). Panel **C:** CMR-Perfusion imaging in the short axis showing heterogeneous perfusion of the mass (vascular structure). Panel **D:** CMR-Late Gadolinium enhancement imaging in the short axis view showing heterogeneous contrast enhancement typical of a vascular mass. LV: left ventricle; RV: right ventricle.

Discussion

Polymorphous adenocarcinoma (PAC), previously called polymorphous low-grade adenocarcinoma (PLGA), of the salivary glands is a rare cancer and reports on heart metastasis are virtually unknown. Population-based studies report an annual incidence of 0.051 cases per 100 000 individuals [4]. It is the second most common minor salivary gland tumor found in the oral cavity [5]. Its incidence in a major salivary gland is much lower [6].

PAC was initially described as a low-grade histology with low metastatic potential and excellent survival [7]. However, a recent update made by the salivary gland section of the World Health Organization (WHO) decided to remove the term low-grade due to emerging evidence showing recurrence rates of up to 19% and cases of transformation to high-grade malignancies [8].

In a Memorial Sloan Kettering experience, 450 patients with minor salivary gland tumors of the head and neck were evaluated [9]. The 5-year predicted overall survival was 86% and the disease specific survival was 94% at 5 years. AJCC stage and pathology were the most predictive variables across all outcomes. In line with previously published studies, a cohort of 54 patients has excellent survival, with a 5- and 10-year survival rate of 88%–79%, and a 5- and 10-year Disease-Specific Survival (DSS) of 98% and 94%. The survival difference between survival rate and DSS highlight the indolent behavior of this carcinoma, as the majority of deaths are not cancer related [10]. Despite being exceedingly rare, cardiac tumors form an important component of cardio-oncology practice in which diagnosis and management are vital. Neoplastic lesions can be further classified into primary and secondary tumors (i.e., metastasis to the heart). Up to 90% of primary neoplastic tumors may originate from the pericardium or myocardium. However, metastatic cardiac masses represent the most common tumor of the heart, being 20 to 40 times more common than primary cardiac tumors [11]. Hemodynamic impact and symptoms largely depend on localization and the extent of the cancer.

The advent of advanced cardiovascular imaging modalities has allowed the non-invasive diagnosis of such formations. In particular, contrast-based cardiac CT and Cardiovascular Magnetic Resonance (CMR) represent the gold standard imaging modalities for the identification of cardiac masses, allowing the precise morphological characterization, the evaluation of hemodynamic impact and the possible involvement with extracardiac structures. Our case exemplifies as these advanced cardiac modalities, being non-invasive in nature, fast and reliable, should be implemented whenever a cardiac involvement is suspected as they provide incomparable information in respect to the most common traditional imaging modalities.

Conclusions

Salivary gland carcinoma are considered a very rare disease, as evidenced by a review of nearly 7000 salivary gland tumors [12], that found an incidence of only 1% of polymorphous low-grade adenocarcinoma. This case of minor salivary gland PAC report herein is of particular interest for its aggressive nature and the occurrence of large cardiac metastasis. The last decade has seen the discipline of cardio-oncology come into its own. Our aim in presenting this case is to increase awareness of intracardiac metastases and to stress the importance of taking advantage of advanced cardiac imaging to characterize the disease.

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