Pleural effusion as the presenting feature of palatal adenocystic carcinoma recurrence

T. Fevereiro*, R. G. Matos***, A. Ferreira**, P. Morais***, A. Ferreira de Almeida****

Abstract

The authors present the case of a 68 years-old female patient with a right side pleural effusion, without cardiovascular or infectious disease complaints.

The final diagnosis was of a secondary pleural tumor: an adenocystic carcinoma with the primary location on the hard palate, which had been diagnosed and treated 8 years previously, with no subsequent local recurrence.

The authors highlight the rarity of this diagnosis and the fact that whilst pulmonary metastization with pleural involvement has been described, it is extremely rare to find solitary pleural metastasis from this primary tumor.

Key words: secondary pleural tumor, adenocystic carcinoma of the hard palate, pleural effusion.

Introduction

Uni- or bilateral pleural effusion is a common presentation of cancer, and is expected to occur in around 50% of cases of malignant neoplasms, in any stage of their evolution.[1]

Malignant pleural effusion is the second most common cause of pleural exudate, while the first is bacterial infection, especially tuberculous pleurisy. The neoplasms most frequently involved are lung carcinoma (30%), breast neoplasm (25%) and lymphoma (20%).[1]

The approach to this situation, with the aim of identifying the primary lesion, ranges from a correct and exhaustive anamnesis to the correct hierarchy of the supplementary diagnostic methods, which include imaging exams and invasive techniques. In this situation, the integrated intervention of various medical and surgical specialties is extremely useful, even compulsory.

Case Report

A 68-year-old female patient is hospitalized for clarification of a profile of symptoms with evolution of about 2 months, with progressive exacerbation, non-productive cough that is worse in the supine position, dyspnea on exertion, non-selective anorexia, unquantified weight loss, asthenia and adynamia.

The patient denied fever, excessive sweating, thoracalgia or haemoptysis. There was no history of pulmonary tuberculosis or known contact with patients with active pulmonary tuberculosis, nor was there any known history of respiratory pathology, complaints compatible with chronic bronchitis or bronchial asthma. Smoking habits and known professional or recreational exposure to pollutants were also excluded.

The patient had no known history of hypertension, and had no other complaints suggestive of heart failure. Nor were there any complaints related to other organs or organ systems.

In the individual history, there was an adenoid cystic carcinoma of the hard palate diagnosed about 8 years before the current hospitalization, which was removed by surgery (tumorectomy with partial resection of the hard palate), with the histological examination having revealed the presence of neoplasm at the edges of the excision, lying horizontally, with no evidence of this neoplasm at greater depths. The patient was later submitted to 10 radiotherapy sessions (method and dose unknown), and did not present local recurrence of the carcinoma, according to the attending physician.

*Resident to the Internal Medicine Supplementary Internship

**Resident to the Dermatology Supplementary Internship

***Internal Medicine Hospital Assistant

****Head of Service

Medicine III Service of the Hospital de Santo António dos Capuchos, Lisbon

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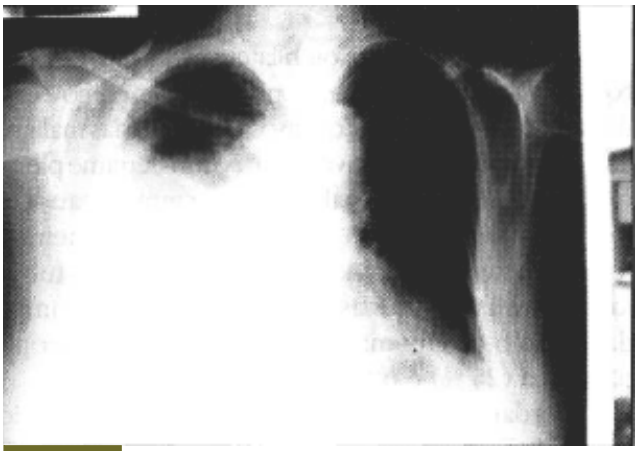

FIG. 1

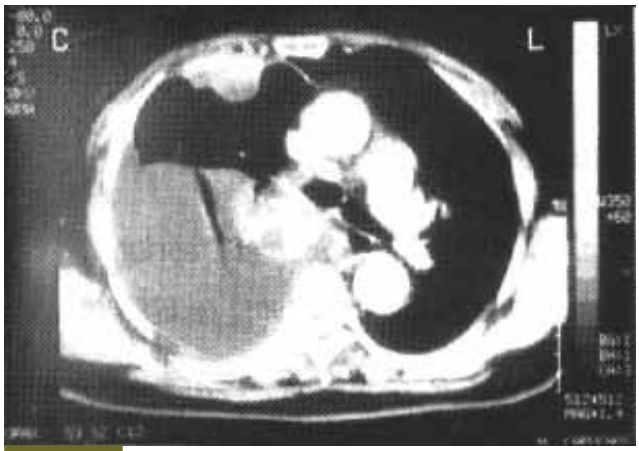

FIG. 2

FIG. 3

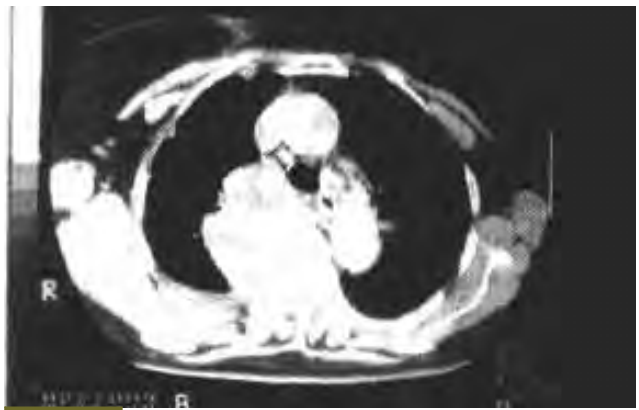

FIG. 4

Objective examination

Aspects emphasized in the observation of this patient were good general condition and nutritional state and vital parameters within normal range, except for polypnoea associated with mobilization.

Semiological evaluation of the respiratory system revealed alterations compatible with pleural effusion, affecting the lower two thirds of the right hemithorax, without semiological alterations on the left. The palate, breasts and abdomen did not present alterations, and neither did the gynecological and anorectal examinations.

Supplementary diagnostic tests

Chest teleradiography confirmed the semiological evaluation (*Fig. 1*).

Thoracocentesis was carried out with collection of serohaematic fluid, laboratory evaluation of which revealed an exudate, with abundant signet ring cells. The bacteriological examination, both direct and of culture, was negative.

Serum tumor marker dosages revealed high CEA, CA 19.9 at the upper limit of normal and remaining markers within the normal range of values. The Mantoux test, performed with 1 unit of tuberculin PPD, was negative. Thoracic CT scan (*Fig. 2*) revealed the presence of a 5x3 cm mass, forming an obtuse angle with the pleura, in which it was not possible to discern the organ of origin: lung vs. pleura. The left lung and the mediastinum did not exhibit any alteration, particularly aspects compatible with neoformative lesions.

It was decided to perform echography-guided biopsy, histological analysis of which revealed aspects compatible with mesothelioma, where it was not possible, in the anatomical pathologist's opinion, to rule out a hypothesis of an adenocarcinoma. The mammography and the abdominal and pelvic echo-

graphy, as well as the abdominal and pelvic CT, cranio encephalic CT and bone scintigraphy, were negative, both in the localization of a possible primitive tumor, and in the detection of metastases.

After interdisciplinary clinical discussion, the patient was submitted to a thoracotomy with excisional biopsy, which revealed the presence of a mass, located in the right hemithorax, sessile and mamillated, exclusively pleural, not involving the pulmonary parenchyma, the histological analysis of which (*Fig. 3*) was compatible with metastatic adenoid cystic carcinoma of the minor salivary glands.

Another therapeutic option, namely radiotherapy, was excluded due to the poor response that this type of tumor exhibits when metastasized. In making this decision, we considered the limited benefit of this adjuvant therapeutic approach vis-à-vis the probable loss of quality of life.

The patient is currently in a stable clinical situation, ambulatory, and undergoing periodic tomographic control. The most recent examination, carried out two months after the thoracotomy (*Fig. 4*), reveals multiple pleural metastases on the right, without involvement of the pulmonary parenchyma and with mediastinal adenopathies, indicating the lymphatic pathway as the most likely to be involved in this unusual dissemination.

Discussion

Minor salivary gland tumors represent about 15% of salivary gland tumors,[2-3] responsible for 0.34% of all malignant tumors[4] and for about 3 to 4% of head and neck tumors,[5-6] and their most frequent histological type is the adenocystic carcinoma.[7] They are most often located in the hard palate, in the upper lip and in the cheek mucosa, and habitually appear as slow--growing masses, sometimes taking a decade to be detected, or else they can take the form of painful or ulcerated masses.[3-4, 8-10]

The metastization of these tumors is performed by lymphatic or neuronal pathway, or by contiguity to the neighboring bone.[3-4, 8-10]

The usually recommended treatment is surgical excision, followed by local radiotherapy.[3-4, 8-10]

The bibliography that we consulted refers to the metastization of these tumors in about 40% of cases for the lung, with possible secondary involvement of the pleura.[10] Isolated pleural metastization is not described as possible, and we did not find any account in the literature of clinical cases with this form of secondary presentation in the bibliography.

We emphasize, in this very common form of pathological presentation, the rarity of the final diagnosis, the limited specificity of some supplementary diagnostic methods, such as the pleural fluid analysis, and even the echography-guided biopsy, the importance of anamnesis and the need, which is always pressing in Medicine, for interdisciplinary. ■

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