Solitary pulmonary metastasis of mucoepidermoid carcinoma of the palate 43 years after the initial treatment

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Abstract

This report describes the case of a 71-year-old female presenting with a metastatic mucoepidermoid carcinoma (MEC) in the lung 43 years after the initial treatment for the primary tumor. This case represents a very long period between initial diagnosis and distant metastasis with a pathological examination of both the primary and metastatic tumors. Metastatic tumor should be considered for the differential diagnosis of a pulmonary nodule in a patient who has a history of this type of oral tumor.

Keywords: Histology (mucoepidermoid); Metastasis

1. Introduction

In 2007, a 71-year-old female was admitted to the hospital for further evaluation of single 16 mm pulmonary nodule in the right lower lobe (Fig. 1), which was detected on a screening chest CT to assess a slight chest discomfort. Surgery was performed, based on the suspicion of lung cancer. The intra-operative pathological diagnosis revealed that the tumor was malignant, but no definitive diagnosis could be obtained. Therefore, a lower lobectomy was performed as a standard procedure for primary lung cancer. Macroscopically, the resected tumor was firm and white with a well-defined edge.

The past medical history indicated a malignancy in the oral cavity. In 1964, the patient presented with a small (~5 mm), firm, and painless swelling on the palate at the age of 32. The patient underwent a local tumor excision at a primary care physician without pathological diagnosis. A 5 mm tumor recurred at the same place in 1967, and the patient underwent a re-excision at a cancer hospital. The tumor was pathologically diagnosed as mucoepidermoid carcinoma (MEC) of the palate. In 1970, the tumor recurred again at the same place. The patient underwent an extended tumor resection including bone and palate. Since then, the patient has been healthy without any recurrence.

Hematoxylin-eosin staining of the specimen showed non-keratinizing squamous cells with ample eosinophilic cytoplasm that grew in a trabecular pattern and scattered mucous cells were observed with PAS staining (Fig. 2a, b). Suspecting a metastatic MEC, the tumor sample resected in 1970 was obtained and compared to the pulmonary nodule. The cellular and histological appearance of the oral tumor was identical to the pulmonary nodule (Fig. 2c, d). In addition, both tumors were positive for CK34bE12 but negative for CK7 or CK20. Based on these findings, the lung tumor was diagnosed as a solitary metastasis from the MEC in the oral cavity 43 years after the initial treatment. No additional treatment was offered, and the patient remained healthy for 24 months.

2. Comment

MEC is the most common salivary gland malignancy [1]. Approximately, half of tumors occur in minor salivary gland, especially in the palate. Most patients with this disease have a favorable outcome after a complete resection [2, 3]. However, the disease may recur in distant organs in a small subset of the patients during long-term follow-up periods. A review of 173 salivary gland MECs [3] noted that distant metastases affected 16 patients (9.2%), most frequently in the lungs. Although late recurrence is not rare in MEC, this case had an exclusively long interval between the initial treatment and distant metastasis. Furthermore, this is thought to be the longest interval among the case reports where a histological examination for both primary and metastatic tumor could be performed.

MEC is characterized by squamoid, mucus producing and cells of an intermediate type. MECs are usually classified as low-, intermediate-, or high-grade malignancies based on the histological variables including necrosis, mitosis, perineural invasion, and dominant type of cells [1, 4]. This case was classified into an intermediate-grade malignancy. From the clinical point of view, the repeated local recurrences suggest that the malignant potential of this disease is relatively high.

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Besides the salivary glands, the lung is one of the primary sites where MEC may develop [3]. However, most of the pulmonary MECs arise from bronchial glands in the central airways. Since the tumor in this case was located in peripheral lung parenchyma with clear defined margin, the disease was diagnosed metastatic MEC.

The question when the tumor began to grow and formed a metastatic lesion is interesting but cannot be answered. Tumor cells might have been disseminated and present in the lung when the primary and local recurrent tumor was treated. This kind of phenomenon is often explained by cancer dormancy, in which residual disease is present but remains asymptomatic or undetectable [6]. Although the precise mechanism of this hypothesis remains poorly understood, various factors have been identified as possible contributors.

In summary, a metastatic tumor should be considered for the differential diagnosis of a pulmonary nodule in a patient who has a history of this type of oral tumor.

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References