

CASE REPORT

Primary Pleomorphic Adenoma of the Lung

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Introduction

Despite being the most common type of benign salivary gland lesion, pleomorphic adenomas are encountered rarely as primary pulmonary tumors. Fewer than twenty cases have been presented in the literature.¹⁻⁵ When described in the lung, these tumors tend to be centrally located and associated with a major or secondary bronchus.^{1,4,5} Since they are so rare, there is no standard-of-care for the treatment of these lesions. However, similar to their salivary gland counterparts, the consensus is to resect them surgically with clear margins. Their incomplete excision may be associated with local recurrence.¹ We present a rare benign pulmonary tumor that was diagnosed as a primary pleomorphic adenoma.

Case Report

A 61-year-old woman was evaluated in the outpatient setting for a new lung mass. With a previous history of breast cancer, the patient underwent a routine chest x-ray that illustrated a right-sided lung density. On computed tomography (CT) scan of the chest, there was a 3.5 cm hilar mass associated with post-obstructive atelectasis (Figure 1). Aside from breast cancer, she denied any other medical co-morbidities. She had never smoked and had no symptoms of weight loss, chest pain, dyspnea, cough, or hemoptysis. She had no salivary gland symptoms such as mandibular pain, swelling, or facial nerve palsy. Her physical examination was unremarkable.

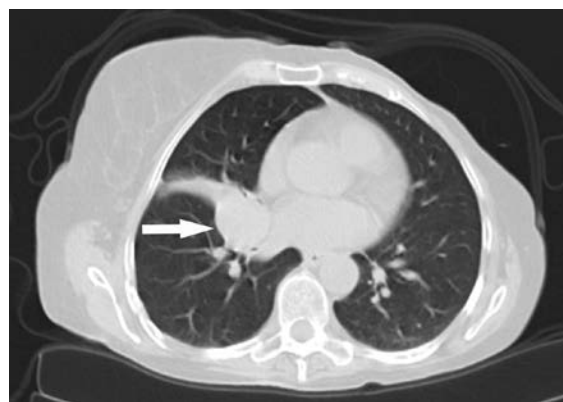


Figure 1. CT scan of the chest illustrating a 3.5 cm right hilar mass (arrow).

On initial work-up, peripheral blood counts were normal. A full body positron emission tomography (PET) scan showed hyper-metabolism in the right hilum with a standard uptake value of 5.1 consistent with the location of the lesion noted on the CT scan. There were no other regions of hyper-metabolism, including the salivary glands. Additionally, there was no evidence of mediastinal adenopathy. Because of its proximity to and involvement of the right hilum, a bronchoscopy with biopsy of the lesion was conducted. The pathology illustrated positive immunohistochemical staining for pankeratin, vimentin, smooth muscle actin, glial fibrillary acidic protein, and S-100. These findings substantiated the diagnosis of primary pulmonary pleomorphic adenoma.

Pulmonary function testing demonstrated: vital capacity (VC) = 3.26 L, %VC

= 98%, forced expiratory volume (FEV1) = 2.54L, %FEV1 = 103%, carbon monoxide diffusing capacity (DLCO) = 19.3mL/mmHg/min, %DLCO = 104%. Surgical resection was recommended and a right posterolateral thoracotomy was performed. At the time of surgery, the tumor was found to encroach into the bronchus intermedius. A right middle and lower lobectomy had to be performed. The patient tolerated surgery without any complications.

The final pathology following her lobectomies illustrated a pleomorphic adenoma measuring 4.5 x 3 x 3 cm in size with invasion into the bronchus intermedius (Figure 2). The bronchial resection margin was free of tumor, and the six identified regional lymph nodes were free of invasion.

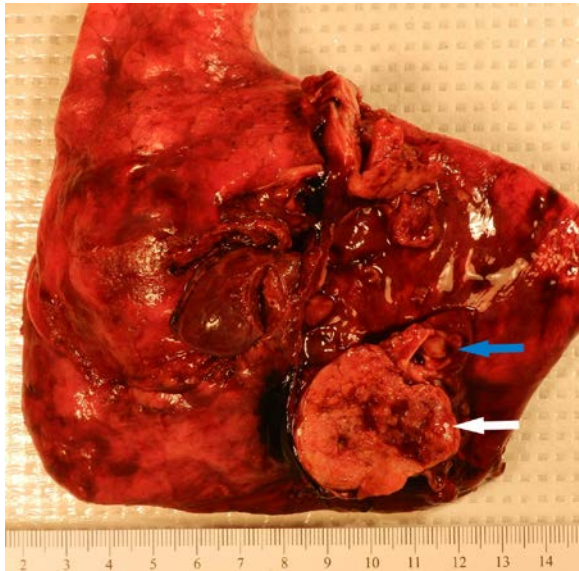


Figure 2. Photograph of the gross pathologic specimen illustrating the pleomorphic adenoma (white arrow) with invasion into the right bronchus intermedius (blue arrow).

The patient was dismissed from the hospital on post-operative day three. Her follow-up in the outpatient setting was continued with routine chest imaging

because of her previous history of breast cancer. It also allowed for screening any possible recurrence of her pleomorphic adenoma. At eighteen months of follow-up, she was without complications or recurrence.

Discussion

Pleomorphic adenoma rarely presents as a primary pulmonary lesion. When it does, it typically arises from seromucous glands of the trachea and major bronchi.^{3,5} Because of their rarity, the true incidence of these lesions is unknown. In addition, the prognosis of these tumors in this setting is unclear. They exhibit similar microscopic and immunohistochemical characteristics as those seen in salivary gland tumors.³ In the absence of metastases, the current consensus is to treat these lesions with complete surgical resection, including negative margins. Long-term surveillance is recommended because of the possibility of local recurrence, which, as in the case of their salivary gland counterparts, may present years following surgical excision.¹ While long-term surveillance is recommended, there are no specific guidelines on the type or frequency of imaging.

This was a rare case of a primary pleomorphic adenoma of the lung in a patient who was asymptomatic. Tissue diagnosis was used to guide her overall treatment. The patient remained healthy without any further evidence of recurrence or surgical complications. The appreciation for the presence of these tumors in the lung allow for appropriate surgical therapy. It also provides an opportunity for more expansive dialogue that may lead to definitive standard-of-care guidelines for treatment and surveillance follow-up.

References

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