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Pulmonary Hamartoma with Left Hilar Endobronchial Extension in a 63-year-old Female: A Case Report

Rollin P. Tabuena

M.D., FPCP, FPCCP; Dhenver S. Fenis, MD; Ray V. Olac, MD

ABSTRACT

The most common benign lung tumor is pulmonary hamartoma, though its incidence varies between 0.025% and 0.032% in the adult population. This case introduces a 63-year-old female Filipino has a history of paroxysmal coughing fits. The patient initially requested a chest X-ray; it showed a pulmonary mass in the upper left lobe. A computed tomography (CT) scan of the plain chest was then taken, showing an inhomogeneously enhancing pulmonary mass in the left upper lobe with endobronchial and hilar extension. Additional diagnostic evaluations included immunohistochemical test, CT-guided core needle biopsy, transbronchial needle aspiration biopsy, and bronchoscopy. Every investigation consistently found pulmonary hamartoma and found no evidence of cancer. This case highlights how important a thorough diagnostic work-up and a multidisciplinary approach are to the successful treatment of pulmonary hamartoma.

Keywords: Pulmonary hamartoma, pulmonary mass, endobronchial, bronchoscopy, biopsy, immunohistochemistry test, malignancy, case report

INTRODUCTION

The word "hamartoma" comes from the Greek word "hamartia," which means "erroneous" or "faulty." Hamartomas are typically benign tumors that can develop in the skin, breast, heart, lungs, and other anatomical locations. Usually consisting of two or more mesenchymal components, pulmonary hamartomas are benign lung malformations. These could be leiomyomatous, chondromatous (the most prevalent kind), or both. Rarely, they might be primarily made up of fatty tissue or be lipomatous.

In adults, pulmonary hamartoma is the most common benign lung tumor and the third most common cause of isolated pulmonary nodules. Its prevalence in the adult population is, however, comparatively low, ranging between 0.025% and 0.032%. Male-to-female ratios are estimated to be between 2:1 and 3:1, with the condition being more common in people in their fifth and sixth decades of life. While about 10% of cases present endobronchially, the majority of pulmonary hamartomas are found in the peripheral lung parenchyma.

The lesions are frequently discovered by chance on chest imaging, and the majority of patients with pulmonary hamartomas do not exhibit any symptoms. On radiography, they show up as distinct, lobulated, or round masses. Most of these tumors grow slowly and have a diameter of less than 4 cm, though they can occasionally grow to more than 10 cm.[11] The location of the tumor has a significant impact on how

pulmonary hamartoma presents clinically. While endobronchial hamartomas can cause symptoms like coughing, hemoptysis, pneumonia, or dyspnea, parenchymal hamartomas are usually asymptomatic.
A case involving a 63-year-old woman whose primary complaint was paroxysmal cough is presented in this report. Following a battery of chest imaging tests and biopsy procedures, the results were in line with pulmonary hamartoma involving the endobronchial region. About 2% of all lung neoplasms are benign endobronchial tumors combined.[14]

CASE REPORT

The case is about a 63-year-old Filipino woman who was recently diagnosed with pulmonary tuberculosis and bronchial asthma. She has a history of hypertension and type 2 diabetes mellitus, which does not require insulin. Coughing fits were the patient's initial symptom.
According to the patient's history, the start of paroxysmal coughing fits and sporadic expectoration of whitish to yellowish phlegm occurred four months before the patient was admitted. The patient saw a private doctor two months before admission due to a persistent productive cough and sporadic expectoration. She was prescribed Montelukast once daily and Prednisone for ten days, but no symptomatic improvement was observed.
A month before being admitted, the patient sought advice from a different private doctor because their symptoms had not improved despite previous treatments. They were once more prescribed Montelukast, but their symptoms persisted. When a chest radiograph was ordered, it was said to show pleural effusion and bilateral pneumonia. After starting Cefixime, N-Acetylcysteine, and Montelukast, the patient showed some improvement in their symptoms. The patient has sent a sputum specimen to DOTS, which was found to be negative as claimed. In the interim, a follow-up chest x-ray was done, revealing regression of pneumonia. However, a mass is noted in the left upper lung, as claimed. One week prior to admission, the patient sought another consultation with a private physician and was started on Prulifloxacin, Azithromycin, Rifampicin and Isoniazid, Methylprednisolone, and Montelukast.
On the day of admission, the patient was conversant, showed signs of cardiopulmonary distress, and had a Glasgow Coma Scale (GCS) score of 15. On physical examination, the left mid- to upper lung fields showed reduced breath sounds, bibasilar rales, and good air entry. Vital signs were as follows: blood pressure of 130/80 mmHg, oxygen saturation of 92% on room air, tachycardia of 110 beats per minute, tachypnea of 28 cycles per minute, and afebrile at 36.1°C. According to arterial blood gas analysis, there was mild hypoxemia and fully compensated metabolic alkalosis, requiring an inspired oxygen fraction (FiO_2) of 17%. A nasal cannula was used to put the patient on oxygen support at a rate of two liters per minute.
After a chest radiograph was taken, pneumonia with consolidation in the left upper lung was officially diagnosed. Although there was a pulmonary mass, pulmonary tuberculosis could not be ruled out. For additional analysis, correlation with computed tomography (CT) imaging was advised.
Ticarcillin-Clavulanic acid was used to start antibiotic therapy, and Rifampicin and Isoniazid were kept up. When a plain chest CT scan was ordered on the first day of the hospital, it showed an inhomogeneously enhancing pulmonary mass in the left upper lobe with hilar and endobronchial extension. The right lung's compensatory hyperaeration was also observed. A histopathological analysis was recommended. A transbronchial needle aspiration biopsy and bronchoscopy were carried out on the fifth day of the hospital stay. A pulmonary mass in the left upper lung with an erythematous and obstructed lining and irregular mucosal surfaces were among the perioperative findings. Results from a CT-guided core needle biopsy on

the seventh day of the hospital stay were in line with pulmonary hamartoma. Immunohistochemistry testing was performed on Hospital Day 10 and showed negative results for S100 and positive results for thyroid transcription factor-1 (TTF-1) and Desmin. Immunohistochemistry was used to confirm the diagnosis of pulmonary hamartoma. The patient was appraised for surgical intervention but was undecided and opted to go home and will discuss plans with surgery service on an OPD basis.

DISCUSSION

Pulmonary hamartoma is the most common benign tumor of the lung, accounting for 3% of all lung tumors. According to two comprehensive autopsy-based studies, its incidence was 0.025% and 0.032%.[5] One autopsy series revealed that pulmonary hamartomas were found in 2 out of 7,972 cases (0.025%).[8] Another large autopsy study carried out in South Africa discovered pulmonary hamartomas in 152 out of 47,635 male coal miners, which translates to an incidence of 0.32%.[9]

The nature of the mass can be described using a variety of diagnostic techniques. Usually small, well-circumscribed nodules or masses with smooth or lobulated margins are visible on computed tomography (CT) scans or chest radiographs. Lesions vary in size; most are between 2.5 and 4 cm in diameter, though some may be larger than 10 cm. About 60% of lesions are composed of fat, and 20–30% have calcification or ossification, which is frequently referred to as "popcorn-like." The average volume doubling time is more than 400 days, and growth is typically slow. Even though CT imaging is still the best method for assessment, other diagnostic procedures might be necessary. A fluorodeoxyglucose-positron emission tomography scan can help determine the rate of fluorodeoxyglucose uptake and, therefore, the metabolic rate of lesions with an indeterminate risk of malignancy.[13]

A biopsy becomes mandatory to rule out underlying malignancies in the case of lesions with an absent adipose component or those lacking the characteristic calcification pattern.

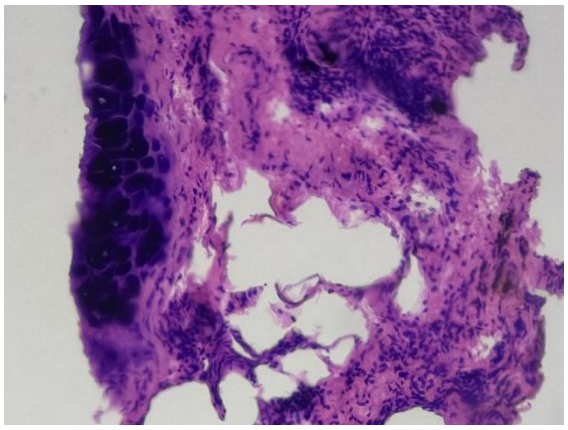

Figure 1. Pulmonary Hamartoma biopsy. The section shows hyaline cartilage tissues adjacent to mature adipose tissues. The periphery contains compressed pulmonary cells with associated lymphocytic inflammation.

Bronchoscopy with fine needle aspiration is commonly the strategy of choice, though aspirations can be scant due to the density of the lesions.[13] Masses with the typical coin appearance that fulfil CT criteria for hamartoma, including less than 4 cm in size, well-defined edges, detectable calcification, or fatty component, should remain subject to conservative follow-up with periodic observation. Resection is

reserved for fast-growing or symptomatic masses or those in which the possibility of malignancy cannot be excluded.

Pulmonary hamartoma is indicated for surgery in case of 1) a solitary pulmonary lesion with a diameter above 2.5 cm; (2) it is an overweight psychic burden for a patient; (3) having the tendency of expansion or recurrence; (4) pulmonary symptoms unresponsive to drug treatment; and (5) the lesion cannot be differentiated from malignancy.[9]

In this case, a chest CT scan revealed an inhomogenously enhancing pulmonary mass (7.0cm x 5.1cm x 5.6cm), in the left upper lobe with hilar and endobronchial extension; thus, further diagnostic workup was done. Bronchoscopy and transbronchial needle aspiration biopsy revealed a pulmonary mass in the left upper lobe. Histopathologic diagnosis also revealed negative for malignancy and consistent with pulmonary hamartoma. The patient was appraised for surgical intervention after revealing the diagnosis to the patient.

Surgery remains the only definitive curative option available. In the event of surgery, preservation of functional lung tissue is the primary goal. Therefore, enucleation and wedge resections are the most common surgical choices, with more radical lobectomy or total pneumonectomy reserved for intense lesions, multiple or large lesions that make wedge resection impossible, or lesions adhering severely to the hilum of the lung. Obtaining intraoperative frozen sections is generally recommended to avoid overlooking underlying malignant potential. Most pulmonary hamartomas are smaller than 2.5 cm, and resection is typically unnecessary if the nodule exhibits classical signs of a hamartoma or if follow-up shows no growth. However, if the lesion is very large, resection may be required due to respiratory symptoms or the risk of malignancy. Previous case reports have documented large hamartomas ranging from 8 to 20 cm, with patients experiencing significant symptoms (e.g., dyspnea and massive hemoptysis). The prognosis for patients with endobronchial pulmonary hamartoma is generally favorable.[7] Lesions are slow-growing, and surgery is curative in cases where symptoms are present and persistent. Once removed, the chances of the recurrence of endobronchial hamartoma are very low.[14] Beyond the possibility of airway obstruction, with subsequent atelectasis or recurrent pneumonia, pulmonary hamartomas have, in rare cases, been noted to bear potential for sarcomatous transformation. Common signs of malignant alteration are the rapid growth of the lesion and systemic symptoms, including weight loss, weakness, or fatigue.

CONCLUSION

Despite being acknowledged as the most prevalent form of benign lung tumor, pulmonary hamartoma is still a relatively uncommon clinical entity with a low incidence rate in the general population. Epidemiological data and autopsy-based studies show that, although it is common among benign pulmonary neoplasms, its occurrence is rare. The size, location, and accompanying symptoms of pulmonary hamartomas, which are made up of mature but disorganized lung tissues, frequently determine their clinical significance.

Based on their anatomical location, pulmonary hamartomas can be divided into two main subtypes: parenchymal and endobronchial. In the peripheral lung fields, the parenchymal subtype is much more prevalent and usually manifests as a single pulmonary nodule. On the other hand, endobronchial hamartomas are uncommon, making up only about 10% of all cases that are reported. Because of its tendency to produce recurrent respiratory symptoms, such as cough, wheezing, dyspnea, and, in certain situations, obstructive pneumonia, this endobronchial localization is clinically significant.

The initial assessment of pulmonary hamartomas heavily relies on radiographic evaluation. However, radiographs of the chest might not show anything noteworthy, especially if the lesions are small or in the middle. As a result, more sophisticated imaging techniques, like computed tomography (CT) scans, are frequently needed to define the features of the lesion. In addition to showing internal fat or calcifications, which are characteristics suggestive of hamartoma, CT imaging usually shows a well-circumscribed mass with smooth or lobulated margins.

Bronchoscopy is the preferred diagnostic procedure when an endobronchial mass is suspected due to clinical presentation or imaging results. Bronchoscopy makes it easier to sample tissue for histopathological confirmation in addition to enabling direct visualization of the lesion. In these situations, a biopsy is thought to be necessary in order to rule out the possibility of an underlying cancer, especially if there are atypical features or the lesion exhibits irregular morphology or rapid growth.

Pulmonary hamartomas rarely undergo malignant transformation, and the majority of peripheral lesions without unusual characteristics can be treated conservatively. To check for changes in size or morphology in these situations, observation combined with sporadic follow-up imaging may be adequate. However, when the lesion is symptomatic, especially when endobronchial hamartomas are obstructing airways, or when histopathological analysis shows abnormalities suggestive of malignancy, surgical resection is still the only option. After total excision, recurrence is rare and resection is usually curative.

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Ethical Consideration

The authors declared that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

Statement of Authorship

All authors certified fulfillment of ICMJE authorship criteria.

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