Pulmonary hamartoma and squamous cell carcinoma: a very rare coexistence

Aynı oğuda pulmoner hamartom ve skuamöz hücreli akciğer kanseri

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Pulmonary hamartoma is the most frequent benign neoplasm of the lung. Although they are benign, malignant transformation or accompaniment by primary lung cancer may rarely be seen. A 38-year-old woman was admitted with complaints of chronic cough and shortness of breath. A computed tomography scan of the thorax showed a 3x2-cm mass at the lower lobe of the left lung, surrounding the left main bronchus. At thoracotomy, frozen section analysis of the mass revealed malignancy, so a left lower lobectomy was performed. Histopathologic diagnosis was hamartoma and squamous cell carcinoma in situ. The patient was asymptomatic during a year’s follow-up.

Key words: Carcinoma, squamous cell; hamartoma/surgery; lung neoplasms/surgery.

Pulmonary hamartoma is the most common benign neoplasm in the lung, and accounts for 7-14% of all solitary lung nodules.[1] The majority of hamartomas are parenchymal and often located in the periphery of the lung. These tumors grow slowly. They tend to be asymptomatic and are usually found incidentally on chest radiographic examination.[2]

CASE REPORT

A 38-year-old woman was admitted with complaints of chronic cough and shortness of breath. She had a history of chronic bronchial asthma for which she received treatment for more than four years. On physical examination, there was only a widespread inspiratory rhonchus in the lungs. Laboratory data were within the normal limits. A chest roentgenogram demonstrated a nodular density in the lower lobe of the left lung. A computed tomography scan of the thorax showed a 3x2-cm mass surrounding the left main bronchus concentrically in the paramediastinal site of the lower lobe of the left lung (Fig. 1). The patient underwent bronchoscopy under local anesthesia, which demonstrated narrowing of the superior segment of the lower lobe like a fissure by a mass effect. Bronchoalveolar lavage cytology and mucosal biopsy were found benign, and transbronchial fine needle aspiration biopsy showed cells with benign characteristics. Under general anesthesia, the patient was placed in the left thoracotomy position on the operating table. After left posterolateral thoracotomy, a hard, and immobile mass, 4x3x2 cm in size, was palpated in the left lower lobe, surrounding the bronchus completely. A biopsy was taken from the mass for frozen section analysis. Histopathological findings were consistent with malignity, so a left lower lobectomy and mediastinal lymph node dissection were performed. Following hemorrhage control, two drains were placed in the left hemithorax. The layers were covered appropriately. Intraoperative bleeding was minimal. Operative time was 90 minutes. There was no complication in the postoperative period. The patient was discharged on the eighth day of the operation. Histopathological examination showed squamous cell carcinoma in situ in the layer epithelium.

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of bronchial structures accompanied by a hamartoma (Fig. 2a). The patient was asymptomatic during a year’s follow-up.

**DISCUSSION**

Hamartoma is defined as an abnormal mixture of tissue elements, or an abnormal proportion of a single element, normally present in an organ. Pulmonary hamartoma is the most frequent benign neoplasm of the lung, occurring in the parenchyma or sometimes within the bronchi.[3] It consists of an admixture of cartilage, loose collective tissue, fat, bone, epithelial clefts, and gland like epithelial aggregates. Depending upon the predominant component, hamartomas can be divided into various subtypes: chondromatous, leiomyomatous, lymphangiomyomatous, adenofibromatous, and fibro-leiomyomatous. Chondromatous hamartomas are the most common subtype. Hamartoma is also referred to as fibrolipochondroma.

For hamartomas, the age of presentation is 50 to 60 years, with a male/female ratio of 3-5/1. In our case, the patient was a 38-year-old female. The size of hamartomas may range from 1 cm to larger than 10 cm, but they are usually smaller than 4 cm.

Intraparenchymal hamartomas are usually asymptomatic. However, they may cause complaints like chest pain, dyspnea, cough, and hemoptysis. When the tumor becomes large enough, it may obstruct a bronchus causing atelectasis and recurrent or persistent pulmonary infection. It may sometimes be mistaken for chronic obstructive pulmonary disease, pneumonia, and tuberculosis. Our patient had complaints of cough and dyspnea for more than four years and received medication with a diagnosis of chronic bronchial asthma.

Diagnostic imaging methods include chest roentgenogram, computed tomography, magnetic resonance imaging, and bronchoscopy. In general, hamartomas are discovered incidentally on routine chest roentgenograms as an asymptomatic coin lesion. On chest roentgenograms, intraparenchymal hamartoma appears as a sharply outlined, round or oval mass, with lobulation, calcification, peripheral location, and a size smaller than 3 cm. Computed tomography helps make a differential diagnosis. The major radiographic difficulty with pulmonary hamartomas is to distinguish them from inflammatory and metastatic lesions. Hamartomas must be differentiated from small primary bronchogenic carcinoma and solitary pulmonary nodules. Specific popcorn-type calcification is almost pathognomonic for pulmonary hamartoma. In our patient, computed tomography of the thorax showed a 3x2-cm mass surrounding the left main bronchus concentrically at the paramediastinal site of the lower lobe of the left lung. The mass, descending aorta, and left inferior pulmonary vein were separated by fat plans, suggesting that the mass was not attached to the major vessels. With these computed tomography findings, the initial diagnosis of the mass included pri-

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Fig. 1. Computed tomography of the thorax showing a mass in the lower lobe of the left lung.

Fig. 2. (a) Hamartoma and in situ squamous cell cancer (H-E x 2). (b) Severe atypical squamous metaplasia and squamous cell carcinoma in situ areas in the bronchial epithelium (arrow) (H-E x 100).
mary lung cancers, bronchial carcinoid tumors, vascular
tumors, or benign neoplasms of the lung.

Parenchymal hamartomas are not visible on bron-
choscopy. However, indirect tumor findings can be
visualized by bronchoscopy. In our patient, computed
tomography, bronchoscopy, and transbronchial fine-
needle aspiration biopsy were performed preoperatively.
Yet, the precise diagnosis was made after histopatho-
logical examination.

Surgical treatment is the gold standard in intraparen-
chymal hamartomas including enucleation, lobectomy
or sleeve resection, wedge resection, segmentectomy,
and pneumonectomy. Endobronchial hamartomas can
be removed successfully through bronchoscopy.[4]

We could easily reach the mass through a left pos-
terolateral thoracotomy in our patient. The mass was
firm, fixed, and immobile. Upon detection of malignan-
cy in frozen section analysis, left lower lobectomy and
mediastinal lymph node dissection were performed.

Although hamartomas are benign tumors, they may
rarely assume malignant characteristics and may be
invasive to surrounding tissues.[5] Besides, some other
lung pathologies may rarely accompany pulmonary
hamartomas such as bronchiectasis, primary lung can-
cer, tuberculosis, and metastatic tumors of the lung. It
has been recommended that patients with hamartoma
should be thoroughly evaluated and closely followed-
up with respect to the risk for associated synchronous
malignancies.[6,7]

In addition to hamartomatous lesions in the fibro-
muscular stroma, squamous cell carcinoma in situ
was determined in the epithelial layer of the bronchial
structures. The tumor was composed of cartilage, fibro-
myxoid stroma, and adipose tissue with incorporated
bronchiolar epithelium. Areas of severe atypical squa-
mous metaplasia and squamous cell carcinoma in situ
were seen in the bronchial epithelium which was sur-
rounded by the basal membrane (Fig. 2b). Hemorrhage
and atelectasis sites were noted in the lung parenchyma
around the lesion. No pathology apart from intensive
anthracsosis was observed in lymph node specimens.

Based on these findings, the stage of the tumor was
rated as $T_{is}N_{0}M_{0}$.

Tojo et al.[8] reviewed some 50 reports of lung cancer
accompanying chondromatous hamartoma and sum-
marized some common features as follows: men past
middle age, adenocarcinoma, and lung cancer and
chondromatous hamartoma present in the same lobe.
In our case, the tumor accompanying hamartoma was
squamous cell cancer and it was in situ stage.

In conclusion, hamartomas can be seen together with
lung malignancies. However, the question whether the
accompanying malignancy is a coincidental occurrence
or is associated with malignant growth in the existing
hamartoma layer remains uncertain. It should be kept
in mind that hamartomas greater than 4.5 cm, firm,
and fixed on manual examination during operation
should raise suspicion for malignancy. In such cases,
perioperative histopathological examination is required
to determine the existence of malignancy and the extent
of surgical procedure.

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