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***Case Report** / Olgu Sunumu*

Spontaneous massive hemothorax as an atypical presentation of bronchial carcinoid

Bronşiyal karsinoidin atipik prezantasyonu olan spontan masif hemotoraks

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ABSTRACT

Bronchial carcinoid tumors can present with symptoms of distal airway obstruction, local airway irritation, or hemoptysis. Presentation with a spontaneous massive hemothorax has never been encountered before. In this article, we present a case of spontaneous massive hemothorax caused by the rupture of an atypical carcinoid tumor in the right upper lobe. The tumor appeared on the chest radiograph with a massive hemothorax surrounding the atelectatic lung. Spontaneous hemothorax associated with lung cancer is an exceedingly rare condition.

Keywords: Carcinoid tumor, hemothorax, lung, spontaneous.

ÖZ

Bronşiyal karsinoid tümörler distal hava yolu tıkanıklığı, lokal hava yolu irritasyonu veya hemoptizi semptomları ile ortaya çıkabilir. Daha önce spontan masif hemotoraks ile prezantasyon görülmemiştir. Bu makalede sağ üst lobda bulunan karsinoid tümörün rüptürüne bağlı spontan massif hemotoraks olgusu sunuldu. Tümör, göğüs grafisinde massif hemotoraks ve atelektazi ile ortaya çıktı. Akciğer kanseri ile ilişkili spontan hemotoraks son derece nadir görülen bir durumdur.

Anahtar sözcükler: Karsinoid tümör, hemotoraks, akciğer, spontan.

Spontaneous hemothorax is defined as a subcategory of hemothorax with blood accumulation in the pleural space but without thoracic trauma and other causes. Its incidence is 5% among all hemothorax cases, and it is a life-threatening condition.[1] There are many factors in its etiology; the most common reason is the rupture of a bulla localized in the apical area of the lung and the separation of the accompanying pleural adhesion.[2] Neoplasms are a rare cause of spontaneous hemothorax that should be considered in the differential diagnosis.[1] In the literature, cases of pulmonary, mediastinal, and intra-abdominal tumors, including angiosarcoma, hepatocellular carcinoma, germ cell tumor, pulmonary choriocarcinoma, and lung cancer, presenting with hemothorax have been reported.[1,2]

To our knowledge, this is the first case report of a typical carcinoid tumor-associated spontaneous massive hemothorax in the literature. Herein, we discuss the diagnosis of this rare condition and present a review of the literature.

CASE REPORT

A 61-year-old male patient applied to the emergency service of another hospital with a complaint of cough. Thorax computed tomography (CT) scan revealed a 60×55 mm lesion in the right upper lobe and

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ground-glass opacity around the mass (Figure 1). The patient, who had a pharyngeal swab taken with a prediagnosis of coronavirus disease 2019 (COVID-19) pneumonia and was sent home with the recommendation of isolation, came back to our clinic with complaints of increased chest pain and shortness of breath after three days. The patient's chest X-ray showed homogeneous opacity compatible with massive pleural effusion in the right hemothorax (Figure 2). The pleural fluid sampled by diagnostic thoracentesis was found to be hemothorax, and right tube thoracostomy was performed. Since there was approximately 4000 mL of hemorrhagic drainage from the chest tube in the first 24 h and the patient's blood hemoglobin value decreased from 12.6 g/dL to 9.8 g/dL, two units of erythrocyte suspension were administered to the patient, and he was transferred to the intensive care unit.

The patient, who did not have any additional diseases and did not use drugs, had no history of smoking. In a thorax CT scan taken in 2010, a 25×20 mm oval and well-circumscribed hypodense nodule localized in the right upper lobe was observed, and it was radiologically followed for two years (Figure 3). In this period, no progression was observed in the lesion, and no diagnostic intervention was performed. The lesion, considered to be a hamartoma, was not followed radiologically in the following years.

At the physical examination, the patient had a blood pressure of 120/80 mmHg, a rhythmic pulse of 108/min, a body temperature of 36.1°C, a respiratory rate of 20/min, mild dyspnea, and the breathing sounds could not be obtained from the right hemithorax in the auscultation. Heart sounds were normal, rhythmic, and tachycardic.

Laboratory parameters were as follows: hemoglobin, 9.8 g/dL, hematocrit, 29.8%; red blood cell count, 3.32 10^6/uL; white blood cell count, 12,600/mm^3; platelet count, 334,000/mm^3; C-reactive protein, 30.21 mg/dL; glucose, 130 mg/dL; blood urea nitrogen, 18 mg/dL; creatinine, 0.96 mg/dL; sodium, 138 mEq/L; potassium, 4.4 mEq/dL; calcium, 7.8 mg/dL; aspartate aminotransferase, 17 U/L; alanine aminotransferase, 22 U/L; lactate dehydrogenase, 330 U/L; ferritin, 179.9 ng/mL; troponin I, 0.01 ng/mL; D-dimer, 1,848 ng/mL; prothrombin time, 13.7 s; international normalized ratio, 1.13. Blood gas test values were as follows: pH, 7.52; pCO_2, 31.5 mmHg; pO_2, 57.8 mmHg; saturation, 92.9%. Pleural fluid values were as follows: hemoglobin, 4.3 g/dL; hematocrit, 14.2%; lactate dehydrogenase, 230 U/L; glucose, 55 mg/dL. Meanwhile, the COVID-19 PCR test was reported as negative.

In the positron emission tomography (PET/CT) taken after tube thoracostomy, a 60×55 mm hypodense lesion in the right upper lobe posterior segment with minimal fluorine-18-fluorodeoxy-D-glucose (FDG) involvement (maximum standardized uptake value [SUV_{max}]: 2.9) was observed at the periphery, while

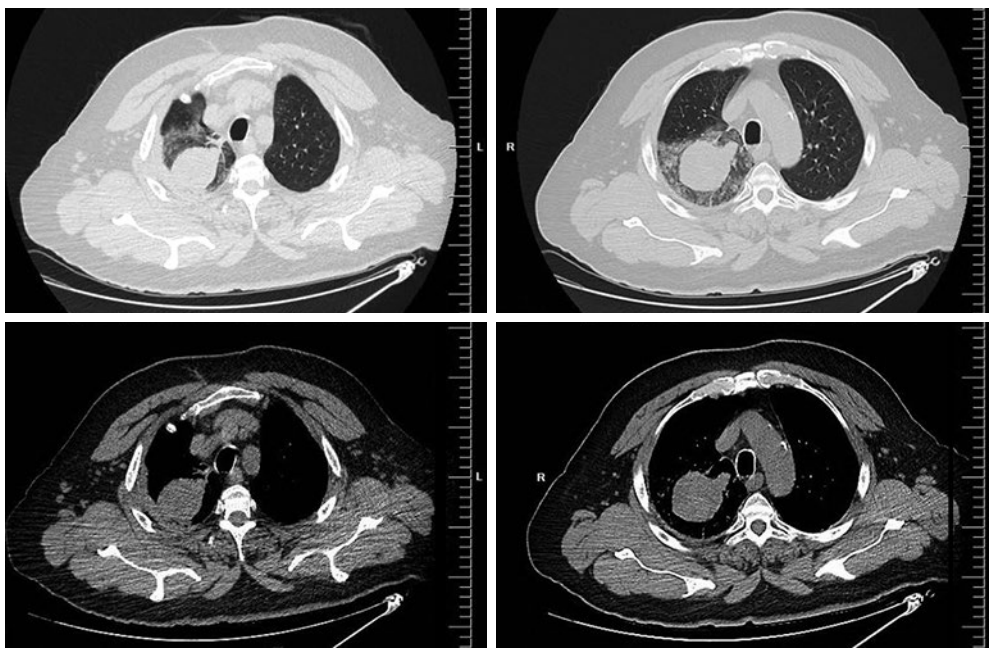

Figure 1. A 60×55 mm mass lesion is observed in the right upper lobe posterior segment in the thorax computed tomography axial sections, while ground-glass opacity is observed around the mass.

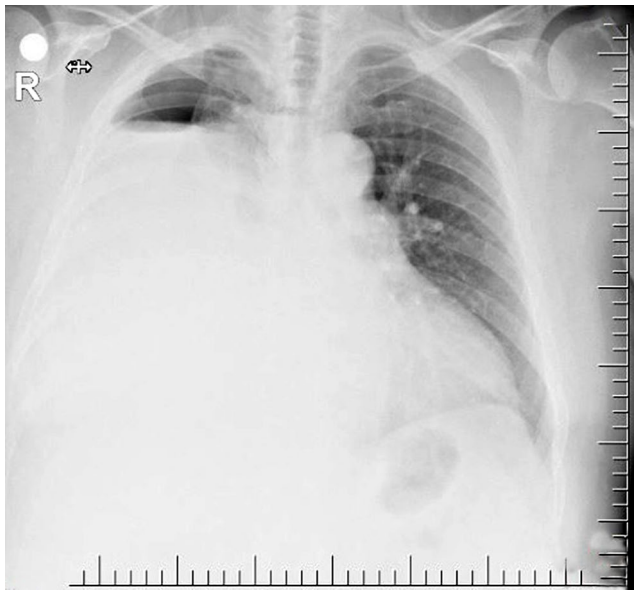

Figure 2. A homogeneous opacity consistent with massive pleural effusion that almost completely fills the right hemithorax is observed in the chest radiograph.

pleural fluid with 63 mm thickness (SUV_{max}: 2.4) was detected in the right hemithorax (Figure 4). Metastasis was not observed in the brain magnetic resonance imaging.

The clinically stabilized patient underwent fiberoptic bronchoscopy and right posterolateral thoracotomy under general anesthesia the next day. The endobronchial lesion was not observed in fiberoptic bronchoscopy. The thoracotomy revealed a defibrinated hematoma in the thorax and a 60×50 mm cavitary lesion with a hematoma in the right upper lobe. After cleaning inside the cavity, an incisional biopsy was taken from the lung parenchyma that seemed to be suspicious, and a frozen section analysis was carried out. Upon pathology report stating malignancy, right upper lobectomy and mediastinal lymph node dissection were performed. The patient was discharged on the sixth day with an uneventful postoperative follow-up. Histopathological examination of the lobectomy material revealed a

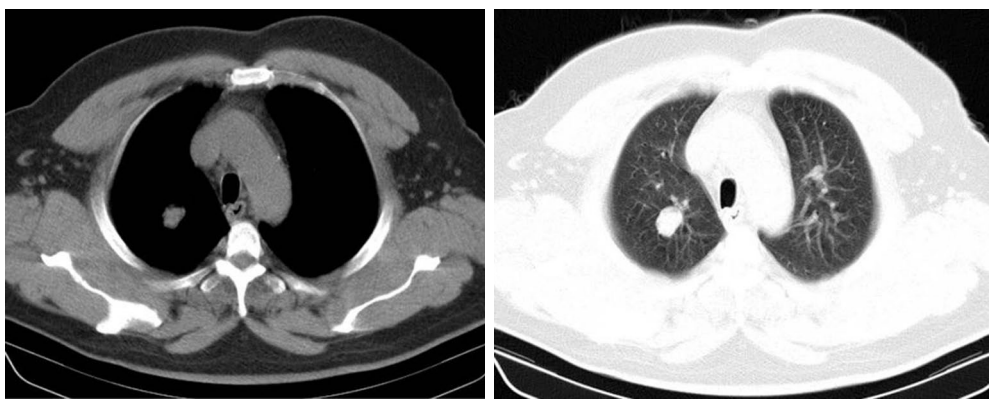

Figure 3. A 25×20 mm hypodense nodule with an oval shape and smooth margins is observed to be localized in the right upper lobe in the axial sections of thorax computed tomography scan performed in 2010.

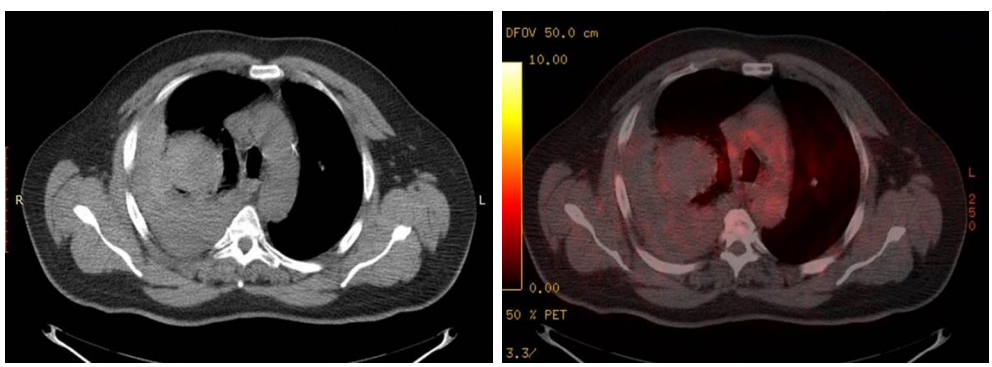

Figure 4. In PET/CT, a 60×55 mm hypodense lesion in the right upper lobe posterior segment with minimal FDG involvement (SUV_{max}: 2.9) is observed at the periphery, while pleural fluid with 63 mm thickness (SUV_{max}: 2.4) is observed in the right hemithorax.

PET: Positron emission tomography; CT: Computed tomography; FDG: Fluorine-18-fluorodeoxy-D-glucose; SUV_{max}: Maximum standardized uptake value.

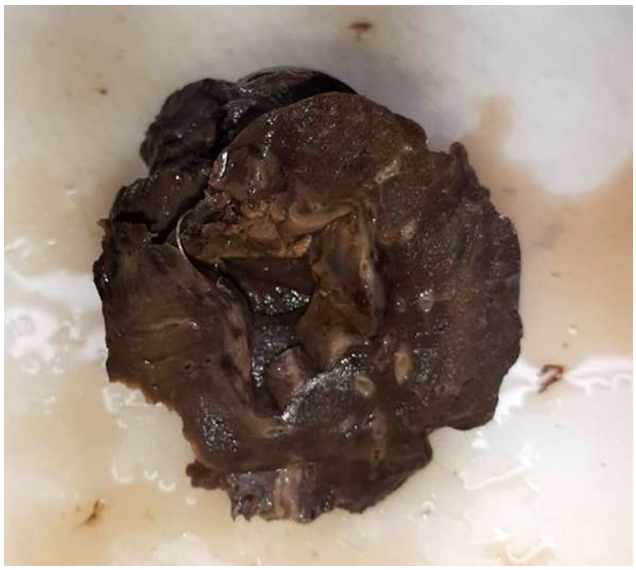

Figure 5. A cavitary lesion is observed in the macroscopic view of the lobectomy material of the lung parenchyma.

typical carcinoid tumor (T2bN0M0, Stage IIA), and no metastatic lymph node was observed (Figure 5).

DISCUSSION

Spontaneous hemothorax is a type of hemothorax encountered in cases where there is no trauma or other causes, and it is rarely observed in lung cancer. The cause of hemothorax in lung cancer is bleeding into the pleural cavity as a result of the visceral pleural invasion and vascular invasion within the lung parenchyma due to the tumor.[1,2] Although there were cases of pneumothorax associated with bronchial carcinoid tumors and only one case of hemothorax in the literature, we did not find a case of massive hemothorax.[3-5]

Bronchial carcinoids are tumors from a wide spectrum of neuroendocrine tumors, including typical and atypical carcinoid tumors, large cell neuroendocrine carcinoma, and small cell lung cancer. It accounts for 1 to 2% of all lung cancers, and 85% are localized in the central region (main bronchus or lobe bronchi). It depends on the size of the clinical tumor and its location in the peripheral or central area. Peripherally located tumors are usually asymptomatic, and they are detected incidentally. The tumors located in the central region usually present with symptoms related to airway irritation, such as cough and hemoptysis, and obstruction, as in recurrent pulmonary infections.[3]

In radiological evaluation, particularly in thorax CT scans, centrally located carcinoid tumors are observed to be well-defined margins and hypervascular tumors, while the peripherally located ones are observed as solitary pulmonary nodules.[3] Therefore, peripherally localized tumors, as in our case, have the potential to cause a spontaneous hemothorax, except for the symptom associated with bronchial obstruction. Carcinoid tumors are hypervascular tumors, and they show variable SUV_{max} values depending on the proliferation and mitosis of the tumor in PET/CT. In our case, the peripherally located mass caused visceral pleural invasion and ruptured into the pleural cavity, leading to the development of hemothorax. The fact that the SUV_{max} value of the mass is measured as 2.9 in PET/CT indicates that the tumor is highly necrotic, and the number of viable cells in the mass is low.

Tumor-related hemothorax mechanism is defined as the invasion and ischemic necrosis of the visceral pleura and lung parenchyma by the tumor tissue, causing the rupture of the parenchymal vessels into the pleural cavity.[1,5] In our case, the solitary pulmonary nodule localized in the right upper lobe, which was radiologically detected approximately 10 years ago, became a mass over the years, and massive hemothorax developed as a result of the visceral pleural invasion and bleeding into the pleural cavity.

The prognosis is poor in cases of cancer-related hemothorax.[1] However, in bronchial carcinoid tumors, the prognosis will be excellent with the surgical treatment performed after the patient's clinic is stabilized. Ordinarily, lobectomy is preferred as the treatment method for bronchial carcinoid tumors, and the five-year survival rate for localized diseases without lymph node metastasis is 90%.[3]

In conclusion, spontaneous hemothorax associated with lung cancer is an exceedingly rare condition, with no previous cases caused by the rupture of atypical bronchial carcinoid tumors in the literature. It should be kept in mind that the underlying disease in patients, although is rare, might be typical carcinoid with a mass in the lung and a developing spontaneous massive hemothorax clinic.

Patient Consent for Publication: A written informed consent was obtained from the patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Conception, design, data collection and analysis, literature review, writer - B.Ç.; Conception, design, data collection and analysis, critical review - H.U.Ç.; Design, data collection and analysis, literature review, critical review - H.K.Ç.

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