Olgu Sunumu (Case Report)

A Rarely Seen Lung Tumor: Inflammatory Myofibroblastic Tumor

Nadir Görülen Bir Akciğer Tümörü: İnflamatuvar Myofibroblastik Tümör

Fatoş KOZANLI[1], Abdulkadir Yasir BAHAR[2]

[1] Kahramanmaras Sutcu Imam University, Medical Faculty Thoracic Surgery Departmant, Kahramanmaras, Turkey
[2] Kahramanmaras Sutcu Imam University, Medical Faculty Patology Departmant, Kahramanmaras, Turkey

Özet

İnflamatuar myofibroblastik tümör (IMT), nadir görülen bir yumuşak doku tümörüdür. Çoğunlukla benign karakterli olmakla birlikte biyolojik davranışı tartışmalıdır.

Öksürük şikayeti ile polikliniğimize başvuran kadın hastayı radyolojik ve histopatolojik olarak değerlendirdik. Sağ akciğer alt lobda tespit ettiğimiz kitle lezyonun histopatolojik tanısı inflamatuar myofibroblastik tümör olarak raporlandı. Radyolojik olarak malign tümörlerden ayırt edilemeyen bu nadir olguyu literatür bilgileri eşliğinde sunmayı amaçladık.

İnflamatuar myofibroblastik tümörlerin radyolojik bulgularla malign tümörlerden ayırt edilmesi güçtür. Total rezeksiyonla hem tanı hem de tedavisi mümkündür.

Anahtar Kelimeler: Akciğer, İnflamatuar myofibroblastik tümör, Kitle lezyon, Wedge rezeksiyon

Abstract

Inflammatory myofibroblastic tumor (IMT) is a rarely seen soft tissue tumor. Although it is mostly benign, its biological behaviour is controversial.

We radiologically and histopathologically evaluated a female patient admitted to our outpatient clinic due to complaint of cough. Histopathologic diagnosis of mass lesion identified at lower lobe of right lung was reported as inflammatory myofibroblastic tumor. We aimed to present this rare case which is not radiologically differentiable from malignant tumors along with literature data. Differentiating Inflammatory myofibroblastic tumor from malignant tumor with radiologic findings is difficult. Both treatment and the diagnosis are possible with total resection.

Key Words: Inflammatory myofibroblastic tumor, Lung, Mass lesion, Wedge resection

Yazışma Adresi: Fatoş KOZANLI, Kahramanmaraş Sütçü İmam Üniversitesi Tıp Fakültesi, Göğüs Cerrahisi Anabilim Dalı Kahramanmaraş, Türkiye

Telefon: +90 505 2231984, **Mail:**opdrfatoskozanli@gmail.com

ORCID No (Sırasıyla): 0000-0001-7664-2657, 0000-0002-6963-3389.

Geliş Tarihi: 29.01.2021

Kabul Tarihi: 18.04.2021

DOI: 10.17517/ksutfd.869381

INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) which is also known as inflammatory pseudotumor is a benign tumor consisted of fibrous tissue, myofibroblasts and inflammatory cell proliferation with indefinite etiology. Making diagnosis of IMT before surgery is difficult due to various radiologic findings. Risk of distant metastasis is low although it has tendency towards local recurrence. Intra-thorax IMTs are typically present as small peripheral tumors and account for 0.7-1% of all lung tumors. Inflammatory myofibroblastic tumor (IMT) behavior is an indistinct, rare, borderline neoplasm (1).

In this paper, we aimed to present the diagnosis and treatment of rarely seen IMT which is frequently confused with malignancy along with literature data.

CASE

A 22-year old female patient admitted to our outpatient clinic with the complaint of cough. She had no history of smoking and there was no significant findings in her physical examination. Computerized tomography imaging was performed after identifying mass lesion at middle zone of right lung on chest roentgenogram **(Figure 1).**

It was observed that there was an irregular parenchymal nodular lesion sized 20x13 mm at the near of fissure in superior of lower lobe of right lung. Fluorodeoxyglucose (FDG) involvement (Suvmax: 5,4) of the lesion sized approximately 2 cm at the superior segment of lower lobe of right lung was consistent with malignancy. Patient was planned for thoracoscopic biopsy. Lesion was excised with wedge resection under uniportal videothorascopic technique. Result of material sent to frozen section was benign. Operation was finished after this result. A gray-white solid lesion sized 2x2x2 cm which was adjacent to parietal pleura and was well-demarcated in lung parenchyma, was identified in macroscopic examination of the material. Fusiform cell proliferation which was rich in vasculature and was mostly in fascicular pattern and partially in storiform pattern accompanied by lymphoid follicles and infiltrates with abundant plasma cells, was observed in microscopic evaluation of paraffine sections of lesion. Fusiform cells had pale eosinophilic cytoplasm, irregular cell demarcation, and vesicular nuclei without atypia **(Figure 2)**. Fusiform cells were immunohistochemically evaluated and resulted as follows: pan-cytokeratin: negative, actin: negative, p53: weak positive, desmin: focal positive, cyclin D1: positive, TTF-1: negative, CD68: focal positive, ALK: positive, CD117: weak isolated positive, CD34: negative, Ki-67 pro-

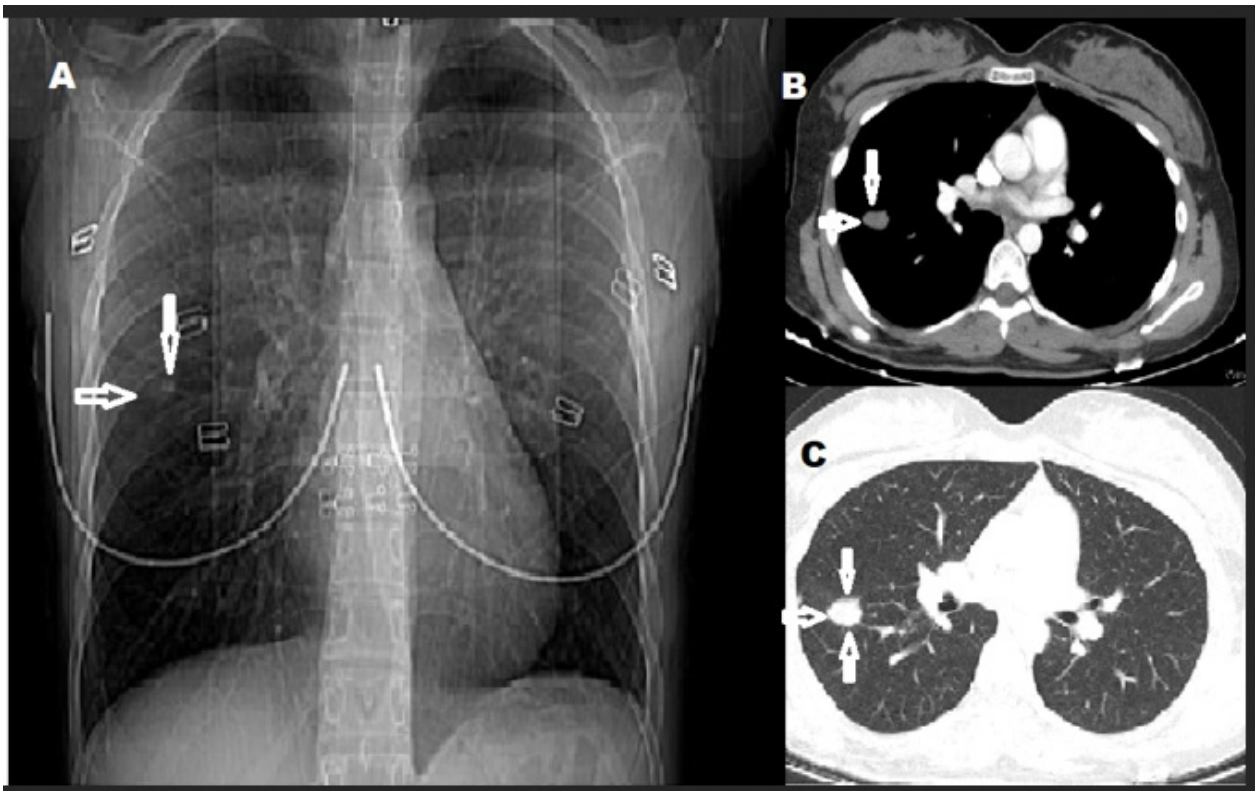

Figure 1. Chest roentgenogram and parenchymal-mediastinum sections of thoracic computerized tomography imaging of case
A. Opaque lesion in the middle zone of the right lung on chest radiography(area marked by arrows). B. An opaque lesion with spicular extension in the lower lobe of the right lung on the mediastinal window in the thoracic tomography (area marked by arrows). C. Opaque lesion with spicular extension in the lower lobe of the right lung in the parenchymal window on computed tomography of the thorax (area marked by arrows).

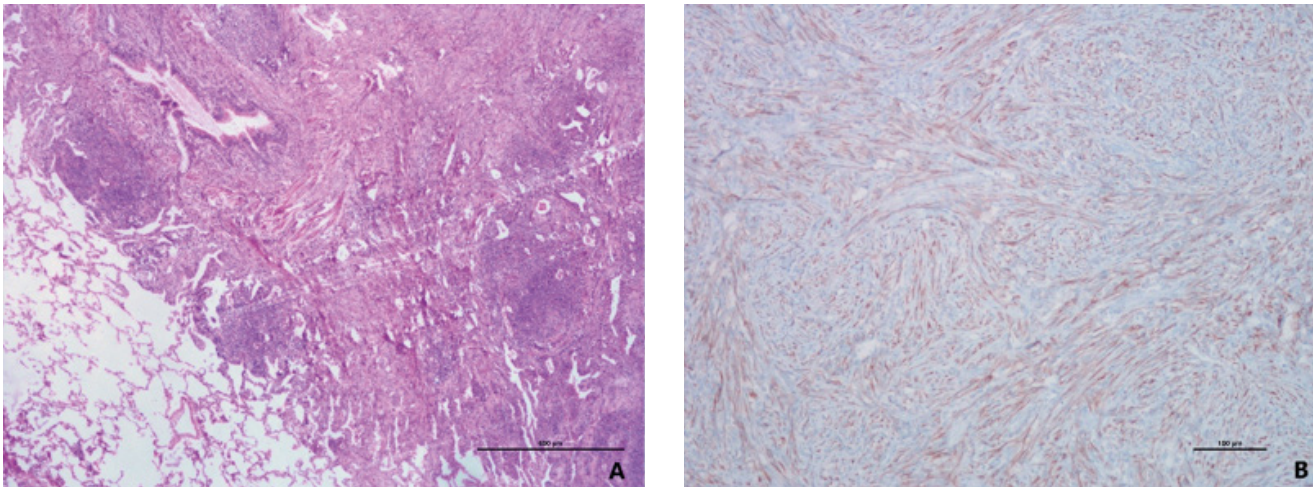

Figure 2. Histopatological views of the lesion
A: Fusiform cell proliferation with chronic inflammatory infiltrate which is rich in lymphoid follicles and plasma cells is seen in adjacent to pleura and normal lung peripheral parenchyma (lower left) (hematoxylin-eosin, 40x magnification).
B: Fusiform cells are immunohistochemically positive for ALK (immunohistochemistry, 100x magnification).

liferation index:1%. The case was diagnosed with IMT. No recurrence or distal organ metastasis were observed after 1 year of follow-up.

DISCUSSION

IMT is a rarely seen lung disease. Incidence rate of IMT among patients undergone lung resection is 0.04% and 26% of patients are under 18 years of age (2). Both genders are affected equally and there has been no reported tendency for geography or ethnicity (3).

About half of the patients are asymptomatic and 26-56% of the patients have complaints of cough, hemoptysis, dyspnea and chest pain. Our case had no symptom except cough. Sometimes weight loss, fever and obstructive airway syndrome may also be seen in some cases (1). In our case, there was no weight loss and obstructive airway symptoms.

Mass lesion is incidentally identified in chest roentgenogram (4). Its slow growth and local invasion pattern have been known (3). In our case, a mass was detected incidentally in the chest x-ray taken due to cough, similar to the literature.

Only 6.3% of IMT cases are diagnosed when based only on analysis of biopsy samples. In addition, distinguishing IMT from other neoplasms in PET scan is generally difficult due to high FDG intake of IMT (5). FDG involvement was 5.4 in the PET scan taken in our case. Pathological FDG involvement was differentiated from malignant tumor. Preferred treatment is surgical resection. Wedge resection with video support or open technique are appropriate for curative objectives. When wedge resection is technically not possible, only other option is major resection (3). In our case, wedge resection performed with uniportal video thoracoscopic technique was sufficient for both diagnosis and treatment. Size of lesion was 2x2 cm and size of resected area was 4x4.5 cm in diameter with clear surgical margin. Prognosis of IMT is dependent on size of tumor (3 cm or smaller) and complete surgical resection. Total 3-year survival rate is approximately 82% and total 5-year survival rate is approximately 74%. Recurrence after complete resection is rare. Outcomes in patient's undergone radical resection are generally good (3).

Tumor has been named with different names as plasma cell granuloma or tumor, xanthogranuloma, plasma cell/histiocytoma complex or post-inflammatory pseudotumor according to dominant cell type in the lesion (2). Matsubara et al. grouped IMT in three groups according to cellular types and main histological features: A) organized pneumonia occurred due to gradually recovered intraalveolar exudation (44%), B) fibrous histiocytoma (44%), C) lymphoplasmacytic type occurred due to aggregation of both plasmocytes and lymphocytes (12%) (4). In accordance with the literature, our case was lymphoplasmacytic IMT which is rich in both plasmocytes and lymphocytes. Diameter of tumor was smaller than 3 cm and it was completely resected. We think that small diameter of tumor and its complete resection may provide beneficial effect on prognosis of patient.

CONCLUSION

Although IMT is rarely seen, it must be considered in differential diagnosis of space-occupying lesions. Its radiological differentiation from malignant tumors is difficult. Total resection for diagnosis and treatment may be beneficial due to its involvement in PET/CT which is mostly consistent with malignancy and its low chance of diagnosis on biopsy materials. It is generally a benign lesion, however, it must be kept in mind that it also has a potential for local invasion and recurrence. Diagnosis and prognosis are dependent on complete resection.

Conflict of Interest and Financial Situation: Our study was not funded by an institution or organization. There is no conflict of interest between the authors in this study.

Author Contribution rate: The authors declare that they have contributed equally to the study.

Informed Consent: Written informed consent was ob-tained from the patient who participated in this case.

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