



Case report

A rare benign tumor of the lung: Inflammatory myofibroblastic tumor – Case report

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ABSTRACT

A fifty year old lady who was operated for thyroid cancer two years ago and completed adjuvant therapy, underwent a computer tomography (CT) of the chest during her follow up. The CT showed a mass lesion in the right lung, located to the lateral segment of the middle lobe. There were no intrabronchial lesions on bronchoscopy. Positron emission CT (PET CT) showed a dense hypermetabolic mass located in the right middle lobe lateral segment and having malignant characteristics. A videothoroscopic wedge resection was performed and the specimen was sent for frozen section, which showed no evidence of malignancy. Pathology report revealed an inflammatory myofibroblastic tumor (IMT). Since IMT is a rare benign tumor of the lung, we herein report this patient along with a discussion of the relevant literature.

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1. Introduction

Inflammatory myofibroblastic tumor (IMT) of the lung, also known as plasma cell granuloma or inflammatory pseudotumor, is one of the rare benign tumors of the lung.¹ It often occurs as a result of excessive inflammatory response. A diagnosis before surgery is difficult because of the variations in radiologic appearance. Complete resection is essential both for excluding malignancy and for curative treatment.^{2,3} A patient who was diagnosed with IMT after videothoroscopic wedge resection is presented.

2. Case report

A fifty year old female patient was operated two years ago for removal of thyroid cancer, and later completed adjuvant treatment. A routine chest CT during her follow up showed a mass in the right middle lobe lateral segment, and the patient was referred to our department. (Fig. 1) She was asymptomatic, and all laboratory studies were normal. There were no pathologic findings on cranial MRI. Respiratory function tests were within normal limits. Her medical history revealed that she had undergone a thyroidectomy

and cesarean section. She was using Levotron tablets once a day since her thyroidectomy, and smoking 5–6 cigarettes a day since 15 years. Her family history was unremarkable. There were no intrabronchial lesions on the diagnostic bronchoscopy performed for the investigation of endobronchial lesions. Cytologic examination of the middle lobe lavage showed abundant bronchial and sparse epithelial cells, alveolar macrophages, rare polymorphic neutrophils and lymphocytes, and mucoid material in the background. The cytologic diagnosis was reported as benign cytologic findings.

On PET CT there was a dense hypermetabolic mass lesion in the right middle lobe lateral segment, suggesting a malignancy (SUV max: 3.9). Since the patient was a smoker, this lesion was primarily considered as a second primary or the lung metastasis of another malignancy. (Fig. 2) The mass was also considered to be inappropriate for transthoracic fine needle aspiration (TTFA) for technical reasons. A wedge resection of the mass was carried out due to the risk of pneumothorax. An intraoperative frozen section was made, and did not reveal any malignancy. Therefore no further treatment was applied. Macroscopic examination reported a nodular lesion in the lung parenchyma, with regular borders and measuring 1.7 cm. The mass appeared elastic, yellow-gray colored, and homogenous on sectioning. Three millimeter thick sections were harvested from the lesion and embedded into paraffin blocks after routine tissue procedures.

The material was examined under light microscopy. Sections showed a storiform or fascicular pattern tumor. Tumor elements consisted of cells with eosinophilic cytoplasm, oval and spindle

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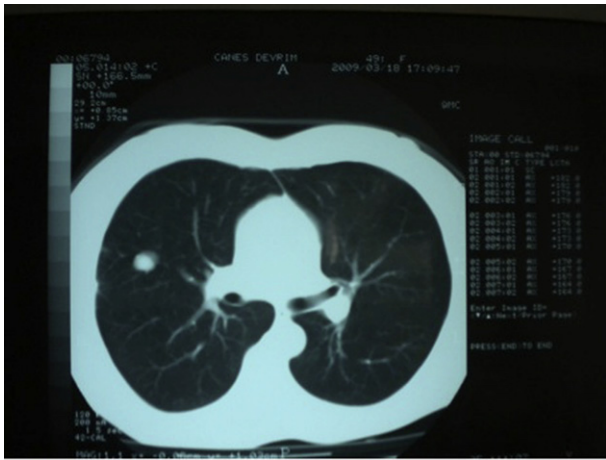


Fig. 1. Computer tomography image shows a mass measuring 1.9 cm and located in the lateral segment of the right middle lobe.

shaped nucleus, thin chromatin distribution, and without distinct nucleoli. There was no cytologic atypia or mitosis. Lymphocytes, plasma cells, and histiocytes were occasionally present. Immunohistochemical studies showed focal positivity for SMA (smooth muscle actin), and there was no staining with antibodies against CD34, CD99, S100, and pancytokeratin. The Ki-67 proliferation index (1%) was low (Figs. 3–9). The pathologic diagnosis was inflammatory myofibroblastic tumor. The patient did not receive any postoperative treatment, and is currently asymptomatic on the postoperative third year.

3. Discussion

IMT is a rare lung disease. The lesion consists of a variable mixture of inflammatory and mesenchymal cells that includes inflammatory pseudotumor plasma cells, histiocytes, lymphocytes, and fibroblasts. It can occur in numerous locations throughout the body, however the most common location is the lung.⁴ Most of the reported cases are between 27 and 50 years of age, and under 40.⁵ Both sexes are affected equally, and no geographic or ethnic predisposition has been reported.⁶ Our patient was a 50 year old female living in the mediterranean area.

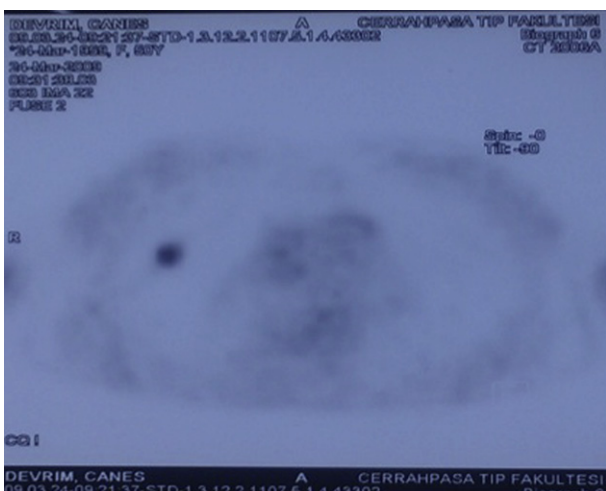


Fig. 2. PET CT shows a mass in the lateral segment of the right middle lobe, showing a malignant character with SUV max: 3.9.

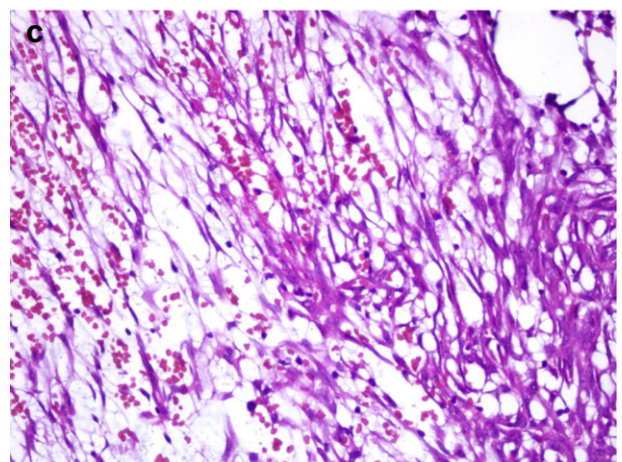
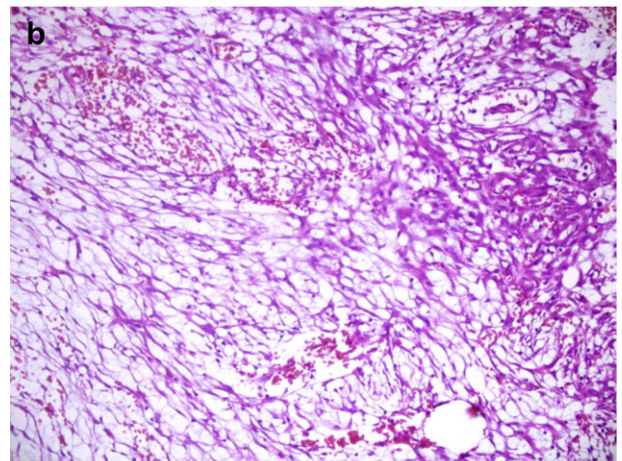
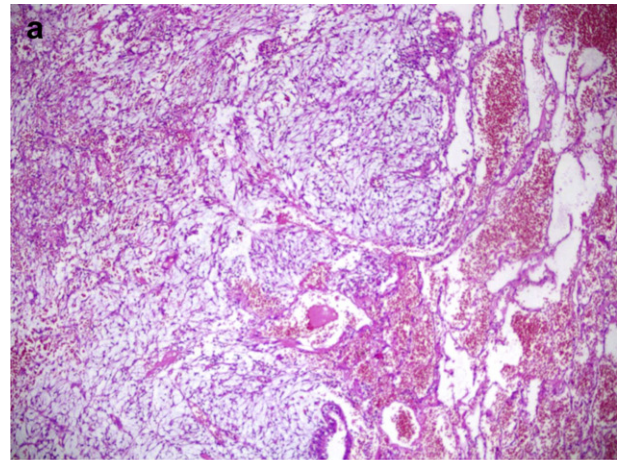


Fig. 3. (a): Storiform and fascicular pattern tumor separated from the lung tissue by smooth borders (H&Ex100). (b): Tumor cells with eosinophilic cytoplasm, oval and spindle shaped nuclei, thin chromatin distribution, and accompanied by lymphocytes, plasma cells and histiocytes in between (H&Ex200). (c): Tumor elements without cytologic atypia or mitotic figures, and inflammatory cells. (H&Ex400).

The frequency of this lesion in the lung is 0.04–1.0% among the general population.^{7,8} Approximately half of the patients are asymptomatic, whereas 26–56% of the patients have symptoms including cough, hemoptysis, dyspnea and chest pain. The mass lesion is discovered incidentally in chest x rays.⁸ Our patient was asymptomatic and the mass was discovered during tumor surveillance.

The radiologic findings of IMT were analyzed by Agrons et al.⁵ Thoracic CT reveals a single nodule and mass in 90% of these

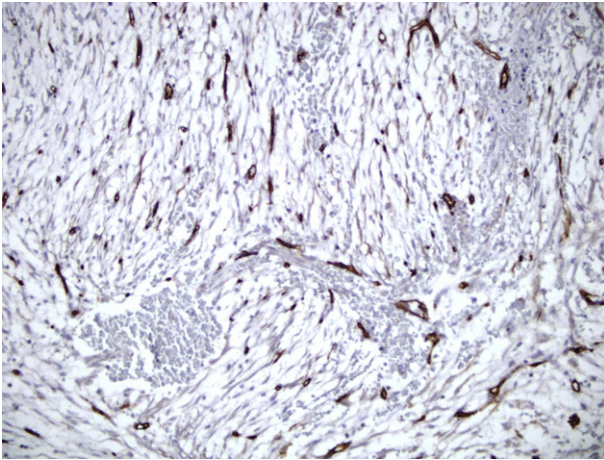


Fig. 4. cd34 staining revealed that the internal control (vascular structures) was positive, whereas the tumor did not show staining. (IHK-cd34x200).

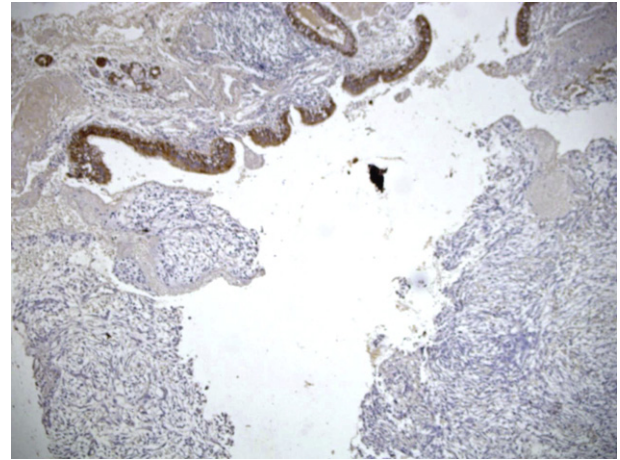


Fig. 7. Pan-ck staining showed that the internal control (bronchial epithelium) was positive, whereas the tumor did not show staining. (IHK pan-ckx100).

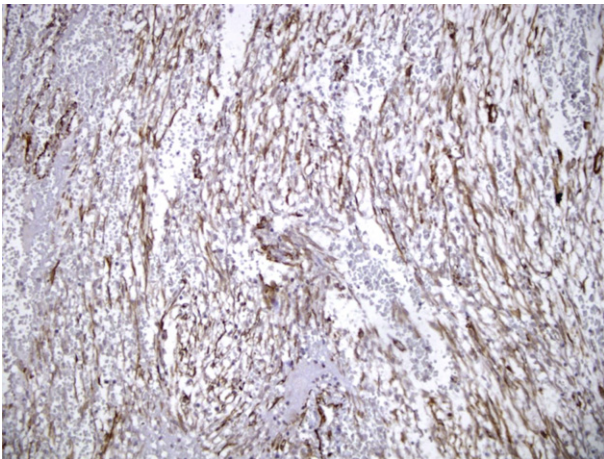


Fig. 5. Positive staining of tumor elements with sma. (IHK-smaX200).

patients, and multiple nodules in 5%.⁵ Secondary infiltration of the hilus, mediastinum and airways were reported in 16% of the patients. The lesion in our patient was also a single nodule detected in chest CT. PET CT is often positive in these cases.⁹ Our patient had FDG uptake in PET CT.

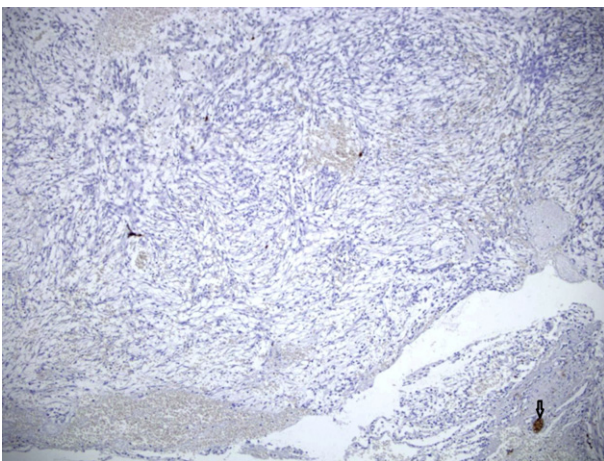


Fig. 6. s100 staining revealed that the internal control (cross section of a peripheral nerve is marked in the lower right) was positive whereas the tumor did not show staining. (IHK-s100x100).

It is difficult to make a diagnosis by bronchoscopic biopsy and transthoracic needle aspiration. Therefore, open lung biopsy or videothoracoscopic resection is often necessary.^{7–10} The diagnosis was made after thoracoscopic wedge resection in our patient.

Matsubara et al. categorized inflammatory pseudotumor into three groups based on cellular types and main histologic properties: A) Organized pneumonia formed by gradual healing of the intraalveolar exudation (44%), B) Fibrous histiocytoma formed by storiform proliferation of plasmocyte and lymphocyte aggregates (44%), and C) lymphoplasmocytic type formed by the aggregation of both plasmocyte and lymphocytes (12%).⁸ A storiform or fascicular pattern tumor was observed in our case. The cells constituting the tumor elements had an eosinophilic cytoplasm, oval, spindle shaped nuclei with thin chromatin dispersion, and with indistinct nucleoli. There was no cytologic atypia or mitosis. There were also lymphocytes, plasma cells and histiocytes.

The treatment of pulmonary IMT is surgical.^{3–11} Recurrences are rare after complete resection.^{12,13} The results are also generally well in patients who undergo radical resection.^{3–11} However, recurrences can still occur years after resection,^{2–14} and deaths related to IMT are reported.^{2–15} The recurrences are caused by incomplete resection.⁷ Since there were no endobronchial lesions in our patient, a parenchyma preserving surgery was preferred and

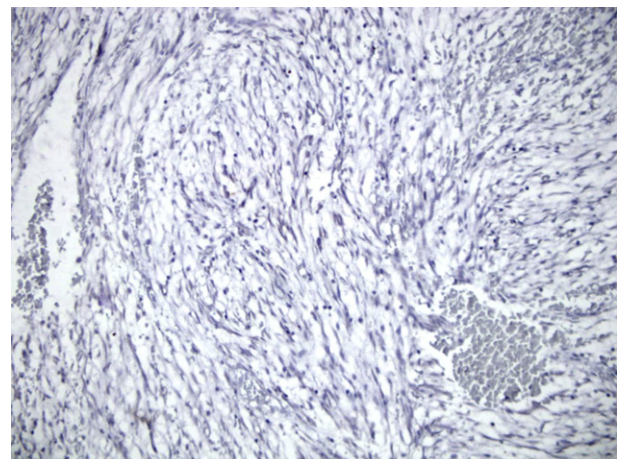


Fig. 8. The external control is positive with cd99, and the tumor is negative. (IHK-cd99x200).

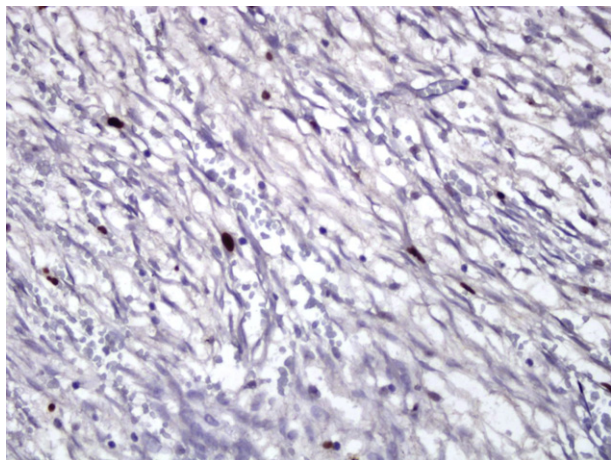


Fig. 9. Scarce nuclear positivity in tumor elements with ki-67. (IHK-ki-67 x400).

videothoroscopic wedge resection was made. There were no recurrences during the three year follow up.

4. Conclusion

Although IMT is rare, it should be considered in the differential diagnosis of lung lesions. It is generally a benign lesion, however it has potential for local invasion and recurrence. The diagnosis and prognosis depend on complete resection.

Conflict of interest

None.

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