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Respiratory Diseases

Inflammatory Pseudotumor of the Lung: About Three Cases

K. Chaanoun[1*], I. Chhen[1], H. Harraz[1], H. Benjelloun[1], N. Zaghba[1], N. Yassine[1]

[1]Respiratory Diseases Department, IBN ROCHD University Hospital, Casablanca

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***Corresponding author:** K. Chaanoun
Respiratory Diseases Department, IBN ROCHD University Hospital, Casablanca

Abstract | **Case Report**

Inflammatory pseudotumors of the lung are generally unifocal lesions of unknown pathogenesis with a benign prognosis. We report the cases of three patients followed in the respiratory diseases department at Ibn Rochd University Hospital in Casablanca. The first patient, a 68-year-old man, was hospitalized for an etiological assessment of a retracted opaque hemithorax revealed by exertional dyspnea and chest pain. The radio-clinical presentation suggested a diagnosis of malignant bronchopulmonary tumor, which was ruled out after further exploration. The second case concerns a 60-year-old woman hospitalized for mild hemoptysis. The clinical-radiological presentation initially raised suspicion of tuberculosis, which was later excluded. The third patient, a 43-year-old woman, was admitted with chest pain and dyspnea. A pulmonary lesion was discovered, again raising suspicion of tuberculosis, which was ruled out after investigation.

Keywords: Inflammatory pseudotumor, xanthoma, histiocytoma, opaque hemithorax, hemoptysis, tuberculosis, pulmonary tumor.

I- INTRODUCTION

Inflammatory pseudotumors (IPTs) were first described in the lung in 1939 under various terms including 'post-inflammatory tumors', 'plasma cell granulomas', 'xanthomas', 'xanthomatous pseudotumors', 'fibrous histiocytomas', or simply 'histiocytomas'. These are rare tumors of unclear pathogenesis and poorly defined treatment. We report three cases of inflammatory pseudotumors diagnosed in the context of different clinical and radiographic presentations.

II- CASE REPORT

A 68-year-old man was admitted for exertional dyspnea associated with burning-type left-sided chest pain. He had a 30-year history of wood smoke exposure but no other toxic habits. On admission, the patient was in good general condition, eupneic, normocardic and afebrile, with normal oxygen saturation on room air (SpO_2 at 96%) and a performance status (PS) score of 1. Clinical examination revealed signs of a left-sided pleural effusion. Chest X-ray showed an opaque and retracted left hemithorax. Laboratory tests revealed a white blood cell (WBC) count of 15.810/mm³ with neutrophil predominance, CRP at 58 mg/L, and ESR of 41 mm in the first hour. Bronchoscopy revealed a protruding, well-vascularized, and well-defined tumor completely obstructing the lumen of the left main bronchus. While awaiting the results of bronchial biopsies, the patient was started on protected amoxicillin for 8 days at a dose of 3 g per day. Histology showed respiratory mucosa with regular epithelium and a dense inflammatory infiltrate composed of lymphocytes, plasma cells, and eosinophils with fibroblasts. A CT scan showed segmental consolidations in the left lower and middle lobes with air bronchogram and mild bronchial distortion. A second bronchoscopy 12 days later revealed diffuse grade 1 inflammation without visible tumor. Follow-up biopsies confirmed a mixed inflammatory infiltrate without malignancy. Repeated tests showed normalized WBC (8.900), CRP (14), and ESR (8). Follow-up chest X-ray was near normal. A diagnosis of inflammatory pseudotumor was made based on clinical, biological, and radiological improvement, along with histological findings.

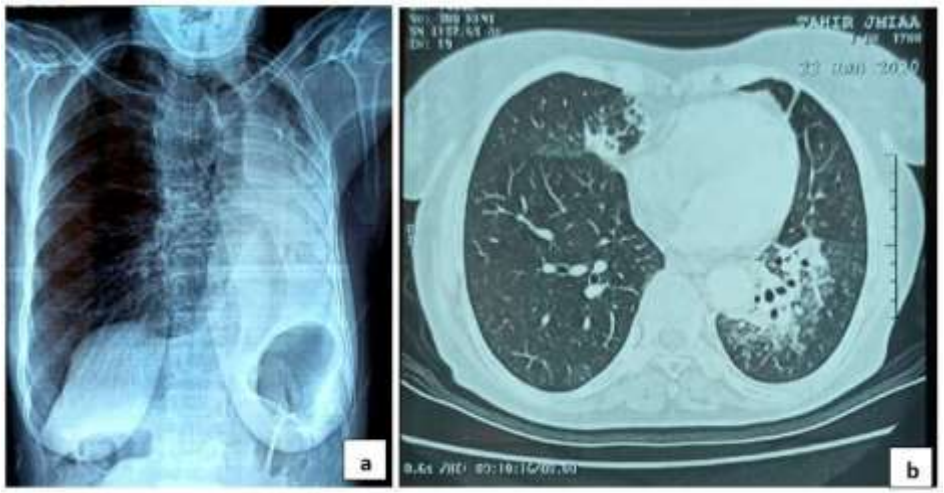

Figure 1: a: Chest X-ray: opaque and retracted left hemithorax

b: Pulmonary CT scan: areas of consolidation in the left lower lobe and middle lobe with slight bronchial architectural distortion.

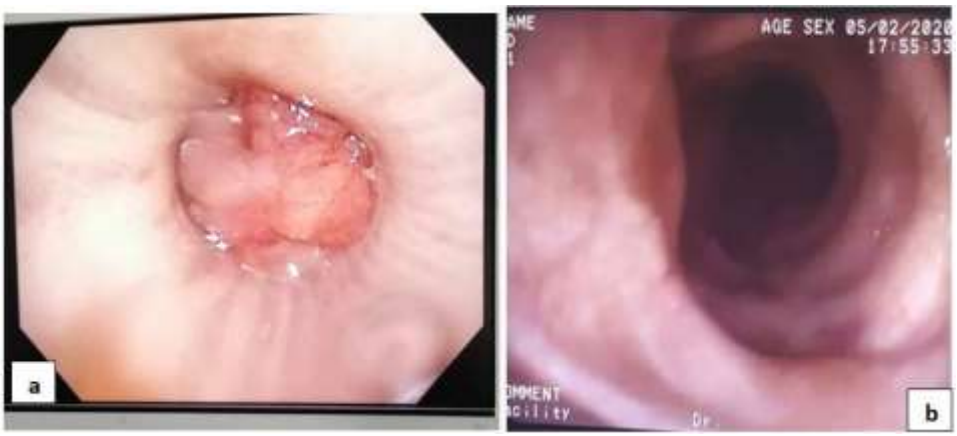

Figure 2: a: Bronchoscopy visualizes a rounded, well-defined, and well-vascularized tumor that completely obstructs the left main bronchus

b: Bronchoscopy visualizes diffuse first-degree inflammation and hyperemic mucosa without a visible tumor in the left main bronchus

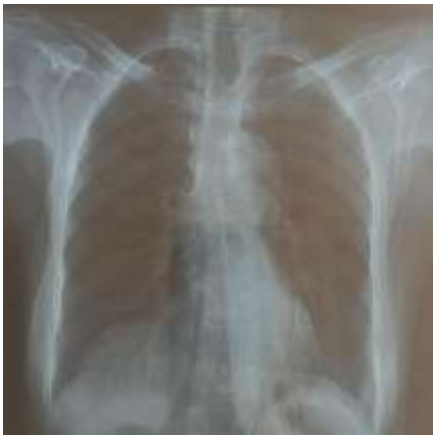

Figure 3: Follow-up chest X-ray showing no abnormalities

III- Case Report 2:

A 60-year-old woman, with a history of treated pulmonary tuberculosis (1989) and 20 years of wood smoke exposure, was admitted for purulent bronchial syndrome, mild hemoptysis, and exertional dyspnea. On admission, the patient had an SpO_2 of 88% on room air,

was polypneic at 22 breaths per minute, tachycardic at 110 beats per minute, normotensive at 120/80 mmHg, and afebrile.

Basal rhonchi were noted. Chest X-ray showed basal infiltrates on the right with blunting of the left costophrenic angle. Laboratory tests showed hemoglobin at 14 g/dL, WBC at 12,280/mm³ (neutrophil predominance), CRP at 24 mg/L and ESR at 46 mm/hr. She was treated with oxygen, hemostatics, and amoxicillin-clavulanate (3 g/day). Bronchoscopy revealed a smooth, reddish tumor fully obstructing the left main bronchus. Histopathology showed minimal lymphoplasmacytic infiltrate without tumor or granuloma. GeneXpert on bronchial aspirate was negative. CT showed tree-in-bud micronodules in the right upper lobe, postero-basal left consolidation with air bronchogram, and left pleural effusion. A second bronchoscopy one month later was normal. Clinically, hemoptysis resolved, bronchial syndrome disappeared, and dyspnea improved. Follow-up labs showed normalized WBC (7,850), CRP (1.6), and ESR (10). CT showed radiological resolution. Inflammatory pseudotumor diagnosis was confirmed by clinical, biological, radiological improvement and histological findings.

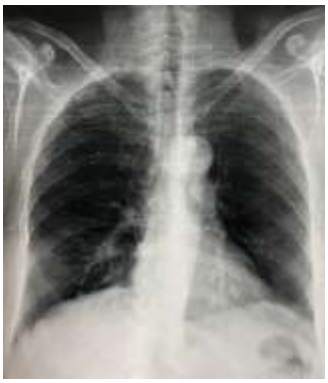

Figure 4a: Chest X-ray showing some right basal infiltrates with blunting of the left costophrenic angle

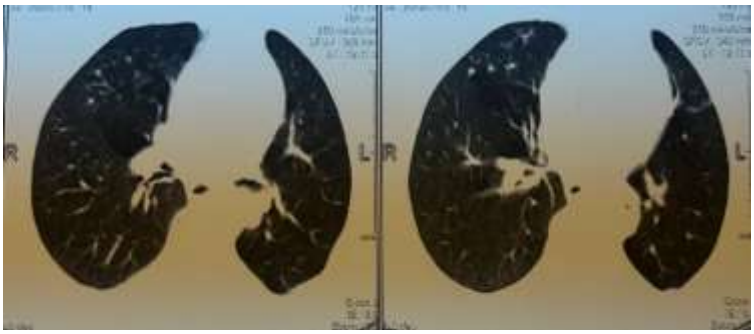

Figure 4b: Chest CT showing micronodular infiltrates in the right upper lobe giving a tree-in-bud appearance, with a postero-basal left consolidation showing an air bronchogram and a left pleural effusion

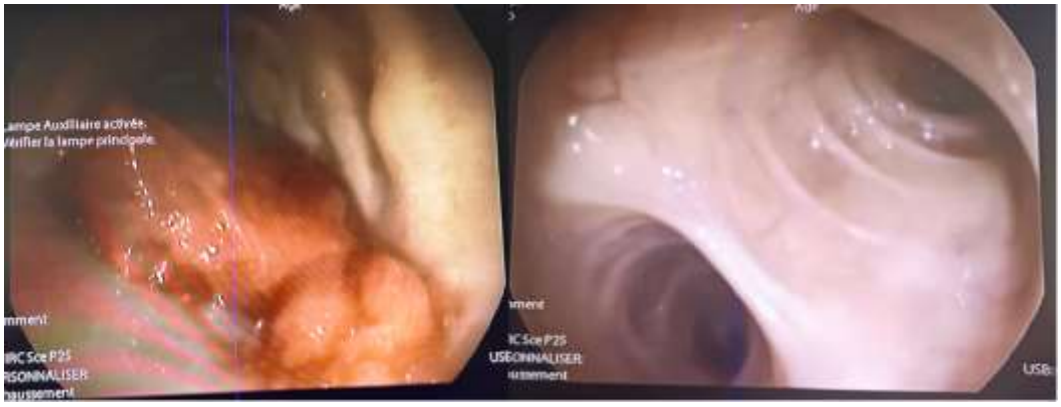

Figure 5a-b:
a: Flexible bronchoscopy revealing a smooth, reddish, budding tumor completely obstructing the left main bronchus
b: Endoscopic appearance essentially normal

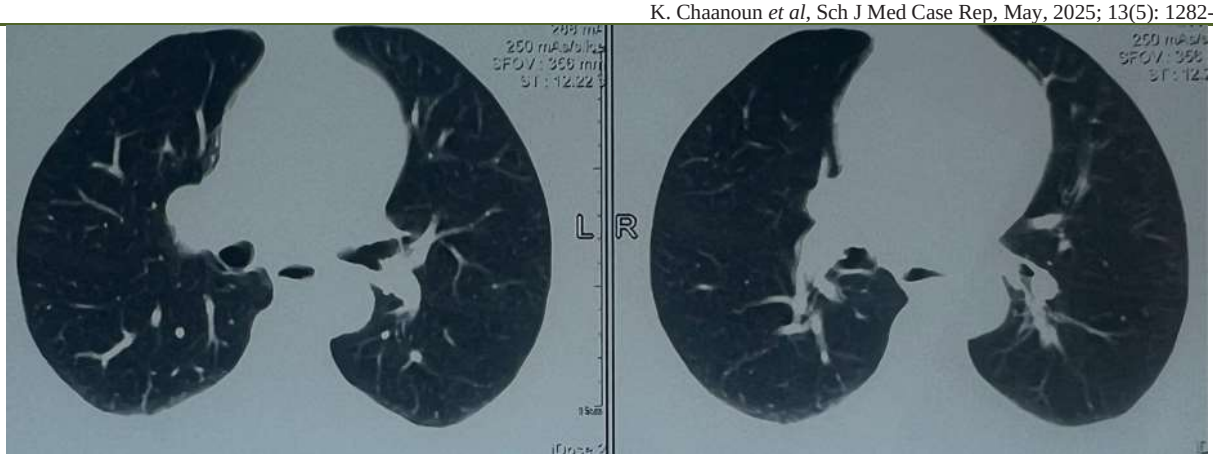

Figure 6: Resolution of the consolidation focus, branched micronodules, and left pleural effusion

IV- Case Report 3:

A 45-year-old woman, with a history of cervical lymph node tuberculosis in 2015 and passive smoking since childhood, was admitted for recent-onset chest pain and exertional dyspnea. On admission, the oxygen saturation was at 98%, the respiratory rate at 17/min, the heart rate at 76 bpm and the blood pressure at 110/80 mmHg. She was afebrile and clinical exam was normal. Chest X-ray showed a right paratracheal opacity. Laboratory tests showed hemoglobin at 12 g/dL, WBC at 5,960/mm³, CRP at 9.4 mg/L, and normal LDH, AFP, and β-hCG. CT revealed a right mediastino-pulmonary lesion. Bronchoscopy was normal. GeneXpert on aspirate was negative. Transthoracic biopsy revealed lymphoplasmacytic granulomatous infiltrate expressing CD30 and CD15 without atypical cells or malignancy. Clinically, dyspnea and chest pain resolved. Follow-up CT at 5 months showed spontaneous resolution. The inflammatory pseudotumor diagnosis was confirmed.

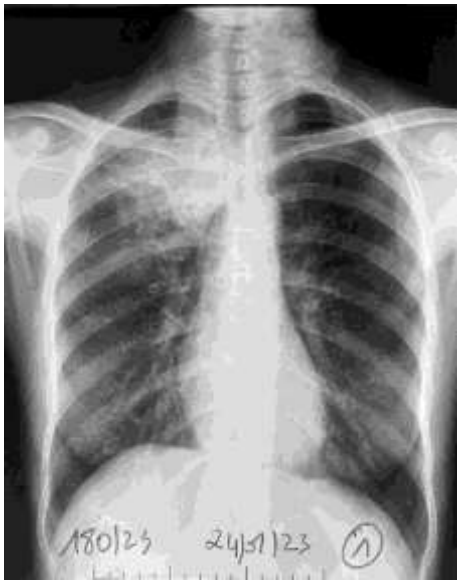

Figure 7a: Chest X-ray showing a right laterotracheal opacity

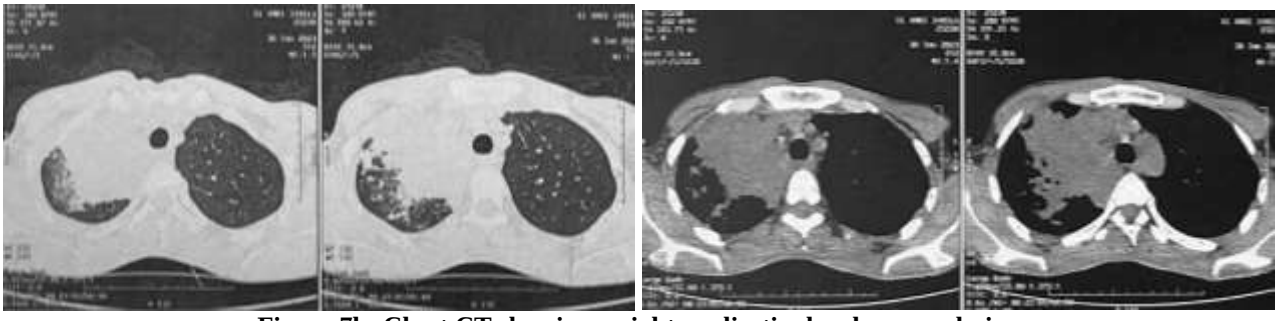

Figure 7b: Chest CT showing a right mediastinal-pulmonary lesion

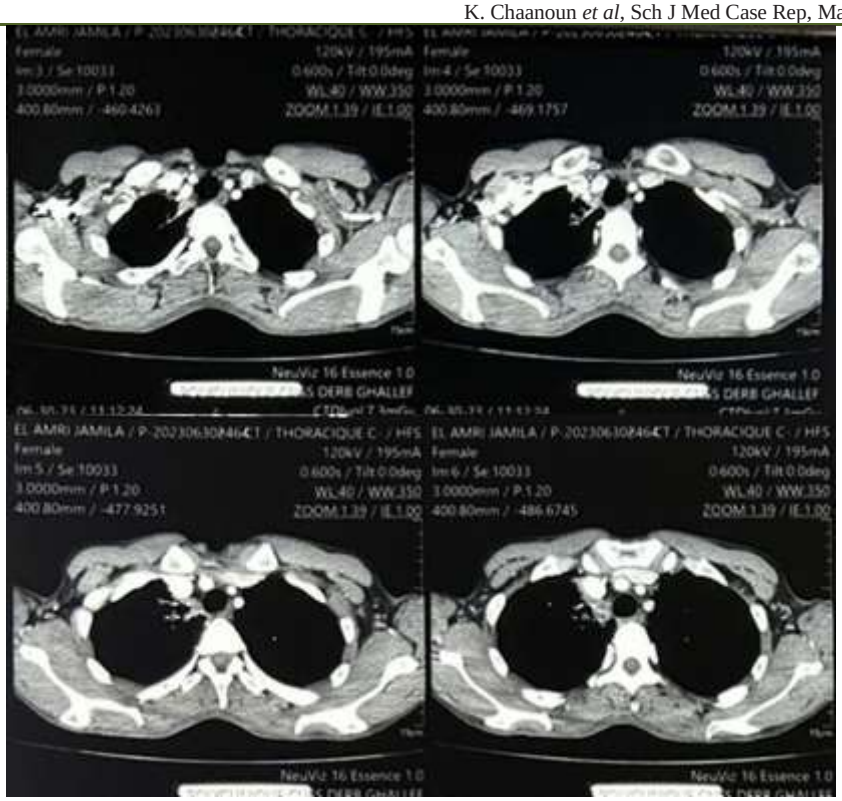

Figure 8: Follow-up chest CT showing spontaneous radiological clearance

V- DISCUSSION

Myofibroblastic tumors are rare, accounting for less than 1% of pulmonary tumors. Known under various names such as inflammatory pseudotumor, xanthogranulomatous tumor, or fibrous histiocytoma, recent cytogenetic studies suggest they are neoplasms rather than reactive lesions. They affect both genders and all ages, more often children and young adults. Around 25% occur before 18 years old, with a second peak between 40–60 years. Our cases were discovered in older individuals. Etiopathogenesis remains unclear, with possible autoimmune or infectious origins (EBV, HHV8, fungi, bacteria like Coxiella, Mycoplasma, Rhodococcus, mycobacteria). Clinically, patients may be asymptomatic (40–75% of cases) or show variable symptoms: cough, fever, chest pain, dyspnea, hemoptysis, recurrent infections, weight loss, anorexia. All our patients had nonspecific symptoms leading to radiological discovery. Imaging is nonspecific and the diagnosis is by exclusion. Radiographs show solitary nodules and lesions, usually well circumscribed, in lower lobes or peripheral zones. Multiple nodules and mediastinal involvement can occur. Calcifications/cavitations are rare. CT shows round or oval parenchymal masses, sometimes with calcifications. Aggressive forms may extend to mediastinum, chest wall, diaphragm. Our findings included opaque hemithorax, consolidations, micronodules, mediastinal-pulmonary masses. Diagnosis is pathological, often requiring surgical biopsy due to cellular heterogeneity. Macroscopically: well circumscribed but unencapsulated firm hemispherical masses. Histology shows inflammatory infiltrate (plasma cells, lymphocytes, histiocytes), fibroblastic or myofibroblastic stroma with collagen. Surgical resection is the treatment of choice with incomplete resection and 60% of recurrence. Resection ranges from segmentectomy to pneumonectomy. Corticosteroids are used if surgery isn't possible. Radiotherapy and chemotherapy may be needed for recurrent, multiple, or inoperable cases with mediastinal invasion. Spontaneous regression has been reported. None of our patients underwent surgery due to favorable spontaneous outcomes. Prognosis is generally good. Five- and ten-year survival post-surgery is 91.3% and 77.7%, respectively. However, IPTs can behave aggressively with mediastinal, pleural, or chest wall invasion, or extrapulmonary spread (brain, spine, muscle, liver). Malignant transformation is rare.

VI-CONCLUSION

The terms inflammatory pseudotumor and inflammatory myofibroblastic tumor likely encompass several entities due to heterogeneous clinical and histological presentations, unclear pathogenesis, and undefined treatment protocols.

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