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Opaque Hemithorax Revealing an Inflammatory Pseudotumor: A Case Report

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Abstract **Case Report**

Inflammatory pseudotumours of the lung are usually unifocal lesions of unknown pathogenesis and benign prognosis. We report the observation of a 68-year-old patient, hospitalised for etiological evaluation of a retracted opaque haemithorax revealed by exertional dyspnoea and chest pain. A radioclinical presentation suggested the diagnosis of a malignant bronchopulmonary tumour, but was refuted immediately after examination.

Keywords: Hemithorax, Inflammatory Pseudotumor, xanthomas.

INTRODUCTION

Inflammatory pseudotumors (IPT) were initially described in the lung, under such varied terms as "postinflammatory tumors", "plasma cell granulomas", "xanthomas", "xanthomatous pseudotumors", "fibrous histiocytomas" or "histiocytomas". These are rare tumors, whose pathogenesis is still unclear and whose treatment is poorly codified.

OBSERVATION

The patient was 68 years old, non-smoker, exposed to wood smoke for 30 years, and was admitted to hospital with recent exertional dyspnea associated with burning left chest pain. On admission, the general examination revealed a patient in fairly good general condition, performance status 1, eupneic, normocardial and apyretic with an oxygen saturation in free air SaO2: 96%. The thoracic examination showed a fluid effusion syndrome of the entire left hemithorax. The rest of the somatic examination was unremarkable. The chest X-ray showed a retracted left hemithorax (Fig 1a). On the biological workup, the white blood cell count was 15,810 mm3 predominantly PNN, the C-reactive protein was elevated to 58 mg/L, the sedimentation rate accelerated to 41 mm at the 1st hour. Bronchial endoscopy (Fig 2a) revealed a well-vascularized, well-limited, rounded tumor that completely obstructed the lumen of the left main bronchus. Pending the results of the bronchial biopsies, the patient was put on antibiotic therapy, a protected amoxicillin for 8 days at a rate of 3 g per day.

The anatomopathological study of the biopsy of the base of the tumor was in favor of a respiratory mucosa with a regular surface coating. The chorion was the site of a diffuse and dense inflammatory infiltrate composed of lymphocytes, plasma cells and some eosinophilic polynuclears with the presence of fibroblasts.

The thoracic CT scan (Fig 1b) performed four days after the bronchoscopy showed foci of systematized condensation in the left lower lobar region with an air and middle lobar bronchogram associated with a discrete bronchial architectural distortion.

A second bronchoscopy (Fig 2b) performed 12 days later showed a diffuse first degree inflammatory state in the left main bronchus with a discrete thickening of the spurs and a hyperhemic mucosa without visible tumor.

The anatomopathological study of the bronchial biopsies was in favor of a bronchial mucosa lined by a regular epithelium largely detached resting on a chorion containing fibrous reorganizations and a polymorphic inflammatory infiltrate made of lymphocytes, plasmocytes, eosinophilic polynuclears

and neutrophils with a vascular hyperplasia by place. No specific lesion or tumor.

A biological check-up performed 10 days later showed normalization of the white blood cell count at 8900 mm3 vs 15810 mm3, C-reactive protein at 14 vs 58 and sedimentation rate at 8 vs 41 mm. The control radiograph (Fig 3) was borderline normal.

The diagnosis of an inflammatory pseudotumor was retained in view of the clinical and biological improvement, as well as the total regression of the tumor under antibiotic therapy and the histological signature.

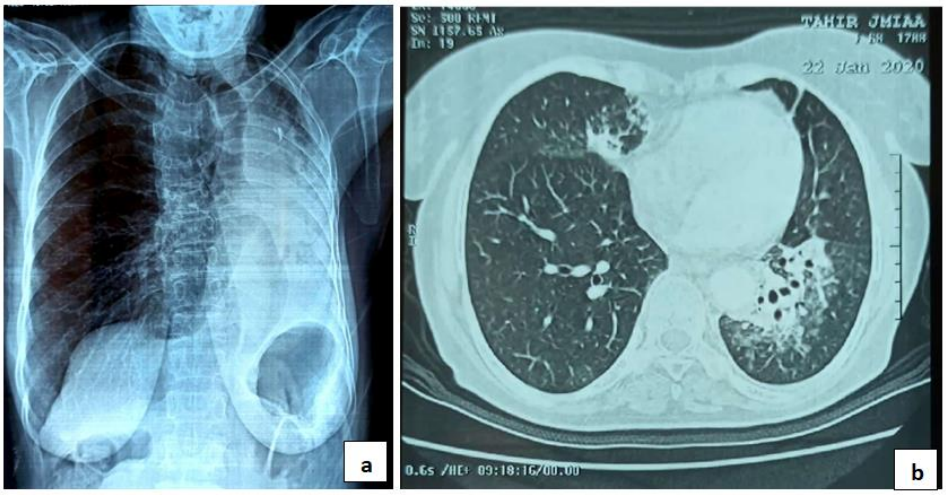

Figure 1: a: Chest radiograph: retracted opaque left hemithorax. b: Chest CT: foci of systematized condensation in the left lower lobar and middle lobar with a discrete bronchial architectural distortion

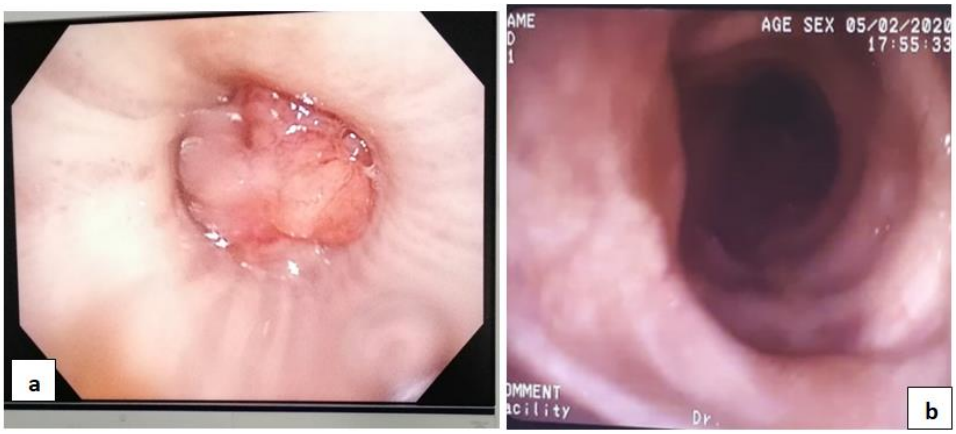

Figure 2: a: Bronchial endoscopy visualized a well-limited, well-vascularized, rounded tumor that totally obstructed the lumen of the left main bronchus. b: Bronchial endoscopy visualizing diffuse first degree inflammation and hyperemic mucosa without visible tumor in the left main bronchus

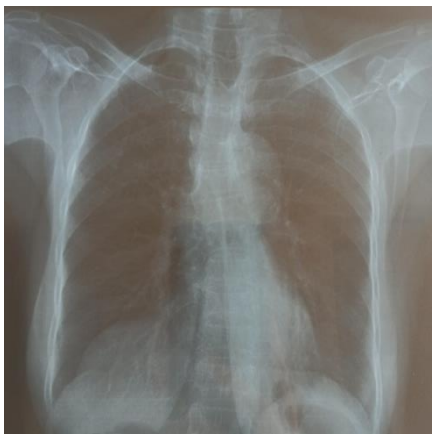

Figure 3: Substantially normal chest radiograph

DISCUSSION

Myofibroblastic tumors are rare. They represent less than 1% of lung tumors [1]. They are known under several terms because of the variability of their cellular composition: inflammatory or xantogranulomatous pseudotumors or fibrous histiocytomas. The latest cytogenetic studies suggest that they are neoplasms and not reactionary lesions [2].

They affect both sexes, at any age, with a predominance of children and young adults in many series [3-5]. The discovery of ITP was made at an advanced age for our patient.

There are many uncertainties about the pathogenesis of ITP. Several hypotheses have been put forward, such as an autoimmune origin, an infectious origin suspected in the presence of a history of viral (Epstein-Barr virus or HHV8 virus), mycotic or bacterial (Coxiella burnetti, Mycoplasma pneumo-niae, Rhodococcus equi, mycobacteriosis) pulmonary infection in 30% of cases, as was the case in our patient, but no infectious agent could be detected in the bronchial samples [5].

A tumor origin has also been suggested by the evidence of clonal chromosomal abnormalities and, more recently, abnormalities frequently involving the 2p23 chromosomal region, which contains the ALK gene, whose oncogenic activity has been demonstrated in anaplastic large cell lymphomas [5].

Clinically, patients may be completely asymptomatic with incidental discovery of the tumor on chest X-ray, or may present with highly variable clinical signs. The majority of patients present with cough, fever, chest pain, dyspnea, hemoptysis or recurrent infections, sometimes associated with weight loss and anorexia [1, 6, 7].

Chest imaging is non-specific and the diagnosis of an inflammatory pseudotumor remains a diagnosis of elimination. This tumor presents on chest radiography as a well-circumscribed nodule or mass, mostly in the lower lobar and peripheral areas [8]. These nodules may be multiple or extend into the mediastinum [6, 9]. Calcifications and cavitations are rare (less than 40% of cases) [1]. In our observation, the inflammatory pseudotumor was revealed radiologically by an opaque hemithorax appearance. Chest CT shows a round or oval parenchymal mass with regular or irregular contours and sometimes calcifications, especially in children (5% of cases) [10]. In aggressive forms, CT may reveal mediastinal, parietal or diaphragmatic extensions [10].

The positive diagnosis of ITP is anatomo-pathological, often requiring a surgical biopsy. Indeed, diagnosis by trans-thoracic or trans-bronchial fine-needle biopsy is difficult due to the varied cellular composition of these tumors [11].

Macroscopically, ITP often appears as a well-circumscribed but non-encapsulated, firm, homogeneous, hemispherical mass with occasional hemorrhagic or necrotic remodeling and areas of bone metaplasia [12]

Histopathologically, ITP consists of an infiltrate inflammatory pattern comprising plasma cells, B and T lymphocytes, histiocytes, and sometimes xanthomatous macrophages. There is an associated fibroblastic and/or myofibroblastic population within more or less hyalinized collagen fibres [12]. In our observation, the anatomopathological study of bronchial biopsies objectified a mucosa with a regular surface coating. The chorion was the site of a diffuse and dense inflammatory infiltrate composed of lymphocytes, plasma cells and some eosinophilic polynuclei with the presence of fibroblasts.

Surgery is the treatment of choice and resection must be complete. A tumor residue signifie a recurrence in 60% of cases [1]. It consists of a segmentectomy, a lobectomy, or even a pneumonectomy because of the invasive nature of the lesion.

Corticosteroid therapy is prescribed in cases where surgery is not indicated [2, 13]. Radiotherapy or chemotherapy is also prescribed for multiple, recurrent or non-operable forms with significant mediastinal invasion. Antibiotic therapy may also be prescribed, as in the case of our patient who was put on protected amoxicillin. Spontaneous regression of ITP has also been reported in the literature [14].

The evolution is usually favorable. Survival after surgery at five and ten years is 91.3% and 77.7% respectively [15]. However, ITPs can behave as aggressive tumors in case of mediastinal, pleural or parietal invasion [16, 17] or in the presence of extra-thoracic localizations (cerebral, spinal, muscular, hepatic) [16, 18]. Malignant transformations are rarely reported [19, 14-20].

CONCLUSION

The terms inflammatory pseudotumor or inflammatory myofibroblastic tumor probably cover several entities, considering the heterogeneity of their clinical and histological presentation, their still unclear pathogenesis and their poorly codified treatment.

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