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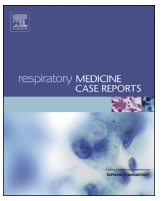

Case Report

Lung consolidation in a young-age active smoker: An unexpected diagnosis

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ABSTRACT

A 29-year-old man presented to Emergency Department with nonspecific symptoms. Through a series of radiological and invasive diagnostic studies we finally reach an unexpected diagnosis of hypersentivity pneumonitis; this is a complex and heterogeneous disease which diagnosis can be challenging as its clinical, radiologic and histopathologic features overlap with those of other interstitial lung diseases (ILDs). Diagnosing an ILD is a dynamic process, and that is the reason why complex cases discussed in a multidisciplinary team may need to be reconsidered in light of evolution of the disease and the results of the performed exams with a flexible approach.

1. Introduction

Hypersensitivity pneumonitis (HP), previously also known as extrinsic allergic alveolitis (EAA) is a complex interstitial lung disease (ILD) caused by exposure to an inhaled antigen. An inhaled antigen can cause the interstitial lung disease (ILD) known as hypersensitivity pneumonitis [1,2]. HP is a heterogeneous disease (ranging from inflammatory self-limiting disease to recurrent or progressive inflammatory disease to chronic fibrotic disease mimicking idiopathic pulmonary fibrosis - IPF) and its phenotype has major impacts on treatment and outcome. Nowadays we are witnessing an increase in the prevalence and incidence of ILDs, as our knowledge has improved. In recent years we have discovered more and more data about pathogenesis and now we certainly have even more means to make a diagnosis. Diagnosing an ILD is a dynamic process, and that is the reason why complex cases discussed in a multidisciplinary team may need to be reconsidered in light of evolution of the disease and the results of the performed exams with a flexible approach. Highlight this concept could be relevant in real-life clinical activity. To our knowledge this is an original case report providing important concepts.

2. Case presentation

A 29-year-old man presented to Emergency Department with chest pain and non-productive-cough. His symptoms had been ongoing for 2 months and were accompanied by night sweat, weight loss (approximately 5 kg in 3 months), but no fever. He was previously a fit and well warehouse worker with no past medical history. He was not taking any regular medications and there was no known allergy. He was an active smoker of 20 cigarettes/day for 4 years and admitted to previous cannabinoids use. He denied any family history for respiratory disease. He did admit to an ongoing mould exposure in his dwelling. An initial chest x-ray showed evidence of para-hilar left upper lobe consolidation with indistinct margins, compatible with inflammatory thickening. He was sent

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home for a presumed respiratory tract infection with antibiotic therapy of levofloxacin with a plan for follow-up Interval chest x-ray. After 7 days the symptoms did not resolve and follow-up chest x-ray after antibiotic therapy was unchanged, so we proceeded to a thorax CT scan with IV contrast. The CT (Fig. 1) showed a marked progression of number, density and dimension of the already present cystic alteration on previous HRTC scan, with evidence of consolidation in the anterior segment of the left upper lobe. He was therefore referred to our respiratory team for further inpatients work-up.

On admission physical examination the patient appeared in no acute distress. Vital signs revealed a temperature of 36.5 °C, peripheral pulse of 68 beats/minute, respiratory rate of 16/min and blood pressure of 130/80 mmHg and oxygen saturation of 98% on room air. Respiratory examination revealed scattered bilateral crackles in absence of wheezes or rubs. Cardiac and abdominal examinations were unremarkable. Laboratory examination of peripheral blood highlighted a borderline increase in inflammatory markers: C-reactive protein was 0,75 mg/dL and procalcitonin was negative; the remaining admission blood exams were unremarkable.

Following up from the CT thorax findings, further imaging was arranged as an impatient in the form of PET scan (Fig. 2). There was significant uptake from the opacification in the left upper lobe (SUV 13.83) and from mediastinal lymph nodes in the aortopulmonary region (SUV 4.46), paratracheal (SUV 4), sub-carinal (SUV 5.34) e hilar regions bilaterally (SUV 4.82). In the first instance, the PET alterations were suggestive for lymphoproliferative disease or a granulomatous disease. Consequently, a lung tissue biopsy of

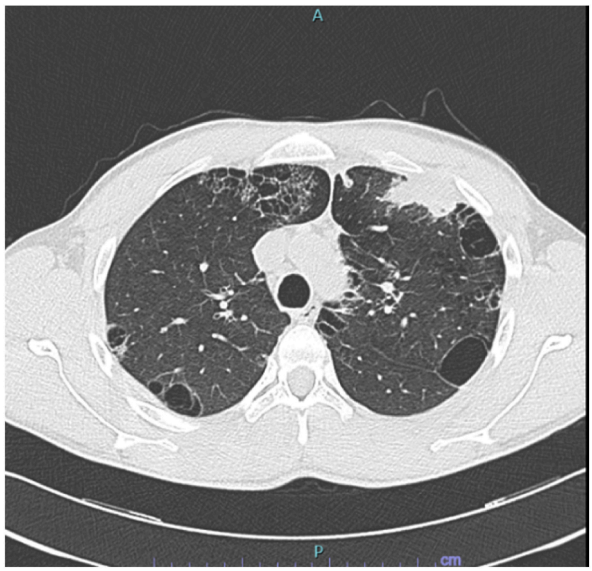

Fig. 1. Chest CT-SCAN of consolidation in the anterior segment of the left upper lobe with already present cystic alteration.

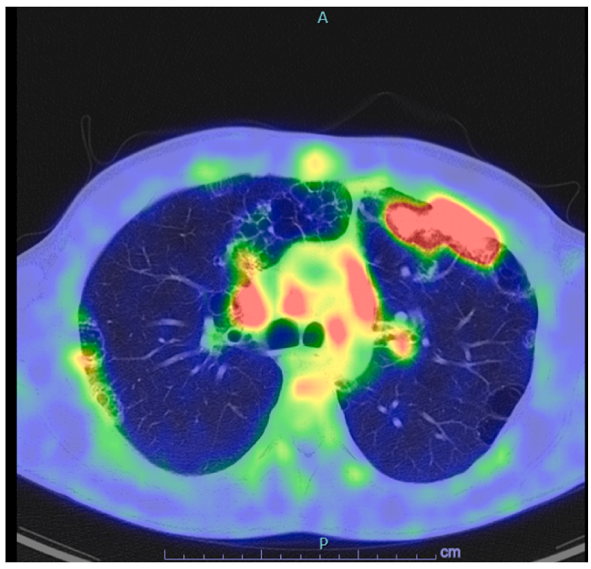

Fig. 2. Chest PET SCAN with signal uptake from the opacification in the left upper lobe (SUV 13.83) and from mediastinal lymph nodes in the aortopulmonary region (SUV 4.46), paratracheal (SUV 4), sub-carinal (SUV 5.34) e hilar regions bilaterally (SUV 4.82).

the apical portion of the left lung in video assisted thoracoscopy was arranged. The histological exam evidenced: inflammatory reactive infiltrate characterised by giant cell foreign body-like cellular components and an exuberant reaction identifying an inflammatory pseudotumor. The microrganisms cultural tests and research were all negative (Mycobacterium tuberculosis and non-tubercular mycobacteria infections were excluded). In addition, the patient underwent an endobronchial ultrasound with transbronchial needle aspiration (EBUS TBNA) in order to biopsy mediastinal lymph nodes which was not conclusive, highlighting lymphocytes aggregation with some bronchial epithelial cells. Contextually to EBUS a bronchoalveolar lavage (BAL) was performed; the cultures from the lavage were negative and tuberculosis infection was again excluded. In the meantime another HRTC Thorax scan was performed with evidence of new appearance of opacities in the right lung.

The case was therefore discussed with a multidisciplinary team composed of respiratory physician, thoracic surgeon, radiologist and pathologist; the MDT agreed the next step would be a lung segment biopsy during video assisted thoracoscopy (VATS) **in order to exclude malignant etiology**. This time the histology exam was conclusive for hypersensitivity pneumonitis (HP). In hindsight the upper lung lobes localisation of disease, the referred exposure to moulds **(supposed to be the causal antigen for developing HP)** and the chest CT scan lesions' tendency to evolve, all support the diagnostic hypothesis of an unusual form of hypersensitivity pneumonitis.

Six months of systemic corticosteroid therapy was completed with gradual resolution of symptoms. Three months after the discontinuation of the treatment, a follow-up HRCT scan (Fig. 3) showed complete resolution of the opacification in the left upper lobe and in the right lung. The cystic alterations and the bilateral hilar lymphadenopathy were stable. There was no evidence of new appearance of ground-glass or solid lesions. At the time of writing, the patient remains under our outpatient follow-up and had no recurrence of disease.

3. Discussion

HP is a complex and heterogeneous disease. Making a diagnosis of HP can be challenging as its clinical, radiologic and histopathologic features overlap with those of other interstitial lung diseases (ILDs) and it may not be possible to identify a culprit exposure [3].

Differential diagnosis mainly concerns with radiologic (fibrosing and not fibrosing) and histopathologic datas2:

For what concerns with histopathology: biopsy should be avoided in patients in whom a confident diagnosis of HP can be made with the available datas. However sometimes, obtaining lung tissue is recommended if warranted based on the risk/benefit for the individual patient. Tissue samples should be reviewed by a pathologist experienced in ILD in the context of clinical and radiologic informations [4]. The usefulness of surgical lung biopsy for the diagnosis of ILD remains controversial, since they are associated with a high morbidity and mortality. VATS is generally considered to be a safer procedure that provides lung tissue samples that are sufficient for a definitive histopathological diagnosis. Often, biopsy is deemed impracticable due to age, disease severity, comorbidities, immunocompromised status, or hypoxemic respiratory failure. In our case description, the patient has no relevant comorbidities and considering the young-age has an optimal performance status; consequently surgical diagnostic tools proved to be conclusive [5].

Managing with patients affected by ILD means to deal with some diagnostic uncertainty and yet have to take diagnostic decisions to best advise the patients about how to manage their disease. Multiple factors can be helpful in this. Above all, multidisciplinary decisions increase the accuracy of the diagnosis and allow it to be based on the consensus of experts rather than on the opinion of individual physicians. MDT has become the gold standard for the diagnosis of many ILD yet.

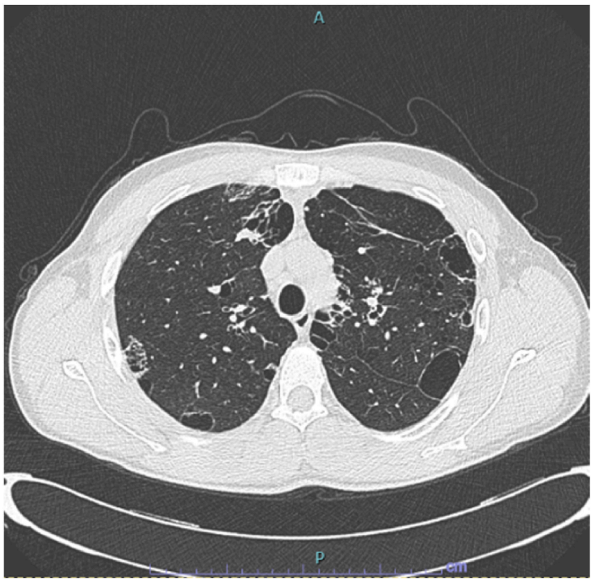

Fig. 3. Chest CT-SCAN showing complete resolution of the opacification in the left upper lobe and in the right lung. There was no evidence of new appearance of ground-glass or solid lesions.

4. Conclusions

- The diagnostic approach to HP has evolved as our knowledge of ILD is improving, but still remains complicated. The currently approved diagnostic criteria depend on a series of findings about the patient and are still under discussion.
- Both surgical and non-surgical biopsy techniques have experienced progress with regards to safety and diagnostic yield in ILD. Further research, continued monitoring, robust patient selection processes and multidisciplinary shared decision-making are some ways to optimise outcomes whilst minimizing risk.
- Diagnosing an ILD is a dynamic process, and that is the reason why complex cases discussed in a multidisciplinary team may need to be reconsidered in light of evolution of the disease and the results of the performed exams, with a flexible approach. Nowadays diagnostic guidelines better correspond to the pragmatic management adapted to real practice and with our case description we want to put paramount importance around the Multidisciplinary discussion and its workup.

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Consent

The patient provided written consent to be included in this manuscript.

CRediT authorship contribution statement

F.R. Bertuccio: Conceptualization, Data curation, Investigation, Methodology, Writing – original draft, Writing – review & editing. **N. Baio:** Data curation. **V. Chino:** Data curation. **V. Ferroni:** Data curation. **S. Montini:** Data curation. **L. Pisanu:** Data curation. **A. Cascina:** Writing – review & editing. **V. Conio:** Writing – review & editing. **A.G. Corsico:** Supervision. **G.M. Stella:** Supervision.

Declaration of competing interest

The authors declare that they have no conflict of interest.

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