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Extrinsic allergic alveolitis. Case report

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ABSTRACT

Extrinsic allergic alveolitis encompasses a group of immunologically mediated entities caused by repeated inhalation of antigens. Its incidence ranges between 0,3 and 0,9 cases per 100 000 inhabitants worldwide. A case of an 18-year-old patient, a carpentry assistant, who started with shortness of breath, cough and fever after cutting wood is presented. Given these symptoms and the finding of crackles at the base of the left hemithorax, it was treated as community-acquired pneumonia. Complementary tests showed increased immunoglobulin G, eosinophilia and a restrictive pattern in spirometry. An extrinsic allergic alveolitis was diagnosed and treatment with oral steroids was indicated, with a satisfactory evolution. Occupational extrinsic allergic alveolitis is a difficult pathology to diagnose due to its low frequency and similarity with other interstitial lung diseases, but once diagnosed it evolves favorably with the use of steroids.

Keywords: Extrinsic Allergic Alveolitis; Interstitial Lung Diseases; Pneumonia.

Hypersensitivity pneumonitis or extrinsic allergic alveolitis encompasses a group of immunologically mediated entities caused by repeated inhalation of antigens that trigger an inflammatory reaction, resulting in granulomatous interstitial pneumonitis[1,2].

This entity was first described in 1932 by Campbell, in a farmer affected by a pulmonary inflammatory reaction after exposure to moldy hay, and it was named farmer's lung disease. However, it was not until 1962 that Pepys introduced the term hypersensitivity. From its first description, farmer's lung disease became the prototype of the disease[3,4].

Its pathogenesis is not yet fully elucidated, but type III (Arthus phenomenon), type IV and type I hypersensitivity mechanisms have been implicated in that order of frequency[3,4].

Nowadays, the actual impact of this disease remains unknown. However, its incidence is reported to be between 0,3 and 0,9 per 100,000 inhabitants worldwide. Some studies suggest that it represents between 2 % and 13 % of interstitial lung diseases[1,5,6]. In the consulted literature, no reference was found on the incidence of this type of pneumonitis in Cuba and only one clinical case with this diagnosis was reported[3].

It should be taken into account that interstitial lung diseases in general and extrinsic allergic alveolitis in particular have a low incidence in the population and they are classified as rare diseases according to the World Health Organization (WHO). The acute presentation of extrinsic allergic alveolitis is misdiagnosed and therefore knowledge of its clinical presentation is essential.

CASE PRESENTATION

An 18-year-old male patient, black skin color, carpentry helper, with only six months of experience, apparently healthy, with no previous hospital admissions, reported a first-degree family history of bronchial asthma and he denied to have any allergies, surgeries or traumas.

The patient reported that one afternoon after an intense day of work cutting wood, he began with shortness of breath, dry cough, decay and he had a fever peak of 39,50 °C. That same day he went to a general hospital where he was administered an intramuscular dose of Dipyrone (1200 mg), but a few minutes later he presented angioneurotic edema,

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Conflict of interests
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for which he was urgently referred to the Provincial Hospital of Cienfuegos.

At the emergency room, physical examination of the respiratory system revealed crackles at the base of the left hemithorax and no other alterations were detected. An initial X-ray showed only inflammatory mottling in both bases and was interpreted as community-acquired pneumonia, so treatment started with intravenous Ceftriaxone (1 g). After its administration, he had a new anaphylaxis reaction, with erythematous rash and 3-4 mm vesicles on the thorax and right upper limb. Subsequently, the presence of marked dyspnea and increased crackles up to 2/3 of both lung fields drew attention. For all these reasons, it was decided to discontinue treatment with Ceftriaxone.

A new posteroanterior chest X-ray was performed, showing diffuse radiopacities of inflammatory aspect up to 2/3 of both lung fields. It was decided to admit him to the Progressive Care Unit with the possible diagnosis of severe community-acquired bronchopneumonia. Meropenem (1g) 1 bulb every 8 hours and Vancomycin (500 mg) 2 bulbs every 12 hours, to be administered in 2 hours, both for 7 days, were administered intravenously. Complementary tests indicated the following results: leukocytes: 18,9 x 10 /L, segmented: 0,58, lymphocytes: 0,36, eosinophils: 0,06, IgG: 22,67 g/L, Sat. PO : 90 % and PO2: 59 mmHg.

Eosinophilia, increased Ig (immunoglobulin) G, decreased oxygen pressure and oxygen saturation were evidenced. In addition, spirometry showed a restrictive pattern and a bronchoscopy was performed where an inflammatory infiltrate with abundant lymphocytes was observed.

The patient remained in the Intensive Care Unit for 7 days, and presented only a discrete improvement, however, he was transferred to the Internal Medicine ward and the diagnosis of bronchopneumonia was reevaluated with the Pneumology service. The possibility of hypersensitivity pneumonitis associated with exposure to wood antigens was raised, with the doubt of not being able to identify in the field the different components with which he worked. Antibiotic treatment was discontinued and he started with prednisone (20 mg) 1 mg/kg/day, not to exceed 60 mg daily. This dose was applied for 15 days and the month of treatment was completed with prednisone (5 mg) 1 tablet daily. After 48 hours of starting steroid treatment the clinical picture of dyspnea, bibasal crackles and low saturation changed radically.

A posteroanterior chest X-ray (PA) was performed on the tenth day of evolution, which reported the presence of bilateral basal hilum infiltrate, without soft tissue or bone alterations. There were no alterations at the cardiomediastinum level (Figure 1A). On the fifteenth day, an evolving PA chest X-ray was performed, which reported remarkable radiographic improvement, without alterations in soft tissue and bone, or at the level of the cardiomediastinum (Figure 1B).

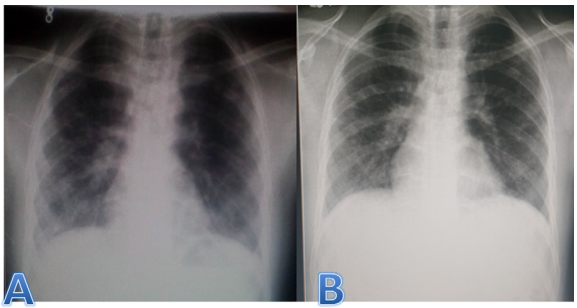

Figure 1. Image A shows a chest radiograph taken on the tenth day of evolution showing bilateral basal hilum infiltrate, without soft tissue or bone alterations. No alterations at the level of the cardiomediastinum. Image B shows an evolutionary PA view chest X-ray performed on the fifteenth day where a remarkable radiographic improvement is observed.

Source: Imaging Department of the "Dr. Gustavo Aldereguía Lima" Hospital, Cienfuegos, Cuba.

The patient was discharged two days later with follow-up by Pneumology and treatment with oral steroids. A simple and contrasted axial computed tomography was performed one month after the end of the steroid cycle, which showed no pleuro-pulmonary lesions (Figure 2). Therefore, his definitive medical discharge was decided.

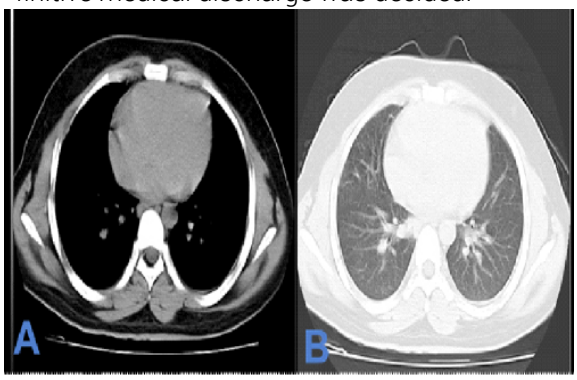

Figure 2. Image A shows a plain chest computerized axial tomography and image B shows a contrasted chest computerized axial tomography performed one month after the

DISCUSSION

Authors such as Miller et al.[7] and Fernández Pérez

et al.[1] coincide in the description of more than 30 different antigens causing this entity, which can be classified as: microorganisms, animal proteins and synthetic compounds of low molecular weight. In this case, the patient was cutting wood, an activity through which he could have been exposed to the triggering antigen; however, the limitation was that it was not possible to verify the causal antigen in the field.

Peña et al.[4] state that there is a close temporal relationship between the exposure to the antigen and the onset of symptoms, between 2 and 3 hours, with a maximum between 6 and 24 hours, which favors the diagnosis. In the analyzed case, there is a history of exposure to an antigen hours before the onset of symptoms.

In the consulted literature, three forms of clinical presentation have been described: acute, subacute and chronic[3,5,7,8,9]. The acute presentation is characterized by the appearance of cough, frequently associated with expectoration, fever, chills, sweating, myalgia and a feeling of prethoracic tightness. The average duration of symptoms is 12 to 48 hours. Physical examination is within normal limits and reveals bibasal crackles[4,5,7]. This symptomatology is also evidenced in the clinical presentation of the case described by Vidal Lacosta[10] and could be observed in the presented case.

The diagnosis of this type of pneumonitis is made in most cases by the presence of a suggestive clinical presentation, history of exposure to a recognized substance, findings on physical examination, a compatible radiograph and the presence of antibodies to the antigen[2,5,7]. Some authors state that the general laboratory data are nonspecific, although elevations of the erythrocyte segregation velocity and of IgG, IgM or IgA isotypes can occasionally be observed, as well as rheumatoid factor positivity, all reflecting an acute or chronic inflammatory process[5,9]. In the presented case only a rise in IgG was observed.

The radiological examination of choice is computerized axial tomography (CAT), where the observed pattern is variable depending on the stage of the disease and is characterized by the presence of patchy and diffuse ground glass pulmonary infiltrates[2,5,7,10]. However, Raghu et al.[5] state that the radiological findings sometimes do not correlate satisfactorily with the pathological anatomy. Recent studies on diffuse interstitial diseases agree with this, showing that the radiological pattern of ground glass only corresponds to inflammation in 65 % of the cases, while in 54 % it translates the existence of fibrosis[2,5,11]. In the presented case, at admission, this study was not performed because the diagnostic possibility of community-acquired pneumonia was evaluated, therefore, it was not possible to observe this pattern in the patient.

As described, lung function in early phases may be normal or show a decrease in forced vital capacity (FVC) and forced expiratory volume in the first second (FEV1), which typically appears in the first 4 to 6 hours of exposure in acute episodes. The ratio (FEV1) / (FVC) is normal or elevated indicating restriction[2,3,5,12]. In this case, it coincides with the scientific evidence since spirometry showed a restrictive pattern.

Anatomopathological studies can be useful to confirm the diagnosis, where changes in the bronchial epithelium, typical of this entity, can be found and evidence of lymphocyte-predominant inflammatory infiltrate in the form of lymphocytosis is an important element. Several studies can be performed such as bronchoalveolar lavage, transbronchial biopsy and surgical lung biopsy[5,9]. In this case, bronchoscopy showed results similar to those described above.

Extrinsic allergic alveolitis should be differentiated from the rest of the interstitial lung diseases, such as acute interstitial pneumonia, cryptogenic pneumonia and acute eosinophilic pneumonia, due to the similarities between them[7]. In the described case, it was necessary to make the differential diagnosis with eosinophilic pneumonia because in the complementary examinations the patient presented eosinophilia. However, De Giacomi et al.[13] state that the diagnosis of this entity is by exclusion and the finding of eosinophils in the lung biopsy is necessary, which was not observed in this case, a reason that allowed ruling out this diagnosis.

Treatment is mainly based on the avoidance of the antigenic source, which in many cases is made difficult by the impossibility of detecting the cause. The drugs of choice , used in the acute and subacute phase, are corticosteroids, in variable doses of 20 to 30 mg per day. In addition, other drugs such as cytotoxic agents can be used, including azathioprine, which is a common treatment in some health centers worldwide[2,7,9]. This patient improved substantially after the use of corticosteroids, even in the subsequent evolution he did not show signs of fibrosis in the CAT, which evidences the efficacy of the therapeutic lines described above.

It should be noted that according to the consulted literature, symptoms, signs and other manifestations disappear in days, weeks or months in most patients, if exposure has not been

CONCLUSIONS

Extrinsic occupational allergic alveolitis is caused by repeated or intense inhalation of various antigens, with a short evolution period, in which it is often not possible to determine the specific causal agent. It is a difficult pathology to diagnose due to its low

frequence and similarity with other interstitial lung diseases, but once diagnosed it evolves favorably with the use of steroids.

AUTHORSHIP

Roine Alberto Pena-Olivera, Ana Laura Navarro-Baldellot, Samuel Sánchez-Sánchez: conceptualization, methodology, resources, supervision, original drafting-drafting, drafting-reviewing, and editing.

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Alveolitis alérgica extrínseca. Informe de un caso

RESUMEN

La alveolitis alérgica extrínseca engloba un conjunto de entidades mediadas inmunológicamente, causadas por la inhalación repetida de antígenos. Su incidencia a nivel mundial oscila entre 0,3 y 0,9 casos por cada 100 000 habitantes. Se presenta el caso de un paciente de 18 años, ayudante de carpintería, que comienza con falta de aire, tos y fiebre después de cortar madera. Ante dicha sintomatología y el hallazgo de crepitantes en base del hemitórax izquierdo se maneja como una neumonía adquirida en la comunidad. Los exámenes complementarios mostraron aumento de la inmunoglobulina G, eosinofilia y un patrón restrictivo en la espirometría. Se diagnosticó una alveolitis alérgica extrínseca y se indicó tratamiento con esteroides por vía oral, con evolución satisfactoria. La alveolitis alérgica extrínseca ocupacional es una patología de difícil diagnóstico por

su escasa frecuencia y similitud con otras enfermedades pulmonares intersticiales, pero una vez diagnosticada evoluciona favorablemente con el uso de esteroides.

Palabras clave: Alveolitis Alérgica Extrínseca; Enfermedades Pulmonares Intersticiales; Neumonía.