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## Letters to the Editor

Anthracostenosis Associated With Exposure to Biomass Smoke and Presence of Mycobacterium tuberculosis  $^{\rm th}$ 

### Antracoestenosis con exposición a humo de biomasa y presencia de Mycobacterium tuberculosis

### Dear Editor,

ARCHIVOS DEBRONCONEUMOLOGÍA recently published a case report by Gómez-Seco et al.,<sup>1</sup> which informed us of the presence of an entity that has been unknown in our setting until now: anthracostenosis or anthracofibrosis. In this letter, we describe a new clinical case in Spain with particularities similar to the other 3 previously mentioned cases.

Our patient is a 66-year-old woman from a rural area, a nonsmoker, with no known history of tuberculosis or exposure to coal, although she had been inhaling wood fire smoke throughout her life. The patient had previously been diagnosed with middle lobe syndrome, and in 2002 she underwent lobectomy of the middle lobe due to recurrent pneumococcal infections.

The patient was sent to our department in 2011 with chronic dry cough. Chest radiography revealed right hilar enlargement and atelectasis-partial condensation of the anterior segmental bronchus of the right upper lobe (RUL). Said findings were confirmed by chest CT, which also demonstrated bronchiectasis in the RUL and a right paratracheal adenopathy measuring 2 cm (Fig. 1A). Given the suspicion for bronchial carcinoma, bronchoscopy was



**Fig. 1.** (A) Partial condensation and atelectasis of the anterior segmental bronchus of the right upper lobe (RUL). (B) Follow-up CT after 6 months of treatment showing partial resolution of the infiltrate together with more evident bronchiectasis in said lobe than at the start of treatment.

performed, which showed a middle lobe lobectomy stump with no signs of neoformation and an extensive black patch at the entrance of the bronchus of the left upper lobe. Due to an important amount of bleeding, only one biopsy sample could be taken. The tissue sample contained bronchial mucosa with macrophages in the *lamina propria* and black cytoplasmic pigment, with no signs of malignancy. The rest of the segmental bronchi of both lungs were impassable due to stenosis, especially in the adjoining airways near the black patch toward the culmen and the lingula. Isoniazid-resistant *Mycobacterium tuberculosis* was isolated in bronchial secretions.

The patient was treated for 6 months with ethambutol, pyrazinamide, and rifampicin. Afterwards, follow-up CT showed partial resolution of the infiltrate in the anterior segmental bronchus of the RUL with normalization of the size of the adenopathy (Fig. 1B). The endoscopic appearance was unchanged despite the treatment received. New bronchial suction and bronchoalveolar lavage samples confirmed the eradication of the Koch bacillus.

Anthracostenosis or anthracofibrosis was first described in 1998 by Chung et al.<sup>2</sup> as an entity that is different from coal workers' pneumoconiosis. It is most frequent in non-smoker women aged between 60 and 70 who live in rural areas of Asia and sub-Saharan Africa. Its etiopathogenesis has not completely been defined, and several hypotheses have been postulated that are not mutually exclusive. The two main hypotheses regarding triggering agents are the inhalation of smoke from incomplete biomass combustion and pulmonary tuberculosis (TB).<sup>1-4</sup> In the former hypothesis, although the ciliary movement eliminates the majority of inhaled particles, there are always residual quantities of accumulated remains at the subdivision points of the airways that activate the alveolar macrophages and alter the cellular immune response, favoring infections.<sup>3</sup> The second hypothesis relates anthracostenosis with TB: it is believed to be an exaggerated immune response to *M. tuberculosis* antigens, with lymph node infarction in the chains adjacent to the bronchi and toward which their caseous content would drain if they ruptured, generating fibrosis.<sup>2,3</sup> Bronchoscopy is the main diagnostic test used, and bronchial stenosis should be observed at several levels with patches of anthracotic pigmentation of the bronchial mucosa.<sup>3</sup> A proper differential diagnosis should rule out bronchogenic carcinoma and go beyond mere endobronchial tuberculosis (with focal affectation of a single segment or lobe and without patchy distribution).<sup>3</sup>

Until the publication of the series of 3 cases by Gómez-Seco et al.<sup>1</sup> in April 2012, no cases of anthracostenosis had been reported in Spanish patients. This fourth case described in our country is of particular interest due to the conjoint presence of the two main hypothetical triggering factors: exposure to wood-fire smoke and TB. It is important not to forget the fact that wood is still extensively used as a fuel source in Western countries, both in rural settings as well as in low-income urban areas, and it is a risk factor for respiratory diseases such as TB, chronic obstructive pulmonary disease, pneumonia, and cancer.<sup>4</sup>

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### **Conflict of Interests**

The authors declare having no conflict of interests.

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# Sleep Apnea-Hypopnea Syndrome and Multiple Symmetrical Lipomatosis $\!\!\!\!\!^{\star}$

### Síndrome de apnea-hipopnea del sueño y lipomatosis simétrica múltiple

### Dear Editor,

We believe that the readers of your journal will find it interesting the association of sleep apnea-hypopnea syndrome (SAHS) with a rare disease such as multiple symmetric lipomatosis (MSL), Madelung's or Launois-Bensaude syndrome. Said association has been reported in several publications in recent years, and we ourselves have witnessed this combination of diseases in one of our patients (Fig. 1). MSL is an uncommon disease of unknown etiology that is characterized by the symmetrical accumulation of fat in the form of non-encapsulated subcutaneous lipomas in different locations, although especially in the neck and shoulders.<sup>1,2</sup> It is most frequent in males between the third and fifth decades of life, and it is frequently associated with alcoholism, chronic hepatopathy and neuropathy. It sometimes causes symptoms caused by the compression of neighboring structures such as the larynx. It can be associated with metabolic diseases and with atherogenic risk factors. Given the therapeutic implications, it should be differentiated from other rare adipose disorders. Diagnosis is usually symptoms-based, and when in doubt either computed tomography (CT) or magnetic resonance imaging (MRI) can be of help. MSL is a progressive disease with an infiltrating behavior. Diet control, alcohol abstinence and lymphatic drainage are recommended. In some cases, surgical reduction of the fat masses is the only treatment that may be effective and is aimed at improving esthetic and psychological consequences as well as treating airway or digestive tract obstruction. if necessary.

Very few cases have been reported of MSL associated with SAHS.<sup>2–6</sup> The presence of large fatty deposits in the cervical area would contribute to the narrowing of the upper airway and could interfere with the normal function of the pharyngeal muscles during sleep, favoring the appearance of SAHS. The frequent association of alcoholism and obesity would likewise increase the risk for SAHS. The diagnosis and treatment of SAHS in patients with MSL is especially important due to the increased risk for cardiovascular and metabolic diseases when both diseases are associated. Although cases have been published in which SAHS has improved after surgical treatment of MSL, when we take into account the limited long-term effectiveness of this therapeutic



**Fig. 1.** Symmetric accumulation of fatty tissue in the neck of a patient diagnosed with multiple symmetric lipomatosis.

option and the success of continuous positive airway pressure (CPAP) treatment in patients with MSL who present SAHS, it seems reasonable to indicate CPAP as a treatment of choice (together with standard diet and hygiene measures) as long as surgery is not indicated for another reason.

In conclusion, in patients diagnosed with MSL, SAHS should be suspected if there are symptoms indicating it, and CPAP treatment should be offered due to the good results obtained to date in published cases.

### **Conflict of Interests**

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