CASE REPORT

Bronchiolitis Obliterans Organizing Pneumonia and Bronchogenic Carcinoma Coexisting in Different Parts of the Lungs

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Bronchiolitis obliterans organizing pneumonia (BOOP) coexisting with nonsmall cell lung cancer in a separate part of the lungs is rare. We report the case of a man diagnosed with BOOP by video-assisted thoracoscopic lung biopsy; the patient also had adenocarcinoma in a different part of the lungs. The BOOP was treated with corticosteroids and the carcinoma was surgically resected. Outcome and clinical course were good after both procedures. The possibility of BOOP associated with bronchogenic carcinoma at a distant part of the lungs, although rare, must be taken into account in the differential diagnosis of radiographic alterations that may present in either of the 2 diseases.

Key words: Bronchiolitis obliterans organizing pneumonia. Bronchogenic carcinoma. Coexistencia de bronquiolitis obliterante con neumonía organizada y carcinoma broncogénico en zonas pulmonares distantes

La coexistencia en un paciente de una bronquiolitis obliterante con neumonía organizada (BONO) y un carcinoma pulmonar no microcítico en zonas pulmonares distantes es un hecho muy infrecuente. Presentamos el caso de un paciente con diagnóstico mediante biopsia pulmonar por videotoracoscopia de BONO, que tenía asociado en una zona pulmonar alejada un adenocarcinoma pulmonar. La BONO recibió tratamiento corticoideo y el carcinoma se resecó quirúrgicamente. Ambos procesos presentaron una buena evolución.

La posibilidad de que se presenten asociados una BONO y un carcinoma broncogénico en zonas pulmonares distantes, aunque muy infrecuente, deberá tenerse en cuenta en el diagnóstico diferencial de las alteraciones radiológicas que se puedan presentar en cualquiera de estas entidades patológicas.

Palabras clave: Bronquiolitis obliterante con neumonía organizada. Carcinoma broncogénico.

Introduction

Bronchiolitis obliterans organizing pneumonia (BOOP) is a clinicopathologic syndrome characterized by excessive proliferation of granulation tissue forming plugs in the small airways and alveolar lumina in association with a surrounding chronic inflammatory cell infiltrate.¹

Areas of BOOP can often be found in regions adjacent to a pulmonary neoplasm,² but bronchogenic carcinoma coexisting with BOOP in a different part of the lungs is exceptional, with few cases reported in the literature.³⁻⁵

We report the case of a man with a histological diagnosis of BOOP associated with lung cancer that was

surgically resected from another part of the lungs. The BOOP diagnosis was confirmed after examining tissue obtained by video-assisted thoracoscopic lung biopsy.

Case Description

The patient was a 60-year-old man with a history of high blood pressure and type 2 noninsulin-dependent diabetes. He was a former smoker of 35 pack-years who had quit 7 years earlier. The patient came to at the emergency department because of confusion and a 5-day fever (up to 40 °C). A chest x-ray showed an infiltrate in the left upper lobe, mainly in the lingula. The white blood cell count was 12 050/ μ L (89% neutrophils). He was diagnosed with community-acquired pneumonia and was hospitalized for 13 days. Treatment with clarithromycin and ceftriaxone was initiated. Sixteen days after discharge he was readmitted due to recurrence of fever without cough, expectoration, chest pain, or dyspnea. Fiberoptic bronchoscopy yielded no significant findings. Cytology of bronchial aspirate, bronchoalveolar lavage, and bronchial brushing was negative for neoplastic cells.

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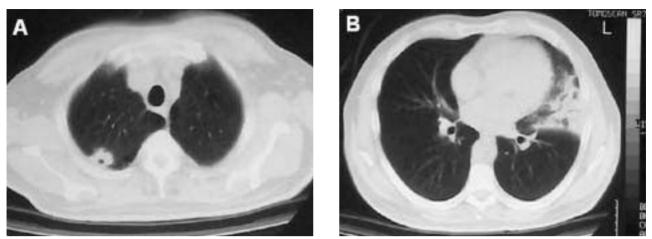


Figure 1. Cavitated lung nodule in the right upper lobe (A) and condensed pattern with air bronchogram in the lingula (B).

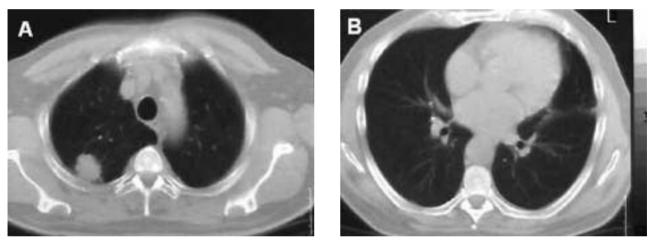


Figure 2. Slight increase in size of the nodule in the right upper lobe (A) and a marked decrease of the infiltrate in the left upper lobe after corticosteroid treatment (B).

Transbronchial biopsy could not be performed. Immunofluorescence for Pneumocystis carinii, auramine stain, standard and fungal cultures, and Löwenstein culture were negative. A diseased lymph node less than 1 cm in diameter in the right paratracheal region and nodes between 1 and 2 cm in the pre-carinal and pre-vascular spaces were observed by computed tomography. A 3×2.5 cm nodular image with polylobulated borders and central cavitation was noted in the posterior segment of the right upper lobe. An infiltrative pattern observed in the left upper lobe by air bronchogram was consistent with pneumonia (Figure 1). The diagnosis was slowly progressing pneumonia in the left upper lobe in a diabetic patient, with a cavitated nodule in the right upper lobe. After antibiotic treatment (ciprofloxacin and teicoplanin) the patient was asymptomatic, although the radiographic images persisted. Given the continued presence of lingular infiltrates and the negative results of diagnostic tests, it was decided to obtain a lung biopsy by video-assisted thoracoscopy in this region. Four lung parenchymal fragments between 0.8 and 1.8 cm were taken, and histology revealed BOOP-like alterations. Upon diagnosis, corticosteroid treatment (prednisone 40 mg/24 h) was initiated. At 2 months

a computed tomographic scan of the chest showed a decrease in the lingular infiltrate (including a decrease in the size of lymph nodes) and a slight increase in the size of the lesion in the right upper lobe (Figure 2). Transparietal needle aspiration was performed to sample the lesion, and cytology was negative for neoplastic cells. Surgical removal of the lesion was recommended. Atypical resection of the nodule in the right upper lobe by video-assisted thoracoscopy was started, but the lesion was seen to be nonsmall cell lung cancer, and the resection was completed by routine open right upper lobectomy and mediastinal lymphadenectomy. The definitive pathologic diagnosis indicated the pulmonary lobe was affected by a moderately differentiated 4 cm adenocarcinoma with a papillary pattern with micropapillary areas. The remaining part of the lobe had areas of interstitial fibrosis and BOOP-like lesions. The mediastinal nodes were uninvolved. The stage was IB (T2 N0 M0). Postoperative recovery was uneventful and the patient was discharged on day 6 after the intervention. He has attended scheduled follow-up outpatient examinations, has reported no persistence of respiratory problems, does not use corticosteroids, and had no sign of tumor recurrence 2 years after the operation.

Discussion

BOOP can be idiopathic and has been described in association with bacterial, viral, parasitic, and fungal infections, connective tissue diseases, drug toxicity, or toxic gas inhalation although cryptogenic types continue to be the most common forms.⁶ BOOP affects both sexes equally between 40 and 60 years of age. Clinical course is under 2 months, with flu-like signs and symptoms including fever, general malaise, and dyspnea.

Radiologic signs of BOOP include a broad spectrum of possibilities. Multifocal, bilateral consolidated patches, often migratory, can be seen on the chest x-ray and computed tomography scan, with solitary pulmonary nodules appearing less frequently.⁷ When patients have an associated solid tumor, BOOP usually presents radiologically as nodular or well-defined masses and with few clinical manifestations.³ However, in our patient BOOP manifested radiologically as a diffuse infiltrate and the clinical effects were very evident from the start.

Large lung biopsy samples should be obtained to establish a diagnosis of idiopathic BOOP and to rule out other possibilities with certainty. The procedure of choice has been lung biopsy by minithoracotomy or video-assisted thoracoscopy. Recently, however, a transbronchial biopsy showing intraluminal organization consistent with BOOP has also been accepted as adequate grounds for initiating corticosteroid treatment, given an appropriate clinical picture.⁸ However, reaching a diagnosis by this technique has limitations, as the sample (which will rarely include bronchioles) must be of good quality and it should be analyzed by an experienced pathologist.

Treatment consists of moderately high doses of corticosteroids (prednisone 1 mg/kg/day) for a long period of time. In cases of intolerance, low doses of macrolides have been used.³

Few studies describe BOOP in patients with neoplastic disease. The literature mainly refers to BOOP associated with osteogenic sarcoma and hematologic neoplasia in children and also in patients with allogenic bone marrow transplants or with breast cancer treated with radiotherapy.⁹ In the latter cases BOOP is usually found on the same side of the chest that received radiotherapy.

A pattern of organizing pneumonia adjacent to bronchogenic carcinoma is also a common finding of pathology.² Bronchogenic carcinoma coexisting with BOOP in topographically distant parts of the lungs, however, is exceptional. The case we report involved areas of BOOP not only in regions adjacent to the tumor but also in the contralateral lung. Only the latter alterations were apparent in diagnostic images, the areas of BOOP adjacent to the tumor becoming evident only upon pathologic examination of surgically excised tissue. Very few reports have referred to this association of BOOP and lung cancer in topographically distant parts of the lungs.³⁻⁵ One report described a series of cases in which BOOP was analyzed in patients diagnosed with underlying cancer. Eight patients with nonsmall cell lung cancer were in the series, but the authors did not specify whether the 2 diseases were concomitant or if BOOP had developed later, in irradiated lung zones for example. That study looked at the relation between type of underlying neoplastic disease and clinical course, radiologic findings, and prognosis, classifying patients with BOOP and cancer in 2 large groups: those who had solid organ tumors (who have a better prognosis, as was the case for our patient) and those with hematologic neoplasias (whose clinical course is less favorable).³

In conclusion we can say that whenever possible a diagnosis of BOOP should be established with a tissue sample obtained by open lung biopsy or video-assisted thoracoscopy because of the therapeutic implications of the diagnosis. The possibility of underlying neoplasia should be considered if abnormal radiologic images persist in a patient with correctly treated BOOP as this association, although rare, may occur and should not remain undiagnosed. Likewise, it should be taken into account that the nodules, masses, or infiltrates that can appear in follow-up radiographs of patients with lung cancer and that are usually attributed to infections or disease progression may instead be BOOP. BOOP, which can simulate any of the aforementioned processes, should therefore be considered in the differential diagnosis. With appropriate treatment, the prognosis is good.

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