Cavitary Bronchiolitis Obliterans With Organizing Pneumonia

Emel Ceylan, MD[1]; Oğuz Kılınç, MD[1]; Neşe Topal, MD[1]; Atila Akkoçlu, MD[1]; M. Şerafettin Canda, MD[2]

[1]Department of Pulmonary Medicine, Dokuz Eylül University Faculty of Medicine, İzmir, Turkey
[2]Department of Pathology, Dokuz Eylül University Faculty of Medicine, İzmir, Turkey

Abstract

A 34 -year-old man was admitted to the hospital because of chronic cough. Pulmonary radiogram showed a cavitary coin lesion and he underwent surgical excision. Histologically bronchiolitis obliterans organizing pneumonia (BOOP) was reported but no etiology is found. After excision, bilateral pulmonary micronodules which had been previously seen on the computer tomography regressed spontaneously. In the differential diagnosis of cavitary solitary lesions, BOOP is a rare entity. The case is discussed with a review of the literature.

Turkish Respiratory Journal, 2004;5:(3):183-6

Keywords: *bronchiolitis obliterans organizing pneumonia, cavitary pulmonary lesion, open lung biopsy*

Introduction

Bronchiolitis obliterans with organizing pneumonia (BOOP) was first described by Lange in 1901; but it was called BOOP by Epler in 1985 and popularized as cryptogenic organized pneumonia. The characteristic histopathological lesion is defined as granulation tissue plugs within the lumine of small airways, sometimes with complete obstruction of small airways, and granulation tissue extending into alveolar ducts and alveoli (1,2,3).

Case Report

A 40-year-old man was admitted to the hospital because of a nonproductive cough for a month, worse at night. It addition, he complained of night and day sweating, subjective fever, and intermittent bad breath. He had a history of contact with a patient having tuberculosis in his workplace about two months ago. Medical history consisted of an operation for appendicitis 14 year ago, using antibiotics frequently for upper respiratory tract infection in winter months for the last ten years and otitis media five years ago. He also said he had a decayed tooth. He had a history of 25 pack-year cigarette smoking and had a budgerigar at home for two years. There was no pathological lymphadenopathy on physical examination. Rales were heard at the right middle region of the lungs, the examination findings being otherwise unremarkable.

Corresponding Author: *Dr. Emel Ceylan*
Dokuz Eylül Üniversitesi
Göğüs Hastalıkları AD
İnciraltı-İzmir, Türkiye
Phone&Fax: +90 (232) 278 33 73
E-mail : ceylanemel@hotmail.com.tr

Sinus radiograph and tomography showed mucosal hypertrophy of the right maxillary sinus and left orbital region. A chest X-ray showed a thin-walled cavity partly thick at the bottom in the right middle zone (Figure 1).

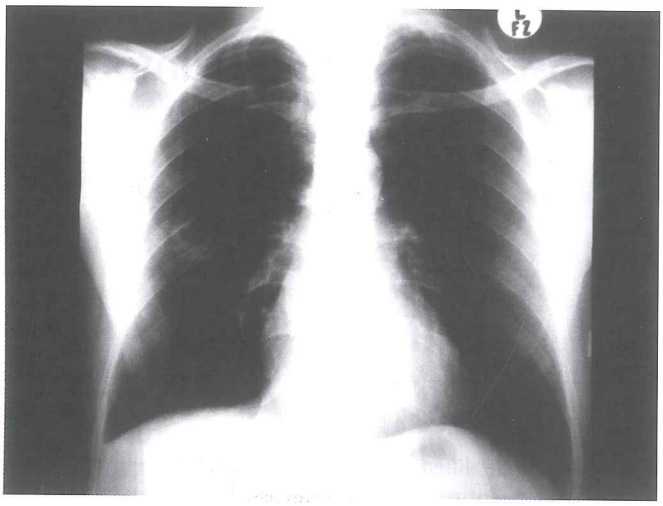

Figure 1. *Chest radiograph on admission showing a cavity in the right middle zone.*

On hospital admission, the patient looked well, with a temperature of 36.5°C and a regular pulse rate of 86 beats/minute. Blood pressure was 100-660 mmHg and the respiratory rate was 22 breaths/minute. Hemoglobin was 12.9 g/dl, leucocyte count was 11600 cells/µl and the erythrocyte sedimentation rate (ESR) was 14 mm/h. All other routine laboratory test results, including liver and renal function tests, and serum proteins, were normal. The result of purified protein derivative skin test was positive.

A chest scan with computed tomography of the thorax revealed lymph nodes below the pathological threshold for significance in the mediastinum. Additionally, parenchymal scarring and fibrotic bands were demonstrated at both apical zones on lung settings. A cavitary lesion, 2.5 cm diameter with irregular margins was seen in the right lower lobe superior segment accompained by soft, irregular and lower density nodulary lesions which appeared to be in the left lower lobe segments (Figure 2 and 3).

The bronchial mucosa of the right lower lobe was oedematous and fragile on bronchoscopy. The microscopic examination of the bronchial washings and sputum for acid-fast bacili was negative as was culture.

The serological test for *Mycoplasma* was negative. Antinuclear antibodies (ANA) and antineutrophil cytoplasmic antibodies (ANCA's) were not found. The serum level of Rheumatoid factor (RF) and CRP (serum reative protein) were normal. Elisa Ig G for hydatid cyst disease was also negative. There were no positive findings for hepatic B, C, or HIV on serological tests. Pulmonary function tests and diffusion capacity were normal.

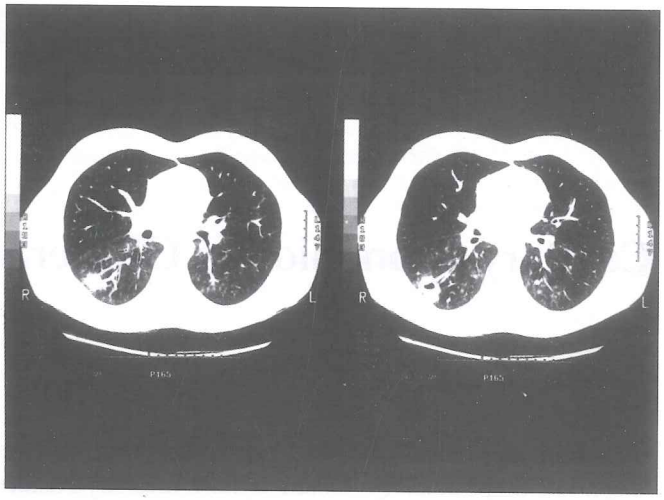

Figure 2. *Computed tomographic scan of the thorax showing a nodulary lesion with a diameter of 2.5 cm, in which its central part has a cavity, at the superior segment of the right lower lobe.*

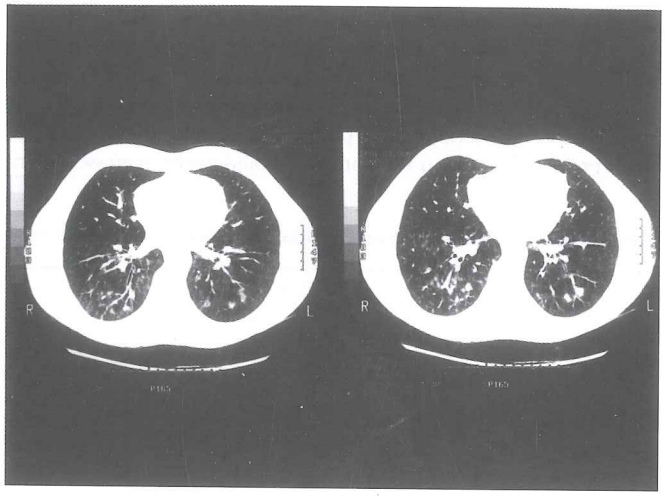

Figure 3. *Computed tomography of the lower lobes of the thorax showing irregular acinary and nodular densities bilaterally.*

Open lung biopsy was performed on the right because the etiology of the cavity was unknown and the possibility of pulmonary malignant disease remained. No further investigations were undertaken, as no malignant pathology was present on the frozen material. A wedge resection was performed. The excised wedge measured 4.5x4x3 cm and was covered by visceral pleura. In serial sections, there were multiple irregular gray-white areas, suggesting an interstitial lung disease. We also noted a 2 cm cavity which was filled with partially mucoid material. Histopathological examination revealed endobronchiolar and intra-alveolar polypoidal organizing connective tissue in patchy areas of the lung parenchyma (Figure 4). It was observed that alveolar septa were thickened by chronic inflammatory infiltration in some areas, as well as focal type II pneumocyte proliferation. On microscopic examination for samples of the cavity, we saw large ulcerated areas covered with a fibrinous exudate on the

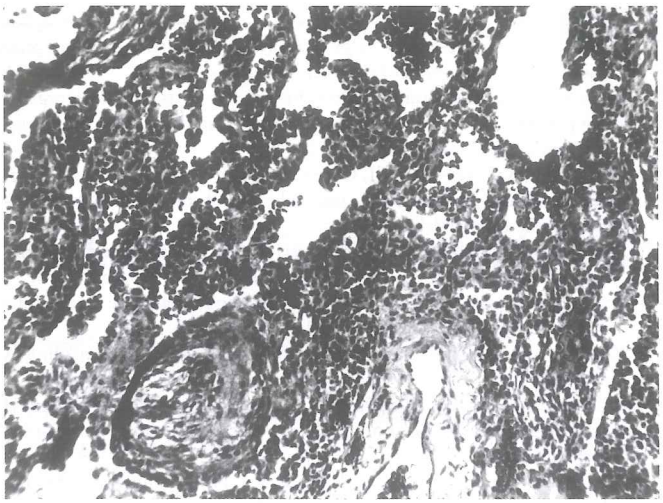

Figure 4. *Open lung biopsy. Luminal obstruction, inflammatory infiltration of the bronchiole and the surrounding interstitium (H&E Stain x 100).*

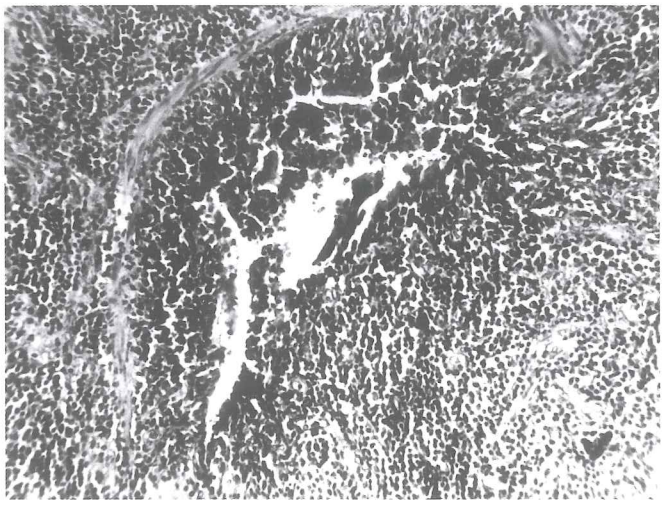

Figure 5. *Granulation tissue plug extending into alveolus (H&E Stain x 100).*

surface. The cavity was thought to be a bronchiol, since there weren't any bronchial cartilage surrounding it (Figure 5). The diagnosis of BOOP was made.

Discussion

Bronchiolitis obliterans with organizing pneumonia is defined as pulmonary inflammatory response against minimal alveolar and bronchial damage. It can be associated with pulmonary infections, tumors, connective tissue diseases, hematologic malignancies (such as lymphoma), vasculitis and hypersensitive reactions or may be idiopathic. Tuberculosis has the first place in the differential diagnosis of our case, because the pathological lesion was in the superior segment of the inferior lobe, where tuberculosis is seen most often. Additional examinations made for other pathologies, including connective tissue disorders, didn't lead us to the diagnosis (2,3,4).

Altough typical pattern of multiple alveolar patchy infiltration which are migratory, and bilateral micronodular opacities in the inferior zones are known, Cordier et al, distinguished three characteristic clinical and imaging profiles in patients with idiopathic BOOP; multiple patchy pneumonia, solitary pneumonia, and diffuse interstitial lung disease. In that study, the pulmonary tissue examined pathologically had been obtained by open lung biopsy in ten cases out of sixteen. In five of these ten cases, surgical excision was performed because of solitary pulmonary lesions and the patients recovered (5). Lung tumour couldn't be ruled out in view of the smoking history, and in our case surgical excision was performed too.

In the literature, idiopathic cases with unusual radiologic patterns are also reported. These include, solitary (6) or bilateral (7) cavitary nodules. The study of Epler described cavitary BOOP in two of 50 patient with idiopathic BOOP. In the series of Epler et al, cavitary lesions and effusions are detected in 5% of patients with BOOP (1,8). Besides a coin lesion there were micronodular alveolar infiltrates in the left lower lobe and around the cavitary lesion in the present case.

Idiopathic cavitary BOOP is not a common entity. Cavities in solitary masses or nodular lesions probably occur after a preceeding infection has caused inflammatory injury and focal areas of necrosis in the lung. As a mechanism of cavitation, the possibility exists that some cavities are secondary to a pneumatocele, or they are preexisting bullous cavities rendered visible by the presence of organizing pneumonia in the adjacent parenchyma (9). Furthermore, these can enlarge the cavity and make it visible by means of a check-valve mechanism as a result of partial small airway obstruction. The possibility of immunological disorders must be kept in mind when multiple cavities are together with the clinical and biological appearence of inflammatory injury. In this situation, BOOP can be a manifestation of covered immunological disorders (9). Safadi et al, reported a case who was diagnosed as having BOOP with percutaneous transthoracic CT guided needle biopsy there was an initial response to steroid therapy. After six months, progression was observed in the pulmonary lesion. Non-Hodgkin's lymphoma was diagnosed in this case when open lung biopsy was subsequently performed because of impairment in the response to steroid therapy and radiological progression (4). In our patient, before the operation, BOOP was not prominent in the differential diagnosis. That's why TBLB was not applied. Detecting BOOP at TBLB should not be accepted as a definitive diagnosis because of other pathologies may be accompained by BOOP and the case would still required a resection procedure.

Haro et al reported a case with bilaterally multiple cavitary nodules whose etiology couldn't be determined and who showed good respond to prednisolone therapy (10). Heller et al. reported two cases with multiple cavitary lesions whose etiologies couldn't be determined. One of these cases showed a good response to six-month prednisolone therapy, but with eventual clinical and radiological progression (11).

We have reported a case of BOOP diagnosed histologically, which manifested as an uncommon cause of a solitary cavitary lesion. In the differential diagnosis of such lesions as determined on chest radiography, it is important to rule out this entity because it has a good response to steroid therapy. In our case after surgical excision of the cavity, regression occured in the other lesions even without therapy. The case is accepted as "idiopathic" because, with the available diagnostic modalities, no infectious and non-infectious cause was established.

References

1. Epled RG, Colby TV, Mc Loud TC, et al. Bronchiolitis obliterans organizing pneumonia. N Engl J Med 1985;312:152-8.
2. Kitaichi M. Differential diagnosis of bronchiolitis obliterans organizing pneumonia. Chest 1992;102(Suppl):44S-49S.
3. Costabel U, Teschler H, Schoenfeld B, et al. BOOP in Europe. Chest 1992;102(Suppl):14S-20S.
4. Safadi R, Berkman N, Haviv YS, et al. Primary non-Hodgkin's lymphoma of the lung presenting as bronchiolitis obliterans organizing pneumonia. Leuk Lymphoma 1997;28:209-13.
5. Cordier JF, Loire R, Brune J. Idiopathic bronchiolitis obliterans organizing pneumonia: definition of characteristic clinical profiles in a series of 16 patients. Chest 1989;96:999-1004.
6. Domingo JA, Perez-Calvo JI, Carretero JA, et al. Bronchiolitis obliterans organizing pneumonia: an unusual cause of solitary pulmonary nodule. Chest 1993;103:1621-3.
7. Froudarakis M, Bouros D, Loire R, et al. BOOP presenting with hemoptysis and multiple cavitary nodules. Eur Respir J 1995;8:1972-4.
8. Epler RG. Bronchiolitis Obliterans Organizing Pneumonia: Definitions and clinical features. Chest 1992;102(Suppl):2S-6S.
9. Cordier JF. Cavitary bronchiolitis obliterans organizing pneumonia [editorial]. Eur Respir J 1995;8:1822-3.
10. Haro M, Vizcaya A, Texido X, et al. Idiopathic bronchiolitis obliterans organizing pneumonia with multiple cavitary lung nodules. Eur Respir J 1995;8:1975-7.
11. Heller I, Biner S, Isakov A, et al. TB or Not TB. Cavitary bronchiolitis obliterans organizing pneumonia mimicking pulmonary tuberculosis. Chest 2001;120:674-8.