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Case report

Development of pulmonary alveolar proteinosis following exposure to dust after the Great East Japan Earthquake

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ABSTRACT

We report a unique case of pulmonary alveolar proteinosis that developed 3 weeks after the Great East Japan Earthquake and the subsequent tsunami. The patient had inhaled dust repeatedly while visiting her devastated neighborhood without wearing a protective mask. Five weeks after the earthquake, lung samples taken from the patient showed foreign particle deposition; however, her serum was negative for GM-CSF autoantibody. The patient's clinical symptoms resolved following whole lung lavage. We conclude that inhalation of fine dust particles after natural disasters may cause the onset of pulmonary alveolar proteinosis.

1. Introduction

The Great East Japan Earthquake deeply impacted respiratory health care in the affected areas. During the acute phase of the disaster, in addition to an increase in the number of patients with "tsunami lung" caused by near-drowning, there was also an increase in the number of patients with common respiratory diseases such as pneumonia and acute exacerbations of COPD and bronchial asthma [1]. During the sub-acute phase of the disaster, patients presented with

Abbreviations: PAP, pulmonary alveolar proteinosis; GM-CSF, granulocyte macrophage colony-stimulating factor; COPD, chronic obstructive pulmonary disease; CT, computed tomography; HRCT, high-resolution computed tomography; BALF, bronchoalveolar lavage fluid; KL-6, Krebs von den Lungen-6; SP-D, surfactant protein D; CEA, carcinobembryonic antigen; TBLB, transbronchial lung biopsy

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allergic lung inflammation, including hypersensitivity pneumonitis and organizing pneumonia. These respiratory conditions occurred not only in the victims of the tsunami but also in the workers engaged to clean up the debris, which contained rubble from buildings and industrial waste material from the sea [2].

Previous studies have shown that dust exposure may be related to the pathogenesis of PAP. Exposure to dust has been reported as a cause of secondary PAP, in which serum GM-CSF autoantibodies are considered negative, but this has not been fully confirmed [3]. Further, recent studies have raised the hypothesis that an inhaled agent may instead be the trigger of the development of autoimmune PAP, characterized by positive GM-CSF autoantibodies [4]. To our knowledge, no increase in the incidence of PAP after natural disasters or the World Trade Center attacks has been reported [5]. However, specific materials contained in the debris from disasters can induce PAP

Here, we report a case of PAP that developed after exposure to dust following the Great East Japan Earthquake.

2. Case presentation

A 63-year-old Japanese woman was referred to our institute for worsening lung infiltrates, dyspnea, and hypoxia. She had never smoked and had a past history of hypertension.

Although she was not otherwise injured her house was completely destroyed in the large tsunami triggered by the Great East Japan Earthquake on March 11 of 2011. After the earthquake, she repeatedly retrieved personal effects from the rubble without wearing a mask. Since large amounts of sludge and burned embers were scattered throughout the area, she was exposed to various kinds of inhaled dust. Three weeks after the earthquake, she developed dry cough and her chest X-ray showed bilateral reticular shadows (Fig. 1Aa). The computed tomography (CT) of her chest showed diffusely-distributed ground glass opacity in the subpleural area (Fig. 1Ab). At the previous hospital, analysis of her bronchoalveolar lavage fluid (BALF) revealed lymphocytosis (lymphocytes: 89.0%, CD4/CD8: 3.6) without turbidity and a transbronchial lung biopsy (TBLB) did not indicate PAP. However, upon re-evaluation, we detected particles within the lung (Fig. 2A), and an electron probe X-ray microanalysis revealed the deposition of silicon, oxygen, and aluminum, while other specific elements were not detected (data not shown). On clinical suspicion of idiopathic interstitial pneumonias, she was treated with prednisolone, cyclosporin, and methylprednisolone pulse therapy. Eight months later, she was referred to our University Hospital as the treatment was not fully effective.

On admission, her blood pressure was 137/98 mm Hg; pulse, 105 beats/min; and body temperature, 36.8 °C. Chest examination revealed slight bilateral inspiratory crackles, and a chest X-ray showed a significant loss of lung volume (Fig. 1Ba). High-resolution computed tomography (HRCT) of the chest showed diffuse ground-glass opacities with superimposed interlobular septal thickening and intralobular lines (Fig. 1Bb). Five weeks after admission, pulmonary function testing showed that the patient had a severe, restrictive pattern (vital capacity, 1.17L; 52.7% predicted) with reduced

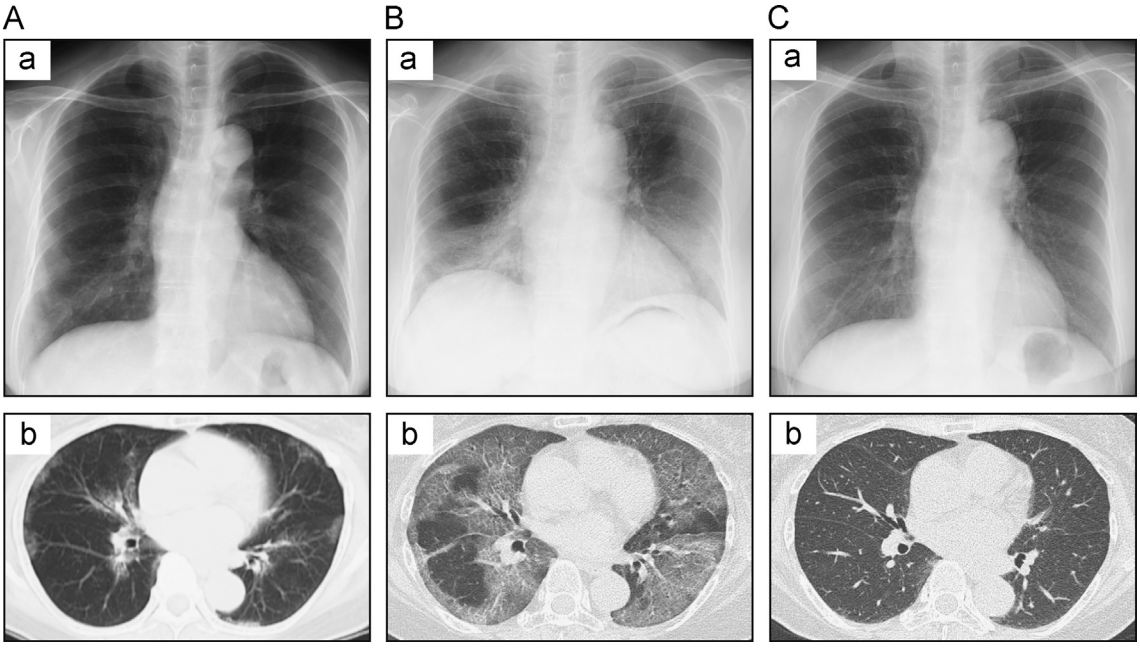

Fig. 1 – X-ray (a) and computed tomography (b) of the chest (A) at the initial visit in the previous hospital (April 2011), showing limited ground glass opacity in the subpleural area; (B) at 3 weeks after admission to our hospital (January 2012), showing significant loss of lung volume and a wide range of ground-glass opacities with superimposed interlobular septal thickening and intralobular lines; (C) at 6 months after the whole lung lavage (August 2012), showing improvements in the lung volume and ground glass opacities.