Bird Fancier’s Lung Which Developed in a Pigeon Breeder Presenting Organizing Pneumonia

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Abstract

Bird fancier’s lung (BFL) is one of the most common types of hypersensitivity pneumonitis. We report a rare case of acute-on-chronic bird fancier’s lung that developed in a pigeon breeder and presented subpleural curvilinear shadow and ground glass opacity on high-resolution computed tomography (HRCT) of the chest. The results of surgical lung biopsy showed mainly intraalveolar organization and alveolitis in addition to the pattern of usual interstitial pneumonia with centrilobular fibrosis. Examination of bronchoalveolar lavage (BAL) fluid revealed an increase in lymphocytes. The results of immunoglobulin (Ig) G and IgA antibodies against pigeon dropping extracts were positive in sera and BAL fluid. Consequently, the patient was diagnosed as having BFL. Avoidance of pigeons and corticosteroid therapy led to rapid improvement.

Key words: hypersensitivity pneumonitis, bird fancier’s lung, pigeon, reversed halo sign, organizing pneumonia


Introduction

Hypersensitivity pneumonitis is an immunologically-mediated lung disease caused by inhalation of a variety of environmental agents, which is manifested by alveolitis with lymphocytic infiltration and the presence of poorly defined non-caseous epithelioid cell granulomas in the peripheral airways (1). Bird fancier’s lung (BFL) is observed in individuals who develop a hypersensitivity response to avian droppings and antigens in feathers (2). The characteristic findings of computed tomography (CT) of the chest in BFL have been reported to be ground glass opacity (GGO), diffuse centrilobular small nodules, and consolidations in the bilateral lungs (3). Furthermore, Ohtani et al (4) reported a few BFL patients presenting with BOOP-like lesions.

We report a patient with acute-on-chronic BFL presenting with organizing pneumonia whose condition deteriorated following intermittent exposure to a large amount of pigeon droppings after his condition had remained unchanged over a period of several decades.

Case Report

A 70-year-old man was admitted to our hospital with the complaint of progressive dyspnea on exertion. He had a smoking history of 20 cigarettes per day for 37 years. He had been breeding approximately 150 racing pigeons on the top of an apartment building next to his house for more than 30 years, and cleaned coops full of pigeon droppings every 2 or 4 weeks. He was exposed to a large amount of pigeon droppings by taking a longer than usual time cleaning coops one week prior to admission at our hospital.

Chest auscultation revealed fine crackles in the bilateral lung bases. Laboratory data on admission showed high levels of KL-6 (1,510 U/mL) and SP-D (149 ng/mL). Results
of arterial blood gas analysis were pH, 7.39; PaCO₂, 38.6 Torr; and PaO₂, 70.3 Torr on room air. The pulmonary function test revealed restrictive impairment (VC, 2.13 L, 66.1% of predicted) with decreased diffusing capacity (DLCO, 9.63 mL/min/mmHg, 53.7% of predicted). Chest X-ray showed infiltrative shadows and GGO in the right middle and lower lung fields and in the left lower lung field (Fig. 1A). Chest CT revealed GGO and consolidation with a subpleural curvilinear shadow in the bilateral lower lobes (Fig. 1B), central GGO and surrounding air-space consolidation of ring shapes, the so-called reversed halo sign, in the left lower lobe (Fig. 1C). Examination of bronchoalveolar lavage (BAL) fluid revealed alveolar macrophages, 23%; lymphocytes, 67%; neutrophils, 6%, and eosinophils, 4%. Total cells were increased, with a low CD4/CD8 ratio, 0.34. Cultures of sputum and BAL fluid were negative for fungal, bacterial, or mycobacterial pathogens. The lung biopsy specimens of the right S'a and S'b obtained by video-assisted thoracic surgery (VATS) revealed peribronchiolar and intraalveolar organization with alveolitis with infiltration of lymphocytes corresponding to GGO, and interlobular septal and subpleural fibrosis (usual interstitial pneumonia; UIP pattern) with centrilobular fibrosis corresponding to subpleural curvilinear shadow and reticulonodular lesions on chest high-resolution CT (HRCT). No granulomas were identified (Fig. 2, 3). Immunoglobulin (Ig) G and IgA antibodies against pigeon dropping extracts (PDE) were positive in sera and BAL fluid. Positive antigen-induced lymphocyte proliferation was observed with a stimulation index of 4.42 (positive ≥2.0). Consequently, the patient was diagnosed as having acute-on-chronic pigeon fancier’s lung. The patient received 1 g/day of intravenous methylprednisolone (mPSL) for 3 days, followed by oral prednisolone (PSL) at 30 mg/day. After these initial corticosteroid treatments, GGO and the subpleural curvilinear shadow in both lungs were immediately resolved (Fig. 4). However, the subpleural reticulonodular lesions remained unchanged. Subsequently, the dose of PSL was gradually reduced and discontinued 6 months later. No relapse has been observed for 2 years after avoidance of contact with pigeons.

Discussion

Hypersensitivity pneumonitis is an immunologically-mediated lung disease caused by inhalation of a variety of environmental agents, which is manifested by alveolitis with lymphocytic infiltration and the presence of poorly defined noncaseating granulomas in the peripheral airways (2). BFL is probably one of the most common types of hypersensitivity pneumonitis and is mainly described among pigeon and budgerigar fanciers (3). The prevalence of BFL ranges from 8% to 30% among pigeon keepers belonging to associations of pigeon fanciers (5). Classically, the clinical presentation has been divided into acute, subacute, and chronic forms depending on the amount of inhaled antigen and repeated exposure (2). Continual exposure may cause an overlapping of these phases. In the present case, we assumed that the CT-pathologic features revealed the overlapping of the acute phase such as organizing pneumonia with alveolitis with the chronic phase indicated by the UIP pattern with centrilobular fibrosis. However, the area of intraluminal organization was very limited in the histopathological specimens of the right S'a. This was thought to be due to the fact that these lung specimens did not include a sufficient amount of lesions corresponding to GGO on the chest HRCT. Also, subpleural reticulonodular lesions in the bilateral lower lobes remained unchanged even with the administration of therapy while GGO and the subpleural curvilinear shadow were immediately resolved after treatment with corticosteroids, indi-
insidious onset with an acute event at this time. Sessions had an insidious onset. However, the present case was recurrent acute episodes, whereas patients with UIP-like lesions, and patients with BOOP-like lesions had histologically diagnosed as BOOP-like lesions, 11 patients as reported by Morell et al (4) reported that 2 of 26 BFL patients were his-
teresting an overlapping of the acute with the chronic phase. Ohtani et al (4) reported that 2 of 26 BFL patients were his-
tologically diagnosed as BOOP-like lesions, 11 patients as UIP-like lesions, and patients with BOOP-like lesions had recurrent acute episodes, whereas patients with UIP-like lesions had an insidious onset. However, the present case was insidious onset with an acute event at this time.

As reported by Morell et al (3), the chest X-ray of patients with BFL showed a predominance of an interstitial pattern in 79% of patients, mainly of the reticular type, with the most frequent chest CT findings being GGO, a mosaic appearance, and/or fine centrilobular nodules. Moreover, in the subacute stage of BFL, Remy-Jardin et al (6) described chest CT findings of diffuse micronodules, focal air trapping

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**Figure 2.** A: Chest HRCT showing GGO and subpleural curvilinear shadow in the right middle lobe. B: Low magnified microscopic appearances corresponding to the right S4a on chest HRCT indicated by circular mark. (Elastic van Gieson stain) (scale bar=1 mm). C: Microscopic appearance of the area indicated by square mark in Fig. 2B shows peribronchial and relatively-widespread intraluminal organization and alveolitis with infiltration of lymphocytes. (Hematoxylin and Eosin staining) (scale bar=250 μm).

**Figure 3.** A: Chest HRCT showing GGO with subpleural curvilinear shadow and reticulonodular lesions in the right lower lobe. B: Low magnified microscopic appearances corresponding to the right S9b on chest HRCT indicated by circular mark. (Elastic van Gieson stain) (scale bar=1 mm). C: Microscopic appearance of the area indicated by square mark in Fig. 3B shows not only interlobular septal and subpleural fibrosis but also centrilobular fibrosis, in addition to scattered intraluminal organization. (Elastic van Gieson stain) (scale bar=100 μm).
or emphysema, and mild fibrotic changes with normal lung volume but impaired diffusing capacity and a predominant lymphocytic alveolitis. To our knowledge, there is no previous description of BFL presenting with a reversed halo sign on CT. The present case showed a reversed halo sign partially in the left lower lobe. However, the reversed halo sign on chest HRCT was understood as relatively specific images to suggest the diagnosis of COP (7, 8). We found a case report described as BFL presenting with a BOOP-like reaction in detail (9). That case was diagnosed as having subacute BFL, with multiple nodules surrounded by a halo of ground-glass attenuation on chest HRCT that histologically consisted of extensive intraluminal granulation tissue surrounded by alveolitis with non-caseous epithelioid cell granulomas. The present case had several of the following characteristics in common with those of BOOP: i) the main histological feature of the disease was organizing pneumonia; ii) the response to corticosteroid therapy was extremely good; and iii) no relapse has been observed after short-term corticosteroid therapy and the avoidance of contact with pigeons.

In conclusion, this is an extremely rare case with acute-on-chronic BFL presenting with biopsy-proven organizing pneumonia that is insidious onset with acute deterioration.

Acknowledgement
The authors would like to thank Dr. Y. Yoshizawa (Tokyo Medical and Dental University Hospital) for measuring IgG and IgA antibodies against pigeon and budgerigar dropping extracts in sera and BAL fluid, and a positive antigen-induced lymphocyte proliferation.

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