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Case report

An autopsy case of bird-related chronic hypersensitivity pneumonitis presenting with repeated acute exacerbation

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ABSTRACT

A 68-year-old woman was admitted to our hospital with a dry cough in 2010. Chest computed tomography showed the appearance of a nonspecific interstitial pneumonia (NSIP) pattern. Video-assisted thoracoscopic surgery (VATS) was performed, and the specimens prominently showed a usual interstitial pneumonia (UIP) pattern. She was diagnosed with bird-related chronic hypersensitivity pneumonitis (BRCHP) on the basis of the detection of antibodies to pigeon dropping extract in her serum and a history of using feather-filled duvets and indirect exposure to birds in her living environment. Even though she was treated with corticosteroids and immunosuppressants and recommended to avoid bird-related antigens, she had a progressive course with repeated acute exacerbation episodes and died of respiratory failure. The autopsy findings showed diffuse alveolar damage superimposed on UIP. Clinicians should be aware that BRCHP patients especially with histopathologically UIP pattern may experience acute exacerbation.

1. Introduction

Hypersensitivity pneumonitis (HP) is an interstitial lung disease caused by the inhalation of a wide variety of antigens. Environmental avian antigens, such as pigeon serum, droppings, and feathers, can induce hypersensitivity pneumonitis called bird-related hypersensitivity pneumonitis, which is one of the most common variants of HP in Japan [1]. Bird-related chronic HP (BRCHP) clinically mimics idiopathic interstitial pneumonias. Here we describe an autopsy case of BRCHP presenting with repeated acute exacerbation.

2. Case report

A 68-year-old woman was admitted to our hospital with a persistent dry cough in 2010. A previous chest X-ray performed at another hospital showed interstitial densities in 2005. She had never smoked, and she used duvets stuffed with pheasant feathers. Oxygen saturation on room air while she was lying was 96%, which dropped to 88% after 6 min of walking. A physical examination revealed fine crackles in both lower lung fields. The serum levels of sialylated carbohydrate antigen KL-6 (KL-6) and pulmonary surfactant protein D (SP-D) were elevated (1111 U/mL, normal, < 500 U/mL and 294 ng/mL, normal, < 110 ng/mL, respectively; Table 1). A chest X-ray showed reticular shadows in both lung fields (Fig. 1). Chest computed tomography (CT) images revealed diffuse reticulation admixed with ground glass opacity without honeycombing, as well as prominent traction bronchiectasis in the right upper lobe (Fig. 2). The cell fractionation of bronchoalveolar lavage was normal. Transbronchial lung biopsy (TBLB) revealed an infiltration of inflammatory cells, predominantly lymphocytes as well as moderate alveolar septal fibrosis.

Based on the above findings, video-assisted thoracoscopic surgery (VATS) was performed, with chronic HP and idiopathic interstitial pneumonia considered as a differential diagnosis. Histologic sections of right upper lobe showed the peripheral and subpleural distribution of the fibrosis, architectural distortion with a microscopic honeycombing, and fibroblastic foci. Granuloma was not observed. These findings indicated the diagnosis of a usual interstitial pneumonia. (Fig. 3A and B). BRCHP was suspected from her environmental history. Serum antibodies to pigeon dropping extract (PDE) were detected (PDE IgG, 0.425 μg/mL, cut-off, 0.36 μg/mL; PDE IgA, 0.583 μg/mL, cut-off, 0.15 μg/

Abbreviations: NSIP, Nonspecific interstitial pneumonia; VATS, video-assisted thoracoscopic surgery; UIP, usual interstitial pneumonia; BRCHP, bird-related chronic hypersensitivity pneumonitis; HP, hypersensitivity pneumonitis; SP-D, surfactant protein D; TBLB, transbronchial lung biopsy; PDE, pigeon dropping extract

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Table 1
Laboratory findings on initial admission.

Hematology		anti-SS-A Ab	(-)
WBC	7790/μl	anti-SS-B Ab	(-)
Neutrophils	61.5%	anti-Scl-70 Ab	(-)
Lymphocytes	30.8%	anti-Jo-1 Ab	(-)
Monocytes	5.4%	anti-RNP Ab	(-)
Eosinophils	1.9%	Anti-CENP-B Ab	(-)
Basophils	0.4%	anti-ds-DNA Ab	(-)
Hb	13.5 g/dl	MPO-ANCA	(-)
Ht	42.7%	IgG	1670 mg/dl
Plt	11.7 10^4/μl	ACE	13.5 IU/l
Biochemistry		sIL-2R	616 U/ml
TP	7.3 g/dl	KL-6	1111 U/ml
Alb	3.3 g/dl	SP-D	294 ng/ml
T-Bil	1.0 mg/dl	Blood gas analysis (room air)	
AST	24 IU/L	pH	7.42
ALT	27 IU/L	$PaCO_2$	37.8 Torr
LDH	227 IU/L	PaO_2	84.4 Torr
ALP	586 IU/L	HCO_3-	24 mmol/l
γ-GTP	64 IU/L	BE	0.3 mmol/l
AMY	10.6 IU/L	Pulmonary function test	
CK	88 IU/L	VC	1.43 L
BUN	14 mg/dl	%VC	64.1%
Cre	0.54 mg/dl	FEV_1	1.17 L
Na	138 mEq/l	FEV_1%	83.5%
K	4.0 mEq/l	$\%DL_{CO}$	46.5%
Cl	106 mEq/l		
Glu	172 mg/dl	BALF analysis	
HbA1c	6.9%	Macrophages	86.7%
Serology		Neutrophils	7.8%
CRP	0.12 mg/dl	Eosinophils	0.2%
ANA	< 40	Lymphocytes	5.3%
RF	9	CD4/CD8	1.06

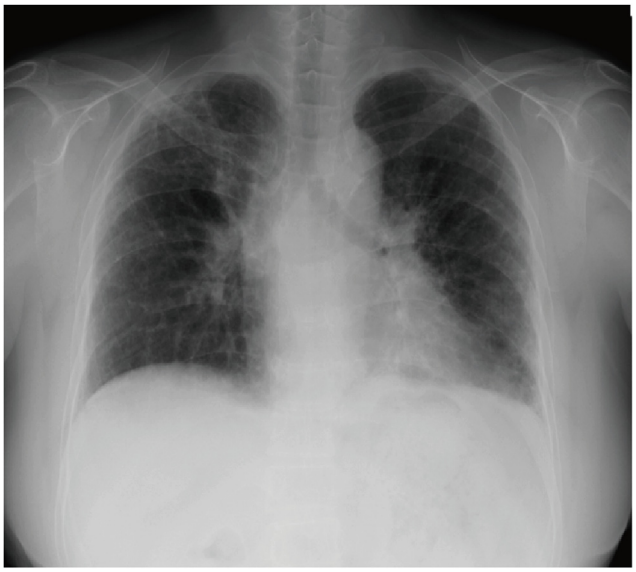

Fig. 1. Chest radiograph on first admission.

mL), while Trichosporon asahii antibodies were not detected; she was diagnosed with BRCHP. Following our recommendation to move to another house and to stop using feather-filled duvets for antigen avoidance, she moved to her daughter's house intermittently. Her symptoms of worsening dry cough, general malaise, and anorexia, which she experienced when at her house, gradually improved when staying at her daughter's house.

In August 2011, she was readmitted to our hospital due to exertional dyspnea with the deterioration of ground glass opacity in bilateral lung fields and desaturation. She was diagnosed with acute exacerbation of BRCHP and administered 10 mg/day prednisolone and 150 mg/day cyclosporine A and discharged on home oxygen therapy.

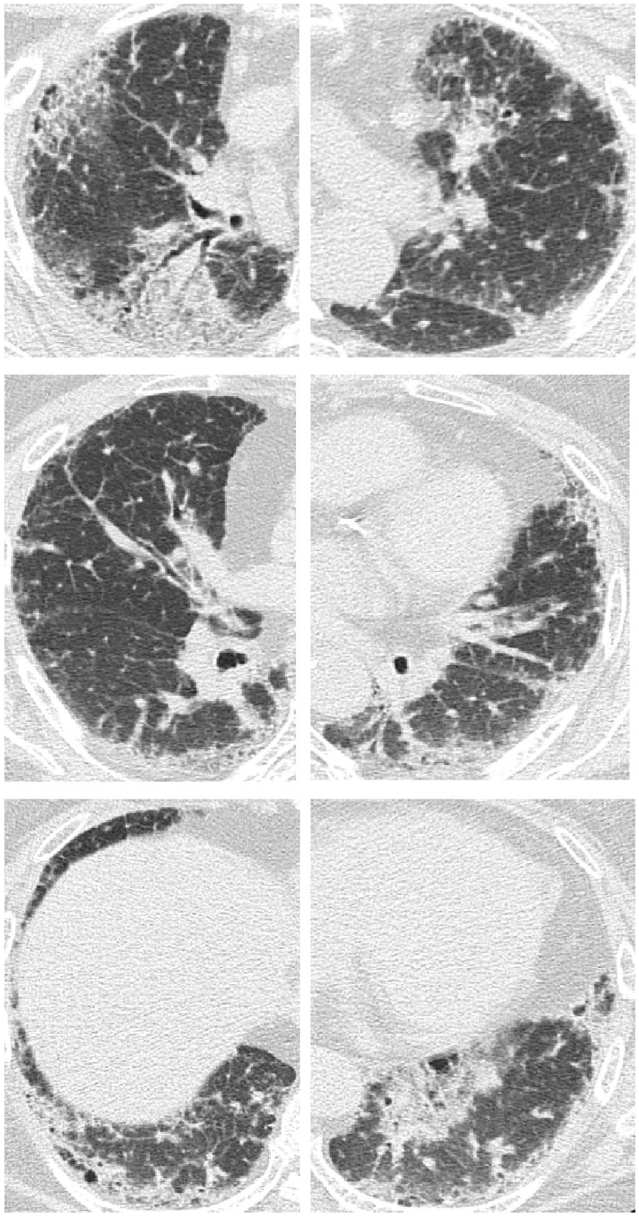

Fig. 2. Chest computed tomography on first admission.

In December 2011, she was again readmitted to our hospital with acute exacerbation (Fig. 4) and died following intensive care treatment with steroid pulse therapy and invasive positive pressure ventilation. An autopsy was performed and the microscopic examination demonstrated a diffuse alveolar damage (DAD) with hyaline membrane superimposed on diffuse interstitial fibrosis (Fig. 5).

3. Discussion

Takemura et al. reported that centrilobular fibrosis, bridging fibrosis, and organizing pneumonia, in addition to bronchiolitis, granulomas, and giant cells, were characteristic features of chronic HP with a UIP-like pattern [2]. However, the characteristic inflammatory findings of lung parenchyma or granulomas may be obscured at the later stage due to structure lobuli changes in patients with insidious BRCHP [3]. In the present case, the histopathological findings of VATS showed a UIP pattern with no evidence of the characteristic inflammatory findings of chronic HP. The sensitivity and specificity of PDE antibodies in BRCHP range from 26 to 79% and 73–93% [4]. The presence of specific

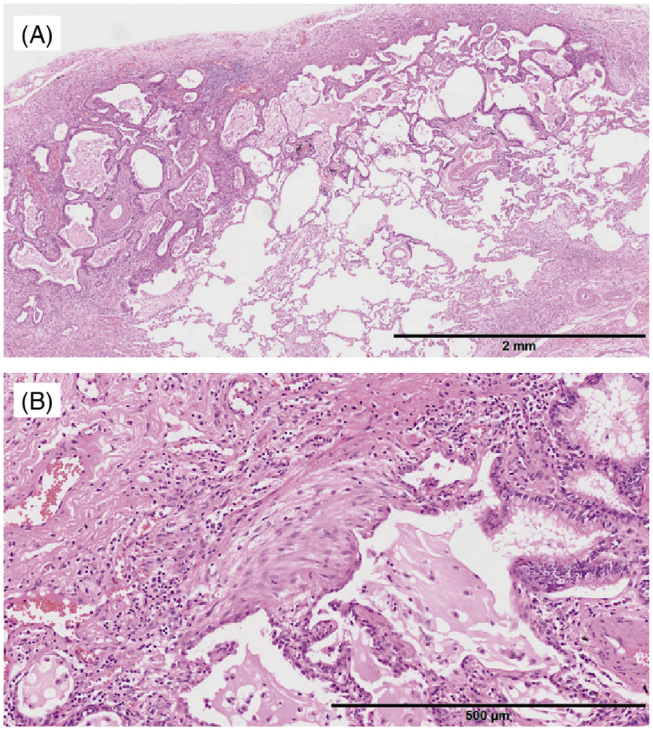

Fig. 3. Histopathological findings of lung specimens from video-assisted thoracoscopic surgery.

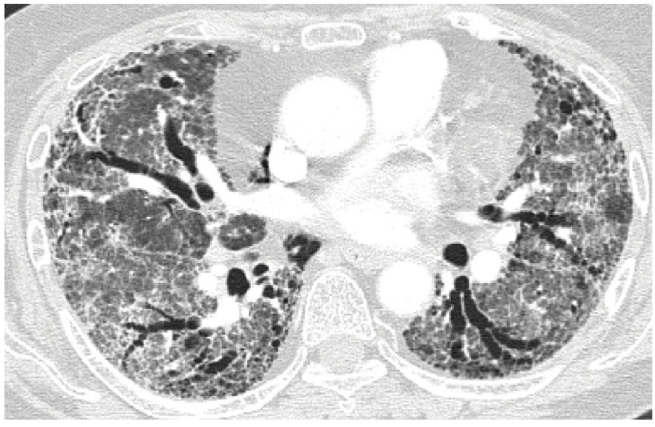

Fig. 4. Chest computed tomography on readmission with acute exacerbation.

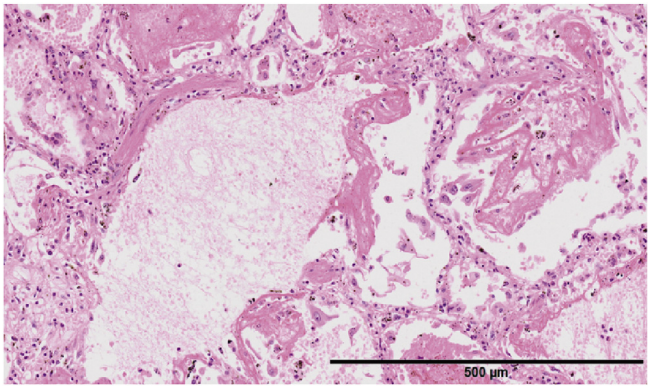

Fig. 5. Postmortem histopathological findings of the lung.

antibodies in the peripheral blood is useful, especially in cases that resemble the radiologic and pathologic features of idiopathic pulmonary fibrosis (IPF). Even though radiologically/pathologically UIP pattern, a multi-disciplinary discussion between clinicians, radiologists, and pathologists is necessary to confirm final diagnosis considering important medical history and clinical laboratory data in such present case.

Acute exacerbation is important as a cause of death in BRCHP. As in the present case, the pathological findings in acute exacerbation of BRCHP include DAD with organized exudates in the airspaces containing hyaline membranes, which are similar to those in acute exacerbation of IPF [5,6]. Clinicians should be aware that BRCHP patients especially with histopathologically UIP pattern may experience acute exacerbation.

Conflict if interest

The authors have no conflicts of interest.

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