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Bird fancier's lung: clinical-radiological presentation in 15 cases

Płuco hodowców ptaków: kliniczno-radiologiczna prezentacja 15 przypadków

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Abstract

Introduction: Bird fancier's lung (BFL) is a type of hypersensitivity pneumonitis occurring in response to avian antigens (usually inhaled proteins in bird feathers and droppings). The diagnosis is based on a combination of clinical, radiological, and biopsy characteristics. The present study was planned to highlight the clinico-radiological presentation in cases of BFL.
Material and methods: The present study is a retrospective analysis of cases of bird fancier's lung diagnosed in a unit of Vallabhbhai Patel Chest Institute over a period of two years, from 2013—2014. The clinico-radiological features of the subjects were analysed. The diagnosis of BFL was made as per criteria laid down by Mark Schuyler and Yvon Cormier.
Results: There were a total of fifteen cases diagnosed with BFL during the study period, comprising twelve females and three males with a mean age of 54.93 ± 14.21 years. All the studied subjects gave significant history of exposure to pigeons and were non-smokers. The period of symptoms prior to presentation varied from one to eight years. The main symptoms on presentation were exertional breathlessness and cough. Radiologically, diffuse centrilobular nodules, ground glassing — diffuse or patchy predominant in upper lobes, fibrosis with or without traction bronchiectasis, honeycombing, and mediastinal lymphadenopathy were seen. Bronchoscopy showed ill-defined granulomas and chronic interstitial inflammation.
Conclusions: BFL can exhibit a wide range of radiological patterns, and a high index of suspicion must be maintained, with particular attention to detailed exposure history in every case of interstitial lung disease.

Key words: bird fancier's lung, hypersensitivity pneumonitis, pigeons, bronchoscopy

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Streszczenie

Wstęp. Płuco hodowców ptaków (BLF, *bird fancier lung*) jest chorobą śródmiąższową rozwijającą się u chorych w odpowiedzi na kontakt wziewny z alergenami ptaków (białka ptasich piór i odchodów). Rozpoznanie ustala się na podstawie charakterystycznego wywiadu, obrazu klinicznego oraz wyniku biopsji płuca. Celem obecnej pracy była analiza przypadków BFL w materiale własnym.
Materiał i metody: Poddano retrospektywnej analizie przypadki BFL rozpoznane w Vallabhbhai Patel Institute w okresie dwóch lat: 2013 i 2014. Rozpoznanie BFL ustalono zgodnie z kryteriami ustanowionymi przez Marka Schuylera i Yvon Cormier.
Wyniki: W omawianym przedziale czasu rozpoznanie BFL ustalono u 15 chorych, dwunastu kobiet i trzech mężczyzn, w średnim wieku 54,93 ±14,21 roku. Wszyscy chorzy zgłaszali narażenie na kontakt z gołębiami, były to osoby niepalące. Czas od początku objawów choroby do ustalenia rozpoznania wynosił 1—8 lat. Głównymi objawami zgłaszanymi przez chorych były duszność wysiłkowa i kaszel. W badaniach radiologicznych stwierdzano rozproszone guzki śródzrazikowe, obszary matowej szyby, dominujące w płatach górnych, włóknienie, niekiedy z obecnością rozstrzeni z pociągania, zmiany o typie plastra miodu oraz powiększenie węzłów chłonnych śródpiersia. W materiale z biopsji płuca pobranej przez bronchofiberoskop stwierdzano słabo uformowane ziarniniaki oraz cechy przewlekłego śródmiąższowego zapalenia.
Wnioski: Płuco hodowców ptaków może powodować szeroką gamę obrazów radiologicznych, dlatego u każdego chorego na śródmiąższową chorobę płuc należy zwrócić uwagę na historię ekspozycji na alergeny wywołujące BLF.

Słowa kluczowe: płuco hodowców ptaków, zapalenie płuc z nadwrażliwości, gołębie, bronchoskopia

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Introduction

Hypersensitivity pneumonitis is a pulmonary disease with symptoms of dyspnoea and cough resulting from the inhalation of an antigen contained in certain organic dusts to which the patient has been previously sensitised. The two most well known types of hypersensitivity pneumonitis are farmer's lung and bird fancier's lung — especially due to pigeons [1]. Bird fancier's lung (BFL) is a type of hypersensitivity pneumonitis occurring in response to avian antigens (usually inhaled proteins in bird feathers and droppings) in susceptible subjects [2] In a population-based study from the United Kingdom, the incidence of hypersensitivity pneumonitis was found to be 1 per 100,000 in the general population [3] while the prevalence of clinical disease in those regularly exposed to pigeons has been estimated to be 1 per 1,000. [4] The diagnosis is based on a combination of clinical, radiological, and biopsy characteristics. The aim of the present study is to assess the clinical, radiological, and pulmonary function testing including diffusion and histology in a sub-group of chronic hypersensitivity pneumonitis patients due to exposure to pigeons.

Material and methods

The present study is a retrospective analysis of cases of bird fancier's lung diagnosed in a unit of Vallabhbhai Patel Chest Institute over a period of two years from 2013–2014. All patients had their detailed clinical history recorded in file and excel sheet. High-resolution computed tomography (HRCT) of the chest and bronchoscopy were performed in all subjects. The diagnosis of bird fancier's lung due to pigeons was based on criteria laid down by Mark Schuyler and Yvon Cormier [5]. The diagnostic criteria followed are discussed in Table 1. Spirometry along with lung volume and diffusion studies were carried out in all the patients, and the results were classified as per ATS/ERS interpretative strategies for lung function testing. Severity of spirometric abnormality was graded based on percentage of predicted FEV_1 as mild (FEV_1 > 70% predicted), moderate (FEV_1 60–69% predicted), moderately severe (FEV_1 50–59% predicted), severe (FEV_1 35–49% predicted), or very severe (FEV_1 < 35% predicted). Severity of diffusion impairment was graded as mild > 60% and < lower limit of normal, moderate 40–60%, and severe < 40% predicted [6]. Bronchoscopy was performed under local anaesthesia with xylocaine 2% and various bronchoscopic procedures performed as per requirement of the case.

Table 1. Diagnostic criteria used for diagnosing bird fancier's lung (modified from [5])

Major Criteria	Minor Criteria
Compatible Symptoms*	Bibasilar crackles
Evidence of antigen exposure**	Arterial hypoxaemia
Compatible HRCT changes***	Low diffusion
BAL lymphocytosis (> 40%)	
Compatible biopsy findings#	
Positive inhalational challenge##	
Diagnosis was considered confirmed if the patient fulfilled four of the major criteria and at least two of the minor criteria, and if other diseases with similar symptoms were ruled out	

The details of each of the diagnostic criteria used in our study
*Symptoms were considered to be compatible with HP if there was exertional breathlessness and/or cough, fever, myalgia
**History and/or positive immediate skin prick test with pigeon feathers/droppings was used as evidence of exposure
***High-resolution computed tomography features considered consistent with diagnosis include any one of the following: centrilobular nodules, ground glassing, predominantly upper lobe, inter/intra lobular septal thickening with or without fibrosis
#Pulmonary histological changes considered compatible include ill-defined granuloma and/or chronic interstitial infiltrates with exclusion of other causes of such histological findings
##Natural challenge test was not done in any case

Results

There were a total of 15 cases diagnosed with bird fancier's lung during the study period. During the same time, 115 cases of interstitial lung disease were diagnosed, thus representing about 13% of interstitial lung disease burden. The age of subjects varied from 29 to 82 years. The details of these patients are summarised in Table 2.

All the studied subjects gave significant history of exposure to pigeons. The majority of these patients were involved in the gaming and rearing of pigeons and regularly used to clean the faecal matter of these birds. Significantly, all the diagnosed subjects were non-smokers.

Most of the patients had a long subdued course of symptoms before presentation, with exertional breathlessness (100%) and dry cough (93.3%) being the most common symptoms. Other symptoms at presentation in patients were low-grade fever and myalgia in 60% of subjects. One of the patients had received a full course of anti-tuberculosis therapy for sputum positive tuberculosis in the past. Of note, there was a mean diagnostic delay of four years from symptom onset.

The pulmonary function tests were abnormal in all except one case. The predominant

Table 2. Baseline characters of 15 patients with bird fancier's disease

Clinical Characteristic	Results
Age (years)	
Mean	54.93 ± 14.21
Minimum–maximum	29–82
Sex	
Male	3 (20%)
Female	12 (80%)
History of exposure to pigeons	15 (100%)
Average duration of symptoms prior to presentation (years)	4 ± 2.8
History of exertional breathlessness	15 (100%)
History of cough	14 (93.3%)
Past history of anti-tuberculosis therapy	1 (6.7%)
Crackles on examination	15 (100%)
Desaturation on 6-min. walk test	11 (73.3%)
Spirometry	
Restriction	12 (80%)
Obstruction	1 (6.7%)
Isolated impairment of diffusion capacity	1 (6.7%)
Normal	1 (6.7%)
Spirometric severity of airway limitation	
No	2 (13.3%)
Mild	5 (33.3%)
Moderate	4 (26.7%)
Moderately severe	3 (20%)
Severe	1 (6.7%)
Very severe	0
Radiology	
Irregular lines	12 (80%)
Centrilobular nodules	6 (40%)
Ground glassing	6 (40%)
Air trapping	5 (33.3%)
Honeycombing	2 (13.3%)
Mediastinal lymphadenopathy	2 (13.3%)
Bronchoscopy	
Chronic interstitial inflammation	13 (86.67%)
Ill-defined granuloma	9 (60%)

abnormality was the presence of restrictive lung disease (12 cases, 80%) while obstruction, isolated impairment of diffusion capacity, and normal lung functions were each seen in one case. The diffusion capacity was reduced in 13 cases with most of the subjects having moderate decrease in diffusion capacity and an average diffusion capacity of 52.86 + 9.77 per cent of predicted value.

Radiologically, diffuse centrilobular nodules — ground glassing diffuse or patchy, predominant in the upper lobes, fibrosis with or without traction bronchiectasis, honeycombing, and mediastinal lymphadenopathy were the various findings observed (Fig. 1). The relative frequency of each finding is summarised in Table 2. The honeycombing, observed in two cases, was limited to the upper lobes with relative sparing of the lower lobe. The mediastinal lymphadenopathy observed in two cases was seen in the pre-carinal and para-aortic group. In one patient with prior history of anti-tuberculosis therapy, multiple right upper lobe cavities were seen.

The tracheobronchial tree was normal on gross bronchoscopic inspection in all cases. Transbronchial lung biopsy was done in all cases, from the most involved areas as suggested by computed tomography. Transbronchial lung biopsies showed ill-defined granulomas in 60% (9/15) of cases and chronic interstitial inflammation in 87% (13/15) cases (Fig. 2).

Discussion

Pigeon-breeder's lung was first reported in 1965 by Reed et al. [7]. The prevalence rates of pigeon-related hypersensitivity pneumonitis was found to be 1–10% in people highly exposed to pigeons [8, 9]. In the present study all the diagnosed subjects gave significant history of exposure to pigeons. Hypersensitivity pneumonitis is known to contribute to 5–15% of ILD burden [10]. Cigarette smoke is believed to have an immunosuppressive effect on alveolar macrophages and thus hypersensitivity pneumonitis is less common in smokers [8]. This is consistent with our study, in which all the diagnosed subjects were non-smokers.

The long duration of symptoms prior to symptom onset (average four years) seen in the present study, as well as in other studies [11], highlights the potential for diagnosing disease in its early stages before irreversible fibrosis sets in. The early symptoms of dry cough and exertional breathlessness along with myalgia are non-specific, and it is essential to take a detailed exposure history in every clinical presentation suggestive of interstitial lung disease to avoid late diagnosis.

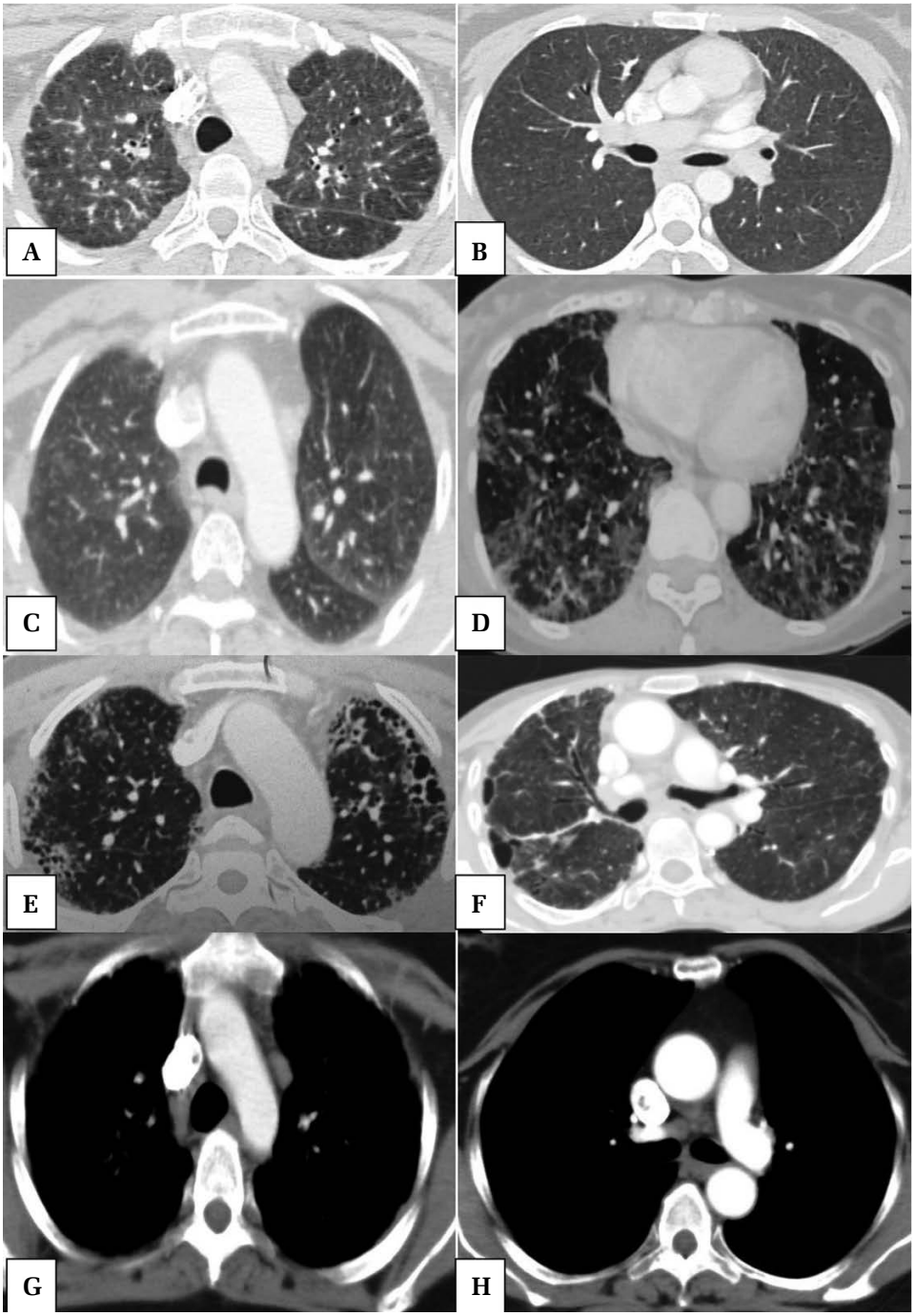

Figure 1. High-Resolution Computed Tomography chest characteristics; **A** — irregular lines; **B** — centrilobular nodules; **C** — diffuse ground glass; **D** — patchy ground glass; **E** — honeycombing; **F** — traction bronchiectasis; **G** — para-aortic mediastinal lymphadenopathy; **H** — pre-carinal mediastinal lymphadenopathy

The pattern on HRCT chest was the characteristic combination of ground-glass opacification, poorly defined centrilobular nodules, and/or irregular lines. The irregular lines representing fibrosis further led to traction bronchiectasis and non-specific honeycombing in two cases. The honeycombing seen in our cases was different from that of idiopathic pulmonary fibrosis by its presence in the upper lobes. An absence of lower lobe predominance has been used to distinguish hypersensitivity pneumonitis from idiopathic pulmonary fibrosis in some studies [11, 12]. The comparison of the radiological findings between the present study and some of the other studies [11, 13, 14] in literature are presented in Table 3.

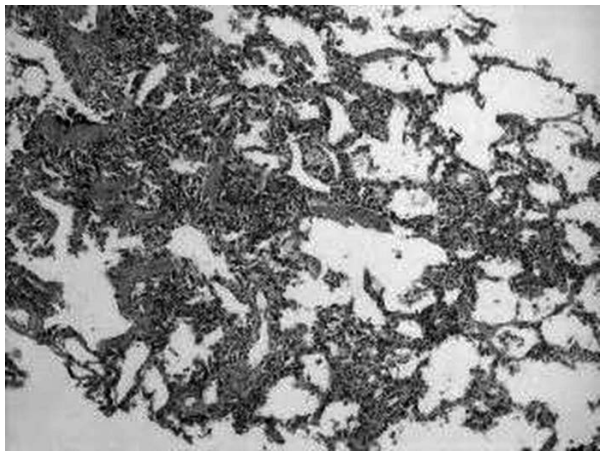

Figure 2. Chronic interstitial infiltrate with ill-defined granuloma

Achieving this objective can sometimes be far from simple. In developing countries like India, pigeons often cohabitate with humans, making antigenic avoidance impossible. Apart from antigen avoidance, corticosteroids in starting doses of 0.5–0.75 mg/kg/day are usually given in tapering doses over 3-6 months with variable effect on the disease progression.

Conclusions

Bird fancier's lung has a wide range of radiological patterns. A high index of suspicion must be maintained and utmost attention paid to taking detailed exposure history in every case of interstitial lung disease. The greatest challenge in hypersensitivity pneumonitis cases, as pointed out by Merrill [16], is for the clinician to consider that hypersensitivity pneumonitis is among the possible diagnoses.

Table 3. Comparison of computed tomographic findings in bird fancier's disease in various reported studies

Radiological Features on HRCT Chest	Morell et al. [10]	Ohtani et al. [12]	Ainslie [13]	Present study
n (%)	41	32	16	15
Micronodules	17 (41)	8 (25)	3 (19)	6 (40)
Ground Glass	28 (68)	30 (93.7)	8 (50)	6 (40)
Traction Bronchiectasis	19 (46)	24 (75)	3 (19)	6 (40)
Honeycombing	3 (7)	17 (53.1)	3 (19)	2 (13)
Irregular lines	12 (29)	–	3 (19)	9 (60)
Lymphadenopathy	–	–	–	(13)

Restrictive ventilator defect was the predominant spirometric abnormality seen in 80% of cases while 6% each showed obstruction, isolated diffusion impairment, and normal lung function. In a study by Morell [11], restrictive defect was seen in 77% of cases, similar to that in our study.

The histology on transbronchial lung biopsy showed chronic interstitial infiltrate in 87% of cases while ill-defined granulomas were seen in 60% of cases. VATS or open lung biopsies are more reliable but seldom necessary [11, 15]. The characteristic histological triad of the following: a) interstitial infiltrates of lymphocytes and plasma cells, b) bronchiolitis obliterans, and c) tiny interstitial loosely-formed non-necrotising granulomas, is specific but is usually not found in chronic hypersensitivity pneumonitis [11]. In a study by Moore et al. [11] the characteristic histological triad was found in only 9% of cases while lymphocytic-histiocytic infiltrate was seen in 67% and poorly formed granulomas in 21% of cases by transbronchial lung biopsy.

The essential treatment in hypersensitivity pneumonitis is avoidance of suspected antigens.

Conflict of interest

The authors declare no conflict of interest.

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