Case report

Bird Fancier's lung: An underdiagnosed etiology of dyspnea

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ABSTRACT

Bird Fancier's Lung is a type of hypersensitivity pneumonitis, an immunologically mediated lung disease due to repetitive exposure of air-borne avian antigen. This was first described in 1965 and is known as one of the most common causes of hypersensitivity pneumonitis. This case highlights this underdiagnosed etiology of dyspnea, disease presentation and exposure variability, and methods of diagnosis.

1. Introduction

Bird Fancier's Lung (BFL) is an immunologically mediated lung disease due to repetitive exposure of air-borne avian antigens [1]. It is a type of hypersensitivity pneumonitis (HP) triggered by exposure to highly antigenic avian proteins excreted in bird droppings and waxy proteins covering feathers of a variety of birds (bloom) which provokes a hypersensitivity reaction in a susceptible host [1,2]. Despite being one of the most common type of hypersensitivity pneumonitis, it is often an under-diagnosed condition [3,4]. Early recognition of the disease and prevention of long-term antigen exposure are necessary to avoid the progression of chronic bird fancier's lung. We present a case of BFL in a patient with history of bird exposure presenting with acute hypoxic respiratory failure initially attributed to heart failure.

2. Case presentation

A 69-year-old Caucasian woman with pertinent past medical history of breast cancer status post chemotherapy and right mastectomy, hypertension, type 2 diabetes mellitus, and morbid obesity presented to the emergency department from an outpatient surgical center for acute hypoxic respiratory failure, with oxygen saturation of 70% on room air. Initially she complained of dyspnea on mild to moderate exertion which progressively worsened since last few months. The night before her scheduled outpatient procedure she reported chest pain, orthopnea, and paroxysmal nocturnal dyspnea. She did not give any history of arthralgia, fever, sore, throat, fatigue, malaise, nausea, vomiting, weight changes, or recent sick contacts. She denied any history of active or passive smoking. Physical exam findings included diffuse rales bilaterally and +3 pitting edema of the lower extremity up to her groin. Notable laboratory results included B-type natriuretic peptide 408 (ref: <125) pg/mL, troponin 0.78 (ref: <0.04) ng/mL, and serum creatinine 0.87 (ref: 0.84–1.21) mg/dL. Transthoracic echocardiogram performed illustrated concentric left ventricular hypertrophy with normal systolic function (ejection fraction 65%), dilated, hypokinetic right ventricle, moderate tricuspid insufficiency with mild pulmonary hypertension [Fig. 1]. The patient was admitted with a provisional diagnosis of acute hypoxic respiratory failure secondary to acute exacerbation of heart failure with preserved ejection fraction.

Intravenous [IV] diuresis was initiated. A left and right heart catheterization performed on third day of hospitalization was consistent with post-capillary pulmonary hypertension [PH]. Despite aggressive diuresis and achievement of euvolemia, the patient continued to require continuous oxygen supplementation prompting further investigations.

High resolution computed tomography [HRCT] chest demonstrated diffuse mosaic attenuation pattern throughout the lung with mild interstitial thickening, without honeycombing or pulmonary fibrosis [Fig. 2]. Additionally, pulmonary function test [PFT] indicated restrictive lung disease due to reduced forced vital capacity, and reduced diffusion capacity for carbon monoxide (38% predicted). These findings prompted a further detailed social history investigation and it was learned that the patient raised doves for most of her adult life and had been breeding pigeons in the compound of her home since several years. With strong history of bird exposure, hypersensitivity panel was sent.
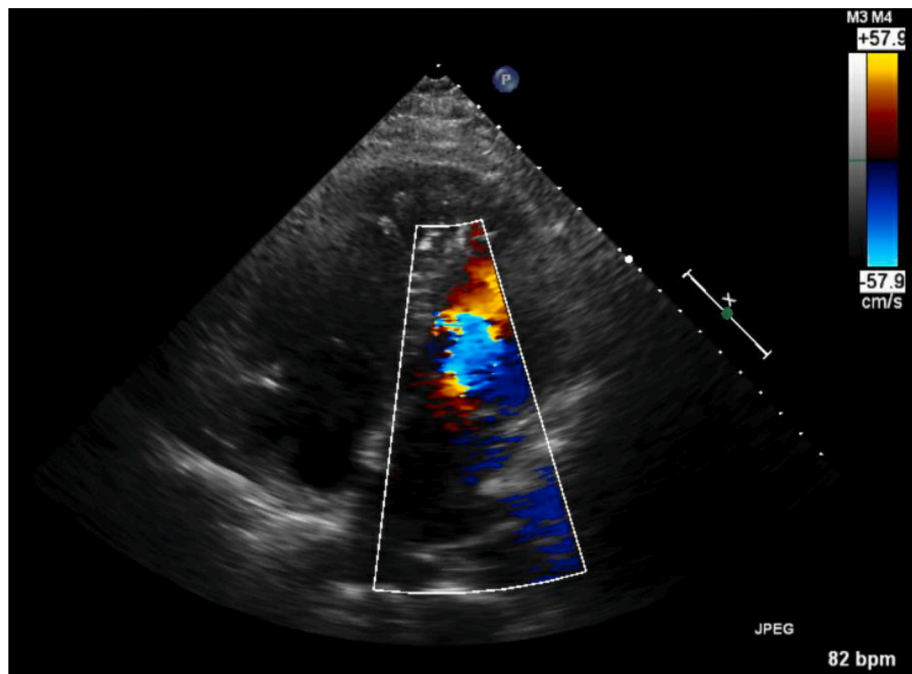
Fig. 1. Transthoracic echocardiogram showing moderate tricuspid insufficiency.
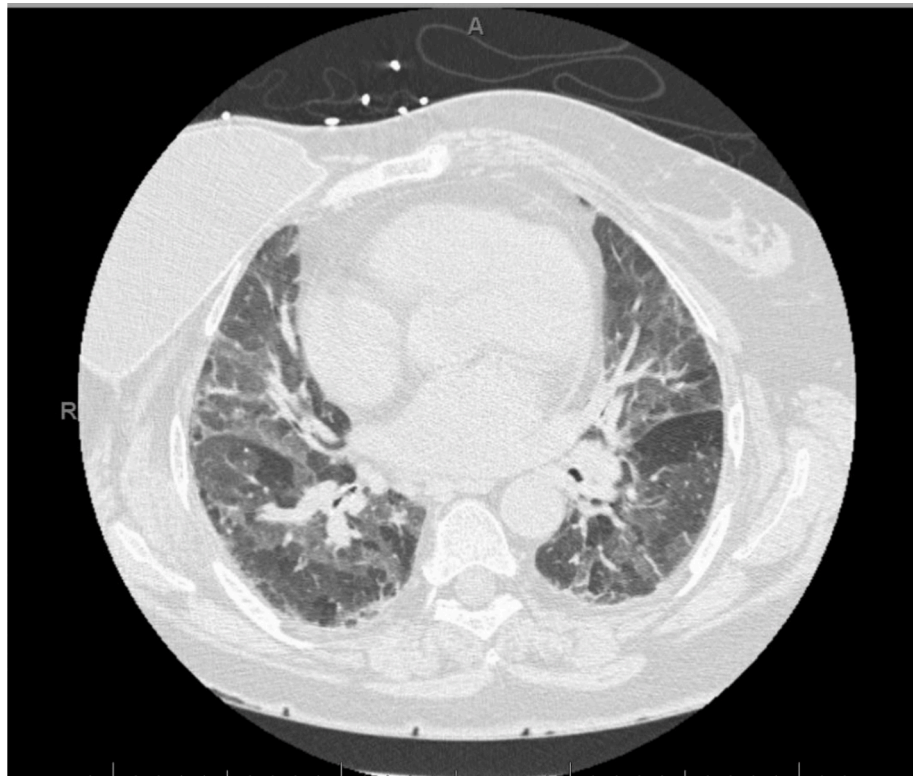

Fig. 2. HRCT chest demonstrating diffuse mosaic attenuation pattern without honeycombing.

Pertinent laboratory results included sedimentation rate 66 (ref: <20) mm/hr, ANA 1:40 (ref: <1:80), as well as ANCA and rheumatoid factor, which were negative. There was no eosinophilia present on complete blood count differential. Endobronchial ultrasound bronchoscopy performed which did not reveal significant findings, however, was negative for malignancy or granulomas. Further, video assisted thoracoscopic surgery [VATS] was performed and biopsies of right upper and lower lung lobes were collected. Following VATS, hypoxemia worsened, necessitating intubation. IV solumedrol 60 mg daily was then initiated. The patient rapidly improved after initiation of steroids and was successfully extubated two days later. Hypersensitivity panel resulted *Aureobasidium pullulans* antibody precipitin detection positive and biopsies were consistent with chronic hypersensitivity pneumonitis [cHP]. The patient was counseled to remove all birds from her home and

discharged with daily prednisone, home oxygen supplementation, and close pulmonology follow up.

3. Discussion

BFL was first described in a case series published in 1965 [3] and is now commonly included in studies describing hypersensitivity pneumonitis [HP]. Initially a disease of Pidgeon breeders, the increasing domestication of birds as pets is now the leading cause of BFL. The reported prevalence of HP among bird fanciers is estimated to range from 20 to 20,000 affected individuals per 100,000 persons at risk [5]. The species of birds raised along with handling practices may affect prevalence, but data regarding such variables are sparse. Parakeets are most commonly implicated; however, many species of birds have been shown to cause HP including a few reported cases caused by hens and canaries. In fact, some cases have even been attributed to feather-filled blankets and pillows. Exposure stems from highly antigenic proteins excreted in bird droppings and bloom [4,6,7]. The reason only a small proportion of exposed individuals develop clinically significant disease process is not known. However, genetic factors imposing immunological abnormalities underlying HP may have an important tole in determining an individual's risk of disease.

BFL, as with all HP, typically presents with cough, dyspnea, fever, and generalized fatigue. Rhinitis and conjunctivitis have also been reported [3,8–10]. Tachypnea, tachycardia, and bibasilar crackles are often present on physical exam, and hypoxemic respiratory failure may occur in severe cases [11]. To help differentiate between different etiologies of HP, a thorough history to uncover possible triggers is crucial. BFL should be considered higher in the differential when there is known bird exposure [3,12]. As it pertains to our case report, patients with bird exposure may be exposed to antigens of various avian proteins and microbial agents. Studies have proposed a "two-hit" hypothesis theory in describing the development of BFL/HP with genetic susceptibility as a "first hit" followed by increased risk for HP after a "second hit" of antigen exposure [13].

HP is commonly classified as either acute (episodic), subacute, and chronic. Several other classification schemes have been proposed, but none are completely satisfactory because of the great variability in the presentation and course of HP [14,15]. One of the diagnostic criteria suggests six major and three minor criteria out of which four major and two minor if present has been suggested for diagnosis of HP [16]. This case fulfilled the four major and two minor criteria as required. (Supplemental file 1). The clinical presentation is based on intensity and significance of antigenic exposure as well as response. Acute HP is characterized by a rapid onset of flu-like symptoms that begin a few hours after exposure, and usually involves cough, chest tightness, and dyspnea [13]. Whereas, Subacute and chronic HP both manifest with an insidious onset of cough, exertional dyspnea, fatigue, and weight loss [17,18]. There is significant overlap between subacute and chronic HP due to significant variation of presentations.

Diagnosis is often difficult due to non-specific lab markers, such as elevation in erythrocyte sedimentation rate [ESR]. Another lab marker which may be useful is lactate dehydrogenase (LDH), which has been noted to be elevated in patients with BFL, similar to levels in farmer's lung [4,19,20]. If a specific antigen is suspected based on detailed history, laboratory testing to confirm the antigen via serum specific IgG antibodies [ssIgG] [4,21] should be done. During symptomatic episodes, PFTs typically show restrictive disease, but an obstructive pattern may also be present [22]. Chest radiograph and CT scan findings can vary based on stage of disease, with mostly nonspecific findings. Specially in acute HP the HRCT may be normal due to fleeting nature of the radiographic opacities [23]. Computer tomography [CT] in subacute and chronic HP often shows ground glass attenuation, centrilobular nodules and a mosaic pattern of air trapping [4,24]. More consistent pattern can be seen in chronic HP secondary to fibrotic changes. Centrilobular nodules and ground-glass attenuation are known to be a reversible entity

in patients who can avoid exposure to the offending agent [25]. Bronchoalveolar lavage (BAL) is the most sensitive tool in identifying the alveolitis in patients suspected of having HP, with lymphocyte values greater than 60% and mast cells greater than 1% considered diagnostic of HP [26]. However, it is not always necessary, particularly in patients with a convincing exposure history and typical high-resolution computed tomography (HRCT) findings. If diagnosis is still unclear, surgical transbronchial biopsies can be performed in patients who remain refractory to therapy [4].

The evolution of lung damage and irreversible disease process in HP is characterized by chronic and persistent antigen exposure. The degree and length of antigenic exposure is directly related to disease progression. Continuous antigen exposure increases the risk for fibrotic development [27]. BFN has been found to progress more often to severe chronic forms than other forms of HP, such as farmer's lung, which can improve spontaneously [16]. The only effective intervention that can halt this chronic evolution is early recognition and control of the causative antigen or exposure. Clinicians must take a detailed occupational and recreational history in any patient presenting with unexplained dyspnea. Our case highlights the importance of a careful recording of environmental history which if done in time could have avoided several invasive diagnostic and therapeutic procedures. The rapid improvement in respiratory status after steroids however, was diagnostic in our case while awaiting hypersensitivity panel results. Perhaps using steroids in challenging cases such as ours where patient present with respiratory distress not amenable to multiple therapies along with history of exposure to birds can help in early identification and treatment for these patients.

BFL is one of the common and preventable causes of hypersensitivity pneumonitis. Antigen avoidance and removal is the most important facet in the management of BFL. Medical therapy in the form of systemic corticosteroids may be useful if HP continues to progress despite avoidance of antigen exposure. Acute or subacute forms may resolve with treatment, however, chronic cases may progress to pulmonary fibrosis where lung transplantation may be required [22].

Ethics approval

Our institution does not require ethics approval for reporting individual cases or case series.

Informed consent

Written and verbal informed consent were obtained from the patient for their anonymized information to be published in this article.

Availability of data and materials

Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

Declaration of competing interest

Neither financial nor non-financial competing interests.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.rmcr.2020.101288.

References

[1] M. Funke, J.-M. Fellrath, Hypersensitivity pneumonitis secondary to lovebirds: a new cause of bird fancier's disease, Eur. Respir. J. 32 (2008) 517–521, https://doi.org/10.1183/09031936.00108507.

[2] V. Chopra, J.L. Joshi, P. Mrigpuri, A.S. Chahal, Pigeon fancier's lung – an under-diagnosed cause of severely debilitating and chronic breathlessness, Egypt. J. Chest Dis. Tuberc. 66 (2017) 557–559, https://doi.org/10.1016/j.ejcdt.2016.08.002.

[3] C. Reed, A. Barbee, Pigeon breeders' lung: a newly observed interstitial pulmonary disease, J. Am. Med. Assoc. 193 (1965) 261–265.

[4] Ferran Morell, Àlex Roger, Leonardo Reyes, Maria Jesús Cruz, Cristina Murio, Xavier Muñoz, Bird fancier's lung. A series of 86 patients, Medicine 87 (2) (2008) 110–130, https://doi.org/10.1097/MD.0b013e31816d1dda.

[5] L.T. Christensen, C.D. Schmidt, L. Robbins, Pigeon breeders' disease–a prevalence study and review, Clin. Allergy 5 (1975) 417–430, https://doi.org/10.1111/j.1365-2222.1975.tb01881.x.

[6] N. Inase, Y. Ohtani, Y. Sumi, T. Umino, Y. Usui, S. Miyake, Y. Yoshizawa, A clinical study of hypersensitivity pneumonitis presumably caused by feather duvets, Ann. Allergy Asthma Immunol. 96 (2006) 98–104.

[7] D.J. Hendrick, J.A. Faux, R. Marshall, Budgerigar-fancier's lung: the commonest variety of allergic alveolitis in Britain, Br. Med. J. 2 (1978) 81–84.

[8] F. Rodriguez de Castro, T. Carrillo, R. Castillo, C. Blanco, F. Diaz, M. Cuevas, Relationships between characteristics of exposure to pigeon antigens, Chest 103 (1983) 1059–1063.

[9] A.W. Van Toorenenbergen, R. Gerth van Wijk, G. Van Dooremalen, P.H. Dieges, Immunoglobulin E antibodies against budgerigar and canary feathers, Int. Arch. Allergy Immunol. 77 (1985) 433–437.

[10] S.J. Bourke, R. Carter, K. Anderson, J. Boyd, S. King, B. Douglas, G. Boyd, Obstructive airways disease in non-smoking subjects with pigeon fancier's lung, Clin. Exp. Allergy 19 (1989) 629–632.

[11] J.V. Hirschmann, S.N.J. Pipavath, J.D. Godwin, Hypersensitivity pneumonitis: a historical, clinical, and radiologic review, Radiographics 29 (2009) 1921–1938, https://doi.org/10.1148/rg.297095707.

[12] M.J. Cruz, F. Morell, A. Roger, X. Munoz, M.J. Rodrigo, Hypersensitivity pneumonitis in construction plasterers (espartosis): study of 20 patients, Med. Clin. 120 (2003) 78–83.

[13] M. Selman, A. Pardo, T.E. King, Hypersensitivity pneumonitis: insights in diagnosis and pathobiology, Am. J. Respir. Crit. Care Med. 186 (2012) 314–324, https://doi.org/10.1164/rccm.201203-0513CI.

[14] G. Boyd, C.P. McSharry, S.W. Banham, P.P. Lynch, A current view of pigeon fancier's lung. A model for pulmonary extrinsic allergic alveolitis, Clin. Allergy 12 (Suppl) (1982) 53–59.

[15] H.B. Richerson, I.L. Bernstein, J.N. Fink, G.W. Hunninghake, H.S. Novey, C. E. Reed, J.E. Salvaggio, M.R. Schuyler, H.J. Schwartz, D.J. Stechschulte, Guidelines for the clinical evaluation of hypersensitivity pneumonitis. Report of the Subcommittee on Hypersensitivity Pneumonitis, J. Allergy Clin. Immunol. 84 (1989) 839–844, https://doi.org/10.1016/0091-6749(89)90349-7.

[16] Y. Lacasse, M. Selman, U. Costabel, J.-C. Dalphin, M. Ando, F. Morell, R. Erkinjuntti-Pekkanen, N. Muller, T.V. Colby, M. Schuyler, Y. Cormier, HP Study Group, Clinical diagnosis of hypersensitivity pneumonitis, Am. J. Respir. Crit. Care Med. 168 (2003) 952–958, https://doi.org/10.1164/rccm.200301-137OC.

[17] R.S. D'souza, A. Donato, Hypersensitivity pneumonitis: an overlooked cause of cough and dyspnea, J. Community Hosp. Intern. Med. Perspect. 7 (2017) 95–99, https://doi.org/10.1080/20009666.2017.1320202.

[18] D.P. Schlueter, Response of the lung to inhaled antigens, Am. J. Med. 57 (1974) 476–492, https://doi.org/10.1016/0002-9343(74)90141-7.

[19] A. Rask-Andersen, Allergic alveolitis in Swedish farmers, Ups. J. Med. Sci. 94 (1989) 271–285.

[20] S.P. Matusiewicz, I.J. Williamson, P.J. Sime, P.H. Brown, P.R. Wenham, G. K. Crompton, A.P. Greening, Plasma lactate dehydrogenase: a marker of disease activity in cryptogenic fibrosing alveolitis and extrinsic allergic alveolitis, Eur. Respir. J. 6 (1993) 1282–1286.

[21] M. Vasakova, F. Morell, S. Walsh, K. Leslie, G. Raghu, Hypersensitivity pneumonitis: perspectives in diagnosis and management, Am. J. Respir. Crit. Care Med. 196 (6) (2017 Sep 15) 680–689, https://doi.org/10.1164/rccm.201611-2201PP. PubMed PMID: 28598197.

[22] P. Spagnolo, G. Rossi, A. Cavazza, M. Bonifazi, I. Paladini, F. Bonella, N. Sverzellati, U. Costabel, Hypersensitivity pneumonitis: a comprehensive review, J Investig. Allergol. Clin. Immunol. 25 (2015) 237–250, quiz follow 250.

[23] F. Morell, A. Roger, L. Reyes, M.J. Cruz, C. Murio, X. Muñoz, Bird fancier's lung: a series of 86 patients, Medicine (Baltim.) 87 (2008) 110–130, https://doi.org/10.1097/MD.0b013e31816d1dda.

[24] G. Battista, C. Sassi, M. Zompatori, D. Palmarini, R. Canini, Ground-glass opacity: interpretation of high resolution CT findings, Radiol. Med. 106 (2003) 425–442.

[25] M. Remy-Jardin, J. Remy, B. Wallaert, N.L. Müller, Subacute and chronic bird breeder hypersensitivity pneumonitis: sequential evaluation with CT and correlation with lung function tests and bronchoalveolar lavage, Radiology 189 (1993) 111–118, https://doi.org/10.1148/radiology.189.1.8372179.

[26] G. Semenzato, L. Biermer, U. Costabel, P.L. Haslam, D. Olivieri, Extrinsic allergic alveolitis (BAL), Eur. Respir. J. 3 (1990) 945–946.

[27] C. Pereira, A. Gimenez, L. Kuranishi, K. Storrer, Chronic hypersensitivity pneumonitis, J. Asthma Allergy 9 (2016) 171–181.