CASE REPORT

Chronic Hypersensitivity Pneumonia due to Pigeon Breeders' Disease

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Background: Pigeon breeders' disease usually affects adults. Children are more likely to be affected when they share living space with a backyard poultry or pigeon breeding. **Case characteristics:** A 12-year-old girl with persistent cough for 3 years and dyspnea for 2 years. **Obervation:** She was dignosed to be having allergy to pigeon droppings, based on reports of lung biopsy and allergy testing. **Message**: Pigeon breaders' disease should be considered in a child who presents with features of chronic hypersensiticity pneumonitis.

Keywords: *Allergy testing, Lung biopsy, Persistent pneumonia*.

Pigeon breeders' disease (PBD) is one of the most widespread forms of hypersensitivity pneumonia [1], described infrequently in children. We describe a girl with chronic hypersensitivity pneumonia, who presented with prolonged cough and progressive dyspnea.

CASE REPORT

A 12-year-old girl was referred to us with persistent dry cough for 3 years, progressive dyspnea for last 2 years and significant weight loss for last 1 year. She had received anti-tubercular and anti-asthma treatment without any relief. Her past and family history was insignificant, except that they were involved in breeding around 60 pigeons at home. A recent chest *X*-ray revealed bilateral ground glass haziness and pulmonary function tests (PFT) revealed severe restriction (FVC 16.6 %, FEV1 17.9 % , FEV1/ FVC 1.07), with reduced diffusion capacity of lung for carbon monoxide (DLCO) (19%).

On examination, she was cachexic and dyspneic at rest, with tachycardia and tachypnea. Her S_pO_2 was 82% at room air that improved to 95% with supplemental oxygen. Respiratory system examination revealed use of accessory muscles of respiration, with pectus excavatum and fine basal crepitations. Cardiovascular system examination revealed loud P2, and normal jugular venous pressure and hepato jugular reflux. She had no hepatomegaly. She weighed 23 kgs (< -3 SD), with a height of 144 cm (-2 to -3 SD) and body mass index of 11.11 (< -3SD). She was in tanner stage II of sexual maturity.

On investigation, arterial blood gas revealed PO_2 of 70 mmHg, with saturation of 91%. Echocardiography revealed dilated pulmonary artery, dilated right ventricle and moderate tricuspid regurgitation (gradient 30 mm of Hg) suggestive of pulmonary artery hypertension (PAH). Work up for tuberculosis, immunodeficiency (primary and secondary), sarcoidosis, connective tissue disorders, celiac disease, tropical pulmonary eosinophilia, allergic bronchopulmonary aspergillosis, cytomegalovirus and mycoplasma were negative. Fibreoptic bronchoscopy was grossly normal. Cytology of bronchoalveolar lavage and flow cytometry were inconclusive. High resolution computed tomography (HRCT) of chest (***Fig.* 1**) revealed diffuse mosaic pattern and multiple ill-defined centrilobular nodular lesions in both upper lobes and interstitial thickening in the apical segment of left lower lobe. Histopathology of lung biopsy specimen revealed classical features of hypersensitivity Pneumonia (***Web Fig.***1) [2,3]. Test for pigeon dropping allergy in serum was positive.

She was administered intravenous methylprednisolone (30 mg/kg/day) for 3 days, followed by oral prednisolone and inhaled budesonide. Bosentan (BOSENTAS 62.5 mg twice daily) [4] was also added in view of the pulmonary hypertension. At day 45, she was discharged on home oxygen, and advised to avoid exposure to pigeons. Her weight at discharge was 24 kg. She was having dyspnea only on exertion, with SpO_2 of 92% at room air and FEV1 and FVC of 29% and 23%, respectively. Oral steroids and bosentan were continued. Diuretics were not administered as she had no features of congestive heart failure. She was followed-up with monthly monitoring of respiratory rate, SpO_2, pulmonary function and liver function test. Bosentan was stopped after two-dimentional echocardiography at eight weeks

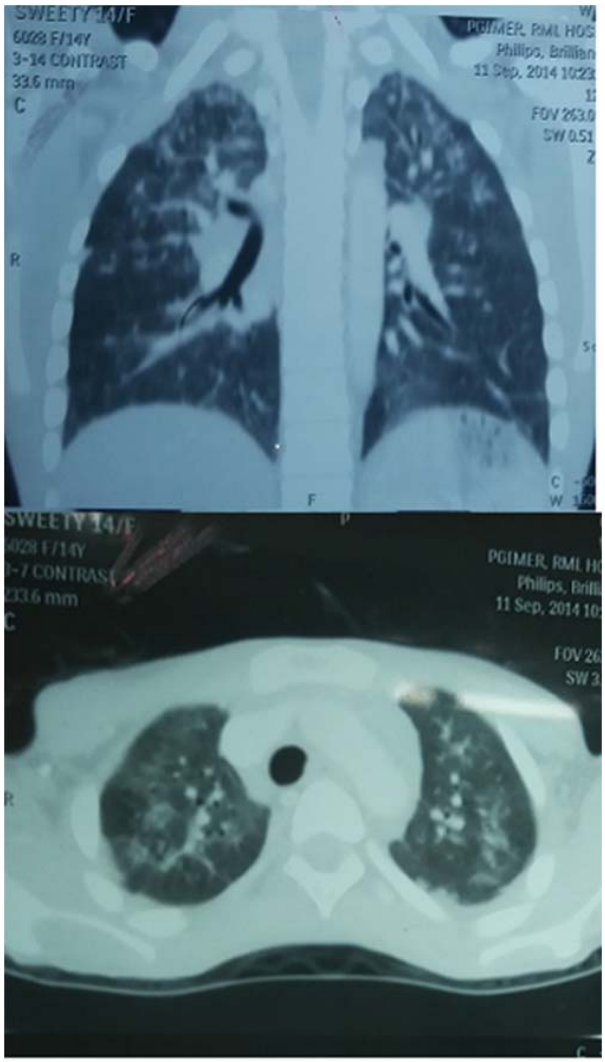

FIG. 1 *Patchy areas of ground glass haze seen in both upper lobe with air trapping (white arrow) giving rise to mosaic pattern and multiple ill-defined centrilobular nodular lesions in both upper lobes (black arrows) and interstitial thickening in the apical segment of left lower lobe.*

showed no evidence of pulmonary hypertension. She was continued on low dose prednisolone. At 10 months follow-up, she was on room air, attending school; FEV1 and FVC were 33% and 37%, respectively.

DISCUSSION

Hypersensitivity pneumonia is an immune-mediated lung parenchymal injury occurring in response to repeated inhalation of an antigen. Antigens implicated include animal proteins, fungi, amoeba, bacteria, medications and chemicals. Avian antigens are one of the most common causes of hypersensiticvity pneumonia [2]. Both type III and type IV hypersensitivity responses have been implicated in the disease process [5]. The chronic form of hypersensitivity pneumonia results from long term low-grade exposure, and is characterized by dyspnea, chronic cough, fatigue, anorexia and weight loss. PFT typically reveals a restrictive pattern and a decrease in DLCO. HRCT chest in chronic hypersensiticvity pneumonia reveals fibrotic changes, irregular linear opacities, centri-lobular nodules, honeycombing and traction bronchiectasis [2]. Bronchoalveolar lavage usually reveals a significant increase in the percentage of lymphocytes with decreased CD4+/CD8+ratio [6], On histopathology, the interstitial and alveolar collections of foamy histiocytes are considered to be fairly specific for hypersensitivity pneumonia due to pigeon breeder's disease [3], which was observed in our case. Interstitial fibrosis and interstitial cellular infiltrates that is primarily lymphocytic with large number of plasma cells, with absence of granulomas is also observed in chronic hypersensitivity pneumonia.

Common differentials include other interstitial lung diseases which include immune-mediated collagen vascular diseases, sarcoidosis, langerhans cell histiocytosis and malignancies [7-8]. Diagnostic criteria for hypersensitivity pneumonia include six major and three minor criteria [9]. Establishing diagnosis requires the presence of at least 4 major and 2 minor criteria. The present case had 5 major, and all 3 minor criteria. Treatment for chronic hypersensitivity pneumonia includes oral prednisolone over several months, depending on the response to improvement in symptoms and functional abnormalities [10].

Early treatment leads to complete reversal in acute and sub-acute hypersensitivity pneumonia. Chronic form may proceed to irreversible lung damage in spite of treatment and avoidance of the offending antigen. Our case continues to have restrictive changes at 10 months follow-up. The clinicians should have a high index of suspicion in order to make early diagnosis and avoid disease progression and irreversible lung damage.

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REFERENCES

1. Grech V, Vella C, Lenicker H. Pigeon breeder's lung in childhood: Varied clinical picture at presentation. Pediatr Pulmonol. 2000;30:145-8.
2. Kurup VP, Zacharisen MC, Fink JN. Hypersensitivity pneumonitis. Indian J chest Dis Allied Sci. 2006;48: 115-28.
3. Riley DJ, Saldana M. Pigeon breeder's lung. Subacute

course and the importance of indirect exposure. Am Rev Respir Dis. 1973;107:456-60.
4. Abman SH, Hansmann G, Archer SL, Ivy DD, Adatia I, Chung WK, *et al*. Pediatric Pulmonary Hypertension Guidelines from the American Heart Association and American Thoracic Society. Circulation. 2015;132:2037-99.
5. Du Marchie Sarvaas GJ, Merkus PJ, De Jongste JC. A family with extrinsic allergic alveolitis caused by wild city pigeons: a case report. Pediatrics. 2000;105:e62.
6. Ratjen F, Costabel U, Griese M, Paul K. Bronchoalveolar lavage fluid findings in children with hypersensitivity pneumonitis. Eur Respir J. 2003;21:144-8.
7. Fan LL, Deterding RR, Langston C. Pediatric interstitial lung disease revisited. Pediatr Pulmonol. 2004;38:369-78.
8. Deutsch GH, Young LR, Deterding RR, Fan LL, Dell SD, Bean JA, *et al*. Diffuse lung disease in young children: application of a novel classification scheme. Am J Respir Crit Care Med. 2007;176:1120-8.
9. Schuyler M, Cormier Y. The diagnosis of hypersensitivity pneumonitis. Chest. 1997;111:534-6.
10. Fink JN, Zacharisen MC. Hypersensitivity Pneumonitis. *In:* Adkinson NF, Yunginer JW, Busse WW, Bochner BS, Holgate ST, Simons FE. Middleton's Allergy: Principles and Practice. 6th ed. Philadelphia: Mosby and Co; 2003. p. 1373-90.