Extrinsic allergic broncholitis in a bird fancier

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ABSTRACT A patient in whom a severe systemic illness was characterised by weight loss, arthralgia and breathlessness was exposed to her pet bird. This pragmatic approach reproduced the features of her illness. The changes that occurred in her pulmonary physiology and histology differed from those seen in extrinsic allergic alveolitis in many important respects.

The term extrinsic allergic alveolitis is used to describe the abnormalities in lung pathology and chest radiography that result from the inhalation of dusts to which the patient had developed circulating antibodies. Extrinsic allergic alveolitis caused by bird antigen was reported by Plessner in 1960.1 We report here a case with symptoms due to exposure to a pet cockateal caused by an extrinsic allergic broncholitis. There are important differences between extrinsic allergic broncholitis and alveolitis that could lead to diagnostic error.

Case report

A 41 year old woman presented in May with a six month history in increasing shortness of breath. She had had episodic cough with sputum since the age of 18. After a haemoptysis in 1965, a bronchogram was performed and showed minimal bronchiectasis. There was no history of asthma or chest infection in childhood. Her mother had kept budgerigars until the patient left home aged 22.

Two years before her admission to hospital she had acquired a cockateal that was allowed to fly about the house; it died in June 1976. In September 1976 she bought another cockateal and in December of that year she began to wake each morning feeling generally unwell. By January 1977 she had developed shortness of breath and palpitations on minimal exertion with night sweats and arthralgia. She had lost 28 lbs (12.7 kg) during the previous year. In March 1977 she was admitted to another hospital for one week and improved, but within five hours of returning home she had relapsed. She was then admitted to Brompton Hospital where an open lung biopsy was performed (figs 1 and 2).

On examination she was short of breath on any exertion and centrally cyanosed, with clubbing. Her chest was hyperinflated and she had dry inspiratory crackles throughout both lung fields. The remainder of her physical examination was normal. She was non-atopic.

The following investigations were performed: Hb 15.5 g/dl, white cell count 7.4 x 10^9/l, 2% eosinophils, ESR 32 mm in one hour, platelets normal numbers, LE cells none seen, ANF negative, sputum culture negative, rheumatoid factor negative, sputum examination for fungi negative, and urea, electrolytes, albumin, globulin, liver function tests, and complement C3 normal. Anti-DNA antibody concentration IU/ml was also normal. Calcium 2.35 mmol/l, phosphate 1.31 mmol/l, total IgE 50 IU/l (normal), thyroid function test normal, sweat sodium 53 mmol/l (normal), and immunoglobulins IgG 162 IU, IgA 182 IU, IgM 321 IU. Slight rise in IgM, others normal. Viral serology negative, sputum for AFB negative, sputum cytology negative. The chest X ray film showed a little bronchial wall thickening, particularly at the bases, suggestive of bronchiectasis. The lung fields were otherwise clear. Precipitating antibody to avian serum protein positive (one line). Prick testing to budgerigar serum and feathers negative.

LUNG FUNCTION TESTS
Lung function tests were done on admission, during the bronchial provocation test, and at intervals after she had given away her bird. Spirometry and flow volume loops were performed using an Ohio dry piston spirometer. Lung volume measurements were made by body plethysmograph (Fenyves and Gut, Basle). Alveolar volume and 10 second single breath carbon monoxide gas transfer were measured using a PK Morgan respirimeter.

The table shows test results on admission and after one month away from her bird. Arterial blood
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Lung function in a bird fancier immediately before admission, after one month at home after removal of the bird, and after one year

<table>
<thead>
<tr>
<th></th>
<th>Predicted</th>
<th>On admission</th>
<th>One month later</th>
<th>One year later</th>
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<tbody>
<tr>
<td>FEV₁ (ml)</td>
<td>2970</td>
<td>1040</td>
<td>1120</td>
<td>1370</td>
</tr>
<tr>
<td>FVC (ml)</td>
<td>3590</td>
<td>1950</td>
<td>2410</td>
<td>2770</td>
</tr>
<tr>
<td>Ratio %</td>
<td>53</td>
<td>46</td>
<td>46</td>
<td>49</td>
</tr>
<tr>
<td>TLC (ml)</td>
<td>5380</td>
<td>5490</td>
<td>4940</td>
<td>5580</td>
</tr>
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<td>RV (ml)</td>
<td>1810</td>
<td>3340</td>
<td>2840</td>
<td>2730</td>
</tr>
<tr>
<td>Vₐ (ml)</td>
<td>5380</td>
<td>3340</td>
<td>3620</td>
<td>3690</td>
</tr>
<tr>
<td>VC (ml)</td>
<td>3590</td>
<td>2150</td>
<td>2100</td>
<td>2850</td>
</tr>
<tr>
<td>Raw (kPa/l/s)</td>
<td>&lt;0·2</td>
<td>0·3</td>
<td>0·34</td>
<td>0·58</td>
</tr>
<tr>
<td>SGaw (kPa/s)</td>
<td>1·3–3·6</td>
<td>0·88</td>
<td>0·87</td>
<td>0·51</td>
</tr>
<tr>
<td>DLCO (mmol/min</td>
<td>8·76</td>
<td>3·1</td>
<td>2·91</td>
<td>6·21</td>
</tr>
<tr>
<td>kPa)</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>KCO (mmol/min/ kPa/l)</td>
<td>1·79</td>
<td>0·93</td>
<td>0·80</td>
<td>1·68</td>
</tr>
</tbody>
</table>

gases at rest while breathing air showed PO₂ 55 mm Hg, PCO₂ 38 mm Hg, pH 7·43. After exercise at 200 kpm/min the PO₂ rose to 68·5 mm Hg and PCO₂ fell to 30 mm Hg.

BRONCHIAL CHALLENGE TESTING

On 20 July the patient was exposed to her pet bird for one hour in a closed ventilated chamber measuring 6 m³. FEV₁ and FVC were measured with a dry wedge spirometer (Vitalograph) before challenge, every five minutes for 30 minutes after challenge, and then every 10 minutes for another 30 minutes. From then on measurements were made every hour until 10 pm and at 2 am, 4 am, and 6 am, then again 72 hours after challenge. Control measurements were made on a separate day in the same way. Lung volumes and gas transfer were also measured before challenge and at four, eight, 24, and 72 hours. Eleven hours after challenge she was short of breath, and the crackles audible on the first admission were present again. Her white cell count rose from 11·5 × 10⁹/l to 18·4 × 10⁹/l, owing to a neutrophilic leucocytosis. Estimation of plasma histamine performed before challenge, 11 hours after, and 24 hours after was 6·3 ng/ml, 6 ng/ml, and 6·2 ng/ml respectively as measured by an automated fluorometric method.² There was no change in the level of prostaglandin breakdown products. FEV₁ and FVC fell progressively over 24 hours returning to prechallenge levels after two days (fig 3). Total lung capacity remained constant, and the fall in vital capacity was reflected in a rise in residual volume. At the same time DLCO and Vₐ fell but KCO remained unchanged (fig 4). Airways resistance rose and specific conductance fell. All these changes were maximal at 24 hours and reverted by 72. No such changes occurred when the same measurements were made on a control day without challenge. Histology on the open lung biopsy was reported as follows: "Sections of lung show chronic bronchiolitis with infiltration of the walls of several terminal and first order respiratory bronchioles by many neutrophils, eosinophils, lymphocytes, plasma cells, and histiocytes (fig 1). There are neutrophils among the cells of the bronchiolar lining epithelium, and also within the lumen. Some interstitial histiocytes are collected together in small groups suggestive of granuloma formation, and one multinucleated giant cell is seen (fig 2). There are patches of alveolitis present in places, but the more striking change is..."
chronic bronchiolitis. The chantes are compatible with an allergic reaction to bird allergens affecting bronchioles more than alveoli."

**Discussion**

The combination of breathlessness and cracks on auscultation in a bird owner immediately suggests allergic alveolitis. Nevertheless, the hyperinflated chest x-ray film with clear lung fields in combination with obstructive spirometry caused diagnostic difficulty. Crackles, airflow obstruction, and a normal chest x-ray film are the features of bronchiolitis in children and adults and in this case led to a clinical diagnosis of extrinsic allergic bronchiolitis.

Lung function testing showed reduced flow rates at low lung volumes in association with a residual volume and normal total lung capacity. This was interpreted as indicating airflow obstruction occurring in both large and small airways. The low vital capacity that might have indicated restriction was due to this airflow obstruction with gas trapping. During bronchial challenge FEV₁ and FVC fell proportionately and this could well have been misinterpreted as being due to an acute alveolitis, particularly as the changes were accompanied by the development of cracks on auscultation. Similarly the fall in DLCO during provocation testing was due largely to a drop in alveolar volume secondary to airflow obstruction. This is shown in the relative constancy of the KCO. If spirometry and DLCO alone had been measured the changes would have been attributable entirely to an acute alveolar reaction. It is important to emphasise that the physiological changes show that airflow obstruction was the dominant abnormality but do not exclude a coexisting alveolitis.

The low KCO that falls a little farther on provocation indeed suggests that an alveolar component doses exist. Nevertheless, the rest of the lung function tests, the rise in PaO₂ on exercise, and the histology all show that this component is small.

The histological appearances of chronic bronchiolitis with a suggestion of granuloma formation are consistent with what may be termed as "extrinsic allergic bronchiolitis." The presence of bronchiolitis is well recognised in association with allergic alveolitis in the acute disease. It may result in airways obstruction in the chronic stage, or it may resolve. The unusual feature of the present case is the mild nature of the alveolitis, compared with the severe bronchiolitis.

The importance of this case lies in the likelihood of misdiagnosis. The clinical, radiological, and physiological patterns are all sufficiently different from classic extrinsic allergic alveolitis for the diagnosis to be missed. If similar patients are misdiagnosed they will be treated inappropriately while remaining in contact with their birds and so develop progressive irreversible respiratory impairment. As birds are common pets there are probably other patients deteriorating under the stultifying label of chronic obstructive lung disease.

Our patient's pre-existing mild bronchiectasis may be the reason for her atypical disease. In theory this will reduce flow rates, especially in the smaller airways. Any inhaled particulate antigen will then
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tend to settle in airways rather than passing out into the alveoli. A bronchiolitis rather than alveolitis would then be expected. In this connection it is interesting that smokers appear to be protected from extrinsic allergic alveolitis.\textsuperscript{5} When flow rates are reduced by smoking the same argument will apply. This suggests that the protection is more apparent than real. Smokers may avoid farmer's lung by having an unrecognised bronchiolitis instead. Any resulting worsening in their airflow obstruction will naturally be blamed on smoking. In the cupboard of chronic obstructive lung disease there may be many such skeletons.

Requests for reprints to Dr MG Harries, Northwick Park Hospital, Harrow, Middx.

References


