CASE REPORTS

Pulmonary aspergillosis in two families

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REPORTS of more than one case of pulmonary aspergillosis occurring in a family are rare.

Hertzog, Smith & Goblin (1949) described a brother and sister aged 5 and 11 who died of pneumonia within a month of each other, necropsy in one of them showing multiple granulomata from which *A. fumigatus* was isolated.

More recently, Strelling *et al.* (1966) reported two fatal cases of the invasive type of aspergillosis in two sisters, aged 4 years and 14 months respectively. In both cases the lungs were full of nodular lesions tending to become confluent, and *A. fumigatus* was cultured from lungs, and tracheobronchial lymph glands, which were enlarged.

Because familial incidence of the allergic eosinophilic infiltration type of aspergillosis has not previously been reported, four such cases, occurring in two families are described.

Case reports

Family A—Country dwellers and poultry keepers.

Case 1 (Fig. 1). R.W., aged 5½ years, suffered from recurrent bronchitis with sinusitis. Two years later he developed pneumonia in the left upper lobe associated with hilar adenitis, followed a year later by a fresh lesion in the right lower lobe, which failed to resolve completely. Bronchography showed suppurative bronchitis rather than bronchiectasis. Two years later he suffered his third episode of pneumonia, in the right upper lobe, followed 18 months later by a fourth episode, in the left upper lobe. He was investigated at Brompton Hospital, where aspergillosis of lungs and sinuses was diagnosed based on positive prick skin tests, bronchial sensitivity tests, and the isolation of *A. fumigatus* from bronchial secretions and antral washouts, together with a blood eosinophilia. He has developed cor pulmonale.

FIG. 1. Family A, case 1. Tuberculin negative. Infiltration in left upper lobe with well-marked left hilar adenitis.

Case 2 (Fig. 2). M.W., aged 8 years, had pneumonia in the right upper lobe, which cleared, but was followed by fresh infiltration in the left upper lobe, and left lower lobe, at intervals of 2 and 4 years. Tomography after the last illness showed distended bronchi in the right upper lobe, and bronchoscopy an inflamed right upper lobe orifice. He was transferred to Brompton Hospital with his brother, where aspergillosis of lungs was confirmed by positive prick skin tests, bron-

FIG. 2. Family A, case 2. Right upper tomogram showing pus-filled segmental bronchi in right upper lobe.

FIG. 3. Family B, case 3. Total collapse of left lung due to simultaneous mucus obstruction of smaller bronchi.

FIG. 4. Family B, case 4. Infiltration left lower lobe.

chial sensitivity tests and the isolation of *A. fumigatus* from bronchial secretions. Purulent sinusitis was also found, but *A. fumigatus* was not cultured from antral washouts. He died of cor pulmonale a year later.

Family B—Farmers.

Case 3 (Fig. 3). H.E., farmer's wife, aged 60 years, had pneumonia in the right lower lobe with a blood eosinophilia which cleared rapidly. Eight years later she became suddenly breathless with thick sputum, and chest X-ray showed total collapse of the left lung. Aspergillosis was confirmed by positive prick skin test, the presence of serum precipitins and the isolation of *A. fumigatus* from bronchial aspirate, with a blood eosinophilia. Eventual re-expansion of the lung was achieved by bronchoscopic suction and intensive postural drainage and percussion. She has remained well since moving away from the farm.

Case 4 (Fig. 4). E.E., farmer's son, aged 35 years. Experienced breathlessness and chest tightness without wheeze. X-ray showed pneumonia

in left lower lobe, with blood eosinophilia, which had not completely cleared 4 months later when he developed fresh infiltration in the right dorsal lobe. Aspergillosis was confirmed by positive prick skin test, the finding of serum precipitins, and isolation of *A. fumigatus* from sputum. He has remained well since wearing a mask during threshing and grinding operations.

Discussion

These cases show that when allergic aspergillosis occurs in more than one member of a family, all the main radiological features may occur, including transient pulmonary exudations, pus-filled bronchi, and atelectasis of segments or of the whole lung.

Although allergic aspergillosis is more commonly associated with asthma, there was no such history in these families.

The two boys in the first family (cases 1 and 2) both developed staphylococcal sinusitis necessitating repeated antral wash-outs from which *A. fumigatus* was also cultured in one of them. It is known that the upper respiratory tract may be the site of aspergillosis, as described by Montreuil (1955), Savetsky & Waltner (1961), Hora (1965) and others, but in none of their cases were the lungs affected.

In case 1 of this series, I believe that the sinuses were secondarily infected from the bronchi, and that they should always be screened for aspergillosis in such cases.

The presence of well-marked hilar adenitis in one tuberculin-negative boy, during episodes of pulmonary infiltration, is interesting, and is not seen in adult aspergillosis. Neither has it been described as part of the radiological picture in children, though it concurs with the necropsy findings of Strelling *et al.* (1966).

Neither boy had true bronchiectasis, though both showed the typical bronchographic appearance of suppurative bronchitis. It is likely that they re-infected one another, and the aspergillosis may have originated in the poultry meal.

Since the death of case 2 from cor pulmonale a year after aspergillosis was diagnosed, his brother has had no further infiltrations, but has latterly developed cor pulmonale himself.

I conclude that the aspergillosis in these two

boys was a late event, secondarily imposed upon long-standing upper and lower respiratory infection.

The farming family show a different picture. The mother (case 3), developed the unusual and serious complication of total collapse of the lung after previous infiltrations, and has been reported in an earlier paper (Ellis, 1965). Normally fit and well, both mother and son became ill following exposure to corn dust during threshing and grinding which was probably the source of infection. This impression is unsupported by the isolation of fungus from samples from the farm, or by high spore counts inside the buildings, but both have remained well since taking precautions against dust exposure.

The fact that other unrelated farmworkers were equally exposed without developing aspergillosis, raises the question of hereditary predisposition to the disease in this family. This would be rendered more likely should the farmer himself reveal evidence of aspergillosis during one of his regular exacerbations of bronchitis.

The demonstration of serum precipitins against mouldy-hay antigens in the daughter-in-law and a cowman on this farm without symptoms and signs of farmer's lung, illustrates the close association between this disease and aspergillosis, and indicates that serological surveys in farming communities might yield information of epidemiological interest with regard to these two diseases.

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