

## CASE REPORT

# A case of allergic bronchopulmonary aspergillosis in a 43-year-old farmer following single high level exposure to organic dust with symptomatic remission and radiological resolution after early diagnosis and treatment

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## Abstract

Allergic bronchopulmonary aspergillosis (ABPA) is an uncommon lung condition associated with development or worsening of asthma symptoms, distinctive radiological and serological findings. It is thought to be the consequence of chronic colonization of the airways in individuals with pre-existing atopic conditions. We present a case unique in the literature of the development of ABPA in an individual without pre-existing atopic disease following a single high level exposure to organic dust. Early treatment was associated with complete symptomatic remission and significant resolution of bronchiectatic changes.

## INTRODUCTION

Allergic bronchopulmonary aspergillosis (ABPA) is an uncommon lung condition associated with an allergic immunological response to airway *Aspergillus fumigatus*. It is normally seen in those with pre-existing asthma or cystic fibrosis occasionally other atopic disease [1]. It is associated with radiological changes including focal infiltrates, segmental lung collapse and bronchiectasis, microbiological and serological evidence of *Aspergillus*. These together form criteria for the diagnosis of this condition [2]. Little is known of the natural history of ABPA but, whilst infiltrates and collapse are associated with eosinophilic inflammation and are reversible changes, bronchiectasis and asthma, once established, are thought to be irreversible [3].

Here we report a case of a patient without pre-existing asthma or atopic disease developing ABPA after a single exposure to high concentration of fungal spores. Treatment including steroid and antifungal therapy was associated with complete resolution of asthma symptoms allowing treatment withdrawal and with partial resolution of bronchiectatic changes.

## CASE REPORT

A 43-year-old farmer presented to outpatients with a 3-week history of wheeze, shortness of breath and cough productive of firm rubbery mucous plugs. His symptoms started following a single exposure to clouds of dust produced during the disposal of piles of rotting hedge clippings. The cough and wheeze were

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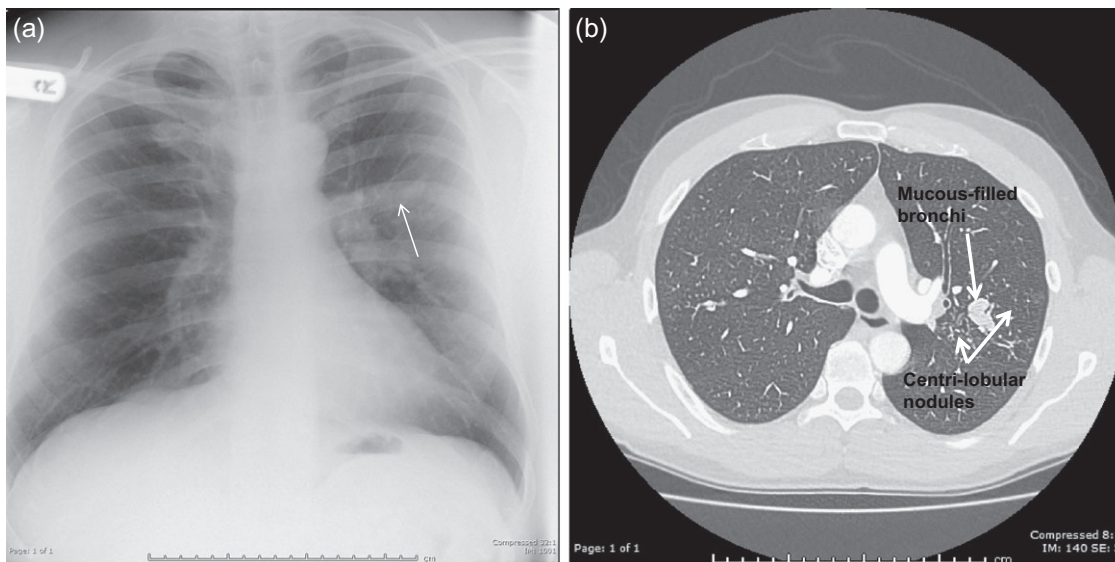


Figure 1: (a) CXR showing 'gloved finger' change (arrow). (b) CT thorax showing centri-lobular nodules and mucous-filled bronchi corresponding to CXR changes

worst at night and in the morning. There was no previous history of respiratory or atopic disease.

On examination there was widespread audible expiratory wheeze. His peak expiratory flow was 400 L/min (63% predicted). CXR showed bilateral upper zone infiltrates with left upper zone changes consistent with the 'gloved finger' change (Fig. 1).

CT thorax showed findings consistent with the diagnosis of ABPA: bilateral central bronchiectatic changes with multiple mucous-filled airways in the upper lobes corresponding to the CXR abnormality and associated with peripheral centri-lobular nodules (Fig. 1).

Blood tests showed high total IgE levels 1454 kU/L (ULN 81.0), strongly positive *A. fumigatus*-specific IgE 40.5 kU/L (ULN < 0.35) and elevated eosinophil count  $1.1 \times 10^9/L$  (ULN  $0.4 \times 10^9/L$ ), *Aspergillus* precipitins were negative but sputum culture grew *A. fumigatus* and cytological examination of bronchoalveolar lavage fluid showed branching fungal hyphae.

Based on diagnostic criteria the diagnosis of ABPA was confirmed and treatment started with oral Prednisolone 30 mg and Itraconazole 200 mg bd. His symptoms resolved rapidly with improvement in lung function PEF 700 L/min (111% predicted) FEV1 4.5 L/s (111% predicted) and clearance of CXR changes.

*Aspergillus* serology showed progressive improvement (Fig. 2) and total IgE decreased substantially to 333 kU/L.

After 12 months of therapy Prednisolone, Itraconazole 200 mg bd and inhaled corticosteroid therapy were stopped with no deterioration in symptoms or lung function.

A follow-up CT thorax at 30 months showed complete resolution of the centri-lobular nodules, infiltrates and mucous plugging with partial resolution of bronchiectatic changes (Fig. 3). The bronchiectasis persisted in the LUL bronchi most affected by mucous plugging.

## DISCUSSION

The diagnosis of ABPA is based on a number of criteria including asthma, total IgE > 412 IU/l, elevated *Aspergillus* species specific IgE and IgG, CXR infiltrates, central bronchiectasis and *Aspergillus* cultured from sputum and/or fungal hyphae seen in mucous plugs [2]. Our patient met all criteria barring IgG

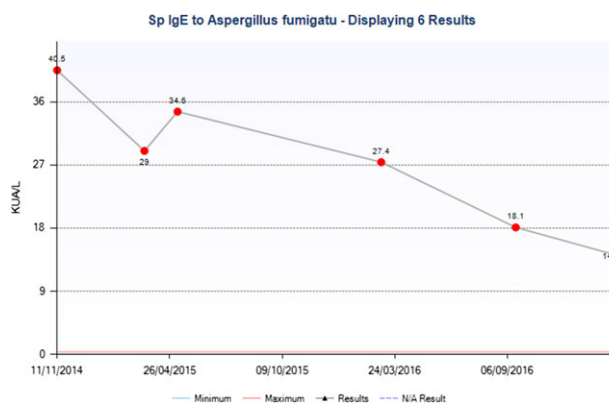


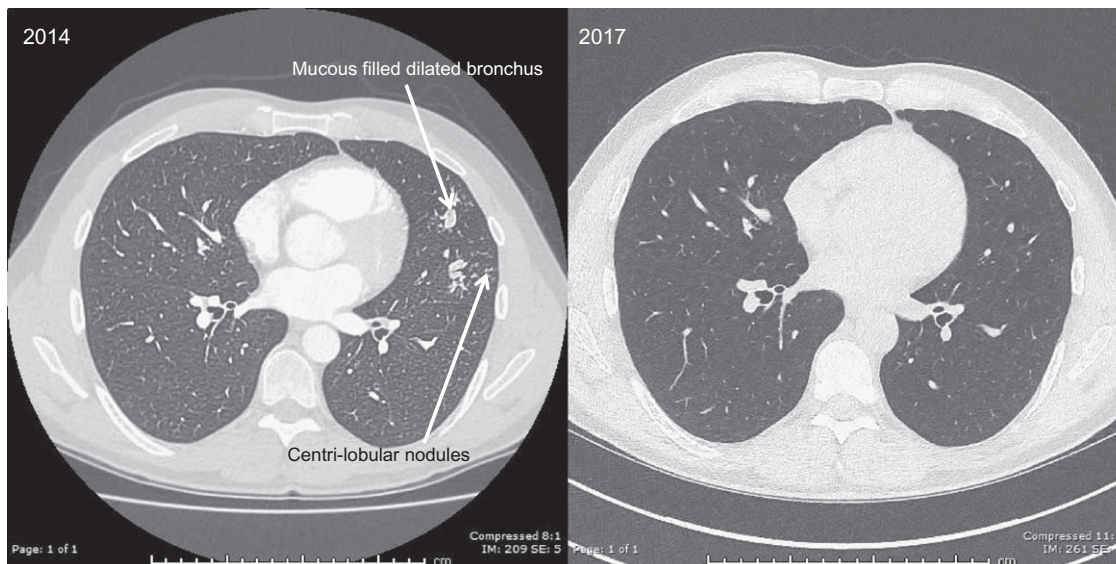
Figure 2: Time course of *Aspergillus fumigatus*-specific IgE titre before, during and after treatment

specific antibodies and in the context of his illness the diagnosis of ABPA seems secure. He described no symptoms suggesting pre-existing atopic lung disease and his Primary Care record did not suggest undiagnosed disease for example there was no history of recurrent chest infection or previous elevated blood eosinophil count.

Chronic airway colonization with *aspergillus* is thought to create an immunological response leading to ABPA [1]. *Aspergillus* is ubiquitous in the environment and index exposure is rarely identified. There are occasional case reports identifying a potential source of chronic exposure [4–6] but this is the first reported case of a single high level exposure leading to abrupt onset of symptoms.

Exposure to sensitizing allergens is known to cause asthma in those with no pre-existing atopic disease [7] but this is the first report of ABPA arising in an individual with no history of pre-existing atopy, asthma or cystic fibrosis.

Little is known of the natural history of ABPA but consistent with other conditions associated with asthma and with bronchiectasis, ABPA is thought of as a chronic disease with a relapsing remitting course and no cure [8]. Although treatment with oral steroids and Itraconazole improves symptoms and slows disease progression in many [9], once bronchiectatic or



**Figure 3:** CT thorax of lower lobe changes before (2014) and after (2017) treatment showing resolution of centri-lobular nodules and mucous impaction/bronchiectasis

fibrotic changes are established, they are not thought to be reversible. Here we present for the first time evidence that early diagnosis and intervention is associated with complete symptomatic remission of asthma with reversal of bronchiectatic change. Although *Aspergillus*-specific serological tests are markedly diminished and continue to fall, they are still elevated, some bronchiectatic change persists and the potential for future relapse remains. Our patient, however, remains symptom-free over 12 months post treatment cessation and complete cure with normalization of serology and continued resolution of bronchiectasis is possible.

## CONCLUSION

This case reports a number of new findings in ABPA. It is the first reported case of ABPA associated with a single high level exposure to *A. fumigatus*. It is the first report of ABPA occurring in an individual with no prior history of atopic disease. It is the first reported instance of complete and sustained symptomatic resolution, partial resolution of bronchiectatic change and potential ABPA cure. It highlights the potential importance of early diagnosis and treatment to the natural history of this condition.

## CONFLICT OF INTEREST STATEMENT

There is no conflict of interest.

## FUNDING

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## ETHICAL APPROVAL AND CONSENT

Patient consent was obtained.

## GUARANTOR

P.F.P. is the guarantor of this article.

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