

CASE REPORT

‘Fungal soup’: Report of two cases of tumour-like blocked pulmonary cavities with liquid content infected with aspergilli, a rare form of pulmonary aspergillosis

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*Tuberculosis and Chest Unit, Grantham Hospital, Aberdeen, Hong Kong, China***‘Fungal soup’: Report of two cases of tumour-like blocked pulmonary cavities with liquid content infected with aspergilli, a rare form of pulmonary aspergillosis**WONG CF, YAN SW, CHAN KW. *Respirology* 2008; 13: 306–308

Abstract: Two cases of a rare and uncommonly described form of *Aspergillus* lung disease were diagnosed from incidental CXR abnormalities. This strange presentation has been described in the literature as ‘tumour-like blocked pulmonary cavities with liquid content infected by *aspergilli*’. The details of these two cases are reported together with a discussion of the diagnostic features of the disease and its position in the spectrum of pulmonary diseases caused by *Aspergillus*.

Key words: aspergillosis, diagnosis, pulmonary.

INTRODUCTION

Aspergillus is one of the common aetiological agents for fungal infection in human subjects. It is well known that lung infection by this fungus displays a wide spectrum of manifestations.^{1–4} We report two cases of *Aspergillus* lung infection that did not fit any of the common and well-described forms of pulmonary aspergillosis (PA).

CASE 1

A 40-year-old housewife presented with a persistent shadow on CXR. This CXR abnormality had been noted for over 5 years, and the patient had been prescribed antituberculosis treatment twice at two different institutions despite the absence of bacteriological or histological evidence of tuberculosis. She had been asymptomatic over this whole period. Physical examination and preliminary investigations were unremarkable. Sputum culture for fungus and serology

for aspergillus precipitin antibody were negative. The CXR showed a homogenous lesion with a well-defined border over the left upper lobe (Fig. 1a). CT of the thorax (Fig. 1b) revealed a cyst-like lesion in the left upper lobe that probably contained fluid. Percutaneous transthoracic needle aspiration (PTNA) of the lesion yielded 10 mL of greyish pus-like fluid, which on histological examination revealed necrotic debris and inflammatory cells. On Grocott staining, slender septate hyphae consistent with *Aspergillus* were identified. Culture of the fluid also revealed the presence of *Aspergillus* species.

A course of antifungal treatment in the form of amphotericin B was administered, followed by oral itraconazole. Repeat thoracic CT after 3 months of itraconazole therapy showed a smaller left upper lobe lesion, but repeat PTNA still yielded pus-like fluid that was positive for *Aspergillus* on fungal staining and culture. The patient declined surgical resection of the lesion. Itraconazole therapy was continued for 6 months and then stopped. The patient was placed under observation and the lesion remained static with slight fluctuation in size on subsequent CXR. About 2 years later, the patient began to experience intermittent self-limiting bouts of coughing with mouthfuls of greyish sputum. Despite a smaller upper zone shadow on CXR, there were new left lower zone infiltrates. Clinical suspicion of spillage of the contents of the cavity into the lower lobe was later confirmed by thoracic CT. Another course of itraconazole was administered and the left lower lobe infiltrate resolved.

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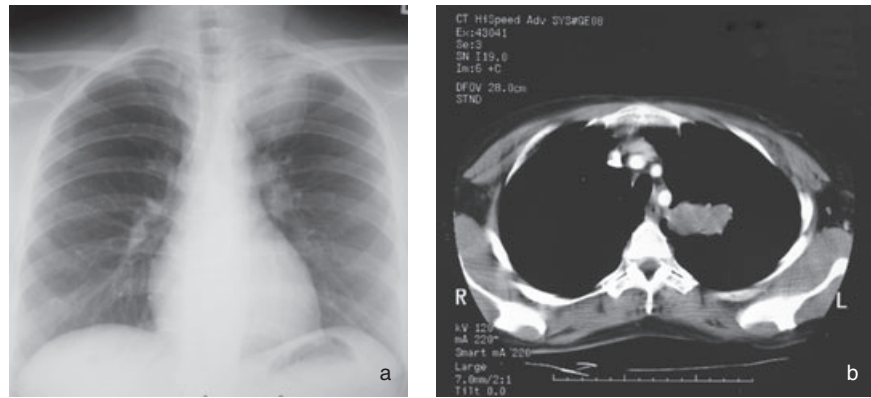


Figure 1 (a) CXR of case 1 showing a homogenous lesion in the left upper zone. (b) Thoracic CT of case 1 showing a cyst-like structure with fluid content in the left upper lobe.

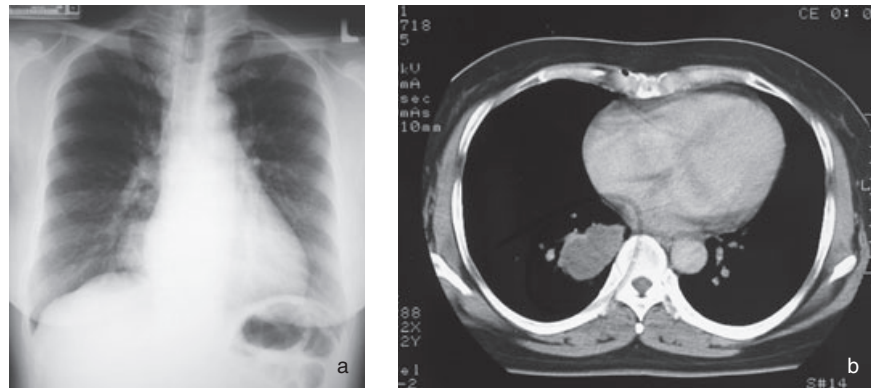


Figure 2 (a) CXR of case 2 showing a well-circumscribed lesion in the right lower zone. (b) Thoracic CT of case 2 showing a mass-like lesion in the right lower lobe.

The patient finally acquiesced to surgery and a lobectomy was performed by VAT surgery. Gross examination of the lung specimen showed a $5 \times 4.5 \times 3$ cm cavity with a thick brownish lining and consolidation of the lung tissue surrounding the cavity. There was no gross communication between the cavity and the bronchus. Microscopic examination showed an ectatic bronchus with intact epithelial lining and mild non-specific chronic inflammation of the bronchial wall. There was no evidence of fungal infection of the cavity wall or the surrounding lung.

CASE 2

A 41-year-old Philipino woman presented with an incidental CXR finding of a left lower zone shadow (Fig. 2a) during a routine health check. She had an unremarkable history and was completely asymptomatic. Thoracic CT showed a 4.4-cm mass-like lesion with well-defined borders at the postero-basal segment of the right lower lobe (Fig. 2b). Blood tests were unremarkable and sputum culture for fungus was negative. PTNA of the lesion yielded several millilitres of greyish pus-like fluid. On microscopic examination neutrophils and fungal hyphae were identified. Culture of the fluid revealed *Aspergillus fumigatus*. Itraconazole therapy was commenced and a right lower lobe segmentectomy was subsequently performed. Gross examination of the resected

specimen showed a $4 \times 3 \times 3$ cm cyst containing thick brownish turbid fluid. There was no gross communication between the cavity and the bronchus. Microscopic examination revealed an ectatic bronchiole with lining composed of ciliated pseudostratified cells. Below the epithelial surface, the cyst wall contained fibrous tissue, macrophages and inflammatory cells. A small focal group of hyphae was present on the cyst wall. There was no evidence of *Aspergillus* infection of the cavity wall or the subjacent lung.

DISCUSSION

Aspergillus is an ubiquitous soil-dwelling fungus. Human infections are usually acquired by inhalation of airborne spores from inanimate sources. PA can present as different clinicopathological entities, including pulmonary aspergilloma, chronic necrotizing pulmonary aspergillosis (CNPA), invasive pulmonary aspergillosis (IPA) and allergic bronchopulmonary aspergillosis (ABPA), depending on the atopic and immune status of the host and the site of involvement within the respiratory system.

The two cases of PA described in this report showed rare and unusual presentations. Both patients were middle-aged women who had no underlying systemic illness or chronic local lung disease. Because of their immunocompetent status IPA was unlikely. Neither patient had chronic respiratory symptoms on

presentation but both showed well-defined homogenous opacities on CXR. The absence of pre-existing cavities or an air-crescent indicated these were not typical cases of aspergilloma. There were also no features of allergic reactions suggesting ABPA. The lesion contained a fluid substance shown on examination to be infected with *Aspergillus*. Pathologically, there was no communication with the bronchi. There was no invasion of the subjacent lung tissue or vessels. This 'non-invasiveness' differs from CNPA and IPA.

On review of the literature, these two cases did not fit any of the PA syndromes. However, this condition was described in 1978 by Rzepecki *et al.* who reported a surgical case-series of 10 patients with 'tumour-like blocked pulmonary cavities with liquid content infected with aspergilli' (TBLA).^{5,6} Four characteristics of the condition were highlighted. First, it appeared as a tumour-like mass on CXR (thoracic CT was not readily available at that time). Second, it was a pulmonary cavity with blocked bronchus which was responsible for the round or oval shape of the distended cysts with their sharp contours and homogenous appearance on CXR. Third, it contained liquid and finally, the liquid invariably grew *Aspergillus*. It suggested that the basic pathological feature was the occlusion of the bronchus forming a cystic structure. Deficient drainage and access to oxygen resulted in the formation of a fluid-filled cyst rather than a fungal ball growing in a pre-existing cavity. The condition was thought to be a variant form of pulmonary aspergilloma, but the many differences from ordinary pulmonary aspergilloma justified it being regarded as a distinct entity.^{5,6} The natural course, prognosis and optimum management were unclear due to the rarity of the condition.

From our experience with these two cases, it appears that *Aspergillus* cannot be eradicated simply

by the administration of antifungal agents. Although there was no demonstrable communication, the cyst may empty its contents into the airway owing to excessive pressure within the lesion. Therefore, the most effective treatment for TBLA is likely to be surgical resection with antifungal coverage, as demonstrated in case 2.

We have reported two cases of an interesting and unique form of PA, first described as TBLA by Rzepecki *et al.*^{5,6} Its exact position in the spectrum of PA is not well defined. Whether it is a rare 'fungal soup' variant of pulmonary aspergilloma or a distinct form of *Aspergillus* lung infection needs further exploration.

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