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Endobronchial zygomycosis

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Zygomycosis is an opportunistic fungal infection, some forms of which may also be known as mucormycosis; the older term for these conditions, phycomycosis, is obsolete.[1] The most characteristic clinical form is the rhinocerebral type, which is often associated with diabetes mellitus. Pulmonary zygomycosis may also be associated with diabetes, though leukaemia and lymphoma, particularly when they follow cytotoxic chemotherapy, are also frequent predisposing factors.[1] We present a case in which zygomycosis, limited almost entirely to an airway, was an incidental necropsy finding.

Case report

An 83 year old woman, a sufferer from idiopathic Parkinsonism, was transferred to Manchester Royal Infirmary for neurosurgical evaluation after head injury. She had fallen down a flight of stairs at home and, after a period of lucidity, her neurological condition deteriorated rapidly, so that she was unconscious on admission. She died two days later.

External examination at necropsy showed numerous haematomas and abrasions, including some over the head. There were fractures of the pelvis and of several ribs, but the skull was intact. A recent large right subdural haematoma was present. The brain showed widespread contusions and focal softening of the right occipital lobe and the left cerebellar hemisphere. The other organs were unremarkable, apart from a plug of firm green material in a small bronchus in the upper lobe of the right lung. The infective nature of this lesion was not appreciated at this stage, so culture was not attempted.

Microscopic examination of the pulmonary lesion showed a tangled mass of fungal hyphae occupying the lumen at a bronchial bifurcation. The hyphae were broad, but irregularly so, ranging in diameter from about 5 to 20 μm. There were very few septa, and branching was randomly spaced and at right angles throughout. Many hyphae had an irregular and "kinked" outline (figure). There was superficial ulceration of the bronchial mucosa, with only very shallow hyphal penetration of the bronchial wall. Many sections were examined, with no evidence of vascular invasion apart from a few hyphae in a small vessel in the angle at the bifurcation of the affected bronchi.

The histological diagnosis of zygomycosis was thus established before formal neuropathological examination of the brain could be carried out, and so we initially assumed that some or all of the damaged areas within the brain would prove to be infarcts or abscesses induced by embolic spread of zygomycosis from the lung. Despite careful examination of many histological sections of each of the macroscopically abnormal areas in the brain, however, we could find no evidence of zygomycosis.

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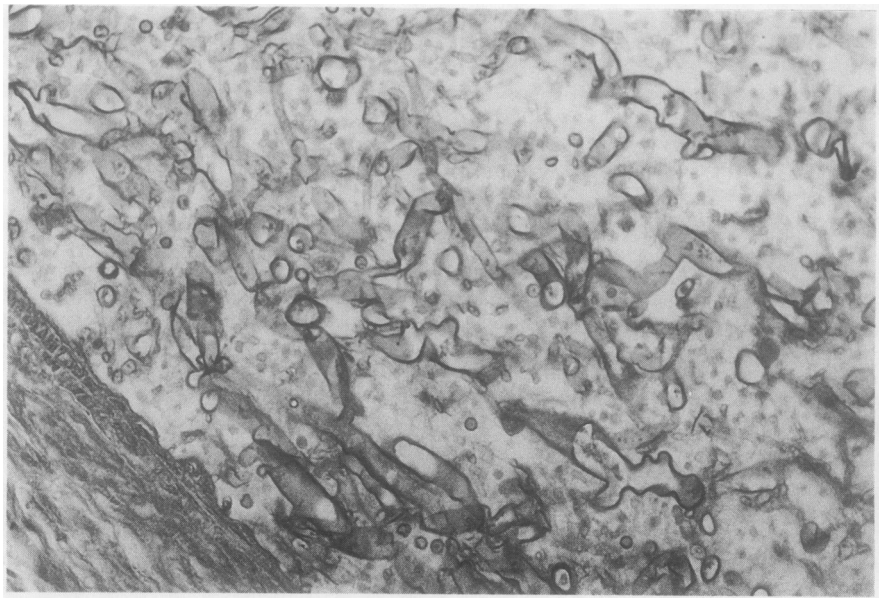
Discussion

The main differential diagnosis of the endobronchial mass was an aspergillus fungus ball. In that condition the hyphae form neat, fan-like arrays with regular dichotomous branching and frequent septa. Zygomycosis is characterised by tangled masses of hyphae that are more variable in diameter and generally broader than those in aspergillosis; septa are scanty, and branching is irregular and roughly at right angles. Gross hyphal kinking and irregularity of outline are often seen.

Confirmation of zygomycosis in life may be carried out on culture or biopsy material, and preferably on both. Unfortunately, culture is often unsuccessful. Some recent reports have included details of serological confirmation,[2–4] but this is not yet widely available; skin testing has no useful role.

Zygomycosis of the lower respiratory tree is usually in the form of patchy pneumonia or of abscesses; there are very few reported examples in which the fungal infection has been largely limited to the airways or a pre-existing peripheral pulmonary cavity.[2 5–9] In all those examples the fungal infection was clinically important, and so our case is unusual because the endobronchial zygomycosis was a chance finding of no immediate clinical relevance. Most of the previous cases were associated with diabetes mellitus[5–7]; one followed multiple urinary tract infections in an infant,[8] and another occurred in a cavity apparently left after lobectomy for tuberculosis many years earlier.[2] Our patient's blood sugar concentration was not found to be raised during her short period of clinical observation, and so her glucose tolerance was not assessed. Diabetes mellitus cannot therefore be excluded.

The few published reports suggest that endobronchial zygomycosis may present with a cough that may be productive[2 5] or non-productive.[7] Haemoptysis may occur, and may range in severity from trivial[2] to fatal.[6 9] In one bizarre case tracheal zygomycosis caused acute airways obstruction,[8] and in the only paediatric case on record zygomycotic bronchial stenosis led to rapidly progressive hyperinflation of one lung.[9] The prognosis is poor, for half the patients in reported cases died, either of massive haemoptysis[6 9] or after surgery.[2] All the survivors had had local resection,[5 7–9] usually supplemented with amphotericin B.



The fungal mass in the bronchus: at this point the bronchial epithelium has been eroded, but there is no invasion of the wall. (Grocott stain.)

Invasion of adjacent tissue, such as bronchial wall[5] or pulmonary arteries,[6 9] occurs frequently and readily. It is therefore imperative, if the diagnosis is established in life, that resection should be carried out as soon as possible. Amphotericin B is the only antifungal agent that is ever of clear benefit in systemic zygomycosis, and it should be used promptly.

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