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Isolated sphenoid sinusitis due to *Pseudallescheria boydii*

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Abstract

A case of sphenoid sinusitis due to *Pseudallescheria boydii* is described in a 52-year-old non-immunocompromised woman. Treatment should always involve surgical drainage, and antifungal chemotherapy may be of benefit if there is histological evidence of invasion of surrounding tissue. For *P. boydii* infection miconazole should be the agent of choice, rather than amphotericin B. For this reason it is important to obtain culture and histological examination of sinus contents if fungal infection is suspected.

Key words: Sphenoid sinus; *Pseudallescheria boydii*

Introduction

Isolated sphenoid sinusitis is rare, with disease due to fungal infection being even more unusual. Lew *et al.* (1983) calculated the incidence at 2.7 per cent, and in their series of 30 cases none were due to fungal infection. Stammberger (1991) reports the incidence of mycotic infection as almost 10 per cent in all patients requiring surgical management for sinusitis. *Aspergillus fumigatus* is the most common species implicated in paranasal sinus infection (Romett and Newman, 1982) with other species being rarely reported. To our knowledge there have been only two previous reported cases of *Pseudallescheria boydii* (*Petriellidium boydii*, *Allescheria boydii*) infection of the sphenoid sinus (Mader *et al.*, 1978; Bryan *et al.*, 1980), and one case of sphenoethmoid sinusitis (Salitan *et al.*, 1990). All these cases presented in the USA and we believe this to be the first report of such a case in the United Kingdom.

Case report

A 52-year-old woman was referred with a 12 month history of right facial pain. She described the pain as being in the frontal and maxillary regions, with radiation to the temple and ear. Her only other symptom was mild post-nasal drip. On examination the only abnormality was crepitus over her right temporomandibular joint, and in particular there were no signs of cranial nerve palsy. In view of the history, a CT scan of her sinuses was arranged, and this showed opacity of the right posterior ethmoid and sphenoid sinuses (Fig. 1). The bony margins of the right sphenoid sinus appeared to be sclerotic. Repeat nasendoscopy after the scan did reveal a few small polyps behind the middle turbinate on the right side.

Admission was arranged and she subsequently underwent an endoscopic ethmoidectomy with opening of the right sphenoid sinus. At operation the anterior ethmoid sinus was normal, but the posterior ethmoid air cells were full of granulation tissue. The sphenoid sinus was opened to reveal 'inspissated debris' with an appearance like that of cholesteatoma.

At operation fungal infection was suspected, and this was confirmed on histological examination which showed the tissue from the sphenoid sinus was a fungal ball. The fungus had septate and branching hyphae (Fig. 2). The surrounding tissue contained a substantial infiltrate of plasma cells, together with some neutrophil and eosinophil polymorphs. There was no evidence of fungal invasion into surrounding tissue or blood vessels. Culture subsequently showed the fungus to be *P. boydii*.

At follow-up, two weeks after surgery, her symptoms had almost resolved. Six weeks after surgery she was well, and nasendoscopy revealed healthy mucosa in the open sphenoid and ethmoid sinuses. There was no obvious residual fungal infection. Repeat CT scan three months after surgery showed a well aerated sphenoid sinus with no evidence of residual disease (Fig. 3).

Discussion

P. boydii is a saprophyte frequently isolated from soil, manure

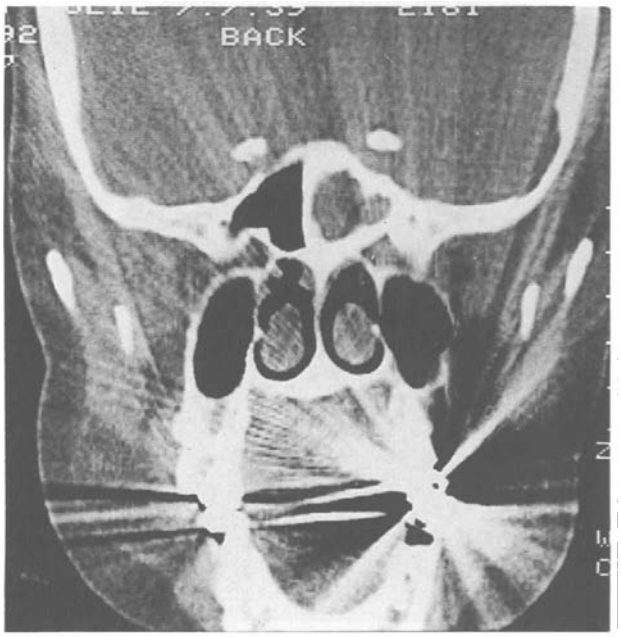

FIG. 1

Coronal CT scan showing an opaque right sphenoid sinus with sclerosis of surrounding bone.

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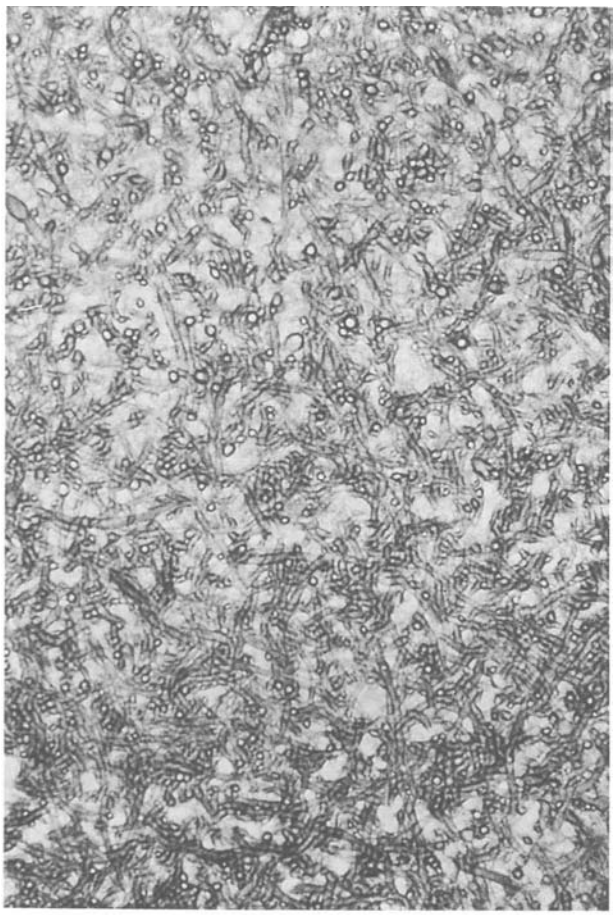

FIG. 2
Contents of sphenoid sinus showing fungus with septate and branching hyphae (PAS).

and decaying vegetation (Bennett, 1990). It enters the body through a penetrating injury or by an airborne route. In subcutaneous tissue the fungus grows in visible clumps and the resulting chronic infection is referred to as mycetoma. Other sites of infection include pulmonary cavities and ectatic bronchi in patients with chronic obstructive lung disease. Joints may be infected with *P. boydii* following penetrating injury and the eye may develop infection after corneal trauma. *P. boydii* can also grow in the external auditory canal and cause a form of otitis externa called otomycosis. When there is fungal invasion or dissemination from any site the condition is referred to as Pseudallescheriasis.

Diagnosis of *P. boydii* relies on culture of the fungus, as on histological examination it may be impossible to distinguish the hyphae of *P. boydii* from those of *Aspergillus* (Mader *et al.*, 1978). Both have septate, branching hyphae which may show areas of bulbous swelling. However, histological examination is important in detecting invasion of surrounding tissue or blood vessels. This case illustrates the importance of obtaining prompt tissue culture and histological examination when findings at operation are unusual.

Unlike *Aspergillus* and *Mucoraceae*, *P. boydii* is rarely invasive and most human infections result in a mycetoma or 'fungal ball'. Our case was of this form of infection, although two of the previous cases involving the sphenoid sinus did show invasion with intracranial extension (Bryan *et al.*, 1980; Salitan *et al.*, 1990). *Aspergillus* is now recognized as causing an allergic form of sinusitis (Jonathan *et al.*, 1989) but as yet no other fungi have been found to cause this condition. However, one report of allergic bronchopulmonary disease suggests that *P. boydii* may be responsible for an allergic form of fungal disease (Lake *et al.*, 1990).

Treatment of *P.boydii* infection of the paranasal sinuses is along similar lines to that of Aspergillosis. However, unlike *Aspergillus*, response to antifungal chemotherapy agents is poor, so adequate surgical drainage should be the principal treatment in all cases (Winn *et al.*, 1983). If there is no evidence of fungal invasion of surrounding tissue on histological examination, then no further treatment is required. Should invasion be present then adjuvant chemotherapy may be of some benefit. However, in one case where bony erosion was noted at operation, cure was achieved by surgery alone (Winn *et al.*, 1983). When chemotherapy is used, miconazole should be the agent of choice as *P. boydii*, unlike *Aspergillus*, is relatively resistant to amphotericin B (Lutwick *et al.*, 1979; Salitan *et al.*, 1990). For this reason it is important to distinguish between the two fungi with culture, rather than relying on a diagnosis based on histological appearance.

As with *Aspergillus*, the invasive form of infection with *P. boydii* is more often seen in patients who are immunocompromised. Examples of invasive sinus disease have been reported in a diabetic (Gluckman *et al.*, 1977) and a patient with haematological malignancy (Hecht *et al.*, 1978). Our patient had no evidence of immune deficiency and the fungal infection was of the mycetoma form. However, *P. boydii* has been found to be invasive in non-compromised patients in two cases (Bryan *et al.*, 1980; Salitan *et al.*, 1990), with the earlier case resulting in the death of the patient.

Sphenoidal disease is difficult to diagnose due to its inaccessability. The classic symptom of sphenoid sinus pathology is pain localizing to the vertex, though this may be absent, or obscured if complications are present. This may be illustrated by our patient, whose only symptom was facial pain. This could have been due to irritation of the trigeminal nerve, though formal examination of the nerve was normal. Due to the lack of nasal symptoms, isolated sphenoid sinusitis may not present until complications have occurred (Deans and Welch, 1991). These complications are often serious due to the close proximity of important structures. Disease may spread to both intracranial and orbital compartments, and the initimate relationship to cranial nerves II to VI results in sphenoid sinusitis often presenting to the neurologist rather than the ENT surgeon (Brockbank and Brookes, 1991). As in our case, the diagnosis is often not made until a CT scan is performed, and this should be the definitive investigation in sphenoid sinus disease. Plain X-ray alone is

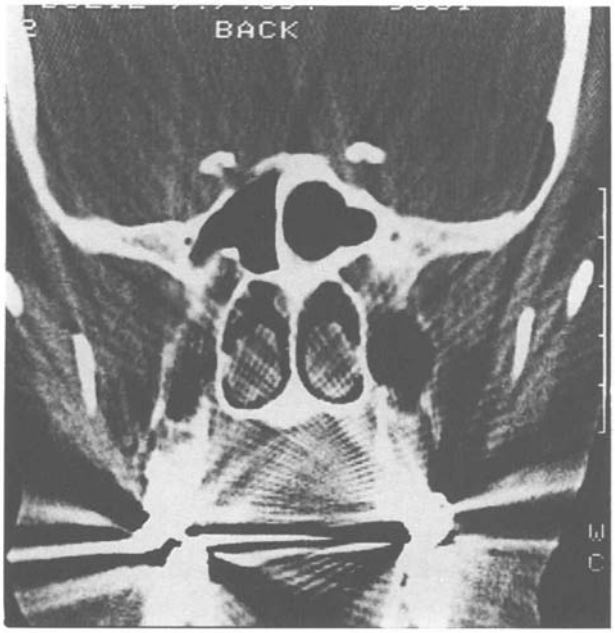

FIG. 3
Post-operative coronal CT scan showing a well-aerated sphenoid sinus.

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unreliable (Deans and Welsh, 1991). Furthermore, with the advent of CT scanning and nasendoscopy as a routine part of outpatient assessment of sinus disease, all forms of fungal sinusitis are likely to become increasingly recognized.

Conclusion

Isolated sphenoid sinusitis is difficult to diagnose, but should be considered in the differential diagnosis of anyone complaining of facial pain or headache. Fungal disease should always be treated by surgical drainage and aeration of the sinus, and it is important to obtain a histological examination and culture of the sinus contents. Further treatment with antifungal chemotherapy can be considered if invasion of surrounding tissue is present, and the choice of chemotherapy agent will depend on the fungus involved. *P. boydii* shows a significant degree of resistance to amphotericin B, so miconazole should be used instead. In view of this it is important to distinguish between *P. boydii* and *Aspergillus*, which is a more common fungal pathogen in the paranasal sinuses. This relies on culture of the fungus rather than histological appearance.

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