Symptomatic perineural extension of fungal sinusitis in an immunocompetent person: imaging features

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ABSTRACT

Perineural spread is a rare mode of spread for fungal infection, even more so in immunocompetent individuals. We report a 21-year-old immunocompetent man in which symptomatic perineural extension of aspergillosis along the maxillary division of trigeminal nerve was accurately diagnosed on imaging.

Keywords: aspergillosis, computed tomography, fungal sinusitis, magnetic resonance imaging, perineural spread, trigeminal nerve

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INTRODUCTION

The severity of aspergillus infection is dictated largely by the underlying immune status of the patient.[1,2] Clinical spectrum ranges from noninvasive sinusitis or fungal ball in immunocompetent patients, to fulminant invasive forms in immunocompromised patients.[1] Although invasive aspergillosis typically occurs in hosts with lowered resistance, a few reports have emphasised its occurrence in immunocompetent individuals.[3-5] In its invasive form, aspergillosis typically spreads by osseous erosion or vascular invasion. Perineural spread of aspergillosis is very rare, with less than ten cases reported in the literature.[3] We report a case of invasive aspergillosis with symptomatic perineural involvement in an immunocompetent patient, which to the best of our knowledge, has been reported only once in the literature.[3] The imaging features are highlighted and a brief review is presented.

CASE REPORT

A 21-year-old immunocompetent man, a farmer by occupation, presented with a six-month history of painful right eye proptosis, along with lid swelling and paraesthesia involving the right side of the face. The symptoms were gradually increasing. Ophthalmological examination revealed proptosis of the right eye with conjunctival congestion and painful ocular movements. Laboratory investigations including haemogram and blood glucose levels were normal, and human immunodeficinecy virus (HIV) serology was negative. The patient was further evaluated with computed tomography (CT) and magnetic resonance (MR) imaging.

CT revealed a soft tissue lesion in the right posterior ethmoid and sphenoid sinuses, with erosion of medial orbital wall. Contiguous extension into the orbit and pterygopalatine fossa was present, along with soft tissue causing widening of the foramen rotundum, vidian canal and proximal palatine canal (Figs.1 & 2). Hyperdense areas were present within the mass on non-contrast CT (Fig. 1). Subsequently, contrast-enhanced MR imaging was performed, which showed a T1- and T2- iso- to hypointense, enhancing mass in the right posterior ethmoid and sphenoid sinus with contiguous extension into the right orbit, causing enlargement of the medial and inferior rectus muscles (Fig. 3). The lesion extended into the orbital apex and via the inferior orbital fissure into the pterygo-palatine fossa, from where there was further retrograde extension through the foramen rotundum into the middle cranial fossa (Fig. 3).

Imaging findings, including hypointense signal on T2-weighted images and hyperdense areas on non-contrast CT, suggested fungal infection. The patient underwent an endoscopic biopsy that revealed tissue invasion by multiseptate fungal hyphae, suggestive of aspergillosis. Proptosis, pain and eyelid swelling resolved with itraconazole treatment. At one year follow-up, the patient has some residual numbness; however, the tingling sensation has resolved completely. He is therefore on expectant management in view of the good clinical response.

DISCUSSION

Aspergillus infection of the paranasal sinuses and orbit can be noninvasive or invasive. Noninvasive aspergillus infection manifests as allergic sinusitis or mycetoma, is confined to the sinuses and is usually seen in immunocompetent individuals. It may cause bony expansion, but there is no invasion of tissue or

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Fig. 1 (a) Unenhanced axial CT image shows a heterogeneous lesion with hyperdense areas in the right posterior ethmoid and sphenoid sinus that is extending into the orbit. At a more caudal level (b,c), soft tissue extension is seen within the foramen rotundum (long arrow in Fig. 1b), pterygopalatine fossa (star in Fig. 1c) and in the vidian canal (arrowhead in Fig. 1c). The involved foramina are widened compared to the normal side (foramen rotundum: short arrow in Fig. 1b, pterygopalatine fossa: curved arrow Fig. 1c). (d) Unenhanced coronal CT image taken at the level of the orbital apex shows contiguous extension of the lesion via the inferior orbital fissure (long arrow) into the pterygopalatine fossa and the sphenopalatine foramen (star). The right inferior orbital fissure (long arrow) is widened compared to the left side (short arrow).

bone destruction. Invasive infection, on the other hand, pursues a fulminant course. It may extend contiguously from the sinuses into the orbit and brain by causing bone erosion, and less commonly, can affect multiple organs through haematogeneous spread.[1]

Invasive aspergillosis is typically encountered in immunocompromised patients, primary risk factors being neutrophil defects and corticosteroid use. Other predisposing factors include HIV infection, diabetes mellitus, trauma, use of prosthetic devices and possibly, advanced age. Invasive aspergillosis is being increasingly reported in immunocompetent patients as well.[3-5] It is postulated that hot and dry environmental conditions, poor hygiene and low socioeconomic status may contribute to tissue invasive diseases in immunocompetent patients.[4] Our patient was a farmer of low socioeconomic status, and who, although immunocompetent, may have had excessive environmental exposure.

Paraesthesia or anaesthesia in the area of the fifth cranial nerve distribution is one of earliest symptoms of perineural extension. Although published articles on perineural extension of fungus are few, a large series reported facial numbness as the first symptom in 7% of patients with mucormycosis.[6] The authors emphasised paraesthesia as a less recognised but an early manifestation of fungal infection. The importance of anaesthesia was also stressed in another study, and resolution of anesthesia was reported as a clinical marker of response to therapy.[7] Our patient had paraesthesia of the right side of the face in the distribution of the maxillary division of the trigeminal

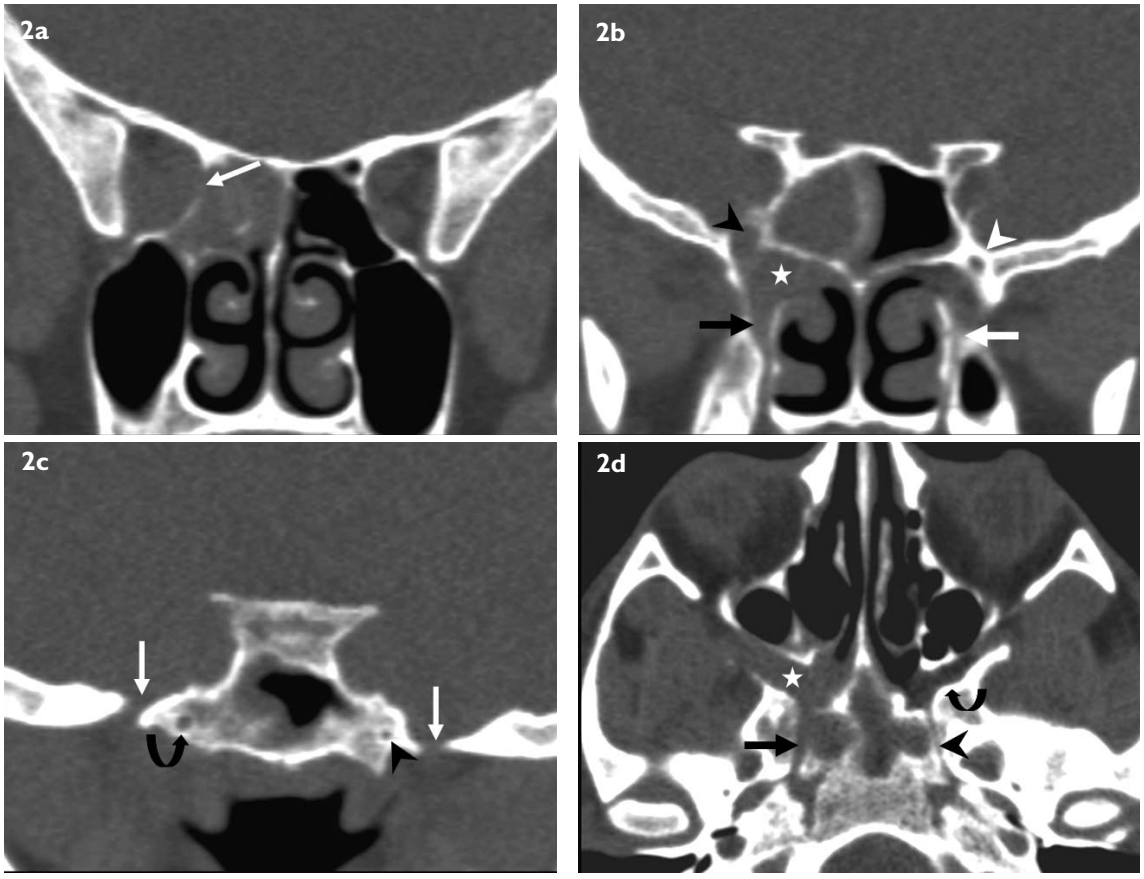

Fig. 2 Coronal (a–c, anterior to posterior) and (d) axial CT images taken with bone window setting show erosion of the medial orbital wall (arrow in Fig. 2a), soft tissue causing distension of the pterygopalatine fossa/sphenopalatine foramen (star in Figs. 2b & d), foramen rotundum (black arrowhead in Fig. 2b), proximal portion of the palatine canal (black arrow in Fig. 2b) and the vidian canal (curved black arrow in Fig. 2c and black arrow in Fig. 2d). Normal calibre pterygopalatine fossa (curved black arrow in Fig. 2d), foramen rotundum (white arrowhead in Fig. 2b), palatine canal (white arrow in Fig. 2b) and the vidian canal (arrowhead in Figs. 2c & d) are seen on the left side. The foramen ovale are indicated by white arrows in Fig. 2c.

nerve, which however did not respond to antifungal treatment.

In the largest series describing invasive fungal infection in immunocompetent patients, 25 patients were assessed.[4] However, none of these had perineural extension. The majority of these patients had bone erosion and showed a T2-hypointense mass with homogeneous enhancement. Aspergillus infection is characterised by hyperdensity on non-contrast CT and has typical MR imaging signal characteristics that are well exemplified in our case. The lesion is typically hypointense on T2-weighted images, appears hypo- to isointense on T1-weighted images, and shows homogeneous enhancement. These features in the appropriate clinical setting are helpful in early diagnosis and management of aspergillosis of sinonasal origin in immunocompetent hosts.[3,5] T2 hypointensity possibly results from iron accumulation by the fungus.[5]

Perineural spread is a well-recognised mode of spread in head and neck malignancies, in which the tumour disseminates to noncontiguous regions along the endoneurium or perineurium. It is rarely described as a mode of spread in invasive aspergillosis,[8-11] with only one published report in an immunocompetent patient.[3] Both CT and MR imaging can help detect perineural spread. Although MR imaging is the modality of choice because of its superior soft-tissue contrast,[5,8,9] multidetector CT with its higher, isotropic spatial resolution and superior quality multiplanar reformats, helps to delineate the small neural foramina better, as in our case. The direct signs of perineural spread are enlargement, irregularity and excessive enhancement of the nerve, with obliteration of fat in the foramina.[8] In our patient, widening and soft tissue attenuation within the bony foramina was seen clearly on CT (Figs.1 & 2). The replacement of fat in the pterygopalatine fossa, thickened and enhancing maxillary division of the trigeminal nerve on MR imaging, further confirmed perineural involvement (Fig. 3).

The treatment of invasive sinonasal aspergillosis is combined surgical debridement and antifungal therapy.[12] The use of preoperative administration of itraconazole improved outcome.[4] Our patient reported significant improvement on treatment with itraconazole. Proptosis and conjunctival congestion disappeared, although patient still had paraesthesia

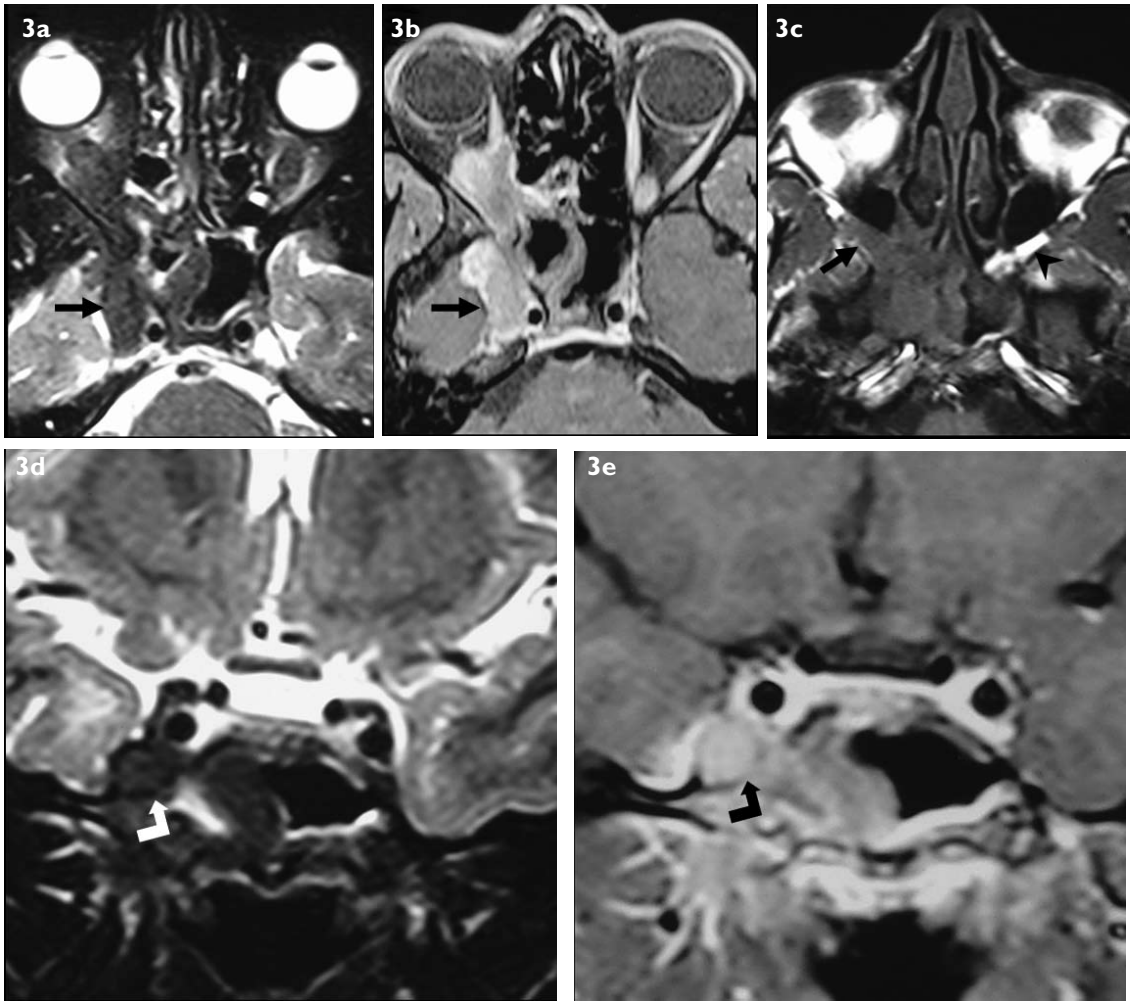

Fig. 3 Axial (a) fat suppressed T2-W, and (b) corresponding contrast-enhanced T1-W MR images show a T2-hypointense, enhancing lesion in the right orbit and sphenoid sinus with intracranial extension along the foramen rotundum (arrow). (c) Axial T1-W MR image at a more caudal level shows replacement of fat in the right pterygopalatine fossa by isointense soft-tissue (arrow). The left side pterygopalatine fossa shows normal (fat) signal (arrowhead). (d) Coronal fat suppressed T2-W, and (e) corresponding contrast-enhanced T1-W MR images show T2-hypointense, homogeneously-enhancing soft tissue causing enlargement of the foramen rotundum (block arrow).

for which he was awaiting surgical debridement. In conclusion, this report is unique because it emphasises the simultaneous occurrence of two rare manifestations, namely: tissue invasion by aspergillus infection in an immunocompetent patient, and perineural spread; both were accurately diagnosed on imaging. It is important to recognise these uncommon features of aspergillus infection and their imaging manifestations, so as to institute appropriate therapy at the earliest opportunity.

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