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Case report

Brain lesion in a recreational drug user: Isolated cerebral mucormycosis

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ABSTRACT

Isolated cerebral mucormycosis is a relatively rare and unique variant of mucormycosis which is seen most commonly in patients with intravenous recreational drug use. While this invasive fungal infection in the brain is thought to spread from the sinuses or the lungs in other hosts such as diabetics and those with malignancy, hematogenous spread and seeding has been attributed in the pathogenesis of isolated cerebral mucormycosis. Clinical features and radiological findings may be non-specific and hence, heightened clinical suspicion for a prompt diagnosis and early medical and surgical intervention is paramount for a favorable outcome in such rare, but potentially fatal infections.

Introduction

Mucormycosis is an aggressive and potentially fatal infection, commonly encountered in the severely immunocompromised hosts [1]. Patients with recreational intravenous drug use are at risk for a unique entity of mucormycosis – isolated cerebral infection, where basal ganglia tend to be preferentially affected [1,2]. Since this presentation is not very common and the clinical features are not distinguishable from some other infectious and non-infectious entities, suspicion for this cerebral mucormycosis is critical so that prompt and appropriate intervention can be instituted [2,3].

Case

A 36-year-old male with medical history of active polysubstance use (marijuana and cocaine) and chronic hepatitis C was brought to the hospital for altered mental status. Upon presentation, his vital signs were remarkable for a temperature of 38.8 C, heart rate of 100 beats/min and blood pressure of 136/61 mm Hg. On physical examination, he was lethargic, aphasic, and had a right facial droop with decreased right upper extremity strength. Laboratory data revealed white blood cell count was 15,700 cells per mm^3, hemoglobin 14 gm/dL and platelet count was 176,000 cells per mm^3. Urine toxicology showed presence of cocaine and the HIV test was negative. The patient was admitted to the neurosurgical intensive care unit as he needed to be intubated for airway protection. Further work-up included a CT scan of the head which revealed an extensive white matter hypodensity in the left frontal and temporal lobes including capsular white matter with a subtle ring-like region in the posterior left basal ganglionic/left thalamic region with a 5 mm left to right mid-line shift (Fig. 1). MRI imaging showed abnormal T2 signal intensity of left basal ganglia, left internal capsule and left thalamus without enhancement and with some hyperintense diffusion weighted imaging (DWI) signal intensity (Fig. 1). The radiological imaging was suggestive of an infiltrative tumor with DWI hyperintensity thought to be from hypercellularity; abscess was in the differential. The patient was started on ceftriaxone, vancomycin and metronidazole as well as dexamethasone, while work up including blood cultures was in process. Chest x-ray showed no cardiorespiratory abnormality and the CT and MRI head imaging did not reveal any sinus or orbital pathology.

The patient failed to improve on antimicrobials and his neurological examination remained unchanged 48 h into admission. The blood cultures were negative. Repeat MRI imaging of the brain showed an expansile left basal ganglionic/thalamic mass now with expansion across midline with 6–7 mm of left to right midline shift again with patchy DWI signal intensity (Fig. 1). He was taken to the operating room for resection of the left basal ganglia mass. On-site evaluation of frozen section showed inflammatory changes and fungal (hyphal) elements (Figs. 2, 3). Given these findings, further resection was not pursued, and the cranium was replaced. Diagnosis of cerebral mucormycosis was

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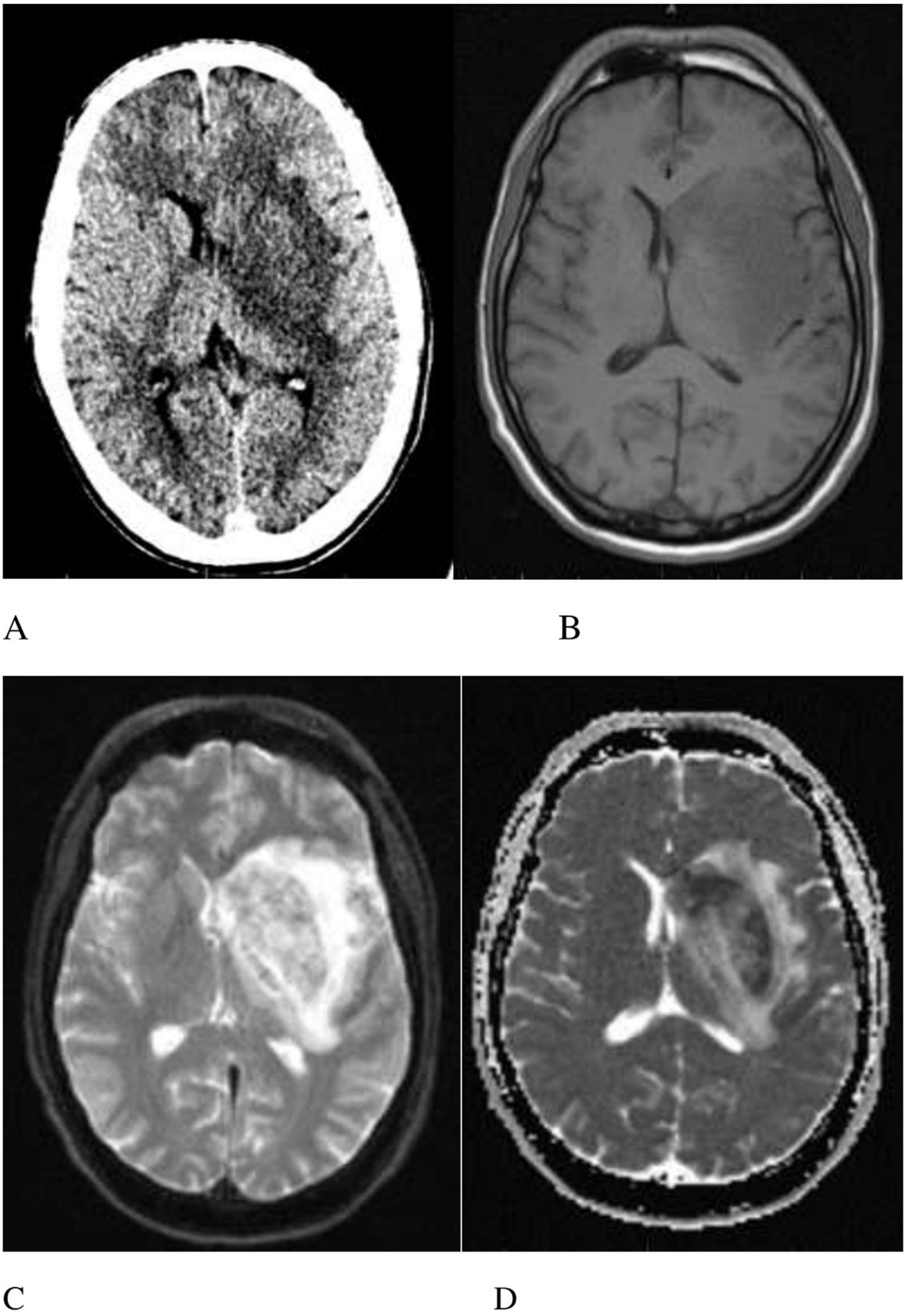

Fig. 1. An axial CT of the brain shows a mass like region in the posterior left basal ganglionic and left thalamic region (panel A). A T1-weighted MRI with gadolinium shows a hypodense expansile mass with patchy minimal enhancement (panel B). T2- weighted MRI demonstrates a hyperintense lesion with hypointense central region (panel C). Diffusion-weighted MR imaging (DWI) shows a central necrotic lesion with ring-enhancement is seen in the left basal region on DWI (panel D).

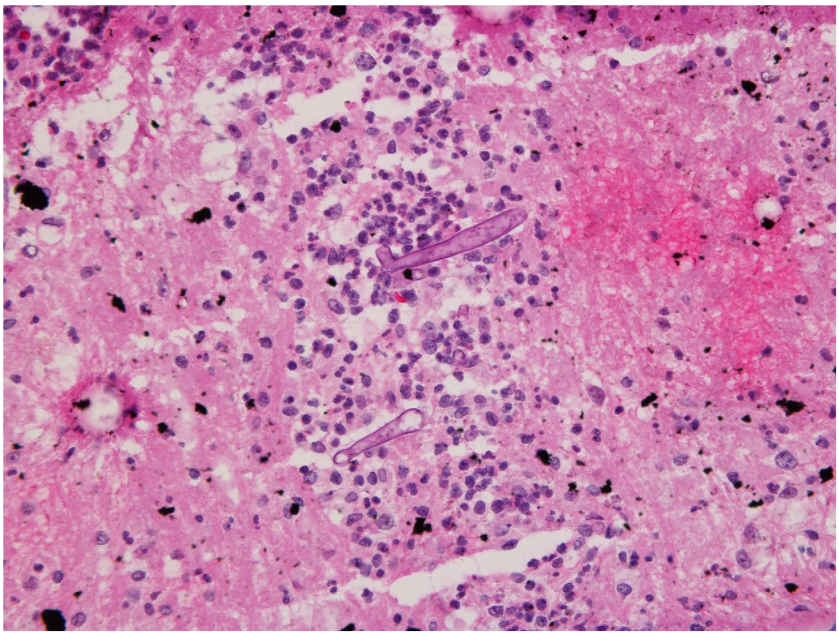

Fig. 2. Aseptate hyphae amidst inflammatory cells.

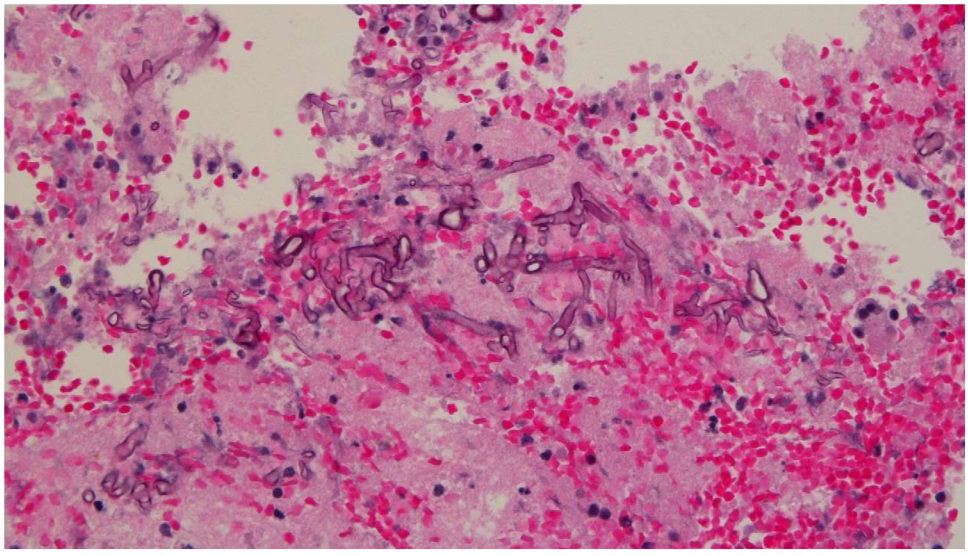

Fig. 3. Ribbon shaped, obtuse angle branching, pauci-septate hyphae.

made based on the characteristic morphological features on histopathology. Cultures were not sent. The patient was started on liposomal amphotericin B, but his clinical status worsened within hours post-operatively with new onset seizures. This was attributed to post-resection edema and increased intracranial pressure which was not amenable to aggressive medical or surgical management. The patient progressed to brain herniation and unfortunately passed away.

Discussion

While typical risk factors associated with mucormycosis are uncontrolled diabetes, immunocompromised state such as transplant recipients and those with malignancy undergoing chemotherapy, some other well identified (albeit relatively less commonly encountered) predisposing factors are use of intravenous drugs and deferoxamine therapy [1]. The site of involvement tends to vary based on the host population [1]. In a large review of close to 1000 patients with zygomycosis (now termed mucormycosis), Roden et al. reported association of certain underlying medical conditions with higher predilection to certain organ involvement [1]. Cerebral involvement was the most common presenting manifestation in patients with intravenous drug use (IDU) [1]. And in this subset of patients, there was primary involvement of the brain without concomitant sinus infection, much in contrast to patients with diabetes where rhino-cerebral infections are often seen [1]. The exclusive, 'isolated', cerebral involvement in IVU has been attributed to hematogenous seeding of the fungal spores that are introduced into the blood stream during episodes of intravenous injection of contaminated recreational drugs [1,4]. These spores enter the systemic circulation and have been seen to preferentially seed the basal ganglia [1,4]. This phenomenon has been demonstrated in animal models [5].

Clinical manifestations of isolated cerebral mucormycosis tend to be non-specific [3]. Patients may present with headache and focal neurological deficits [6]. Stroke like features may be seen since this invasive fungal infection tends to cause vessel thrombosis and hemorrhage of the parenchyma [6]. Since the involvement of the brain parenchyma is not secondary to spread from other regions such as the facial, oral or orbital areas, absence of characteristic necrotic lesions of mucormycosis are not seen in these body sites. This can make the clinical diagnosis challenging. Additionally, cerebrospinal fluid analysis may suggest a profile for bacterial meningitis and antibiotics, without empiric antifungal therapy, may be empirically initiated [6]. Radiological imaging may be mistaken for a neoplastic process [2]. Contrast enhancement may or may not be seen, and that finding per se does not help with the diagnosis [3]. What seems to be most suspicious on brain imaging is the involvement of basal ganglia [1–4]. While this area of the brain could be affected by other infectious (Creutzfeld Jacob Disease) and non-infectious (hypoglycemia, uremia, hypoxic-ischemic injury) processes, history of IDU must heighten the suspicion for cerebral mucormycosis [6]. Also, other conditions that affect the basal ganglia tend to do this symmetrically, while in the initial cases of mucormycosis, unilateral involvement may be seen [3,6]. This, however, could spread to the contralateral side [3,6]. Hence, diagnosis of this entity is challenging for a few reasons and tends to delay diagnosis.

Treatment of this potentially fatal infection requires combination of prompt medical and surgical intervention [3]. Amphotericin B (liposomal formulations being less toxic compared to the conventional one) is considered the drug of choice [3]. Neurosurgical intervention would be required for both diagnostic and therapeutic purposes, though challenges may be encountered for source control in deep seated infections [3,7]. Outcomes generally tends to be dismal, particularly if prompt diagnosis and early medical and surgical interventions are not instituted [3,7]. This case underscores the need for heightened suspicion for this uncommon entity so that appropriate therapy can be implemented.

Credit author statement

All the authors – Ngoc-Tram Ha, Megan Lowery, Jean Woo, Yatin Mehta, and Nitin Bhanot contributed to the writing of the manuscript of the case report.

Declaration of Competing Interest

None of the authors have any potential conflict of interest to disclose

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Consent

Not applicable. The patient passed away, unfortunately. We have ensured to not report any potential identifying information in the manuscript.

Author contribution

All authors were equally involved in data gathering and manuscript writing

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