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Fungal granuloma in immunocompetent patient masquerading as olfactory groove meningioma

Amr El Mohamad, Mohamad Alhoobi, Ahmed Eid, Ali Ayyad, Issam Albozom, Firas Hammadi

ABSTRACT

Introduction: Intracranial fungal infection is uncommon condition with high morbidity and mortality. Recently, the incidence has increased owing to the increase of the number of immunocompromised patients (e.g., acquired immunodeficiency syndrome, uncontrolled diabetes mellitus, immunosuppressive drugs, and organ transplant). Furthermore, intracranial fungal granuloma is very uncommon in immunocompetent patients. Unfortunately, it may be masked as it may mimics meningioma clinically and radiologically which may further delay the diagnosis and treatment especially in absence of the risk factors of immunosuppression. **Case Report:** We will present a case of intracranial fungal granuloma that mimics olfactory grove meningioma both clinically and radiologically in absence of risk factors of immunosuppression with no paranasal sinuses involvement. In addition to that we will discuss the suspicious radiological signs that might help in making fungal granuloma more possible than meningioma. **Conclusion:** Preoperative diagnosis of intracranial fungal granuloma is challenging in immunocompetent patients, in the absence of evidence of nasal infection. There are some radiological clues that could help us to differentiate between fungal granuloma and meningioma preoperatively.

Keywords: Immunocompetent, Intracranial fungal granuloma, Olfactory groove meningioma

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INTRODUCTION

Intracranial fungal infection is uncommon with reported mortality rate of 13–50% involving foci in paranasal sinuses and even higher mortality rates (80–100%) in patients of immunocompromised status [1]. Mass forming granuloma is rare, even in high volume centers [2]. The incidence of fungal infection of central nervous system is rising recently owing to improvement of the diagnostic modalities, such as imaging studies and pathology tests [2]. It may present as acute and rapidly fatal illness like encephalitis, meningitis or brain abscess, or as chronic illness like cerebral fungal granuloma [2]. Aspergillosis is the most common organism causing intracerebral fungal granuloma [3]. Diagnosis is challenging as it might mimic brain neoplasms. Early diagnosis and treatment improve outcome better. Herein we will present a case of cerebral fungal granuloma that was diagnosed and treated as olfactory groove meningioma, and we will discuss some clues to raise the suspicion of isolated fungal granuloma in immunocompetent patients.

CASE REPORT

A 40-year-old male Indian patient previously healthy presented to the emergency department (ED) after 1 episode of generalized tonic-clonic seizure. His roommate

called the Emergency medical services who found him confused at home and they brought him to the ED. In the hospital the patient was complaining of headache. No tongue biting, no urinary incontinence, no vomiting, no blurred vision, no motor weakness were seen. The patient mentioned that there was headache of one-week duration. On physical examination Glasgow Coma scale was 15. Pupils were bilaterally equal 3 millimeters and reactive to light. Normal cranial nerves examination, except for left anosmia, no sensory or motor deficit was found. Another attack of tonic-clonic seizure in the ED associated with up rolling of the eyes which was aborted in 1 minute without giving any medication.

Investigations

Hematological and biochemical blood investigations were normal. White blood count was 10.5 $10^3/\mu L$, with neutrophils 75.2%, eosinophils 2.2%, coagulation profile was normal, creatinine was 81 micromole per liter (umol/L) (normal value: 62–106), and electrolytes were normal: Sodium 140 millimole (mmol), potassium 3.7 mmol, and chloride 103 mmol. Computed tomography (CT) scan without contrast of the head showed large well defined hyperdense lesion in the left frontal region (Figure 1A). It appeared as extra axial, with mass effect on the underlying brain parenchyma (effacement of the left lateral ventricle and rightward midline shift by 7 mm). There was also overlying left frontal bone hyperostosis. Magnetic resonance imaging (MRI) of the brain showed a large left inferior frontal parasagittal extra-axial mass, over the floor of the anterior cranial fossa. It had lobulated outline and measured $4.6 \times 5.6 \times 6.5$ centimeter (cm) in anteroposterior, mediolateral, and caudocranial planes, respectively. It had low signal intensity on T1 W1/T2W1 (Figure 1B–D). No diffusion restriction was seen. It showed heterogeneous post-contrast enhancement, with wide contact with the overlying convexity meninges and anterior falx cerebri (Figure 1E and F). It extended inferiorly through the cribriform plate of the ethmoid bone to the left ethmoid air cells. Paranasal sinuses were normal with no evidence of involvement. The impression was that it is left olfactory groove meningioma with malignant changes in the posterosuperior aspect.

Treatment

As the patient presented with seizure and was harboring anterior cranial fossa lesion that intensely enhanced and meningioma was the first differential. Surgery was discussed with the patient in details and patient agreed for surgery. Under general anesthesia, microscope and neuronavigation were used. Left frontal craniotomy was done. Left frontal sinus was opened. Cranialization was done and packed with Gelfoam, muscle, and bone wax. Then dura was opened and reflected over the base. The tumor was identified, devascularized, dissected, and resected using a piecemeal technique. It was whitish and firm rubbery tumor, attached to the dura of the anterior cranial fossa, infiltrating the convexity dura and the brain posteriorly. The dura infiltrated by the tumor was resected. The dura of skull base has been reconstructed. Postoperatively patient was doing well with no complains expect for left anosmia that was present preoperatively. Magnetic resonance image of head was done postop day 1 showed near total resection of the previously seen left frontal intracranial mass with no definite extra-axial definite component (Figure 2A–C). Histopathology showed brain tissue as well as sclerotic/fibrotic tissue that were heavily infiltrated by ill-defined destructive granulomas having numerous multi-nucleated giant cells (Figure 3A) surrounded by lymphocytes as well as macrophages, some of these granulomas showed shadows of branching fungal hyphae (Figure 3B). Grocott's methenamine silver stain (GMS) and Periodic acid–Schiff (PAS) stains for fungi highlighted the fungal hyphae which turned to be acutely branched and septate fungal hyphae (Figure 3C) which were negative by mucin stain. Glial fibrillary acidic protein (GFAP) highlighted the end infiltrated brain parenchyma between the granulomas. There was no evidence of malignancy. Therefore, the diagnosis was fungal granuloma. Intravenous (IV) amphotericin B liposomal 375 milligrams (mg) per day was started for two weeks, then oral voriconazole 400 mg every 12 hours for two doses followed 200 mg twice a day for four weeks.

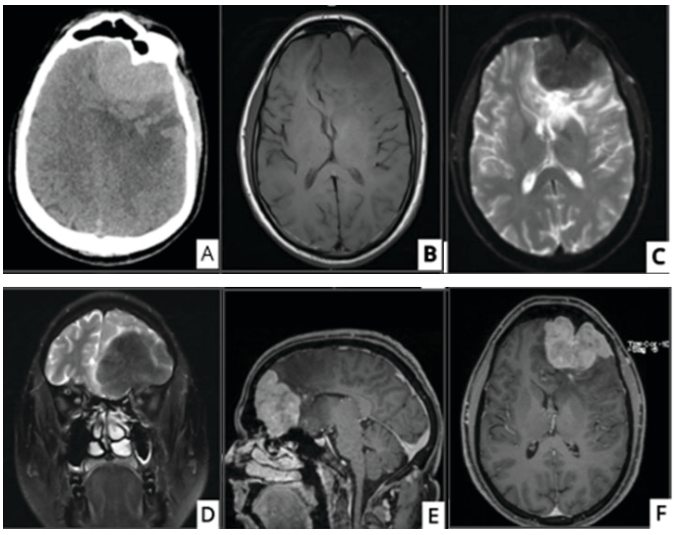

Figure 1: (A) Computed tomography scan of the head showed large well-defined hyperdense lesion in left frontal region, mostly extraaxial. (B) MRI of the head T1W1 without contrast, axial view showing hypointesnse lesion in left inferior frontal area. (C) MRI of the head T2W1 without contrast, axial view showing hypointesnse lesion in left inferior frontal area. (D) MRI of the head T2W1 without contrast, coronal view showing hypointesnse lesion over the floor of the left anterior cranial fossa. (E) MRI of the head T1W1 with gadolinium, sagital view showing heterogeneous post-contrast enhancement over the floor of the left anterior cranial fossa. (F) MRI of the head T1W1 with gadolinium, axial view showing heterogeneous post-contrast enhancement.

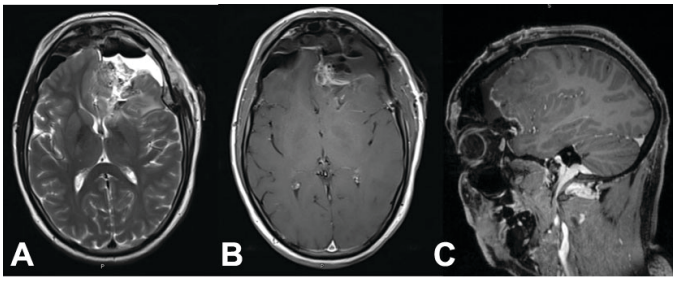

Figure 2: (A) MRI axial view, T2 sequence showing post-operative changes and no extra axial residual. (B) MRI axial view, T1 sequence with contrast showing near total resection of the mass. (C) MRI sagittal view, T1 sequence with contrast showing near total resection of the mass.

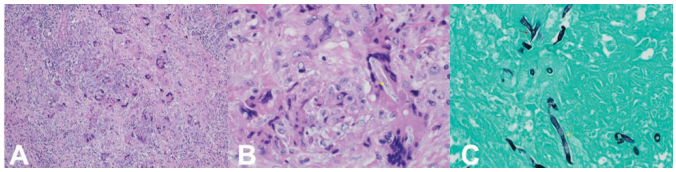

Figure 3: (A) Light microscopic view showing several destructive necrotizing granulomas infiltrating the brain tissue with numerous multi-nucleated giant cells (H&E ×100). (B) High microscopic image showing granuloma engulfing fungal hyphae-yellow arrow (H&E ×400). (C) Gomori's methenamine silver stain highlighting acutely branched (red arrow) and septate (yellow arrow) fungal hyphae (GMS ×600).

Outcome and follow-up

The patient had several episodes of hypokalemia as side effect of amphotericin which was corrected by potassium injection. Computed tomography of the chest, abdomen, and pelvis showed no abnormalities. As well as other workup for immunosuppression including Hepatitis B and C, human immunodeficiency virus and interferon-γ release assay (IGRA) were negative. Echocardiography was normal except for congenital redundant interatrial septum. The patient was seen in neurosurgery clinic multiple times and he is doing well with no headache or neurological deficit.

DISCUSSION

Aspergillus spreads to cranium either by hematogenous route from primary lung disease or, it may directly invade the cranium from paranasal sinuses by producing skull base osteomyelitis and may appear as granulomatous mass at the skull base [4]. Bony affection is not a must, as the fungus could spread along the vessels [5]. Intracranial fungal granuloma is seen mostly in India, Africa, and California in the United States, may be due to hot and dry climate with high content of fungal spores in agricultural dust [6]. Our patient is an Indian and living in Qatar, he is the first patient diagnosed with intracranial fungal granuloma in Qatar.

While fungal granuloma frequently found in immunocompromised patients, it is up to 50% of cases that show no clear evidence of risk factors for immunosuppression [6].

This could be explained by subclinical qualitative cellular or subcellular immunodeficiency [5–7]. In addition, our patient had no risk factor for immunosuppression that could be identified which includes diabetes mellitus, human immunodeficiency virus, tuberculosis, and Hepatitis B and C. In addition to that he had normal paranasal sinuses; normal CT scan of chest, abdomen, and pelvis; and normal echocardiography without any evidence of primary infection.

The most common presenting symptom is headache (in approximately 60% of patients), and the most common sign is cranial nerve involvement in 52% of case [2]. Seizure was the presenting symptom in 10% of cases and anosmia in 2.2% of cases [2]. Our patient presented for seizure, headache of two weeks duration, with left sided anosmia.

Aspergillosis is the most common fungal agent causing fungal granuloma [3]. Mishra et al. [2] found that aspergillosis accounts for 47.8% of cases followed by zygomycosis 14.4%, and in 13% of cases no specific organism was found. In our case histopathological results were positive for fungal infection with no specific organism detected, and fungal culture was not done because meningioma was suspected preoperatively. The anterior cranial fossa and the frontal lobe are the most common affected areas (53.3%), followed by middle cranial fossa and the temporal lobe (20%) [3].

Radiologically, there is no specific pattern of intracranial fungal granuloma on CT of the brain, but fungal infection usually appears as irregular iso/hyperdense lesion with faint heterogenous contrast enhancement with perilesional edema [3], which is not specific for fungal granuloma. In addition, the presence of paranasal sinusitis, bone destruction, and infarction due to arteritis suggests fungal infection [3]. On MRI scan, intracranial fungal granuloma appears as hypointense on T2W1, and isointense on T1W1 and there is homogeneous gadolinium enhancement according to one study [3]. As per Naik et al. MRI findings differ according to the type of the organism, for example, cryptococcoma shows hypointensity on T2W1; however, aspergilloma shows isointensity on T2W1 [6]. In our case, CT scan findings fit with the literature results (Figure 1A), but on MRI scan the mass was hypointense on T1W1 and T2W1 (Figure 1B–D) and there was a heterogenous instead of homogeneous contrast enhancement (Figure 1E). Furthermore, with the absence of paranasal sinus infection which makes radiological diagnosis challenging (Figure 1D and E).

The gold standard treatment is the combination of radical surgical excision of the granuloma in addition to antifungal medications [5]. Aggressive surgical treatment showed decrease in mortality from 63% to 39% of aspergilloma cases [5]. Voriconazole, itraconazole, 5-flucytocin, fluconazole, and amphotericin-B are used

for the treatment of intracerebral fungal granuloma [5]. Amphotericin was the drug of choice for aspergilloma, but it had a lot of side effects [5]. Liposomal amphotericin B was used to decrease side effect, and a clinical trial done showed its efficacity and its lower percentage of side effects comparing to amphotericin B [5].

In our case the patient received IV liposomal amphotericin-B for two weeks followed by oral voriconazole of four weeks duration. The patient had hypokalemia, which was corrected, and he had a temporary slight increase in creatinine level which was also corrected. The outcome with aggressive surgical treatment and appropriate medications was good in our patient.

CONCLUSION

Central nervous system fungal granuloma is uncommon condition that usually affects immunocompetent patients, furthermore it is rarer in immunocompetent. It might mimic clinically and radiologically brain neoplasms, such as meningioma including olfactory groove meningioma. High index of suspicion is essential in diagnosis to prevent delay in the diagnosis with its devastating sequelae. Low signal intensity in T2 weighted images, MRI makes the lesion more likely to be fungal granuloma than meningioma. More studies needed to understand the pathophysiology, mechanism of infection, and radiological characteristics of the intracerebral fungal granuloma in immunocompetent patients.

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Author Contributions

Amr El Mohamad – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Mohamad Alhoobi – Design of the work, Acquisition of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Ahmed Eid – Conception of the work, Design of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Ali Ayyad – Conception of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Issam Albozom – Conception of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Firas Hammadi – Conception of the work, Design of the work, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

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Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest
Authors declare no conflict of interest.

Data Availability
All relevant data are within the paper and its Supporting Information files.

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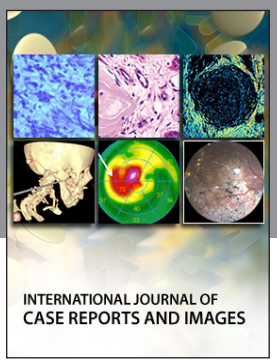

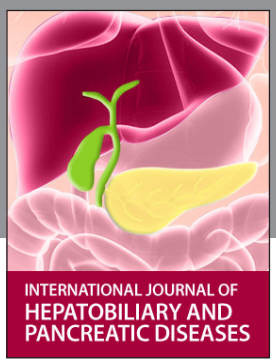

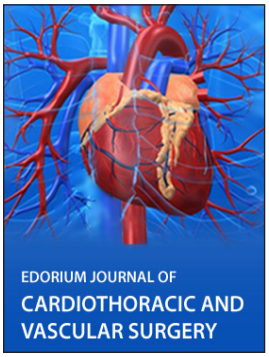

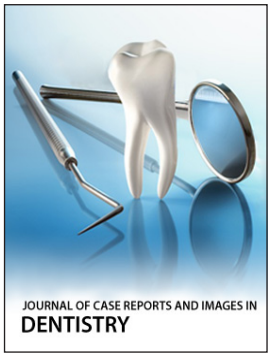

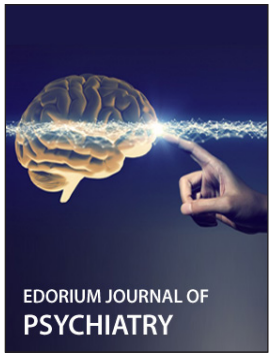

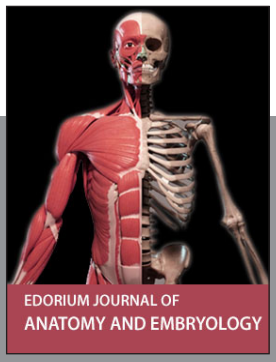

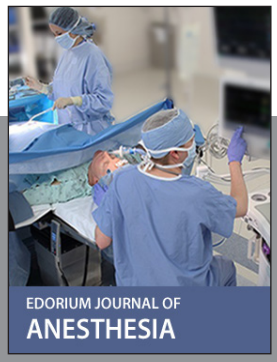

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