IDCases 18 (2019) e00603

Contents lists available at ScienceDirect

IDCases

journal homepage: www.elsevier.com/locate/idcr

Case report

A case of rhino-orbital mucormycosis in an immunocompetent patient following Hurricane Irma

Folusakin Ayoade[a,*], Christina Cloke[a], Tanya Quiroz[b], Youley Tjendra[a]

[a] University of Miami Miller School of Medicine, 1120 NW 14th Street, Miami, FL 33136, USA
[b] Jackson Health System, 1600 NW 12th Avenue, Miami, FL 33136, USA

ARTICLE INFO

Article history:
Received 9 July 2019
Received in revised form 18 July 2019
Accepted 19 July 2019

Introduction

Exposure to molds following a major hurricane has been associated with opportunistic fungal infections especially in immunocompromised hosts [1–4]. Most of these mold infections are usually aspergillosis, typically involving the lungs. On the other hand, only a few cases of mucormycosis (or mucor) have been linked epidemiologically to a major hurricane, predominantly in transplant patients [5,6]. In this report, we present a case of rhino-orbital mucormycosis in an immunocompetent, non-diabetic patient one year following hurricane Irma and highlighted the challenges encountered with making the correct diagnosis.

Case presentation

A 67-year-old woman presented with two weeks of headache and diplopia in her right eye. Three days prior to presentation, her condition progressed with almost total vision loss in the same eye. She had no previous history of significant eye problems or recent eye trauma. She denied any significant sinus pain or pressure, nasal or ear discharge, neck stiffness, fevers, nausea or vomiting. Her past medical history is significant only for hypertension, hypothyroidism and aortic stenosis status post bio-prosthetic aortic valve repair a year earlier. One week before her symptoms started, she underwent a dental procedure prior to which she received prophylactic antimicrobials.

She lived in the Florida Keys and suffered damage to her home from hurricane Irma 12 months prior to presentation. The damage caused flooding and significant debris in her home unit. It took a few months to completely clean up her home which she did mostly by herself accompanied by her husband. She reported exposure to extensive amounts of mold during the cleaning process. She smoked 1 pack of cigarette per day for several decades but denied any history of intravenous drug use. Her medication consisted only of levothyroxine 25 μg once daily.

Her vital signs at presentation were within normal limits. Examination at presentation showed both pupils were equal, round, and reactive to light and accommodation. She was unable to abduct her right eye. There was no periorbital swelling, hypopyon or hyphema seen in both eyes and dilated fundus examination was essentially unremarkable. Evaluation of the nasal mucosa was also unremarkable.

She subsequently underwent magnetic resonance imaging (MRI) study of the head and both orbits which revealed mucosal thickening with fluid levels in the ethmoid air cells and air-fluid levels in the sphenoid sinus (Fig. 1a and b). There was also a heterogeneous T1/T2 hypointense enhancing soft tissue effacing fat planes in the right pterygopalatine fossa fat and right orbital apex with encasement of the right optic nerve sheath complex and extension into the anterior aspect of the right cavernous sinus (Fig. 2). The radiologist report describes concern for invasive sinus disease with mucormycosis or invasive aspergillosis as differential diagnoses. Due to these findings, the patient was seen emergently by the Ear, Nose and Throat service and underwent sinus endoscopy which however, showed no evidence of necrosis or significant sinusitis.

As her vision did not improve, a few days later, she underwent right maxillary antrostomy and right sphenoidotomy. Findings at surgery showed evidence of right sphenoid sinusitis as well as

* Corresponding author.
E-mail addresses: fxa375@med.miami.edu (F. Ayoade), cxc1323@med.miami.edu (C. Cloke), tquiroz@jhsmiami.org (T. Quiroz), youley.tjendra@jhsmiami.org (Y. Tjendra).

https://doi.org/10.1016/j.idcr.2019.e00603

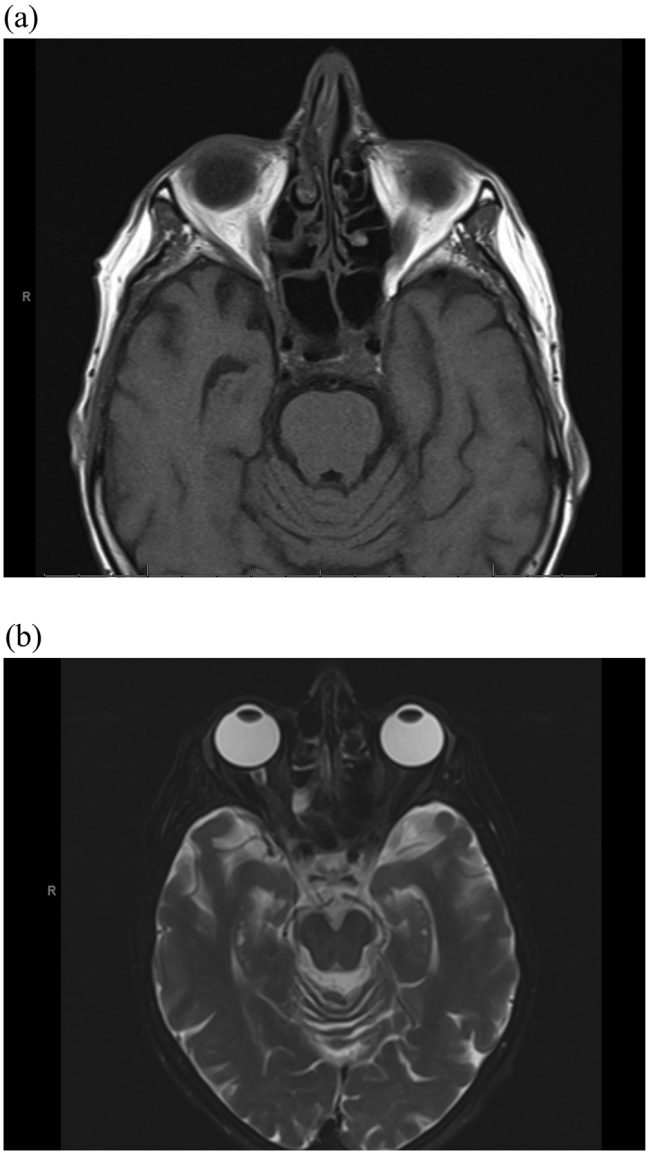

Fig. 1. (a) Head MRI without contrast (Coronal view) showing thickening of the right sided ethmoid and sphenoidal sinuses compatible with sinusitis. (b) Head MRI with contrast showing air fluid levels in the right ethmoid and sphenoidal sinuses.

some purulent drainage. Direct examination and evaluation of the nasal cavity and the other sinuses at surgery were however, normal. Biopsy of the right pterygopalatine fossa, maxillary and sphenoid sinuses was obtained and sent for direct microscopy, fungal stains, including immunohistochemistry, bacterial and fungal culture and histopathology. Based on the findings of inflammation and purulence at surgery, she was commenced empirically on ampicillin/sulbactam and vancomycin. The cultures from the purulence material obtained from the sphenoidal sinuses only grew *Staphylococcus epidermidis.* Given the absence of findings suggestive of fungal infection from endoscopy, operative findings and frozen section stains, antifungal therapy was initially withheld.

Few days later, preliminary histopathology result noted chronic rhinosinusitis with rare non-necrotizing granulomas. Confined within the granulomas were scant degenerative appearing fungal hyphae that highlighted on Grocott-Gomori methenamine silver (GMS) stains. There were no fungal elements identified outside of the granulomas and no evidence of angioinvasion or significant tissue necrosis. Due to the degenerative nature of the fungal

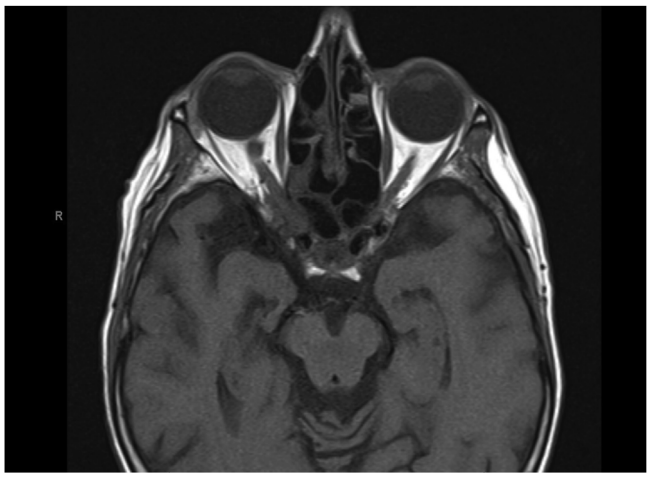

Fig. 2. Head MRI without contrast showing a heterogenous T1/T2 hypointense enhancing soft tissue in the right pterygopalatine fossa and right orbital apex, encasing the right optic nerve complex.

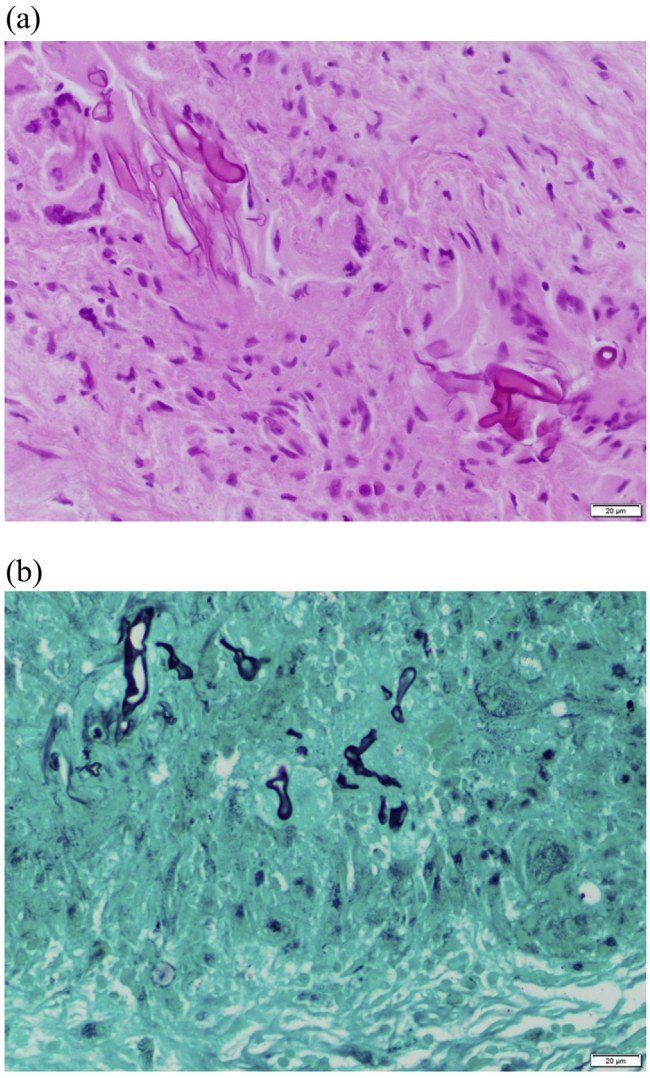

Fig. 3. (a) Hematoxylin and Eosin stains of histologic sections of the right lateral sphenoid recess showing invasive fungal sinusitis with associated foreign-body giant cell reaction and necrosis. Mag. 400×. (b) Grocott-Gomori methenamine silver stain of histologic sections of the right lateral sphenoid recess highlights numerous fungal non-septate branching hyphae, morphologically consistent with mucormycosis. Mag. 400×.

hyphae, speciation was difficult based solely on Hematoxylin and Eosin (H&E) and GMS stains. At this point, voriconazole was added to her empirical therapy. The rationale was, since the patient was immunocompetent, and the histopathologic findings were mostly consistent with chronic granulomatous invasive fungal sinusitis, aspergillus infection was favored based on frequency of occurrence. Other antimicrobials were discontinued after one week and she was discharged home on oral voriconazole 300 mg twice daily with follow-up appointment in the Infectious Disease clinic.

After a week of being at home, the patient was requested to return to the hospital for readmission based on further pathology results which noted: a negative Aspergillus histochemistry stain and findings of hyphae confined to granulomas with pauci-septated and irregular branching, more consistent with a Zygomycete rather than Aspergillus species. She was promptly readmitted and started on lipid complex amphotericin B. She underwent more surgeries including right total ethmoidectomy, right frontal sinusotomy, right orbital wall decompression and right extended maxillary antrostomy. Findings at surgery this time showed 'abnormal tissue' in the right lateral sphenoid recess and pterygoid palatine fossa, and right dehiscent optic nerve extending into the right optico-carotid recess. Further biopsies were obtained for cultures and histopathology. Histopathology a week later from the right lateral sphenoid recess demonstrated again hyphae which are pauci-septated and irregular branching as well as chronic rhinosinusitis with eosinophils (>10/HPF) (Fig. 3a). GMS stain was positive again for fungal organisms, morphologically consistent with mucor (Fig. 3b). Immunohistochemistry for Aspergillus was again negative. Fungal cultures were negative from all the specimens from both surgeries. The patient received lipid complex amphotericin B for a total of ten weeks. Post treatment, she had an excellent response with almost complete vision restoration. Serial follow-up MRI of the brain and eyes showed no evidence of disease recurrence six months later.

Discussion

Increase in the incidence of cutaneous and invasive fungal infections has been described following natural disasters including hurricanes and flooding [3,4]. Hurricane Irma was a category 4 storm when it hit the Florida Keys in September of 2017 with catastrophic flooding. Mucor infection has been previously described in immunocompetent hosts but generally rare [7–10]. None of these reports however were associated with natural disaster or flooding. As far as we know, this is the first case of rhino-orbital mucormycosis related to a major hurricane in an immunocompetent patient.

The diagnosis of mucormycosis in immunocompetent hosts is often a challenge due to a general low index of suspicion, poor growth characteristics in culture media and unavailability of reliable serum or tissue markers [8]. We encountered similar diagnostic challenges in this case. For example, fever, periorbital swelling, the classic black eschar, nasal ulcerations or necrosis often described on sino-nasal endoscopy of rhino-orbital mucormycosis were not present in our patient [11]. In addition, several culture samples obtained yielded no growth of mucor. Histopathology is often the main method of diagnosis and played a pivotal role in the accurate diagnosis for our patient. When available, newer techniques such as polymerase chain reaction and matrix- assisted laser desorption ionization–time of flight may be helpful in diagnosis [12–14].

Even though we cannot directly prove that in our patient, the exposure to molds related to hurricane and flooding was primarily responsible for mucor infection, the epidemiological association is rather strong and relevant as previously described [1,5]. The fact that she presented with symptoms 12 months after the natural disaster is unusual but not surprising as she lived in the same flooded home for several months after the hurricane. Professional mold treatment of her home after the flooding was also not pursued. Our case highlights the importance of including mucormysosis as a differential diagnosis of vision problems in immunocompetent patients following hurricane situations. This is a reasonable approach especially when there is evidence of potential mold exposure. Our case demonstrates a success story of cure of rhino-orbital mucormycosis which typically has a high overall mortality ranging from 25 to 62% [15]. A high index of suspicion, multiple attempts at making the accurate diagnosis, adequate source control and a good follow up plan were elements contributing to success in this case.

Author statement

FA contributed to the conceptualization, writing, review & editing of the manuscript. CC contributed to the review, editing and proofreading of the manuscript. TQ contributed to the review, proofreading and editing of the manuscript. YT contributed to editing the manuscript as well as the production and description of the pathology photo slides.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Acknowledgement

None.

References

[1] Kouadio IK, Aljunid S, Kamigaki T, Hammad K, Oshitani H. Infectious diseases following natural disasters: prevention and control measures. Expert Rev Anti Infect Ther 2012;10(January 1):95–104.
[2] Benedict K, Park BJ. Invasive fungal infections after natural disasters. Emerg Infect Dis 2014;20(March 3):349–55.
[3] Kontoyiannis DP, Shah EC, Wurster S, et al. Culture-documented invasive mold infections at MD Anderson Cancer Center in Houston, Texas, pre- and post-hurricane harvey. Open Forum Infect Dis 2019;6(April 4)ofz138.
[4] Sridhar J, Lam BL, Pasol J, Sternau L. Neuro-ophthalmic manifestations of fungal disease associated with posthurricane environment. J Neuroophthalmol 2012;32(September 3):197–201.
[5] Kueht K, Villarreal JA, Reece E, et al. Cutaneous mucormycosis in solid organ transplant recipients after hurricane harvey: short- and long-term management. Plast Reconstr Surg Glob Open 2019;7(January 1)e2041.
[6] Webb BJ, Blair JE, Kusne S, et al. Concurrent pulmonary Aspergillus fumigatus and mucor infection in a cardiac transplant recipient: a case report. Transplant Proc 2013;45(March 2):792–7.
[7] Rashid S, Ben Abid F, Babu S, et al. Fatal renal mucormycosis with Apophysomyces elegans in an apparently healthy male. Aging Male 2019;(March):1–4.
[8] Gupta N, Kumar R, Soneja M, et al. Mucor menace in an immunocompetent young male after dental manipulation. J Family Med Prim Care 2019;8(February 2):757–9.
[9] Sarkar S, Jash D, Maji A, Maikap MK. Solitary pulmonary nodule: a rare presentation of pulmonary mucormycosis in an immunocompetent adult. Lung India 2014;31(January 1):70–2.
[10] Lineberry KD, Boettcher AK, Blount AL, Burgess SD. Cutaneous mucormycosis of the upper extremity in an immunocompetent host: case report. J Hand Surg Am 2012;37(April 4):787–91.
[11] Yohai RA, Bullock JD, Aziz AA, Markert RJ. Survival factors in rhino-orbital-cerebral mucormycosis. Surv Ophthalmol 1994;39(July-August 1):3–22.
[12] Hammond SP, Bialek R, Milner DA, Petschnigg EM, Baden LR, Marty FM. Molecular methods to improve diagnosis and identification of mucormycosis. J Clin Microbiol 2011;49(June 6):2151–3.
[13] Springer J, Lackner M, Ensinger C, et al. Clinical evaluation of a Mucorales-specific real-time PCR assay in tissue and serum samples. J Med Microbiol 2016;65(December 12):1414–21.
[14] Ling H, Yuan Z, Shen J, Wang Z, Xu Y. Accuracy of matrix-assisted laser desorption ionization-time of flight mass spectrometry for identification of clinical pathogenic fungi: a meta-analysis. J Clin Microbiol 2014;52(July 7):2573–82.
[15] Roden MM, Zaoutis TE, Buchanan WL, et al. Epidemiology and outcome of zygomycosis: a review of 929 reported cases. Clin Infect Dis 2005;41(5):634.