Disseminated mucormycosis (DM) after pneumonectomy: a case report

Qian Wang†, Bo Liu†‡ and Youde Yan‡

Abstract

Background: Mucormycosis is a kind of rare opportunistic fungal disease and the incidence of which has gradually increased. Disseminated mucormycosis (DM) is a life-threatening infection that mostly occurs in immunocompromised patients. The lung and brain are usually involved in disseminated mucormycosis, and other sites are scare. We report the first case of disseminated mucormycosis whose infection sites included lung, skin, liver, vertebra, and spinal cord that ensued after a right lung pneumonectomy in an immunocompetent patient.

Case presentation: A 20-year-old female underwent a right lung pneumonectomy for "lung cancer" presented with an intermittent fever for two years. A computed tomography (CT) scan showed an enclosed outstanding mass in the right chest wall. The patient also suffered from lower limb numbness and weakness, difficulty walking, and dysuria. Medical examination showed superficial feeling of the abdominal wall was decreased from the T7 and T8 level; muscle strength for both lower limbs was decreased; muscle tension of both lower limbs was also diminished. A biopsy through the right chest wall mass and thoracic mass by fistula of chest wall showed broad nonseptate hyphae with right-angle branching, consistent with mucormycosis. With titration of amphotericin B and its lipid complex, the patient recovered.

Conclusions: Our case showed an unusual clinical presentation of disseminated mucormycosis in an immunocompetent patient.

Keywords: Case report, Disseminated mucormycosis, Pneumonectomy, Immunocompetent
outstanding mass in the right chest wall (Fig. 1). Thereafter, the patient was hospitalized three times due to a repeated fever and multiple bump biopsies from the enclosed outstanding mass; however, there was no clear diagnosis. One month before this admission, in addition to a fever, the patient suffered from worsened lower limb numbness and weakness, difficulty walking, and dysuria. In the past, she was healthy and had good nutrition.

A physical examination showed an abnormal and oncotic hard mass (10 cm × 5 cm) that protruded from the right chest wall. A 0.3-cm-diameter external fistula was seen in the central part of the pack (Fig. 2), which oozed a nonodorous, yellowish turbid liquid. Coarse sounds without moist rales or rhonchi were heard with left lung auscultation. Superficial feeling of the abdominal wall was decreased from the T7 and T8 level, while a deep feeling was present. Muscle strength for both lower limbs was decreased: 1° right and 3° left. Muscle tension of both lower limbs was also diminished. The bilateral Babinski sign was positive, but concavity swelling of the lower limbs existed.

Laboratory tests showed a white blood cell count of \(18.21 \times 10^9/L\), a hemoglobin level of 82 g/L, a platelet level of \(509 \times 10^9/L\), a C-reactive protein concentration of 98.00 mg/L, an erythrocyte sedimentation rate of 98.00 mg/L, and negative 1-3-\(\beta\)-D glucan and glactomannan (GM) tests. Post right pneumonectomy, the thoracic CT scan revealed uneven density flake soft tissue shadows in the right chest cavity, multiple sizes of nodules in the left lung, a soft tissue mass (12.5 cm × 5.9 cm) around a small pneumatosis in the right chest wall, and a reduced density in the right

![Fig. 1](image_url) The computed tomography (CT) scan showed an enclosed outstanding mass in the right chest wall 7-month later after the right lung resection.
liver lobe near the diaphragmatic top (January 2015, Fig. 3). The thoracic magnetic resonance imaging (MRI) results showed that a space-occupying lesion existed in the right pleural and chest wall with the T3-6 vertebral body and the right rib damaged, invading the spinal channel and spinal cord (January 2015, Fig. 3).

A biopsy through the right chest wall mass and thoracic mass by fistula of chest wall showed broad nonseptate hyphae with right-angle branching, consistent with mucormycosis (Fig. 4). The mucormycosis was widely disseminated, invading the lung, skin, liver, vertebral, and spinal cord.

The patient quickly developed type II respiratory failure. A noninvasive ventilator assisted breathing, and intravenous amphotericin B (AmB) was administered. When the dose of AmB increased to 0.5 mg/kg/day, with the cumulative dose reaching 150 mg, mental disorders (agitation, fear, etc.) and cardiac arrhythmias occurred. The adverse events disappeared when AmB was changed to liposomal AmB.

**Fig. 3** Results of CT and MRI (January 2015) for the chest. The CT shows a soft tissue mass (12.5 cm × 5.9 cm) around a small pneumatosis in the right chest wall, and a reduced density in the right liver lobe near the diaphragmatic top (a). The MRI shows lesions invaded the spinal channel and spinal cord (b).
After treatment for 28 days, the patient’s body temperature became normal and her respiratory function as well as lower limb sensory and motor function recovered. The size of the right chest wall mass became smaller, and the fistula dried. Laboratory and imaging examinations indicated that the lesions of the lung, liver, and thoracic vertebrae were significantly smaller. The patient was discharged back to the local hospital for further antifungal treatment and had recovered by the 3-month follow-up exam, and plastic surgery was suggested to repair the thoracic wall.

Discussion
In this report, we present a case of DM in the lung, skin, liver, vertebrae, and spinal cord after a right lung pneumonectomy. The lung and brain are usually involved in DM [6]. The involvement of the vertebrae and spinal cord in our case has rarely been reported in DM. DM has a nonspecific manifestation, resulting in difficulties in diagnosis [5]. In our case, it was not difficult to speculate that it first occurred in the lung, even before the pneumonectomy, as a necrotizing granulomatous mass was found. Indeed, pulmonary infection is the second most common form, accounting for more than 30% of infections [7]. Mucorales has a special affinity to endothelial cells, resulting in its invasion and destruction of small arteries. By attracting platelet adhesion and aggregation, Mucorales can also cause thrombosis and fatal bleeding [8, 9]. This might explain the wide dissemination of pulmonary mucormycosis in the present case.

To date, there are no known reliable clinical serological diagnostic methods for the diagnosis of DM. Microbial culture of secretions is time-consuming, with a low sensitivity and a high false-positive rate. The culture of sterile tissue may kill the pathogen because of its grinding process (a step for preparing tissue culture samples), which results in a low rate of positive cultures [10]. Therefore, the gold standard for the diagnosis of DM relies on the characteristic mycelium and pathological changes of the biopsy. The histopathological characteristics for Mucorales is a wide (6–25) μm in diameter, rarely separating hyphae and have irregular or right angle branch. The diagnosis of pulmonary mucormycosis in this report was based on histopathology.

DM generally occurs in severely immunocompromised patients: it represents 1.6% of all invasive fungal infections and is predominant in immunosuppressed patients with risk factors [11]. Recently, the incidence has increased significantly, even in immunocompetent patients [11]. Our report describes DM in an immunocompetent patient. DM is associated with a mortality rate of approximately 100%, but successful treatment has been reported [12–14]. Successful treatment of DM requires a rapid diagnosis, reversal of predisposing factors, aggressive surgical excision, and antifungal therapy [15]. AmB or its lipid complex remains the first choice for treatment [16]. In the present case, after titrating the dose to avoid adverse effects, the patient recovered quickly.

Conclusion
Clinicians need raise awareness that DM should also be regarded in immunocompetent individuals, especially those
with pulmonary mucormycosis. Retrospective studies regarding the morbidity of DM in immunocompetent individuals in the future might be necessary in order to understand its epidemiology.

Abbreviations
AmB, amphotericin B; CT, computed tomography; DM, disseminated mucormycosis; GM, glactomannan; MRI, magnetic resonance imaging

Acknowledgments
None.

Funding
This study was supported by A Project Funded by the Priority Academic Program Development of Jiangsu Higher Education Institutions (PAPD,JX10231802).

Availability of data and materials
All the data supporting our findings is contained within the manuscript.

Authors’ contributions
QW and BL designed the study, wrote the manuscript, participated in literature search, and made figures and tables. YDY conceived of the study, and participated in its design and coordination and helped to revised the manuscript critically for important content. All authors read and approved the final manuscript.

Competing interests
The authors declare that they have no competing interests.

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

Ethics approval and consent to participate
Not applicable.

Author details
1Department of Infectious Diseases, The First Affiliated Hospital with Nanjing Medical University, 300 Guangzhou Road, Nanjing 210029, Jiangsu, China.
2Department of Infection Management, The First Affiliated Hospital with Nanjing Medical University, Nanjing 210029, China.

Received: 29 February 2016 Accepted: 7 June 2016
Published online: 22 July 2016

References
1. Katragkou A, Walsh TJ, Rolides E. Why is mucormycosis more difficult to cure than more common mycoses? Clin Microbiol Infect. 2014;20 Suppl 6:74–81.
2. Camara-Lemarroy CR, González-Moreno E, Rodríguez-Gutiérrez R, Rendón-Ramírez EJ, Ayala-Cortés AS, Fraiza-Hernández ML, et al. Clinical Features and Outcome of Mucormycosis. Interdiscip Perspect Infect Dis. 2014;2014:1–5.
3. Petrikos G, Skaida A, Lortholary O, Rolides E, Walsh TJ, Kontoyiannis DP. Epidemiology and clinical manifestations of mucormycosis. Clin Infect Dis Off Publ Infect Dis Soc Am. 2012;54 Suppl 15:233–34.
4. Lee D-G, Choi J-H, Choi S-M, Yoo J-H, Kim Y-J, Min C-K, et al. Two cases of disseminated mucormycosis in patients following allogeneic bone marrow transplantation. J Korean Med Sci. 2002;17(3):403–6.
5. Gupta KL, Joshi K, Pereira BJ, Singh K. Disseminated mucormycosis presenting with acute renal failure. Postgrad Med J. 1987;63(738):297–9.
6. Pak J, Tucci VT, Vincent AL, Sandin RL, Greene JN. Mucormycosis in immunocompromised patients. J Emerg Trauma Shock. 2008;1(2):106–13.
7. McAdams HP, Rosado de Christenson M, Strollo DC, Patz EF. Pulmonary mucormycosis: radiologic findings in 32 cases. AJR Am J Roentgenol. 1997;168(6):1541–8.
8. Groll AH, Walsh TJ. Uncommon opportunistic fungi: new nosocomial threats. Clin Microbiol Infect Off Publ Eur Soc Clin Microbiol Infect Dis. 2001;7 Suppl 2:8–24.
9. Ribes JA, Vanover-Sams CL, Baker DJ. Zygomycetes in human disease. Clin Microbiol Rev. 2000;13(2):236–301.
10. Yan D, Dong H, Shan B. Progress of epidemiology and diagnosis of invasive fungal infections. Progress of Microbiology Immunology. 2009;37(1):67–71.
11. Pozo Laderas JC, Pontes Moreno A, Pozo Salido C, Robles Arista JC, Linares Sicilia MJ. Disseminated mucormycosis in immunocompetent patients: A disease that also exists. Rev Iberoam Micol. 2015;32(2):63–70.
12. Rickerts V, Atta J, Herrmann S, Jacobi V, Lambrecht E, Bialek R, et al. Successful treatment of disseminated mucormycosis with a combination of liposomal amphotericin B and posaconazole in a patient with acute myeloid leukaemia. Mycoses. 2006;49 Suppl 1:27–30.
13. Mantadakis E, Samonis G. Clinical presentation of zygomycosis. Clin Microbiol Infect Off Publ Eur Soc Clin Microbiol Infect Dis. 2009;15 Suppl 5:15–20.
14. Maury S, Leblanc T, Feuillade M, Molina JM, Schaison G. Successful treatment of disseminated mucormycosis with liposomal amphotericin B and surgery in a child with leukemia. Clin Infect Dis Off Publ Infect Dis Soc Am. 1998;26(1):200–2.
15. Alacacioglu I, Kargi A, Ozcan MA, Piskin O, Solak C, Secil M, et al. Fatal disseminated mucormycosis in a patient with mantle cell non-Hodgkin’s lymphoma: an autopsy case. Braz J Infect Dis Off Publ Braz Soc Infect Dis. 2009;13(3):238–41.
16. Comely OA, Ankan-Akdag il S, Danauel E, Groll AH, Lagrou K, Chakraborti A, et al. ESCMID and ECMM joint clinical guidelines for the diagnosis and management of mucormycosis 2013. Clin Microbiol Infect. 2014;20 Suppl 3:5–26.