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CASE REPORT

Pulmonary mucormycosis mimicking lung tumour in an uncontrolled diabetic patient

Elham Sadat Banimostafavi^{1,2} | Mahdi Fakhar¹ | Zakaria Zakariaei^{1,3} | Ali Sharifpour^{1,4} | Mostafa Soleymani¹

¹Toxoplasmosis Research Center, Communicable Diseases Institute, Iranian National Registry Center for Lophomoniasis (INRCL), Immam Khomeini Hospital, Mazandaran University of Medical Sciences, Sari, Iran

²Department of Radiology, Imam Khomeini Hospital, Mazandaran University of Medical Sciences, Sari, Iran

³Toxicology and Forensic Medicine Division, Toxoplasmosis Research Center, Imam Khomeini Hospital, Mazandaran University of Medical Sciences, Sari, Iran

⁴Pulmonary and Critical Care Division, Imam Khomeini Hospital, Mazandaran University of Medical Sciences, Sari, Iran

Correspondence

Ali Sharifpour and Zakaria Zakariaei, Toxoplasmosis Research Center, Communicable Diseases Institute, Pulmonary and Critical Care Division, Iranian National Registry Center for Lophomoniasis (INRCL), Immam Khomeini Hospital, Mazandaran University of Medical Sciences, Farah-Abad Road, P.O. Box 48471-91971, Sari, Iran. Email: asharifpour0209@yahoo.com and Email: ali.zakariaei@yahoo.com

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Abstract

Mucormycosis is a fungal infection that can affect a variety of organs, one of which being the respiratory system. The most common form of infection with mucormycetes is pulmonary mucormycosis, which has a poor prognosis if infected. The clinical signs and radiological findings of this infection are non-specific and very similar to lung tumours. Here, we describe a 62-year-old obese man with uncontrolled diabetes mellitus who was referred for a tumour-like mass in the left lung, which after bronchoscopy was diagnosed as mucormycosis.

KEYWORDS

diabetes mellitus, mucormycosis, pulmonary, tumour-like

INTRODUCTION

Mucormycosis is a rare and opportunistic fungal infection and is usually seen as an invasive parenchymal consolidation or cavitation but is rarely reported as an endobronchial mass.¹ Mucormycosis has the ability to infect various organs. The most common site of infection is rhinocerebral, followed by cutaneous, lung, disseminated and gastrointestinal tract.²

Pulmonary mucormycosis is the most common form of infection with mucormycetes and has a poor prognosis if infected, unless diagnosed early followed by appropriate treatment.³ Increased blood sugar concentrations stimulate fungal growth, so mucormycosis infection is rare in patients with well-controlled blood sugar levels.⁴

Patients who have dysfunctional cellular immunity, such as those who have had a stem cell transplant, leukaemia, uncontrolled diabetes, a solid organ transplant, neutropenia, corticosteroid and deferoxamine therapy, are more likely to develop infection.^{2,5} The clinical signs and radiological findings of this infection are non-specific and very similar to other lung diseases.⁶ The most common symptoms of pulmonary mucormycosis include fever, dyspnoea and productive cough. Due to low sensitivity and many false positives in the culture method, it is recommended to use histopathological examination to diagnose pulmonary mucormycosis.⁷ Here, we describe an obese old man with uncontrolled diabetes mellitus who was referred for a tumour-like mass in the left lung.

CASE REPORT

On 8 August 2021, a 62-year-old obese man with a history of diabetes mellitus, hypertension, dyslipidaemia and ischaemic heart disease undertreated with suitable medications suffered from a chronic cough with sputum for 2 months, sometimes with mild haemoptysis and lower limb oedema. He was referred to the emergency department with a chief complaint of dyspnoea and orthopnoea. On examination, he had a blood pressure of 140/90 mmHg, a heart

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TABLE 1 Baseline laboratory data

Laboratory data parameter	Result	Normal range
WBC	$18 imes 10^3$	4000-1000/mm ³
RBC	3.9×10^{6}	$4.56\times10^6\text{/mm}^3$
PLT	58×10^3	145,000-450,000/mm ³
Hb	10.7	12.3–15.3 g/dl
Blood sugar	213	90-110 mg/dl
Urea	42	13-40 mg/dl
Creatinine	1.2	0.5-1.3 mg/dl
К	3.9	3.5-5.5 mEq/L
Na	139	135-145 mEq/L
Calcium	10.7	8.5-10.5 mg/dl
Albumin	2.9	3.5–5.5 g/dl
CRP	79.8	Less than 6 mg/L
ESR	88	0-20 mm/h
Poly	88.1	60%-70%
Lymph	10.8	20%-30%
ALT	14	4–36 IU/L
AST	28	10-40 IU/L
ALP	262	44–147 IU/L
РТН	5	10–55 pg/ml

Abbreviations: ALP, alkaline phosphatase; ALT, alanine aminotransferase; AST, aspartate aminotransferase; CRP, C-reactive protein; ESR, erythrocyte sedimentation rate; Hb, haemoglobin; PLT, platelets; PTH, parathyroid hormone; RBC, red blood cell; WBC, white blood cell.

rate of 90 beats/min, a respiratory rate of 20 breaths/min, an O_2 saturation of 95% and a temperature of 37°C. In addition, the patient had a decreasing sound on the left side of the lungs, and there was no organomegaly on the abdomen or pelvis.

Laboratory tests were requested (Table 1). A spiral lung computed tomography scan revealed a complete collapse of the left lung with a right mediastinal shift due to a tumourlike large mass of necrosis invading the mediastinum and into the left bronchus. There were reports of complete obstruction of the left bronchus and mild pleural effusion of the left lung base (Figure 1).

On venous Doppler colour ultrasound, the superficial and deep veins of the lower limbs did not show clots or thrombosis. The patient was admitted to the lung service and had a bronchoscopy performed by a pulmonologist, which showed obstruction of the left bronchus near the carina by a fragile white mass that had bled slightly after multiple biopsies. The samples taken in fibre-optic bronchoscopy were sent to the pathology laboratory to rule out the possible mucormycosis and malignancy. As a result, laboratory findings showed fibro-connective tissue lining and fungal hyphae elements compatible with mucormycosis (Figure 2) and malignancy was not found. A test for galactomannan enzyme immunoassay to rule out Aspergillus infection was requested, which resulted negative.

Finally, the patient was diagnosed with mucormycosis and treated with drug liposomal amphotericin B (a dose of 5 mg/kg; a total dose of 350 mg daily for 6 weeks), and was discharged in good general condition without fever, cough and haemoptysis on oral posaconazole (Noxafil[®], Merck Sharp & Dohme) (400 mg for 4 weeks). At the 2-month follow-up, the patient's pulmonary symptoms, including dyspnoea, orthopnoea, cough with sputum and haemoptysis, resolved, and the mass in the lungs were absorbed. Written informed consent was obtained from the patient for the publication of this case report. This study was conducted according to the Declaration of Helsinki Principles. Also, CARE guidelines and methodology were followed in this study.

DISCUSSION

Mucormycosis is an opportunistic fungal infection usually seen in individuals with immune system disorders. Approximately a quarter of cases of mucormycoses are pulmonary



FIGURE 1 Lung computed tomography scan axial with contrast shows a large necrotic mass (red arrow) in the left hilum with obliteration of the left main bronchus (green arrow), causing complete left lung collapse. The mass invaded the mediastinum and compressed the oesophagus. Moderate plural effusion (blue arrow) is seen in the left hemithorax



FIGURE 2 Photomicrograph showing broad ribbon-like hyphae (black arrows) with haphazard branching (H & E-stained section; $40 \times$ magnification)

mucormycoses, with a mortality rate of more than 50%.⁷ In 1876, Furbinger et al. described the first case of pulmonary mucormycosis.⁸ Most of the patients show predisposing factors, such as diabetes mellitus, blood malignancies, chemotherapy, chronic kidney disease, immunosuppressive drugs, transplantation, HIV and so on.^{9–11} Pulmonary mucormycosis has different characteristics in terms of clinical evidence and imaging. Coughs, fevers, chest pains and dyspnoea are common manifestations of these symptoms.^{10–12}

In our case, the patient presented with a chronic cough with sputum, haemoptysis and various underlying diseases. Fever, cough, dyspnoea and haemoptysis are the most common symptoms of pulmonary mucormycosis. Haemoptysis reveals a poor prognosis, because it is a sign of vascular invasion and intra-alveolar bleeding. To diagnose mucormycosis, samples should be carefully examined.¹ Pulmonary mucormycosis happens due to the aspiration of fungal spores into the airways, which can lead to acute and progressive pneumonia. A definitive diagnosis of pulmonary mucormycosis is based on the observation and identification of hyphae of this fungus in infected tissues.¹⁰

The culture method is not recommended due to low sensitivity and many cases of false positives.⁷ In our case, the diagnosis was confirmed based on histopathological findings in the left main bronchus mass resection. A successful treatment requires an accurate and early diagnosis.¹³ Treatments for mucormycosis include antifungal drug therapy, surgery, removal and improvement of underlying diseases such as blood sugar regulation and neutropenia correction. The drugs of choice as antifungal treatments for pulmonary mucormycosis are liposomal amphotericin B and posaconazole.¹⁴

In conclusion, in immunocompromised patients, particularly those with uncontrolled diabetic mellitus who present with respiratory symptoms, for definitive diagnosis, in addition to para-clinical procedures, fibre-optic bronchoscopy should be considered, and if the diagnosis of mucormycosis is confirmed, appropriate and early treatment is recommended.

CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTION

Ali Sharifpour and Zakaria Zakariaei were involved in the interpretation and collection of data and editing of the manuscript. Mahdi Fakhar and Elham Sadat Banimostafavi were involved in the writing and preparing the final version of the manuscript. Mostafa Soleymani was responsible for collecting data and submitting the manuscript. All authors reviewed the paper and approved the final version of the manuscript.

DATA AVAILABILITY STATEMENT

The data are available with the correspondence author and can be achieved on request.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

ORCID

Mahdi Fakhar ¹⁰ https://orcid.org/0000-0002-4690-6938 Zakaria Zakariaei ¹⁰ https://orcid.org/0000-0003-4835-9349

REFERENCES

- Mahishale V, Patil B, Mahishale A, Malur PR, Avuthu S, Eti A, et al. Endobronchial pulmonary mucormycosis diagnosed by fiberoptic bronchoscope: a rare case report. Med J DY Patil Univ. 2016 Jan 1; 9(1):132.
- Jeong W, Keighley C, Wolfe R, Lee WL, Slavin MA, Kong DC, et al. The epidemiology and clinical manifestations of mucormycosis: a systematic review and meta-analysis of case reports. Clin Microbiol Infect. 2019 Jan 1;25(1):26–34.
- Andrews DR, Allan A, Larbalestier RI. Tracheal mucormycosis. Ann Thorac Surg. 1997 Jan 1;63(1):230–2.
- Agrawal R, Yeldandi A, Savas H, Parekh ND, Lombardi PJ, Hart EM. Pulmonary mucormycosis: risk factors, radiologic findings, and pathologic correlation. Radiographics. 2020 May;40(3):656–66.
- Bansal M, Martin SR, Rudnicki SA, Hiatt KM, Mireles-Cabodevila E. A rapidly progressing Pancoast syndrome due to pulmonary mucormycosis: a case report. J Med Case Rep. 2011;5:388.
- Yang J, Zhang J, Feng Y, Peng F, Fu F. A case of pulmonary mucormycosis presented as Pancoast syndrome and bone destruction in an immunocompetent adult mimicking lung carcinoma. J Mycol Med. 2019 Apr 1;29(1):80–3.
- 7. Luo Z, Zhang L. Diagnosis and treatment of pulmonary mucormycosis: a case report. Exp Ther Med. 2017;14:3788–91.
- Furbinger P. Observations on lungenmycose beinmeschen. Arch Pathol Anat Physiol Klin Med. 1876;66:330–65.
- 9. Hamilos G, Samonis G, Kontoyiannis DP. Pulmonary mucormycosis. Semin Respir Crit Care Med. 2011 Dec;32(6):693–702.
- Wang XM, Guo LC, Xue SL, Chen YB. Pulmonary mucormycosis: a case report and review of the literature. Oncol Lett. 2016 May 1;11(5): 3049–53.
- 11. Zhang L, Tian X, Wang P, Zhang H, Feng R. Recurrent pulmonary mucormycosis after lobectomy in a non-smoking patient without predisposing risk factors. Braz J Infect Dis. 2012;16:590–3.

- 12. Sarkar S, Jash D, Maji A, Maikap MK. Solitary pulmonary nodule: a rare presentation of pulmonary mucormycosis in an immunocompetent adult. Lung India. 2014 Jan;31(1):70.
- Turnbull A, Chembo CL, Leikis M, Pidgeon G, Arnold L, Hay N, et al. A case of pulmonary mucormycosis in a renal transplant recipient. Nephrol Ther. 2017 Aug;22(8):657.
- Dupont B. Pulmonary mucormycosis (zygomycosis) in a lung transplant recipient: recovery after posaconazole therapy. Transplantation. 2005 Aug 27;80(4):544–5.

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