

Case Report

A 17-year-old student with ankle sprain to leg amputation

Xi Yang^{1,2,3}, Yinhan Ding⁴, Yong Liu^{1,2,3}, Yixue Shi^{1,2,3}, Xiaolei Sun^{1,2,3,5,6,*}

¹Department of General Surgery (Vascular Surgery), The Affiliated Hospital of Southwest Medical University, Luzhou 646000, China

²Cardiovascular and Metabolic Diseases Key Laboratory of Luzhou, Luzhou 646000, Sichuan, China

³Key Laboratory of Medical Electrophysiology, Ministry of Education & Medical Electrophysiological Key Laboratory of Sichuan Province, Collaborative Innovation Center for Prevention and Treatment of Cardiovascular Disease of Sichuan Province, Institute of Cardiovascular Research, Southwest Medical University, Luzhou 646000, Sichuan, China

⁴Department of Laboratory Medicine, The Affiliated Hospital of Southwest Medical University, Luzhou 646000, China

⁵Department of Interventional Medicine, The Affiliated Hospital of Southwest Medical University, Luzhou 646000, China

⁶Nucleic Acid Medicine of Luzhou Key Laboratory, Southwest Medical University, Luzhou 646000, China

*Corresponding author. Department of General Surgery (Vascular Surgery), The Affiliated Hospital of Southwest Medical University, No.25 Taiping Street, Luzhou 646000, China. E-mail: sunxiaolei@swmu.edu.cn; sunxiaolei_lg@163.com

Abstract

Cutaneous mucormycosis with ischemic complications is a life-threatening condition with high mortality rates, particularly in immunocompromised individuals. The incidence of mucormycosis has increased during the COVID-19 pandemic due to reduced immunity. We present the case of a 17-year-old high school student who experienced a sprained left lower extremity, followed by worsening pain and swelling due to the topical application of unknown local herbs. Eighteen days after the injury, she was admitted to our department in a comatose state with left lower limb ischemia. The patient had a history of uncontrolled diabetes mellitus and displayed a necrotic lesion on her left ankle, suggestive of invasive infectious fungi disease. Diagnostic procedures, including tissue staining and molecular analysis, identified *Rhizopus oryzae* as the causative organism. Administering amphotericin B yielded marked improvement, but the patient necessitated a mid-thigh amputation to curtail the infection's advance, culminating in her successful discharge post-treatment.

Keywords: fungal infection; cutaneous mucormycosis; immunosuppression; amputation

Introduction

This report describes a critical case of cutaneous mucormycosis in a 17-year-old diabetic student, highlighting the urgency of early diagnosis and aggressive treatment, especially amid the COVID-19 pandemic. It focuses on the diagnostic challenges and treatment strategies for this rapidly progressing infection, particularly in immunocompromised patients.

Case report

A 17-year-old high school student sprained her left lower extremity. Over the 4 days following the injury, topical application of unknown local herbs exacerbated the pain and calf swelling. At 18 days after her ankle sprain, the girl went into a coma and was hospitalized in our department with 'left lower limb ischemia.' The patient had diabetes mellitus for three years without regular medication. Physical examination revealed a black necrotic lesion measuring approximately 15 × 10 cm² on her left ankle, with erythematous blisters and slight edema in the lower leg (Fig. 1).



Figure 1. A black necrotic lesion on the left ankle, erythematous blisters and slight edema in the lower leg.

The lesion skin temperature was abnormally low, and the foot and toes failed to flex dorsally. Digital subtraction angiography revealed total occlusion of the anterior tibial artery, posterior tibial artery and dorsalis pedis artery. The patient underwent a midline amputation of the left lower leg.

Given the reason, the history of uncontrolled diabetes mellitus and sprain, and the extreme progression of ischemia in the

Received: December 28, 2023. Accepted: January 5, 2023

Published by Oxford University Press and JSCR Publishing Ltd. © The Author(s) 2024.

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<https://creativecommons.org/licenses/by-nc/4.0/>), which permits non-commercial re-use, distribution, and reproduction in any medium, provided the original work is properly cited. For commercial re-use, please contact journals.permissions@oup.com

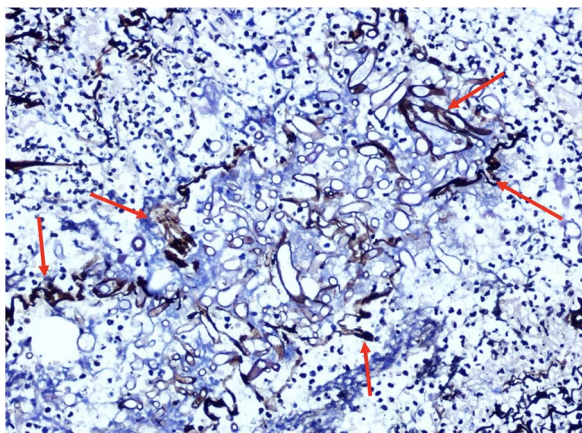


Figure 2. Filamentous fungi were revealed by tissue staining with periodic acid-silver methenamine.

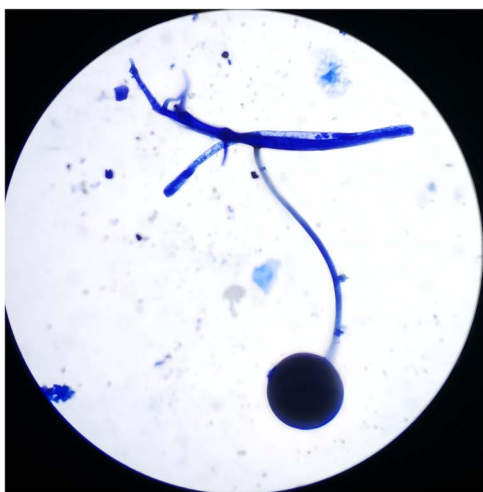


Figure 3. The morphology of the *Rhizopus* genus on the staining with lactic acid phenol cotton after fungi culture.

lower extremities, she was clinically suspected of invasive infectious fungi disease. Tissue staining with periodic acid-silver methenamine demonstrated filamentous fungi with small-vessel invasiveness (Fig. 2). Further staining with lactic acid phenol cotton blue after fungi culture revealed the morphology of the *Rhizopus* genus (Fig. 3), with creeping mycelium, root-like mycelium (pseudorrhiza), sporangiophore and sporangium. In addition, MALDI-TOF-MS analysis identified *Rhizopus oryzae*, with a confirmed score of 2.153, as the causative organism. Within amphotericin B therapy, she was rejuvenated, underwent a secondary mid-thigh amputation and then was discharged successfully.

Discussion

Mucormycosis, a rare but lethal opportunistic fungal infection, is characterized by its sudden onset, rapid progression, diagnostic difficulty and poor prognosis, with an incidence of approximately 1.7 per million and a mortality rate of 40–80% [1]. Although inhalation of airborne spores or direct ingestion are the main

modes of transmission for mucormycosis, spores may penetrate the skin through occult wounds and subsequently invade the vessel wall to produce septal arteritis, causing severe thrombosis or infarction in the cutaneous subtype [2]. Concomitant necrotizing fasciitis is one of the most common skin manifestations of cutaneous mucormycosis [3]. *Rhizopus oryzae* is reported to be the most common human pathogenic fungus causing mucormycosis [4]. This life-threatening infection characterized by a fast onset, rapid progression and high mortality rate (>50% under standard therapy) usually occurs in immunocompromised individuals, as in this case [5, 6]. During the COVID-19 pandemic, the incidence rate surged considerably due to reduced immunity [7]. Early vigorous surgical debridement, as well as high-dose and full-course intravenous antifungal therapy, are critical in the treatment of mucormycosis.

Conflict of interest statement

All authors have no conflicts of interest to declare that they are relevant to the content of this article.

Funding

This study was supported by the International Science and Technology Innovation Cooperation Project of Sichuan Province (22GJHZ0278), the Sichuan Science and Technology Program (2022YFS0614), the Medical Research Project of Sichuan Province (S21020), the Science and Technology Strategic Cooperation Project of Luzhou Municipal People's Government and Southwest Medical University (2021LZXNYD-D10), and the Doctoral Research Initiation Program of the Affiliated Hospital of Southwest Medical University (19041).

References

1. Arjmand P, Bahrami M, Mohammadi ZE, et al. Mucormycosis in pre-COVID-19 and COVID-19 era: a study of prevalence, risk factors and clinical features. *Laryngoscope Investig Otolaryngol* 2022;**7**:1343–50.
2. Sannathimmappa MB, Nambiar V, Aravindakshan R. Storm of a rare opportunistic life threatening mucormycosis among post COVID-19 patients: a tale of two pathogens. *Int J Crit Illn Inj Sci* 2022;**12**:38–46.
3. Walsh TJ, Hospenthal DR, Petraitis V, Kontoyiannis DP. Necrotizing mucormycosis of wounds following combat injuries, natural disasters, burns, and other trauma. *J Fungi (Basel)* 2019;**5**:57.
4. Ibrahim AS, Spellberg B, Avanesian V, et al. *Rhizopus oryzae* adheres to, is phagocytosed by, and damages endothelial cells in vitro. *Infect Immun* 2005;**73**:778–83.
5. Singh AK, Singh R, Joshi SR, Misra A. Mucormycosis in COVID-19: a systematic review of cases reported worldwide and in India. *Diabetes Metab Syndr* 2021;**15**:102146.
6. Rocha ICN, Hasan MM, Goyal S, et al. COVID-19 and mucormycosis syndemic: double health threat to a collapsing healthcare system in India. *Trop Med Int Health* 2021;**26**:1016–8.
7. Furman D, Campisi J, Verdin E, et al. Chronic inflammation in the etiology of disease across the life span. *Nat Med* 2019;**25**:1822–32.