CASE REPORT

Unique case of cutaneous *Cunninghamella* infection in an immunocompromised patient

Mauricio Portillo, Shyam Allamaneni, Richard Goodman

ABSTRACT

Cunninghamella species are an extremely rare cause of fungal infections. The usual mode of transmission is through inhalation; however, rare cases of cutaneous spread have been reported. The objective of this clinical case report is to highlight the uniqueness of which the patient acquired the infection, the progression, and control of it. A 57-year-old male with chronic lymphocytic leukemia was found to have an abscess next to his peripherally inserted central catheter (PICC) line. The abscess culture grew back Cunninghamella and was debrided and treated with isavuconazonium sulfate. The fungal infection was controlled and the total timeframe took 28 days. Rapid recognition and prompt treatment demonstrate the prevention of rapidly progressive angioinvasion and further systemic complications. This case also proves that isavuconazonium sulfate may be appropriate in controlling the spread of Cunninghamella species.

Keywords: Case report, Cutaneous infection, *Cunning-hamella*, Immunosuppression

How to cite this article

Portillo M, Allamaneni S, Goodman R. Unique case of cutaneous *Cunninghamella* infection in an immunocompromised patient. Edorium J Dermatol 2021;3:100003Do2MP2021.

Mauricio Portillo¹, Shyam Allamaneni², Richard Goodman³ <u>Affiliations:</u> ¹Saba University School of Medicine, 27 Jackson Rd., Suite #300, Devens, MA, USA; ²The Jewish Hospital Department of Surgical Oncology, 4777 E Galbraith Rd., Cincinnati, OH, USA; ³The Jewish Hospital Department of Infectious Disease, 4777 E Galbraith Rd., Cincinnati, OH, USA.

<u>Corresponding Author:</u> Mauricio Portillo, Saba University School of Medicine, 27 Jackson Rd., Suite #300, Devens, MA, USA; Email: m.portillo@saba.edu

Received: 16 March 2021 Accepted: 12 June 2021 Published: 16 July 2021 Article ID: 100003D02MP2021

doi: 10.5348/100003D02MP2021CR

INTRODUCTION

Cunninghamella species are known to be an extremely rare fungal infection that can cause mucormycosis in immunosuppressed individuals [1]. Similar to other related fungi, the mode of transmission is predominately through the inhalation of sporangial into the pulmonary system or sinuses [2]. It can be acquired through inhalation, inoculation, or possibly ingestion as the sporangial can be found airborne and on tree nuts and plants [1, 2].

Although extremely rare, *Cunninghamella* has been found to infect individuals through cutaneous means by a breakage in the skin barrier. Some of these cases include a motor vehicle accident, intravenous (IV) drug abuser, insulin injection, and contamination of Elastoplast dressing [1, 3]. Late diagnosis and/or treatment of these deep fungal infections could lead to disseminated infection and a high likelihood of mortality [1]. The uniqueness and extreme rarity of this case is what prompted a write-up.

This case demonstrates a new subtle way in which *Cunninghamella* species can be introduced percutaneously and the timeframe in which the disease progresses as a cutaneous infection. This case also looks to stress the importance of debridement in controlling *Cunninghamella* infections to prevent systemic infection and mortality.

CASE REPORT

A 57-year-old male with chronic lymphocytic leukemia, pancytopenia, and acquired hypogammaglobulinemia was referred to the outpatient care clinic with a painful right arm abscess. On close inspection, there was a skin tear next to the PICC line. The abscess was 2×2 cm area

EDORIUM Journals

Edorium J Dermatol 2021;3:100003D02MP2021. *www.edoriumjournalofdermatology.com*

of erythema with central necrosis as seen in Figure 1A. The abscess was incised, drained, cultured, and packed appropriately with gauze. The patient was provided with follow-up and appropriate wound care instructions.

The pus of the abscess was cultured and the results demonstrated *Cunninghamella* species. The specific species was not identified. The PICC line was removed and IV amphotericin B was prescribed. The patient was admitted to the hospital 16 days after the positive culture results for debridement because the wound continued to increase in size despite antifungal use. Wound care and follow-up were again stressed to the patient and they were later discharged that day.

The patient was readmitted into the hospital for subsequent debridement procedures as a result of nonhealing wound infection. A chest X-ray was performed to assess for pulmonary cause of infection; however, the results proved to be unremarkable. Post-op day 1 imaging after the third debridement is shown in Figure 1B. The patient was discharged 17 days later, on isavuconazonium sulfate and the wound appeared to be healing with granulation tissue. Figure 1C displays the wound on the final day prior to discharge.

Outpatient follow-up visit was performed nine days later to re-assess the wound and recovery. The wound recovery appeared to be rapid and the debridement successful in controlling the cutaneous infection as demonstrated in Figure 1D.



Figure 1: (A) Right arm abscess on first encounter prior to incision and drainage procedure. (B) Post-op day 1 after third debridement procedure. (C) Post-op day 16, demonstrating wound recovery, healing by secondary intention and granulation tissue formation. (D) Outpatient follow-up visit and assessment of wound.

The total amount of days from the initial presentation of the abscess to control of the infection through debridement was 28 days. It was presumed that the infection inoculation was likely due to the skin tear from the PICC line and a harbor of fungal growth on the dressing. There was no evidence of sinopulmonary infection to suggest possible systemic fungemia leading to cutaneous insult.

The patient is currently doing well and there are no reported new lesions or recurrence. Fungal blood cultures came back negative and the infection is presumed to be controlled.

DISCUSSION

Cutaneous instances of *Cunninghamella* infections appear to have rapidly progressive growth patterns as evidenced in this case and others with similar characteristics. High curative rates seem to be achievable if diagnosed and intervened early [4, 5]. Similar to other related fungi, serial aggressive debridements alongside antifungals are the mainstay of treatment [6]. Delay in intervention leads to angioinvasion and a high risk of mortality [1, 2, 6–8].

CONCLUSION

Immunocompromised states are the majority of risk factors for mucor infection. Therefore, a high index of suspicion is required to allow for appropriate and timely treatment. The usual treatment for invasive systemic mycoses is amphotericin B. While amphotericin B demonstrates effectiveness in the treatment of *Cunninghamella*, posaconazole appears to have similar efficacy and fewer side effects. Isavuconazonium sulfate is a new broad-spectrum azole drug that has proven effectiveness against *Cunninghamella* as evident in this case and clinical trials.

REFERENCES

- 1. Gomes MZR, Lewis RE, Kontoyiannis DP. Mucormycosis caused by unusual mucormycetes, non-Rhizopus, -Mucor, and -Lichtheimia species. Clin Microbiol Rev 2011;24(2):411–45.
- Ribes JA, Vanover-Sams CL, Baker DJ. Zygomycetes in human disease. Clin Microbiol Rev 2000;13(2):236– 301.
- 3. Spellberg B, Edwards J Jr, Ibrahim A. Novel perspectives on mucormycosis: Pathophysiology, presentation, and management. Clin Microbiol Rev 2005;18(3):556–69.
- 4. Motohashi K, Ito S, Hagihara M, Maruta A, Ishigatsubo Y, Kanamori H. Cutaneous zygomycosis caused by Cunninghamella bertholletiae in a patient with chronic myelogenous leukemia in blast crisis. Am J Hematol 2009;84(7):447–8.

EDORIUM Journals

Edorium J Dermatol 2021;3:100003D02MP2021. www.edoriumjournalofdermatology.com

- 5. Quinio D, Karam A, Leroy JP, et al. Zygomycosis caused by Cunninghamella bertholletiae in a kidney transplant recipient. Med Mycol 2004;42(2):177–80.
- 6. Ashkenazi-Hoffnung L, Bilavsky E, Avitzur Y, Amir J. Successful treatment of cutaneous zygomycosis with intravenous amphotericin B followed by oral posaconazole in a multivisceral transplant recipient. Transplantation 2010;90(10):1133–5.
- Pastor JF, Ruíz-Cendoya M, Pujol I, Mayayo E, Sutton DA, Guarro J. In vitro and in vivo antifungal susceptibilities of the Mucoralean fungus Cunninghamella. Antimicrob Agents Chemother 2010;54(11):4550-5.
- 8. Donnelley MA, Zhu ES, Thompson GR 3rd. Isavuconazole in the treatment of invasive aspergillosis and mucormycosis infections. Infect Drug Resist 2016;9:79–86.

Author Contributions

Mauricio Portillo – 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

Shyam Allamaneni – 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 Richard Goodman – 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

Guarantor of Submission

The corresponding author is the guarantor of submission.

Source of Support

None.

Consent Statement

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.

Copyright

© 2021 Mauricio Portillo et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.











Submit your manuscripts at

www.edoriumjournals.com







EDORIUM JOURNAL OF





JOURNAL OF CASE REPORTS AND IMAGES IN PATHOLOGY







PSYCHIATRY

EDORIUM JOURNAL OF

ANESTHESIA

JOURNAL OF CASE REPORTS AND IMAGES IN ORTHOPEDICS AND RHEUMATOLOGY







EDORIUM JOURNAL OF

INTERNATIONAL JOURNAL OF CASE REPORTS AND IMAGES





VIDEO JOURNAL OF BIOMEDICAL SCIENCE





INTERNATIONAL JOURNAL OF BLOOD TRANSFUSION AND IMMUNOHEMATOLOGY



