

Acute ulcerated botryomycosis in an immunocompromised patient

Fanny Carolina López-Jiménez¹, Judith Monserrat Corona-Herrera¹,
María José Chavez-Barajas², Marcela Saeb-Lima², Alejandra Aguilar-García³,
Silvia Mendez-Flores⁴

¹National Institute of Health Sciences and Nutrition Salvador Zubirán, Dermatology Department, CDMX, México, ²National Institute of Health Sciences and Nutrition Salvador Zubirán, Pathology Department, CDMX, México, ³Juárez de México Hospital, Internal Medicine Department. CDMX, México, ⁴Dermatology Specialist, at National Institute of Health Sciences and Nutrition Salvador Zubirán, Dermatology Department, CDMX, México

Corresponding author: Judith Monserrat Corona-Herrera, MD, E-mail: jch.fesi.mc@gmail.com

ABSTRACT

Botryomycosis is a rare chronic granulomatous bacterial infection with approx. 200 reported cases worldwide. The most common microorganism associated with this condition is *Staphylococcus aureus*. Botryomycosis manifests itself across a spectrum of lesions with a characteristic histological feature known as the Splendore–Hoepli phenomenon. Herein, we report the case of a 35-year-old female presenting with an ulcerated lesion and a previous diagnosis of lymphoblastic leukemia. Botryomycosis should be considered in immunosuppressed patients with ulcerated lesions. Treatment duration may range from four to eight weeks, with debridement if necessary. It is essential to exclude visceral involvement.

Key words: Botryomycosis, *Staphylococcus aureus*, Lymphoblastic leukemia, Immunosuppression

INTRODUCTION

Botryomycosis, also known as bacterial pseudomycosis or granular bacteriosis, is a chronic granulomatous bacterial infection [1-5]. About 200 cases have been reported around the world [6]. It may be caused by microorganisms such as *Staphylococcus aureus* (40% of cases) [7], *Pseudomonas aeruginosa*, *Proteus spp.*, or *Escherichia coli*. Two clinical forms of the disease have been described: one primarily affecting the skin, the other may manifest in organs such as the liver, lungs, brain, and heart [1-4]. Skin lesions usually take months to years to develop and ulcerate. The histological examination of skin lesions in botryomycosis reveals the development of an eosinophilic rim composed of antigen-antibody complexes, tissue remnants, and fibrin surrounding basophilic granules and bacterial colonies. This is known as the Splendore–Hoepli phenomenon [5,8]. Treatment includes the

administration of antibiotics and surgical drainage if necessary. Antibiotics should be continued from four to eight weeks [9].

CASE REPORT

A 35-year-old female patient was referred to our dermatology department after presenting ulcerated plaques, localized on the head, trunk, and lower extremities (Figs. 1a and 1b). There was no history of a preceding trauma. Her medical background revealed a prior diagnosis of lymphoblastic leukemia, for which she had undergone chemotherapy. She presented with severe neutropenia (0 neutrophils) and fever, leading to treatment with broad-spectrum antibiotics, along with prophylaxis using fluconazole and acyclovir. A biopsy was performed, resulting in a diagnosis of ulcerated botryomycosis (Figs. 2a – 2d). A culture of

How to cite this article: López-Jiménez FC, Corona-Herrera JM, Chavez-Barajas MJ, Saeb-Lima M, Aguilar-García A, Mendez-Flores S. Acute ulcerated botryomycosis in an immunocompromised patient. Our Dermatol Online. 2024;15(3):275-277.

Submission: 19.01.2024; **Acceptance:** 13.05.2024

DOI: 10.7241/ourd.20243.13



Figure 1: Clinical manifestations of the patient. (a) Ulcerated lesion. (b) Close-up views. In dermoscopy, it exhibits white, pink, and orange areas without a structure and perilesional scale.

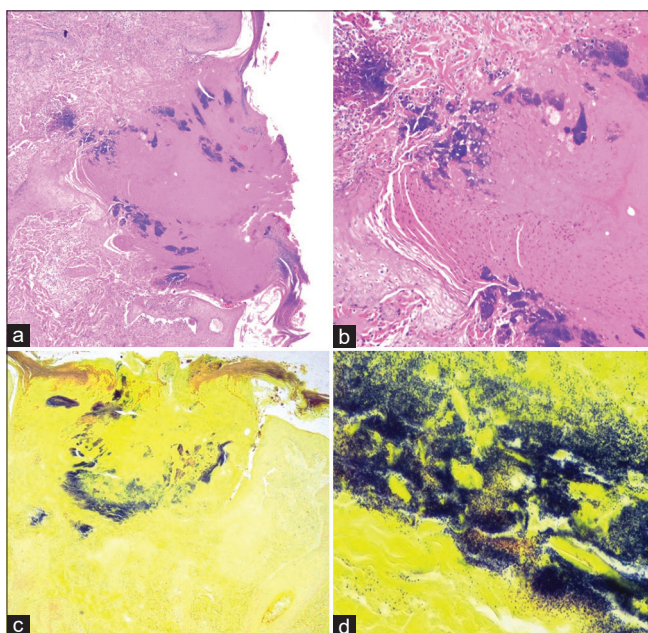


Figure 2: Histopathological findings. (a) Skin biopsy of the left mandible with an ulcer at epidermal level extending to the superficial and middle dermis with abundant fibrin, azurophilic granules, and bacteria (cocci), in addition to extensive acute inflammatory infiltrate with extensive hyalinization of the superficial and deep dermis (H&E; 4x) (b) Higher magnification (H&E; 40x) (c) Gram stain shows abundant bacteria (gram-positive cocci) (Gram; 4x). (d) Higher magnification (Gram; 40x).

tissue showed growth of *Staphylococcus aureus*. She was treated with dicloxacillin and completed a three-week regimen, successfully resolving the lesion.

DISCUSSION

Botryomycosis is an uncommon disease with a worldwide distribution. The cutaneous form of the disease represents approx. 75% of the reported cases [10]. Some conditions related to the host enhance the probability that the disease will develop. Studies suggest that at low concentrations, microorganisms are often phagocytized, yet a high

concentration may lead to necrotic lesions. Another theory has been proposed by Bonifaz suggesting that the development of botryomycosis requires a “delicate balance between the number of microorganisms entering, their low virulence, the patient’s tissue resistance, and a relative impairment of cellular immunity” [5].

The cutaneous form of the disease may manifest as nodules, sinus tracts, and abscesses [8]. These lesions take months to years to develop and ulcerate. However, unlike what is described in the literature, our patient presented with an ulcerated lesion of initial disease onset. Botryomycosis should be clinically distinguished from other conditions such as mycetoma, actinomycosis, nocardiosis, and actinomycetoma [11]. There has been no distinct morphological differentiation established among these conditions.

CONCLUSION

Botryomycosis is a rare yet noteworthy concern among hematologic patients. It should be considered in the differential diagnosis of ulcerated lesions in immunosuppressed patients. There is no consensus on the duration of treatment, yet a period of four to eight weeks is suggested. Thorough assessment is crucial to exclude any potential visceral involvement as well as ascertain the extent of penetration into deep tissues.

Consent

The examination of the patient was conducted according to the principles of the Declaration of Helsinki. The authors certify that they have obtained all appropriate patient consent forms, in which the patients gave their consent for images and other clinical information to be included in the journal.

REFERENCES

1. Hacker P. Botryomycosis. *Int J Dermatol.* 1983;:455-8.
2. Devi B, Behera B, Dash M, Puhan M, Pattnaik S, Patro S. Botryomycosis. *Indian J Dermatol.* 2013;58:406.
3. Machado C, Schubach A, Conceição F, Quintella L, Lourenço M, Carregal E, et al. Botryomycosis. *Dermatology.* 2005;211:303-4.
4. Wimlow DJ. Botryomycosis. *Am J Pathol.* 1959;35:153-67.
5. Bonifaz A. *Micología médica básica. Pseudomycosis por oportunistas.* 5º Ed. México. McGraw-Hill; 2015.487.
6. González-Cardona LP, Alejo Villamil AM, Cortés Correa C, Peñaranda Contreras EO. Exophytic botryomycosis: An unusual clinical presentation. *An Bras Dermatol.* 2022;97:536-8.
7. Arenas R, Torres E. *Micología Médica Ilustrada. Enfermedades por actinomicetos y bacterias.* 6º Ed, McGraw-Hill Interamericana de España S.L.; 2019.
8. Singh A, Cook C, Kollmann K, Rajpara A. Acute cutaneous botryomycosis of the hands. *IDCases.* 2020;19:e00709.

9. Pulido A, Bergón M, Mateos A, Parra V, Suárez R, Carretero F. Cutaneous botryomycosis mimicking ecthyma gangrenosum in a patient treated with ibrutinib. *Ann Hematol.* 2021;100):3065-6.
10. Padilla-Desgarenes C, Vázquez-González D, Bonifaz A. Botryomycosis. *Clin Dermatol.* 2012;30:397-402.
11. Sirka CS, Dash G, Pradhan S, Naik S, Rout AN, Sahu K. Cutaneous botryomycosis in immunocompetent patients: A case series. *Indian Dermatol Online J.* 2019;10:311-5.

Copyright by Fanny Carolina López-Jiménez, et al. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

Source of Support: This article has no funding source.

Conflict of Interest: The authors have no conflict of interest to declare.