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A case of blastomycosis presenting as a non-healing unifocal ulcerative nodule on the jawline

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Abstract

Blastomyces dermatitidis is a dimorphic fungus that can disseminate in the immunocompetent and immunodeficient. Many infected patients display heterogenous cutaneous findings, making it one of dermatology's great clinical mimics. Cases presenting as single lesions are often mistaken for neoplasms or other infections. We report a patient with diabetes mellitus who presented to the emergency department with a two-month history of an ulcerated jawline nodule. Treatment with incision/drainage and doxycycline for presumed bacterial abscess was unsuccessful. Upon re-presentation 10 days later, biopsy and fungal culture of the tumor confirmed infection with *B. dermatitidis*. Chest computed tomography revealed disease dissemination. Halfway through a 6-month course of itraconazole, cutaneous and pulmonary findings were notably improved. Diabetes is an emerging risk factor for dissemination that likely contributed to the severity in our case. Early biopsy can prevent potentially life-threatening treatment delays, highlighting the need for blastomycosis to be considered in the differential diagnosis of non-healing wounds.

Keywords: Blastomyces dermatitidis, dermatology, dimorphic, face, fungal, infectious disease, jawline, skin of color, ulcer, wounds

Introduction

Blastomycosis is a fungal infection caused by *Blastomyces dermatitidis* that can lead to chronic granulomatous and suppurative disease in humans [1,2]. The fungus is endemic to the Mississippi and Ohio River valleys, as well as regions in Africa and Canada [1,3-8]. *B. dermatitidis* grows as a mold in soil and decaying organic matter [1,5] and transforms into a yeast in the human body, propagating through broad-based budding [1,7]. Most infections occur through inhalation of spores, but disease may also occur through direct inoculation with contaminated soil [1,2,5,6,8,9].

In contrast to other dimorphic fungi, *B. dermatitidis* can evade immune defenses and cause systemic disease, even in immunocompetent hosts [1,3,5,8,10]. Up to 50% of those infected remain asymptomatic and mortality ranges from 4-22% in disseminated disease [4-7]. Although pulmonary disease is the most common clinical presentation, 40-80% of patients with disseminated blastomycosis manifest dermatologic findings [1,3-5,7]. Cutaneous involvement occurs by means of lymphohematogenous spread when not caused by primary inoculation [2,3,5,7] and cutaneous findings are often the first clue to an accurate diagnosis [5,8]. Representative cutaneous lesions may appear as hyperkeratotic nodules, papulopustular eruptions, verrucous plaques, or ulcerations with inflamed or violaceous borders. Lesions are often multifocal and commonly affect the face and extremities [1,2,4-6]. The disease presents a significant diagnostic challenge when presenting with atypical cutaneous manifestations. Herein, we report a case of blastomycosis masquerading as a single, non-healing, ulcerative nodule on the jawline of an

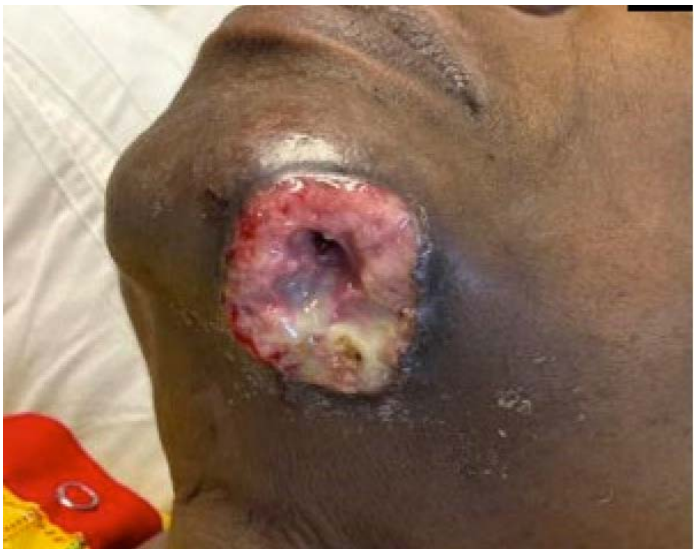

Figure 1. *Initial hospital presentation. Left jawline with 5cm×5cm well-defined, markedly tender, red, ulcerated nodule with indurated borders and white-to-yellow drainage.*

individual with skin-of-color living in the Midwestern United States.

Case Synopsis

A 69-year-old woman from Missouri with type 2 diabetes mellitus (DM) presented to the emergency department with an enlarging, painful, ulcerated nodule on her jawline that had been present for over two months. She reported it began as a pruritic bump believed to be from a mosquito bite. There was no history of trauma. Ten days prior to emergency department presentation, the nodule had been treated as a presumed cyst with incision and drainage and a 7-day course of doxycycline. Despite this, she continued to experience increased pain and mucopurulent drainage. She denied constitutional symptoms, dyspnea, or dysphagia. Physical examination revealed a 55cm ulcerated, draining, pink tumor with raised borders and central depression, without intraoral extension (**Figure 1**). Laboratory results revealed an unremarkable complete blood count and elevated hemoglobin A1c of 11.0%.

Biopsy of the tumor rim showed a suppurative and granulomatous dermatitis with fungal yeast forms demonstrating broad-based budding with multinucleated giant cells on hematoxylin and eosin

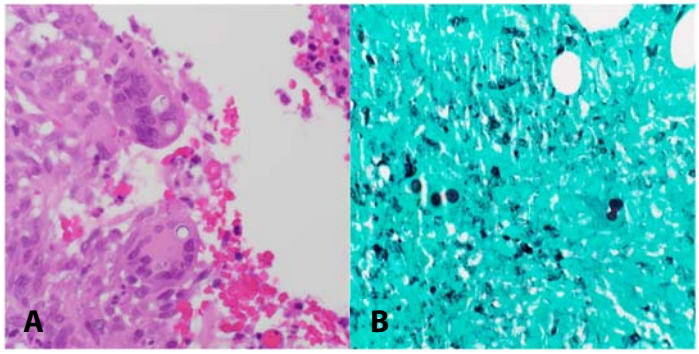

Figure 2. ***A)*** *H&E staining of punch biopsy specimen. Broad-based budding within multinucleated giant cells is visualized within a background of suppurative and granulomatous dermatitis, 400×.* ***B)*** *Grocott-Gomori methenamine silver staining highlights fungal yeast forms demonstrating the characteristic broad-based budding within multinucleated giant cells associated with Blastomyces spp, 400×.*

and on Grocott-Gomori methenamine silver staining (**Figure 2**). Bacterial and mycobacterial stains were unremarkable. Fungal culture grew *B. dermatitidis*. Urine *Blastomyces* antigen was positive at 2.97U/ml. Urine *Histoplasma* antigen was also positive at 1.03ng/ml but was attributed to cross-reactivity given histopathologic and fungal culture results. Blood cultures, β-D-glucan, *Treponema pallidum*, hepatitis C, and HIV tests were unremarkable. Neck computed tomography (CT) showed an irregular rim-enhancing fluid collection. Chest CT revealed multiple pulmonary nodules and a left lower lobe consolidation with ground-glass opacities. Magnetic resonance imaging of the brain was unremarkable.

The patient was started on a 6-month course of itraconazole 200mg daily for disseminated primary pulmonary blastomycosis. At two-month follow-up, the jawline ulceration demonstrated interval improvement and decreased in size to a non-tender, smooth, brown, indurated nodule (**Figure 3**). Chest CT at three months revealed improvement of the left lower lobe consolidation and resolution of the pulmonary nodules.

Case Discussion

Several cases in the literature have reported blastomycosis presenting as non-healing facial ulcers, which may be either multifocal or associated with other dermatologic findings, or less commonly,

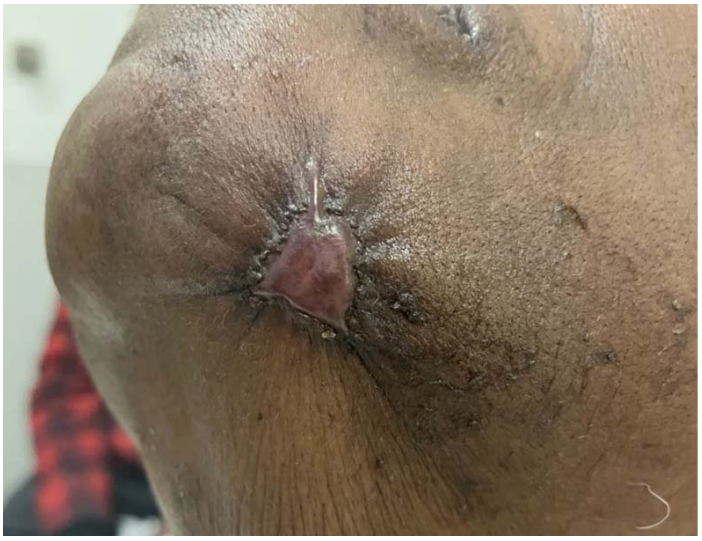

Figure 3. *Presentation after two months of antifungal therapy. Left jawline with one-cm indurated, smooth, brown, non-tender nodule.*

as a single ulcerative nodule [1,2,4,6,9]. Blastomycosis has a heterogenous morphology and ulceration is a less common dermatologic manifestation [1,2,4-7]. Although previous cases have described centrifugal spread of single or multiple papules or pustules, the border of our patient's ulcer was elevated but did not contain pustules, which is a diagnostic clue described by some authors [1,4,5]. When unifocal, these ulcers can be mistaken for other conditions such as pyoderma gangrenosum and malignancies such as lymphoma and squamous cell carcinoma [1,2,4-6]. A unifocal lesion may also mimic a bacterial abscess, which led to the prior incision and drainage and course of doxycycline for our patient. Other preliminary diagnostic considerations in our patient included a mycobacterial infection, other deep fungal infections, syphilis, nocardiosis, and actinomycosis given the jawline location [1,2,4-6].

Previous case reports have shown that pulmonary involvement in blastomycosis is variable and when present, is often discovered only after histopathological diagnosis [1,2,4,6,7,9]. Our patient lacked constitutional symptoms typical of pulmonary blastomycosis and lung involvement was discovered after diagnosis of the cutaneous lesion [5]. In cases of suspected invasive fungal infection, skin biopsy demonstrating a characteristic dermatitis pattern in conjunction with broad-based budding plays a critical role in confirming an etiologic agent early in the diagnostic process and avoiding treatment delays, as fungal cultures may take two to five weeks to result, and cross-reactivity between dimorphic species in urine antigen tests is common [1,3,5-7]. The standard therapy for all patients diagnosed with blastomycosis is antifungal treatment. Although mild to moderate cases are typically managed with itraconazole as the first-line therapy, more severe cases involving disseminated disease, central nervous system involvement, or underlying immunosuppression can be treated with polyene amphotericin B formulations [10-11]. Itraconazole is then commonly used as a step-down therapy after treatment induction with amphotericin B [10-11]. After treatment initiation, serial urine antigen tests are valuable in monitoring treatment response [10, 12]. If cutaneous dissemination does not fully heal with antifungal therapy, surgical intervention may be necessary [1].

Previous studies have found that the incidence of disseminated *B. dermatitidis* is similar for immunocompromised and immunocompetent adults, suggesting that pathogen-related factors may play a larger role than host immune defenses in pathogenesis [1,3,5,8,13]. Alternatively, multiple authors have determined that DM is an independent risk factor for disease, with similar incidence between immunocompromised and DM patients [1,8,13]. Uncontrolled DM may have been a predisposing factor in our patient's presentation and underscores the importance of considering blastomycosis in patients with DM, particularly in endemic regions.

Conclusion

This novel case highlights the importance of including dimorphic fungal infections in the evaluation of ulcerative nodules, even in a patient without a known source of trauma or systemic symptoms, to expedite evaluation of systemic involvement and treatment.

Potential conflicts of interest

The authors declare no conflicts of interest.

References

1. Mason AR, Cortes GY, Cook J, Maize JC, Thiers BH. Cutaneous blastomycosis: A diagnostic challenge. *Int J Dermatol*. 2008;47:824-830. [PMID: 18717863].
2. Elmas ÖF, Uyar B, Kilitçi A. A 25-Year History of Leg Ulceration: Cutaneous Blastomycosis. *Dermatol Pract Concept*. 2020;10. [PMID: 3264230].
3. McBride JA, Sterkel AK, McBride JA, et al. Clinical Manifestations and Outcomes in Immunocompetent and Immunocompromised Patients With Blastomycosis. *Clinical Infectious Diseases*. 2021;72:1594-1602. [PMID: 32179889].
4. Owen WR, Thurs K, Kim DS. Blastomycosis presenting as a nonhealing wound. *Adv Skin Wound Care*. 2012;25:321-3 [PMID: 22713783].
5. Pullen MF, Alpern JD, Bahr NC. Blastomycosis—Some Progress but Still Much to Learn. *Journal of Fungi*. 2022;8. [PMID: 36012812].
6. Ortega-Loayza AG, Nguyen T. Cutaneous Blastomycosis: A Clue to a Systemic Disease. *Blastomicose Cutânea: Um Sinal de Doença Sistêmica*. 2013;88. [PMID: 23739696].
7. Abdelfattah, A. H., Ayub, S., Talib, M., Dawoud, H., & Talib, U. Disseminated Blastomycosis in an Immunocompetent Patient. *Cureus*, 2022;14. [PMID: 36407244].
8. Lemos LB, Baliga M, Guo M. Blastomycosis: The great pretender can also be an opportunist. Initial clinical diagnosis and underlying diseases in 123 patients. *Ann Diagn Pathol*. 2002;6:194-203. [PMID: 12089732].
9. Shah H, Carstensen T, Kmeid M, Bialowas C. Cutaneous Blastomycosis Presenting as a Nonhealing Wound in the Northeast United States: A Case Report. *Int J Low Extrem Wounds*. 2022 [PMID: 36426539].
10. McBride JA, Gauthier GM, Klein BS. Clinical Manifestations and Treatment of Blastomycosis. *Clin Chest Med*. 2017;38:435-449. [PMID: 28797487].
11. Schwartz IS, Kauffman CA. Blastomycosis. *Semin Respir Crit Care Med*. 2020;41:31-41. [PMID: 32000282].
12. Frost HM, Novicki TJ. Blastomyces Antigen Detection for Diagnosis and Management of Blastomycosis. *J Clin Microbiol*. 2015;53:3660-3662. [PMID: 26338856].
13. Villacorta Cari E, Leedy N, Ribes JA, Soria J, Myint T. Risk factors of severe blastomycosis and comparison of diagnosis and outcomes between immunocompetent and immunocompromised patients. *Mycoses*. 2022;65:239-246. [PMID: 34874582]