An Unusual Presentation of Blastomycosislike Pyoderma (Pyoderma Vegetans) and a Review of the Literature

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We report a case of blastomycosislike pyoderma (BLP), also known as pyoderma vegetans, in a 75-year-old woman. The patient initially presented with multiple lesions, some classic and others with highly uncharacteristic morphology. The appearance of unusual purulent hornlike lesions on both cheeks delayed recognition of the disease. The diagnosis of BLP is contingent on the fulfillment of 6 diagnostic criteria and the disease is highly refractory to current treatment strategies. We review the present understanding of disease pathology and available therapeutic modalities.

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The term *blastomycosislike pyoderma* (BLP) was coined by Yaffee[1] in 1960 to describe a lesion resembling cutaneous blastomycosis arising in a tattoo. However, De Azua and Sada y Pons[2] were the first to report the condition in 1903 as *pseudo-épithéliomas cutanés*. Blastomycosislike pyoderma generally presents with verrucous exudative plaques with multiple pustules and elevated irregular borders most often occurring on the extremities or the head and neck regions. It is most commonly reported in immunocompromised patients and is thought to be caused by an unusual inflammatory tissue reaction to a microbial infection.[3] In the past 50 years, there have been numerous case reports of BLP, also commonly referred to as pyoderma vegetans, illustrating the refractory nature of the disease and the difficulty many clinicians have in treating it. Simple excision generally leads to recurrence in a period of a few months; therefore, the search for new and effective treatments continues.

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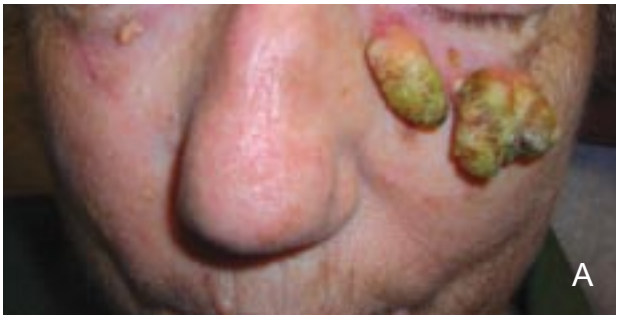
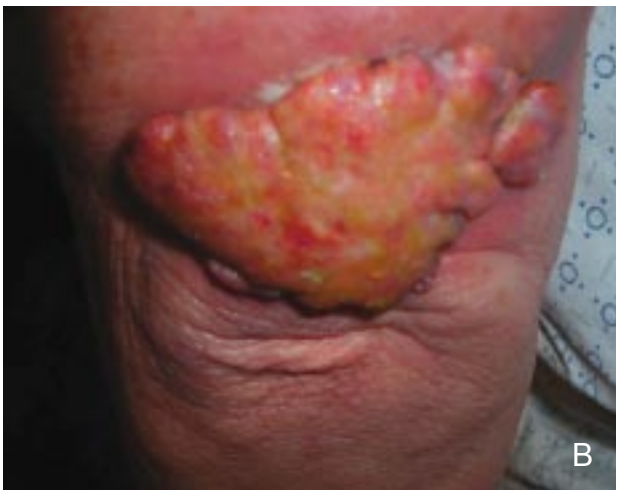
Case Report

A 75-year-old woman presented with a 3-year history of gradually enlarging plaques on her left arm and both cheeks. These lesions had been treated with excision and broad-spectrum antibiotics without improvement. Oral dapsone was initiated at an outside institution for presumed Sweet syndrome and, according to the patient, the lesions initially resolved. Unfortunately, the patient became anemic after 2 weeks on dapsone and the medication was discontinued. Medical records from that time indicated notable interval improvement of the skin lesions and no further treatment was indicated. However, the lesions recurred and progressed several weeks after stopping dapsone therapy.

The patient had a complicated medical history, including a remote history of breast cancer treated with mastectomy and chemotherapy. She had adrenal insufficiency and was on a prolonged prednisone taper. She denied using medication containing halogens. She had no recent travel history and no contact with animals or plants. Review of systems was remarkable for fatigue and night sweats.

Physical examination revealed 2 large, erythematous, purulent cutaneous horns under the left eye and 1 under the right eye (Figure 1A). There also was an erythematous, vegetative, purulent plaque on her left elbow (Figure 1B) and an erythematous nodule on her upper left arm (Figure 1C).





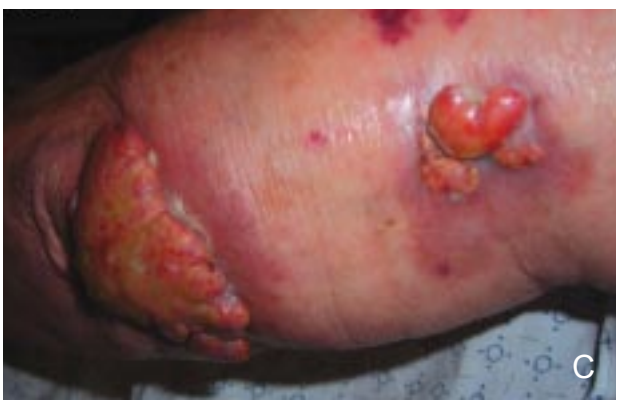
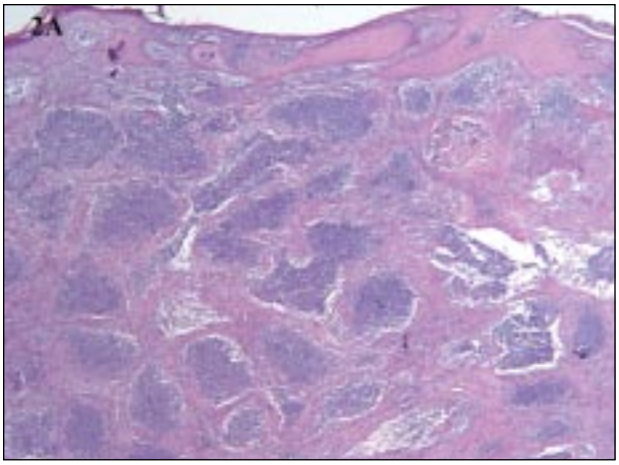
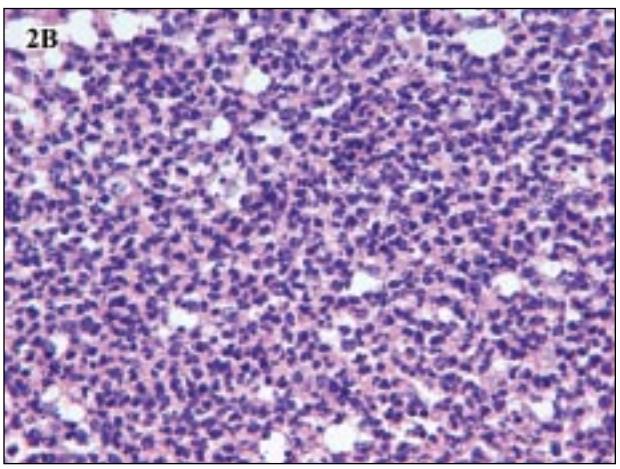


Figure 1. Two erythematous purulent cutaneous horns on the upper left cheek (A). There also was a vegetative purulent plaque on the left elbow resembling coral reef granuloma (B) and an erythematous nodule on the upper left arm (C).

No lymphadenopathy or hepatosplenomegaly were appreciated. A biopsy specimen from the lesion on the left cheek revealed marked pseudoepitheliomatous hyperplasia with coalescing neutrophilic microabscesses (Figures 2A and 2B). Fungal and mycobacterial stains were negative for organisms. Gram stain revealed gram-positive cocci in clusters (Figure 2C). Cultures of tissue biopsies were negative





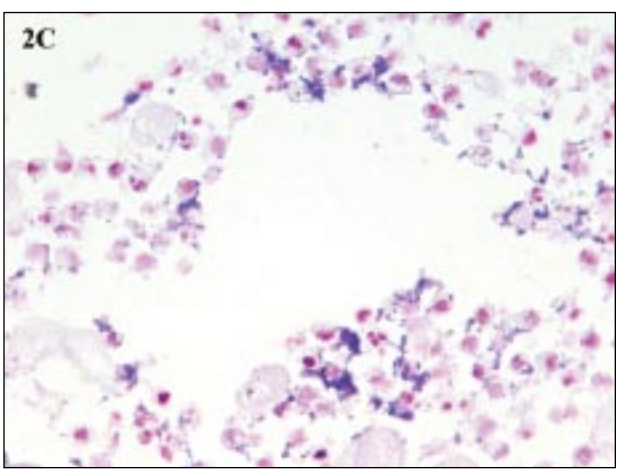


Figure 2. Histopathologic features of blastomycosislike pyoderma. A biopsy specimen from the lesion on the left cheek revealed pseudoepitheliomatous hyperplasia with a dense inflammatory infiltrate (A) and neutrophilic microabscess formation (B)(H&E; original magnifications ×4 and ×40, respectively). Gram-positive cocci in clusters also were present (C)(original magnification ×60). Biopsies from other lesions contained similar histopathology.

for fungi and mycobacteria. Bacterial culture revealed mixed flora, including *Proteus mirabilis*, *Corynebacterium* species, and *Staphylococcus aureus*. An excision biopsy specimen of the left elbow lesion revealed similar histologic findings with Gram stain negative for organisms; cultures of tissue biopsies again revealed mixed flora, including *Enterococcus* species and *Serratia marcescens*. Laboratory testing revealed an elevated white blood cell count of 11,250/uL (reference range, 3280–9290/uL), with a differential of 91% neutrophils, 3% lymphocytes, and 1% each of metamyelocytes and myelocytes. The patient's CD4 lymphocyte count and CD8 lymphocyte count were both substantially depressed with values of 95/mm^3 (reference range, 355–1426/mm^3) and 65/mm^3 (reference range, 255–1090/mm^3), respectively; however, flow cytometry was within reference range. Fungal serologies were negative and iodide and bromide levels were within reference range.

Blastomycosislike pyoderma was diagnosed and the patient underwent reexcision of her lesions and, in consultation with the infectious disease service, was started on culture-directed treatment with amoxicillin as well as trimethoprim-sulfamethoxazole prophylaxis for her low CD4 count. She declined further diagnostic tests to delineate the etiology of the low T lymphocyte counts.

Comment

Blastomycosislike pyoderma is a rare disorder generally characterized by verrucous exudative plaques with multiple pustules and elevated irregular borders. It is primarily seen in immunocompromised patients, with cases reported in patients with chronic myeloid leukemia, underlying neoplasm, diabetes mellitus, pulmonary granuloma, AIDS, local trauma, alcoholism, ulcerative colitis, psoriatic arthritis, immunosuppressive therapy, previous x-irradiation, and a decrease in chemotactic activity of polymorphonuclear leukocytes.[1,3-8] It is important to note that these lesions may be the first manifestations of serious underlying disease; however, at least 10 cases of classic BLP have been reported in immunocompetent patients.[3,5,9,10] In Australia, a variant of BLP has been described and is termed *coral reef granuloma*, which has a similar clinical and histologic appearance to classic BLP but characteristically occurs in areas of severe actinic damage in immunocompetent elderly patients.[11,12] Actinic damage decreases the immune response in skin[13] and may lead to local conditions normally seen only in immunocompromised patients.

It is thought that BLP is caused by an unusual inflammatory tissue reaction to a microbial infection; isolation of a pathogenic microbe is one criterion for diagnosis.[3] *Staphylococcus aureus* is the most commonly isolated pathogen from affected tissue, though cases of *Streptococcus pyogenes*, *Escherichia coli*, *Pseudomonas aeruginosa*, *Proteus* species, *Candida albicans*, diphtheroid organisms, *Bacillus* species, *Clostridium perfringens*, and enterococci have all been noted.[3,14,15] It is common to find multiple species isolated from the same lesion.[3,14,15] Our patient presented with multiple lesions, and the lesion on her arm grew different organisms than the lesion on her face. It is possible that our patient's lesions grew different bacteria because different areas of the body are susceptible to colonization by different microorganisms and these microorganisms may cause infection if a breach in the skin barrier occurs. For instance, areas that are commonly covered are more likely to be colonized with gram-negative bacilli than drier uncovered areas.[16] Our patient's elbow lesion, which she always kept covered, grew *Enterococcus* species and *S marcescens*. Her facial lesion grew *Corynebacterium* species, an organism likely to colonize areas rich in sebaceous glands, and *S aureus*, which most commonly colonizes the nearby nares.[16]

In 1979, Su et al[3] reviewed 7 cases of BLP and proposed 6 diagnostic criteria for the disease: (1) isolation of a pathogenic microbe; (2) clinical presentation of large verrucous plaques with multiple pustules and elevated borders; (3) microscopic findings of pseudoepitheliomatous hyperplasia with neutrophil-rich microabscesses within the epidermis and dermis; (4) negative culture for deep fungi, atypical mycobacteria, and *Mycobacterium tuberculosis*; (5) negative fungal serology test result; and (6) bromide level in the blood within reference range.

Our patient fulfilled all 6 criteria; however, the marked variation in the character of her lesions had not been previously reported for BLP. The patient had classic lesions on her left arm, but the lesions on her face uncharacteristically protruded, resembling large cutaneous horns. These hornlike lesions and the more characteristic lesions responded to our treatment strategies in similar fashions. Culture-driven antibiotics offered limited to no improvement and, as has often been reported, all lesions returned to original size several months after excision. The literature supports our observation that achieving sustained resolution of lesions is difficult. As BLP is an extremely rare diagnosis, there have been no randomized, clinically controlled trials examining the comparative efficacies of different treatment regimens. Certain case reports have demonstrated temporary resolution of lesions with empiric antibiotics while others have reported success with

culture-driven antibiotics combined with other therapies.[3,9,14,17,18] In contrast, as seen in our patient, there are many instances in which organism-specific antibiotics have been unsuccessful.[3,15,19,20] As the disease stems from an overactive immune response, it is possible that antibiotics with anti-inflammatory properties may be more useful than others without this characteristic; case reports of successful treatment with the tetracycline antibiotics and cotrimoxazole have been published.[18,19] Curettage, cryosurgery, acitretin, electrodesiccation, excision, disodium chromoglycate, CO_2 laser debridement, and etanercept also have been attempted with some reports of success, but on the whole, long-term follow-up did not occur.[4-8,14,15,18-20] The use of permanganate soaks, systemic steroids, potassium iodide, and superficial x-ray therapy have been reported with poor results.[15] Our recommendation is to start with long-term culture-driven antimicrobial therapy to prevent the emergence of new bacterial resistance patterns and to eradicate the identified organisms with the utmost efficacy; when applicable, choose a regimen with anti-inflammatory properties to treat the underlying pathophysiologic disturbance at the same time.

Conclusion

This case illustrates the degree to which human disease presentation may vary from one individual to the next. Unfortunately, during a year of follow-up, the patient's lesions continue to recur, illustrating the importance of new treatment strategy development and the continued pursuit of deeper understanding of the disease process.

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