A 69-year-old Caucasian male presented with a 4-month history of a slowly expanding, verrucous plaque of the left upper cutaneous lip. The lesion reportedly began as an abscess and had undergone incision and drainage, followed by multiple unsuccessful courses of oral antibiotics prior to presentation to our clinic. The patient reported that the area was occasionally itchy, but otherwise asymptomatic. Review of systems was negative for any systemic symptoms. The patient denied any preceding illnesses, changes in medications, or previous skin cancers. His social history was significant for an extensive international travel history, though he denied any known exotic exposures. His hobbies included gardening and tree planting near his home in the mountains of western North Carolina, where the patient was residing when the lesion started.

Physical examination revealed a verrucous, erythematous plaque with papillomatous borders and central ulceration on the left upper cutaneous and vermilion lip extending to the nasolabial fold (Figure 1). A small erythematous plaque with overlying serous crust was noted on the patient’s right upper back, which the patient reported had been present for approximately 20 years.

Histopathology

Histopathological examination of multiple punch biopsies showed pseudopodiphleomatous hyperplasia with intraepidermal pustules containing neutrophils and eosinophils. Stains were Periodic acid-Schiff (PAS)-, HSV-, and VZV-negative. A pancytokeratin stain showed no evidence of squamous cell carcinoma. Direct immunofluorescence was negative. Indirect immunofluorescence for skin autoantibodies was negative. Three separate tissue culture specimens showed no bacterial, fungal, or mycobacterial growth.

Leishmania PCR and DNA sequencing was negative. An additional punch biopsy (#7) revealed yeast forms with broad-based budding and refractile walls highlighted with Gomori Methenamine Silver (GMS) stain of the tissue, consistent with cutaneous blastomycosis (Figures 3-5).

Clinical Course

A chest X-ray demonstrated no pulmonary involvement. In collaboration with an infectious disease specialist, the patient was initiated on therapy with itraconazole 200 mg twice daily for a total of 6 months. Side effects during itraconazole therapy included lower extremity edema, a 20-lb weight gain, gastrointestinal upset, blurred vision, and a transient increase in blood pressure, all of which resolved once off the medication. Complete resolution of both the face and back lesions was noted at the completion of the treatment course. At six-month follow-up, residual scarring and alopecia were noted in parts of the previously affected areas of the beard and nasolabial fold (Figure 2).

Discussion

Blastomycosis is a fungal infection caused by Blastomyces dermatitidis, a thermally dimorphic fungus endemic in the soils of the Ohio and Mississippi River Valleys and southeastern United States.1 It most commonly manifests as a pulmonary infection following inhalation of spores, which may be asymptomatic and therefore undetectable. Extrapulmonary disease occurs in ~ 25-30% of patients after hematogenous dissemination from the lungs, with the skin being the most common site of extrapulmonary disease.2 Primary cutaneous blastomycosis is quite rare and occurs due to direct inoculation after trauma to the skin via an infected animal bite, direct inoculation in laboratory settings, or due to injury during outdoor activities involving contact with soil.3 Given our patient’s horticultural hobbies, lack of pulmonary symptoms, and negative radiological examination, primary cutaneous blastomycosis infection is a possibility, though it is difficult to definitively ascertain whether our case represents primary or secondary cutaneous blastomycosis.

Clinically, cutaneous blastomycosis starts as papules that evolve into crusts, vegetative plaques often with central clearing or ulceration. It can be mistaken for squamous cell carcinoma, penicillatus vegetans, leshmaniasis, bacterial pyoderma, and other deep fungal infections, therefore histopathologic examination and tissue culture are crucial to the diagnosis. On histopathology, pseudopodiphleomatous hyperplasia with neutrophilic abscesses is seen. Organisms can be difficult to identify and are often found within histiocytes or abscesses in the dermis. The yeasts are 8 to 15 μm in diameter with thick, double-contoured walls and display broad-based budding.4 Despite stains including GMS and PAS, blastomycosis can be a very difficult diagnosis and it is important to note that a negative result does not exclude the possibility of blastomycosis, as demonstrated in this case. Culture is certainly the most sensitive method for detecting and diagnosing blastomycosis. Growth is typically detected in 5 to 10 days, but can take up to 30 days if few organisms are present in the specimen.1

Though spontaneous remission can occur, it is recommended that all patients with cutaneous blastomycosis be treated to avoid dissemination and recurrence. Itraconazole is currently the treatment of choice.5 Doses are typically 200 to 400 mg per day for 6-12 months.6

References


CASE PRESENTATION

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