Severe Oculo FACIAL Sequelae of Cutaneous Blastomyces dermatitidis

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Abstract: This study reports a case of Blastomyces dermatitidis soft tissue infection resulting in a disfiguring lower eyelid ectropion from cicatricial and postinflammatory cutaneous changes. Primary treatment included intravenous amphotericin B followed by long-term oral itraconazole, which resulted in complete remission of the disease without debridement, after which cicatricial ectropion was repaired surgically with scar release, full-thickness skin graft, and temporary Frost tarsorrhaphy. Cutaneous blastomycosis may cause severe oculofacial sequelae, ranging from eyelid ectropion to widespread facial cicatrix, and may mimic other more common infectious processes, in addition to malignancy. Recommended antifungal therapy includes induction with intravenous amphotericin B and a long course of oral antifungals, preferably coordinated in conjunction with an infectious disease specialist. Ectropion repair should be delayed until the inflammatory response has completely healed. If the ocular surface is compromised or nearby ocular structures are threatened, primary debridement and repair may be indicated.

Blastomyces dermatitidis, the etiologic agent of blastomycosis, causes pyogranulomatous inflammation. Infection is primarily acquired by inhalation of aerosolized spores following disruption of soil to cause pneumonia, which can be followed by lymphohematogenous dissemination to the skin. This pathogen can also cause primary cutaneous disease by a penetrating injury to the skin. This report illustrates a patient with severe facial and periorcular blastomycosis complicated by cicatricial lower eyelid ectropion. Primary treatment and secondary repair are discussed. The medical literature is reviewed for recent clinical and microbiologic updates on blastomycosis. Informed written consent was obtained for the use of clinical photographs from the patient, and this study is Health Insurance Portability and Accountability Act (HIPAA) compliant.

CASE REPORT

A 45-year-old man in August of 2010 was evaluated for bilateral facial lesions (Figure), OS irritation, malaise, and weight loss. He developed periauricular and cervical lymphadenopathy, and purulent nodules involving his face, trunk, and upper and lower extremities 14 months prior to presentation. Over a period of several weeks after initial presentation, several cutaneous lesions spontaneously resolved, but those on his face and buttocks persisted. In 2009, he sought medical attention at an urgent care facility for the facial lesions. He was prescribed clindamycin for 10 days but was unable to complete the antibiotic course due to the development of a drug rash. From December 2009 to August 2010, the patient experienced progression of his facial and buttock lesions. He was diagnosed with cutaneous Blastomycosis based on skin biopsy demonstrating broad-based budding yeast and a positive urine Blastomyces antigen. Culture of the skin lesions grew B. dermatitidis. Following induction therapy with liposomal amphotericin B, which was stopped at 9 days due to development of a morbilliform rash from the amphotericin, he was treated with oral itraconazole solution for 9 months. Itraconazole levels were monitored monthly and were therapeutic (>1 μg/ml) throughout the treatment course. Therapy was discontinued due to side effects of headache, nausea, and fatigue. Surgical debridement of infected tissue was not required to clear the active infection, as appropriate medical treatment yielded dramatic results. (Figure). The verrucous lesions ultimately improved after several months of the above regimen, leaving large cheek and left lower eyelid cicatricial plaques in its wake. The resulting left lower eyelid cicatricial ectropion was repaired in a single-stage procedure 9 months after finishing antifungal therapy. The scar was released. A 35 mm × 17 mm full-thickness skin graft was taken from the right retroauricular area to exactly match and fill the defect, and then a lateral tarsal strip procedure was performed, followed by temporary Frost tarsorrhaphy.

DISCUSSION

Blastomyces dermatitidis grows in the moist soil of wooded areas rich in organic debris of the Mississippi and Ohio River basins, United States, and Canadian regions bordering the Great Lakes and the St. Lawrence River Valley. It is hyperendemic in north-central Wisconsin with an incidence up to 40 per 100,000 in certain counties. The fungus is thermally dimorphic, existing as a filamentous mold in the soil (22–25°C) and as a pathogenic yeast in the tissue (37°C). Following disruption of soil, aerosolized mycelial fragments and infectious spores are inhaled into the lungs and converted into broad-based budding yeast. Once established in the lung, yeast can spread to extrapulmonary sites, most often to the skin, bone, and genitourinary system. Integumentary manifestations occur in up to 80% of patients with disseminated infection, with predilection for the face and exposed areas. Eyelid involvement with systemic blastomycosis infection is rare, occurring in less than 2% of cases. Cutaneous blastomycosis may cause severe oculofacial sequelae, ranging from eyelid ectropion to widespread facial cicatrix. Rarely, intraocular involvement and optic nerve infection have also been reported.

Although integumentary manifestations are common after hematogenous dissemination, direct inoculation of the skin is also possible after a penetrating injury.

Cutaneous and systemic manifestations of blastomycosis may mimic other more common infectious processes and malignancy. Understanding the risk factors associated with acquisition of B. dermatitidis is important for prompt diagnosis. Practice guidelines published by the Infectious Diseases Society of America recommend that patients with severe infection be treated with intravenous amphotericin B followed by a prolonged course of oral antifungals such as itraconazole. Coordination with an expert in infectious diseases is recommended.

Whenever possible, surgical correction of oculofacial sequelae should be delayed until clearance of infection. Delay may not be feasible when nearby ocular structures are threatened or when refractory lagophthalmos or severe retraction threaten to compromise protection of the ocular surface. In these cases, earlier surgical intervention should be considered.
Proptosis Caused by Rhabdomyomatous Mesenchymal Hamartomata Occurring in the Orbit

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Abstract: Two infants were referred for progressive orbital proptosis. MRI in both cases demonstrated a homogenous mass in the orbit adherent to and isointense with a rectus muscle. Histopathology in both cases demonstrated a bland proliferation of spindle cells with entrapped skeletal muscle. Immunohistochemistry demonstrated that the abnormal tissue was of skeletal muscle origin, consistent with rhabdomyomatous mesenchymal hamartoma (RMH). Observation was elected due to the reported benign nature.

REFERENCES


