Primary Cutaneous Histoplasmosis of the Head and Neck

Blake S. Raggio, MD[1,2]

[1]Tulane University Medical Center, Department of Otolaryngology; [2]Ochsner Health System of New Orleans, Department of Otolaryngology

ABSTRACT

OBJECTIVES:
Discuss the clinical presentation, diagnosis, and management of a patient with primary cutaneous histoplasmosis (PCH).

METHODS:
Report a unique case of primary cutaneous histoplasmosis (PCH) and review the salient literature.

RESULTS:
An 80-year-old male farmer presented with a 3-month history of a painful, non-healing, ulcerative pinna lesion refractory to antibiotics and local wound care. The patient also reported a more recent development of similar ulcerative lesions involving the oral tongue and floor of mouth. Biopsy revealed a diffuse granulomatous inflammatory process associated with *Histoplasma capsulatum*. The patient had no clinical or laboratory findings suggesting systemic infection. Itraconazole was begun for treatment of PCH and oral histoplasmosis. The pinna and oral cavity lesions began to improve shortly after treatment initiation; however, the medication was withdrawn 3 weeks into therapy due to kidney failure, which ultimately the patient succumbed to.

CONCLUSION
PCH presents a diagnostic challenge due to its rarity and varied clinical presentation. We present a rare case of primary cutaneous histoplasmosis of the head and neck and discuss its successful diagnosis and management.

CONTACT

Blake Raggio, MD
Tulane University Medical Center
Department of Otolaryngology
1430 Tulane Ave, SL-59
New Orleans, LA 70112
504-235-3994
braggio@tulane.edu

INTRODUCTION

Cutaneous histoplasmosis of the head and neck without evidence of systemic infection is extremely rare. Herein, we report a unique case of primary cutaneous histoplasmosis of the head and neck and describe its clinical presentation, diagnosis, and management.

CASE REPORT

An 80-year-old male farmer with arthritis, anemia, chronic kidney disease, and a history of shingles presented with a 3-month history of a painful, non-healing, ulcerative pinna lesion refractory to antibiotics and local wound care **(Figure 1)**. The patient reported a subsequent development of similar ulcerative lesions involving the oral tongue and floor of mouth **(Figure 2)**. Biopsy of the pinna and oral cavity revealed numerous epithelioid histiocytes containing small, circular yeast cells compatible with Histoplasma capsulatum **(Figure 3A,B)**. Special stains for fungal organisms, including the Gomori's methenamine silver (GMS) **(Figure 3C)** and the periodic acid-Schiff (PAS) **(Figure 3D)** were strongly positive. The patient had no clinical or laboratory findings to suggest systemic fungal infection, thus oral itraconazole was initiated for treatment of primary cutaneous histoplasmosis with dissemination to the oral cavity. Despite a promising initial clinical response, the medication was withdrawn several weeks into therapy due to the patient's worsening renal function. Hemodialysis was initiated; however, the patient's health continued to deteriorate, and he passed away shortly thereafter from renal associated heart and liver issues.

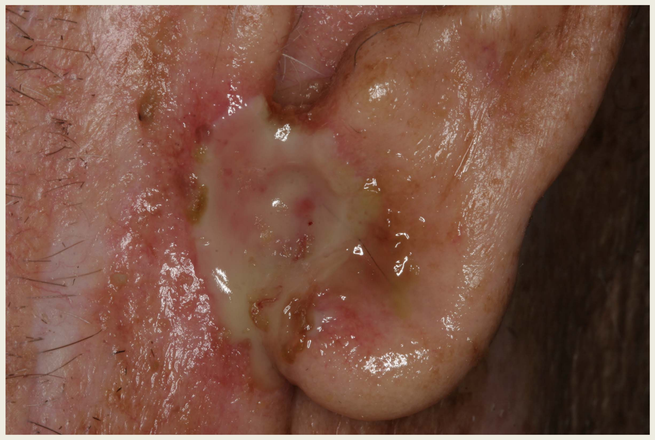

Fig 1. Weeping, ulcero-nodular lesion of the left pinna and pre-auricular area..

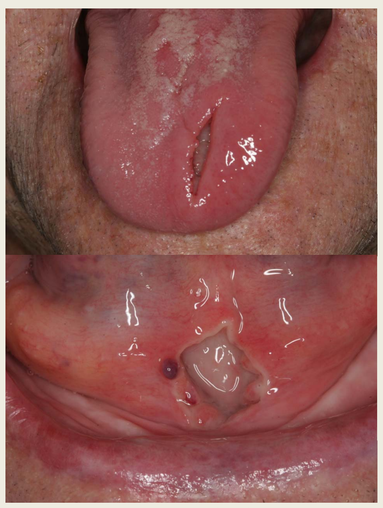

Fig 2. Ulcerative lesion of the oral tongue (top) and floor of mouth (bottom)

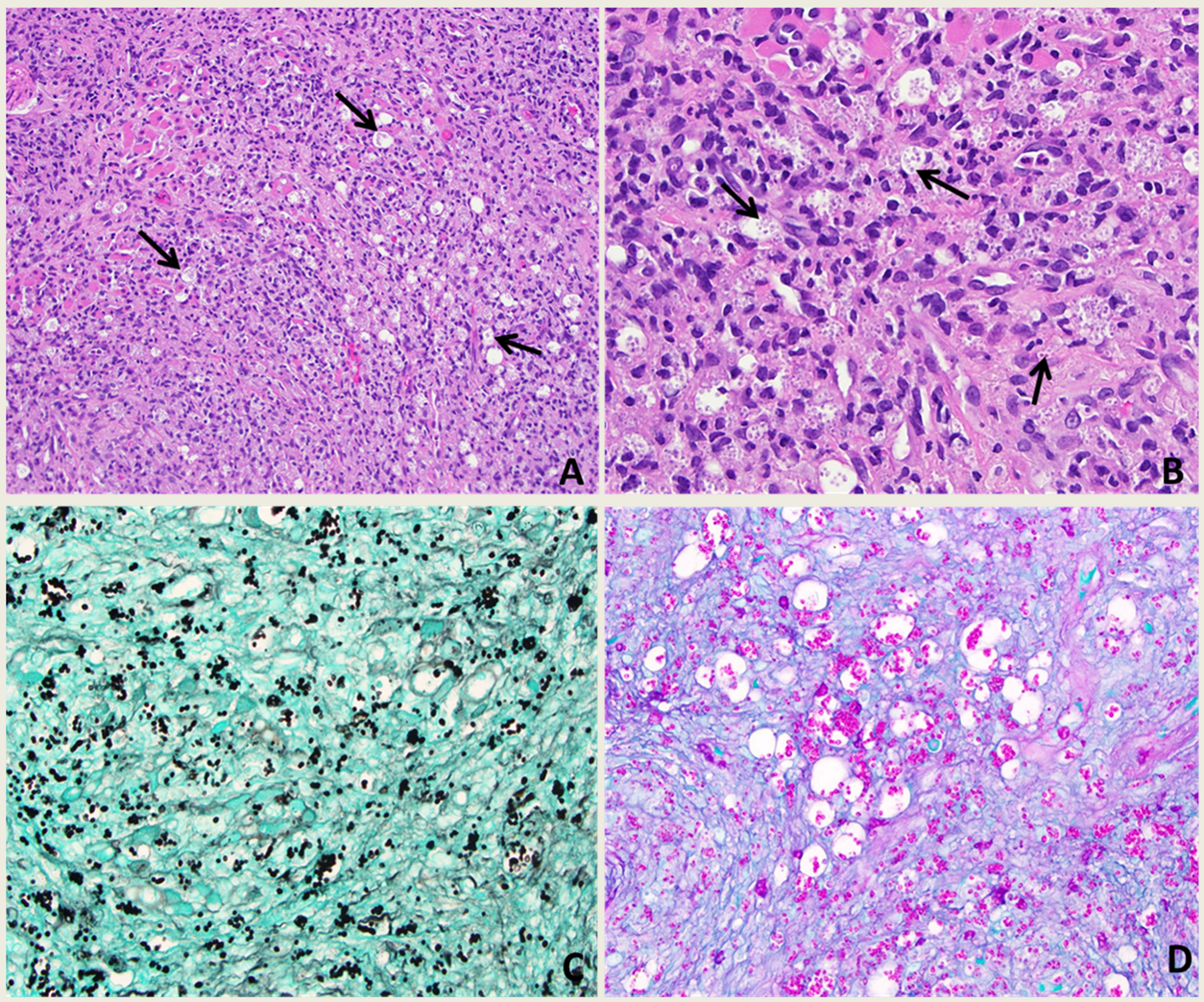

Figure 3
A. Hematoxylin and eosin (H&E) stained section demonstrating numerous vacuolated areas (arrows) surrounded by a diffuse, mixed inflammatory infiltrate (20X magnification).
B. Higher magnification (H&E, 40X magnification) revealing numerous epithelioid histiocytes with small, circular yeast cells (arrows), characteristic of Histoplasma capsulatum.
C. Special stain for Gomori methenamine silver, revealing numerous aggregates and randomly distributed organisms of H. capsulatum (40X magnification).
D. Periodic acid-Schiff stained section, highlighting small, circular yeasts of H. capsulatum (40X magnification).

DISCUSSION

Histoplasma capsulatum is a soil saprophyte endemic to the Ohio and Mississippi River valleys often found in chicken habitats or caves inhabited by bats and birds.[1] Infection by inhalation of aerosolized *H. capsulatum* microconidia primarily affects the lungs of immunosuppressed individuals, though a variety of clinical presentations exist.[2] Cutaneous histoplasmosis, a less common presentation, occurs secondarily from systemic infection with dissemination to the skin[3] or primarily after direct inoculation of *H. capsulatum* via injury to the skin.[4] Primary cutaneous histoplasmosis (PCH) is extremely rare with less than 20 cases reported in the literature.[4-18] We report the first report of PCH among the otolaryngologic literature, and the second case of PCH involving the Head and Neck.[13]

Patients with PCH present with non-specific skin lesions including papules, plaques, ulcers, purpura, abscesses, impetigo, or dermatitis.[19,20] Diagnosis hinges on evidence of fungus in the wound and absence of systemic (disseminated) disease.[4] Tissue biopsy of the cutaneous lesion should be performed promptly. Microscopy may reveal fungal elements, but histopathology showing and distinctive 2-4 µm, oval, narrow-based budding yeasts secures the diagnosis (Figure 2). Granuloma formation may be present. PAS (Figure 3) and GMS stains (Figure 4) help identify *H capsulatum* in the macrophages and within the tissue. Cultures, the gold standard in diagnosis, will show small oval budding yeast. Antigen detection is a sensitive method for ruling out disseminated disease.[2] Therapeutic options for cutaneous histoplasmosis depend on the extent and severity of the disease. Topical amphotericin B and nystatin can be used for limited disease,[4,5] though spontaneous remission has been reported.[13] Systemic amphotericin B[3,15,21] and itraconazole[14] have been proven effective for more widespread lesions, with incision and drainage reserved for lesions refractory to medical therapy.[12]

CONCLUSIONS

PCH presents a diagnostic challenge due to its rarity and varied clinical presentation. We present a rare case of primary cutaneous of the head and neck and discuss its successful diagnosis and management.

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