SKIN

BRIEF ARTICLE

Erythematous Plaque on Right Ear with Extension to Face and Neck in an Older Adult Male

Mara Trifoi[1], Norhan Shamloul[2], Bryan Anderson[2], Ryan Svoboda[3]

[1] Penn State College of Medicine, Hershey, PA, 17033
[2] Penn State University Department of Dermatology, Hershey, PA, 17033
[3] UMass Chan School of Medicine Department of Dermatology, Worcester, MA, 01605

ABSTRACT

Leishmaniasis poses a diagnostic challenge due to its resemblance to many dermatologic conditions. As the incidence of Leishmaniasis has increased in the United States due to a rise in world travel it is imperative for healthcare providers, especially dermatologists, to recognize and properly diagnose Leishmaniasis. In this case report, we present a unique case of New World Leishmaniasis, highlighting key histopathological findings important in diagnosis.

INTRODUCTION

Leishmaniasis presents with clinical manifestations that resemble a wide variety of conditions, thus posing a diagnostic challenge. New world leishmaniasis is found primarily in Latin America. The infection is rarely encountered in the United States with the exception of Southwest Texas and some parts of Florida; therefore, it is often less familiar to North American healthcare providers.[1] In almost half of the documented cases in North America, the family of the patient or the patient first suggested the diagnosis of leishmaniasis.[2] An increase in world travel has been associated with a higher incidence of cases reported, highlighting the necessity for healthcare providers, especially dermatologists, to recognize and properly diagnose leishmaniasis.[3] The varied and often-non-specific clinical presentation of Leishmaniasis creates a diagnostic challenge.

CASE REPORT

A 75-year-old male with a history of latent tuberculosis and diabetes presented with an erythematous plaque on the right earlobe. He reported a history of a bee sting on the helix while in Ecuador, with subsequent development of “cellulitis”. He was treated with doxycycline and amoxicillin-clavulanate which provided relief, but he subsequently developed a progressive indurated plaque with extension from the helix to the cheek, forehead, and post-auricular neck. He sought care in the United States and was treated with multiple rounds of oral antibiotics including topical mupirocin and oral ciprofloxacin, and cephalexin which led to improvement in the degree of erythema and edema but resulted in consistent recurrence after completion of each course. Punch biopsy was performed and revealed a focally ulcerated epidermis and slight epithelial atypia with the dermis presenting with mixed inflammatory infiltrate composed of

neutrophils, lymphocytes, histiocytes and plasma cells. Additionally, an incisional biopsy of the helix performed by Otolaryngology demonstrated mixed inflammatory infiltrate and necrosis of the cartilage which was felt to be suspicious for relapsing polychondritis. The patient was then treated with systemic corticosteroids with notable enlargement and increasing induration of the plaque. He was then referred to our dermatology clinic, and was noted to have a large, red, indurated plaque of the right cheek extending to the frontal scalp, right ear, and post-auricular area, with a rim of normal skin immediately around the posterior auricle (**Figure 1**).

Subsequently, a punch biopsy of the leading edge of the plaque was performed (**Figure 2**). Histopathologic examination revealed a mixed inflammatory infiltrate composed of neutrophils, lymphocytes, histiocytes, and plasma cells. Rare possible intercellular organisms were seen within histocytes and were highlighted by Giemsa, GMS, and CD1A stains. Due to microscopic findings concerning for leishmaniasis, PCR testing was performed which detected Leishmania DNA, Viannia subspecies, confirming a diagnosis of New World leishmaniasis. Otolaryngologic assessment did not reveal any mucosal involvement. Due to the patient's impaired renal function and recent dialysis requirements, the patient was started on fluconazole initially. Red light photodynamic therapy was considered as an additional temporizing measure, but his skin lesion progressed significantly, and he experienced polymicrobial superinfection requiring in-office debridement of his helix, so he was switched to amphotericin B, which resulted in complete clearance of the lesions.

DISCUSSION

The clinical presentation of Leishmania is variable but often features a nodule at the location of the sandfly bite, which subsequently enlarges and becomes ulcerated over the span of a few weeks. Lesions can appear after several months to years due to the long incubation period of the *Leishmania* parasite.[4] The ulcer is well demarcated and often filled with purulence or blood and is most frequently found on the face, hands or other exposed parts.[4,5] The vector species can affect the clinical presentation. Cutaneous leishmaniasis (CL), diffuse CL (DCL), mucocutaneous leishmaniasis (ML) and visceral leishmaniasis (VL) are all possible presentations based on various vector species.[5]

An important distinction in Leishmaniasis diagnosis is between New World and Old World leishmaniasis. New World leishmaniasis typically is the result of *Lutzomyia* sandfly transmission and presents with mucosal involvement.[6] While Old World leishmaniasis is transmitted through the *Phlebotomus* sandfly and is more commonly associated with visceral involvement.[6-7] New World leishmaniasis is endemic to Texas and South America and Old World leishmaniasis is commonly found in the East and Africa.[6,7]

Histologic examination is critical in the diagnosis of Leishmania. There are four key histologic patterns observed differing in terms of type of infiltrate, presence of amastigotes, and stage of granuloma formation. Amastigotes, represented by Leishman-Donovan bodies, are key diagnostic features often presenting within macrophages as round basophilic circles 2-4 um in diameter which usually are only visible under high power.[9] Type 1 –which is observed in 45% of

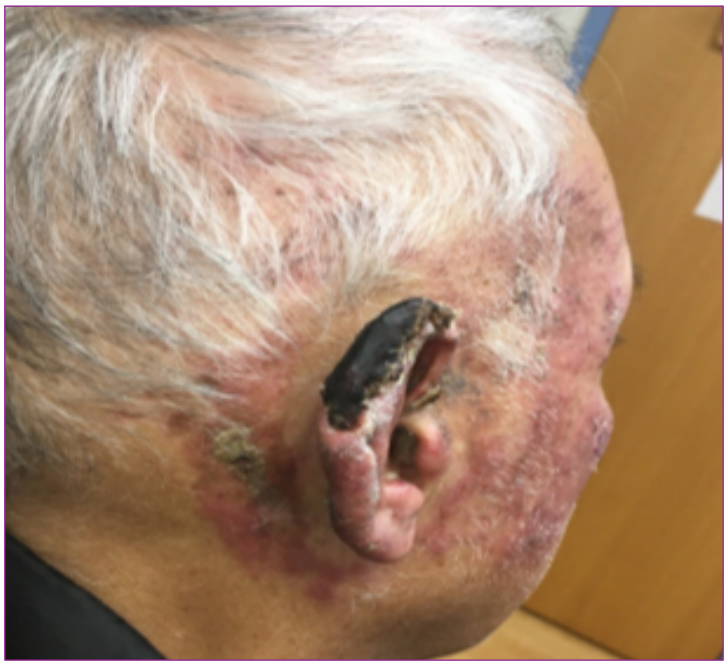

Figure 1. Clinical image showing an erythematous plaque involving right ear helix, lobule and tragus with extension to post-auricular neck, temple, forehead, and cheek. Necrotic eschar on superior helix represents prior failed full-thickness skin graft from incisional biopsy.

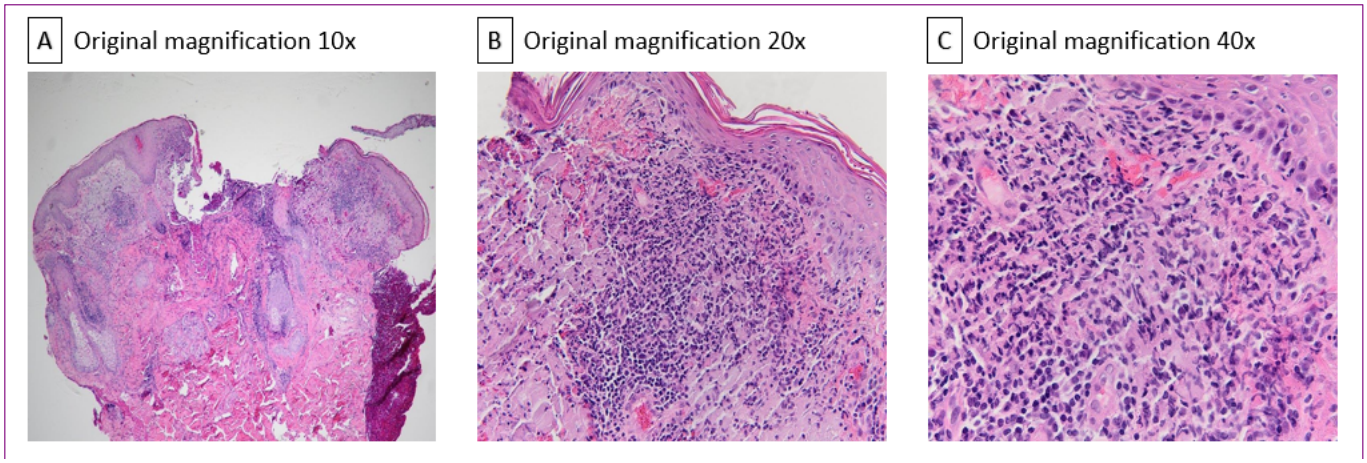

Figure 2. A, Hair follicle showing pittosporum organisms. B, focally ulcerated epidermis with slight epithelial atypia. C, Dermis with mixed inflammatory infiltrate composed of neutrophils, lymphocytes, histocytes, and plasma cells.

all cases—shows abundant amastigotes, while Type 2 –which is only seen in 27.5% of cases—shows a mixture of macrophages, polymorphonuclear neutrophils, and plasma cells with necrosis.[9] Type 3 is seen in 15% of cases and consists of granulomas with epithelioid cells, lymphocytes and plasma cells and Type 4 is seen in 5% of cases and presents with epithelioid granulomas with giant cells.[9,10]

Culture and polymerase chain reaction (PCR) testing are also utilized as essential diagnostic tools. In the United States, the Centers for Disease Control and Prevention provides Novy-MacNeal-Nicolle medium for inoculation and aids in performing analysis.[11] Culture results may be available in 3-8 weeks, but it can take up to 2 months to receive species information.[11] PCR studies allow for species identification within as little as 24 hours, which although it does not allow for sensitivity determination allows for quicker treatment initiation.[12]

Disease involvement and sensitivity to treatment are crucial treatment considerations. Systemic treatment is used in the setting of widespread lesions, mucosal involvement, and large progressive lesions. It is critical for providers to ensure sensitivity of drugs as resistance has become a problem especially in endemic countries.[13] Oral systemic treatment options for leishmaniasis include triazoles such as fluconazole, ketoconazole, or itraconazole, miltefosine, and newer modalities include immunotherapy and chemotherapy.[14] In North America, parenteral options for cutaneous therapy include amphotericin B deoxycholate, pentamidine, and pentavalent antimonial.[14]

This case report presents a challenging diagnosis of New World Leishmaniasis and highlights the need for North American healthcare providers to become more familiar with this infectious disease.

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Corresponding Author:
Mara Trifoi
Penn State College of Medicine, 162A University Manor East, Hershey, PA, 17033
610-607-9642
Email: mtrifoi@pennstatehealth.psu.edu

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