CASE REPORT

Disseminated cutaneous Mycobacterium chelonae infection as a presenting sign of ectopic adrenocorticotropic hormone syndrome

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INTRODUCTION

Mycobacterium chelonae is a rapidly growing nontuberculosis mycobacterium (NTM). Infection with M chelonae typically results from cutaneous injury, injections, or other medical procedures.1,2 Skin infections by the M chelonae complex are heterogeneous and can present as papules, pustules, ulcerating nodules, and deep lesions, such as furuncles and nodules with sinus tracts.3 Disseminated infections have been described in immunocompromised patients, including those receiving systemic steroids and immunosuppressants such as methotrexate and azathioprine.4-6 To our knowledge, a single case report7 previously described disseminated M chelonae in a patient with endogenous hypercortisolemia (Cushing syndrome).2 Here, we describe a case, in which endogenous hypercortisolemia secondary to ectopic adrenocorticotropic hormone (ACTH) syndrome resulted in disseminated M chelonae infection.

CASE REPORT

A 43-year-old Caucasian man presented to our clinic with a 3-week history of erythematous nodules on his left upper arm and right leg and bilateral pitting edema (Figs 1 and 2). The nodules were growing and spreading in a sporotrichoid pattern. No improvement was observed with a 1-week trial of doxycycline. He denied fever, chills, nausea, vomiting, and diarrhea.

Medical history was notable for new-onset hypertension as well as recent herpes zoster and Serratia marcescens skin infection. The patient endorsed a 50-pound weight gain in the preceding 7 months, chronic lower extremity edema, decreased libido, and depression. Physical examination revealed central obesity, dorsocervical fat accumulation, rounded face, violaceous striae on the abdomen and axillae, and muscle weakness.

Evaluation of additional outpatient data, including echocardiogram, complete metabolic panel, urinalysis, thyroid-stimulating hormone, hemoglobin A1c, HIV antibody, and antinuclear antibody, revealed that all of these were unremarkable/within normal limits. A punch biopsy of his right thigh showed granulomatous and suppurative panniculitis (Fig 3, A) with numerous acid-fast bacilli (Fig 3, B). Tissue cultures from his left arm were positive for M chelonae. He was referred to infectious disease for appropriate antibiotic management.

Laboratory evaluation revealed an elevated random cortisol of 46.5 μg/dL (reference range: 4.5-22.7 μg/dL). Cortisol was not suppressed with either low- (1 mg) or high-dose (8 mg) dexamethasone suppression test. ACTH was elevated. The

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24-hour urine free cortisol was 660 μg/24 hours (reference range: 3.5-45 μg/24 hour). No pituitary abnormality was observed on brain magnetic resonance imaging. Inferior petrosal sinus sampling was negative. He underwent a DOTA-TATE positron emission tomography/computed tomography, which revealed a nodule measuring 1.2 cm with mild-to-moderate radiotracer uptake in the lingula, consistent with the source of ectopic ACTH. Given the severity of hypercortisolemia, the patient was admitted to the hospital, and ketoconazole was initiated to inhibit adrenal steroid synthesis while awaiting wedge resection of the lingula. Pathology identified a pulmonary, ACTH-positive, typical carcinoid tumor. Cortisol on postoperative day 1 was 1.4 μg/dL (normal range: 4.5-22.7 μg/dL), suggestive of surgical cure.

Supraphysiologic hydrocortisone replacement was started postoperatively, with plans for a slow taper to physiologic levels. Azithromycin and linezolid were continued for treatment of the M. chelonae infection. One month following surgery, signs and symptoms of hypercortisolemia had dramatically improved, and cutaneous nodules had nearly completely resolved.

**DISCUSSION**

Diagnosis of disseminated NTM requires careful consideration given its heterogeneous clinical presentation. Previous reports of patients with disseminated NTM were initially treated with corticosteroids due to the ability of NTM to present similarly to vasculitis or other autoimmune conditions. Given the lack of unifying features of *M. chelonae*, biopsy and tissue cultures are necessary for definitive diagnosis. In this case, biopsy and identification of NTM prompted concern for an immunocompromised host and led to the diagnosis of hypercortisolemia secondary to ectopic ACTH. Given the severity of hypercortisolemia, the patient was at risk of systemic opportunistic infections. Identification and treatment of hypercortisolemia was potentially life-saving.

Few disseminated bacterial infections secondary to hypercortisolemia are reported in the literature, likely due to the exceptionally high endogenous cortisol levels required for sufficient immunosuppression. Although there is no consensus for what level of hypercortisolemia results in immunosuppression, glucocorticoid excess suppresses both cellular and humoral immunity, increasing the risk of opportunistic infections. Risk is magnified by skin atrophy and fragility. Data suggest that higher cortisol levels increase the risk of infection. Given the degree of immunosuppression associated with disseminated NTM infection in patients with Cushing syndrome and risk of other life-threatening opportunistic infections, patients with disseminated NTM, without a known cause for immunosuppression, should have an expedited work-up for potential underlying etiologies to prevent further complications.

**Conflicts of interest**

None disclosed.
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Fig 3. A, Tiny cavities in the subcutaneous fat were surrounded by neutrophils and macrophages. B, Numerous, beaded acid-fast bacilli were present in the subcutaneous inflammation. Cultures identified Mycobacterium chelonae. (A, Hematoxylin-eosin stain and B, acid-fast stain; original magnifications: A, ×20; B, ×100.)