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Atypical Pyoderma Gangrenosum

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Abstract

Pyoderma gangrenosum is an uncommon neutrophilic skin disorder that varies in presentation. The presence of ulceration is common and diagnosis is often based on exclusion of other entities. A wide variety of treatment options exist; however, cure is often still difficult. The presence of underlying systemic disease must be considered. Herein, we report an atypical case of pyoderma gangrenosum and review current therapeutic modalities.

Keywords: Pyoderma gangrenosum; Atypical presentation; Dermatology

Introduction

Pyoderma gangrenosum is a neutrophilic skin disease that is often characterized with the presence of ulceration. While it commonly is associated with underlying systemic disease, it is most often a diagnosis of exclusion [1, 2]. Cutaneous biopsy may be performed; however, histologic examination is also somewhat nonspecific and clinical diagnosis is typically sufficient. A myriad of topical and systemic treatment modalities exist.

Case Presentation

An 83-year-old Caucasian woman presented with a large multi-focal erythematous plaque with numerous ulcerations on the central and right lower abdomen (Figure 1).

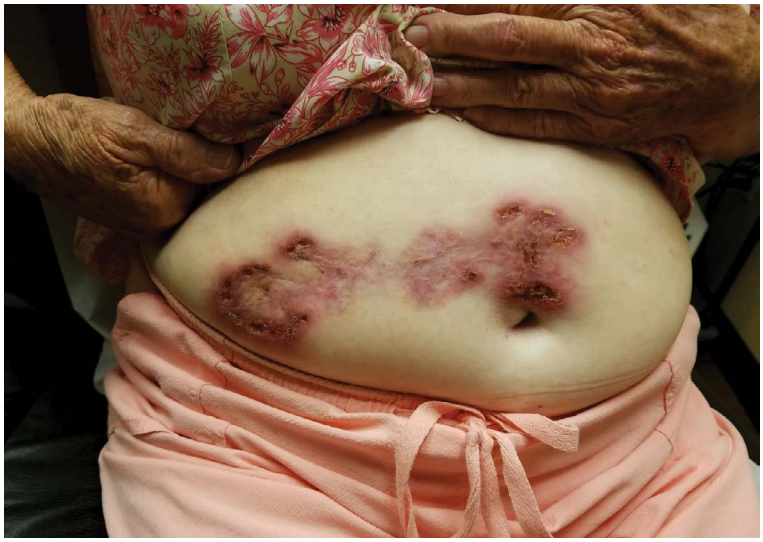

Figure 1: Multi-focal erythematous plaque with numerous ulcerations and scarring on the central and right lower abdomen.

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Past medical and surgical history were pertinent only for hypertension, and specifically negative for ulcerative colitis and Crohn's disease. Clinical suspicion for possible metastatic disease, basal cell carcinoma, calciphylaxis, lupus erythematosus, necrotizing fasciitis, factitial disease, and pyoderma gangrenosum were all considered in the initial differential diagnosis. Multiple cutaneous biopsy sites (including an incisional biopsy) revealed irregular epidermal hyperplasia and a dense inflammatory infiltrate within a fibrotic dermis, composed of a mixed cell type (neutrophils, eosinophils, lymphocytes, and plasma cells). While the histologic findings were suspicious for an infectious etiology, this was ruled out with an absence of organisms on histologic exam (including bacterial, fungal, and mycobacterium cultures and/or stains). This included subsequent infectious histologic evaluation by the Centers for Disease Control and Prevention, which also demonstrated no immunohistochemical or molecular evidence of mycobacteria or fungi. Continued and repeat histologic examination was consistent with a diagnosis of atypical pyoderma gangrenosum. Laboratory examination, to include complete blood count, comprehensive metabolic panel, were within normal limits.

Initial treatment was based on wound care modalities, to include oral doxycycline and topical silver nitrate with moderate benefit. Subsequent therapeutic treatments included topical and systemic corticosteroids. She was later treated with systemic dapsone which provided ultimate resolution.

Discussion

Pyoderma gangrenosum is a rare neutrophilic skin disease characterized by cutaneous ulceration. The disease often presents itself as a papule or nodule that degrades to leave an ulceration. It often mimics an infectious process and is commonly a diagnosis of exclusion as no single diagnostic test is confirmatory. Approximately half of individuals diagnosed with pyoderma gangrenosum have underlying systemic disease such as inflammatory bowel disease, arthritis, autoimmune disease, and/or hematologic disease [1, 2].

Pyoderma gangrenosum affects about 3 to 10 people per million individuals annually worldwide, with a U.S. prevalence of 5.8 per 100,000.[1] Diagnosis is primarily clinical and supported by the Delphi consensus criteria [1]. This criteria calls for 1 major criterion of a biopsy (ulcer edge) that demonstrates a neutrophilic infiltrate, in addition to at least 4 (out of 8) minor criteria that include the following: exclusion of infection, pathergy, history of inflammatory bowel disease or arthritis, a papule (or pustule, vesicle) that ulcerates within 4 days, peripheral erythema with an undermined border at the site of ulceration, multiple ulcers, cribiform scarring, and a decrease in ulcer size within 1 month of immunosuppressive therapy [1]. Utilization of this criteria yields a sensitive of 86% and specificity of 90% [1]. Pyoderma gangrenosum is often associated with conditions such as inflammatory bowel disease, arthritis, or hematologic disorders [2]. Due to trauma or debridement having the possibility of worsening the condition of PG through pathergy, early recognition is critical to avoid unnecessary procedures. Treatment is often challenging, with topical (and systemic) corticosteroids as common initial treatment [3-5]. A multitude of other systemic modalities may be implemented to include dapsone, cyclosporine, azathioprine, mycophenolate mofetil, intravenous immunoglobulin, and targeted biologic medications such as tumor necrosis factor alpha inhibitors (example: adalimumab) [2,5].

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