Pyoderma Gangrenosum and Extensive Aseptic Chest Wall Abscess in a Patient with Inflammatory Bowel Disease

Igor Snast MD, Iris Ostfeld MD, Lev Pavlovsky MD PhD, Emmilia Hodak MD and Anat Gafter-Gvili MD

Departments of Dermatology and Medicine A, Rabin Medical Center (Beilinson Campus), Petah Tikva, Israel
Institute of Hematology, Rabin Medical Center (Beilinson Campus), Petah Tikva, Israel
Sackler Faculty of Medicine, Tel Aviv University, Tel Aviv, Israel

KEY WORDS: aseptic abscess, inflammatory bowel disease (IBD), neutrophilic dermatosis, neutrophils, pyoderma gangrenosum (PG)

Pyoderma gangrenosum (PG) is a rare neutrophilic dermatosis, which presents as a rapidly expanding ulcer with a necrotic undermined border. More than 50% of patients with PG have an antecedent, coincident, or subsequent associated systemic disease, most commonly inflammatory bowel disease (IBD), arthritis, and hematologic disease (i.e., acute and chronic myelogenous leukemia) [1]. Similarly, aseptic abscess (AA) is an inflammatory condition characterized by deep sterile collections of neutrophils, clinically mimicking bacterial abscess [2]. We describe a patient with IBD who presented with a unique combination of an extensive chest wall AA concomitantly with necrotic ulcers characteristic of PG.

PATIENT DESCRIPTION

A 24 year old woman with newly diagnosed IBD, presented with a firm, tender, erythematous nodule located on the proximal clavicle, with sternoclavicular joint pain. At that time, she was being treated with 0.5 mg/kg prednisone and vedolizumab after failing to improve with infliximab, mesalazine, and low-dose prednisone. A chest computed tomography (CT) scan depicted a large subcutaneous chest wall abscess extending within the pectoralis muscle with sternal destruction. An incision was performed with drainage of purulent fluid. Broad-spectrum antibiotics were initiated. Near the chest wound, two satellite rapidly enlarging ulcers with undermined violaceous borders appeared.

Due to progressive chest wall necrolysis on a consecutive CT, radical debridement with excision of pectoralis major muscle was performed, resulting in rib exposure [Figure 1A]. On the thigh, an erythematous nodule appeared, which rapidly underwent necrotic changes, leading to a painful ulcer with violaceous-rolled border [Figure 1B]. Skin biopsies taken from the ulcer base on the thigh and chest wound revealed a diffuse dermal infiltrate of neutrophils extending into chest wall striated muscles, with focal fibrinoid necrosis of blood vessel walls.

Laboratory evaluation demonstrated elevated inflammatory markers; negative serologies for hepatitis B/C, human immunodeficiency virus, cytomegalovirus, and atypical bacteria; negative Mantoux and QuantiFERON tests; negative blood, skin, and purulent fluid cultures including on enrichment media; negative pan-bacterial; and pan-fungal blood polymerase chain reaction.

A diagnosis of chest wall AA with characteristic thigh and chest PG lesions was made based on the clinical presentation, histologic findings, and absence of

Figure 1. [A] Large surgical wound after subcutaneous abscess incision and drainage extensive enough to expose periosteum and muscle, with two inferior satellite ulcers [B] Close up view of pyoderma gangrenosum of left thigh
infectious pathogens [1]. Intravenous pulse methylprednisolone was initiated on 4 consecutive days, followed by oral 1.3 mg/kg prednisone with a concomitant cyclosporine 5 mg/kg. Given the lack of an adequate clinical response and elevation of inflammatory markers when steroid tapering was attempted, intravenous immunoglobulins 3 gr/kg were added. Over 3 weeks, the thigh ulcer healed completely and the chest necrolysis ceased with granulation tissue filling wound-bed. Two weeks later, the patient underwent reconstructive surgery. Exposed ribs were covered with a muscular flap of the remaining pectoralis major so that the entire wound was covered with a split-thickness skin. Prednisone and cyclosporine were slowly tapered. Subsequently, adalimumab was initiated due to IBD exacerbation without skin lesion recurrence.

**COMMENT**

AA represents a new clinical entity characterized by deep neutrophilic abscesses, most commonly of intra-abdominal origin, negative infectious workup, lack of a clinical response to antibiotics, and a dramatic response to steroids [2,3]. Excluding the latter, which could not be assessed as AA was surgically treated, our patient met all of the criteria. Similar to our patient, in the largest cohort to date of 30 patients with AAs, most patients (n=21) had IBD followed by neutrophilic dermatosis (n=6), and relapsing polychondritis (n=3) [2]. Although most cases of AA were shown to involve the spleen. Our case demonstrates that AAs can potentially appear anywhere in the body. Our patient presented with the typical appearance of PG, another rare dermatosis mediated by neutrophils, concurrently with AA. The overlap between the two conditions was also evident in a recent literature review of IBD-associated AA, in which 2 of 37 patients (5.5%) presented with both disorders [3]. This finding suggests that common immunologic pathways may mediate both disorders [4].

**References**