Pyoderma Gangrenosum Affecting the Dorsal Hand: A Case Report

Lucas R. Haase, MD*†
Grant B. Nelson, MD*†
Walter B. Klyce, MD*†
Christi M. Cavaliere, MD, MS‡
Blaine T. Bafus, MD, FAAOS*§

Summary: Pyoderma gangrenosum is an uncommon neutrophilic dermatosis characterized by an ulcerative lesion with a violaceous border. Most frequently, these lesions present in the lower extremity and are associated with underlying immune-mediated comorbidities. Infrequently, these lesions may present in the upper extremity, which presents difficult challenges for upper extremity surgeons as the lesions are frequently misdiagnosed as an infectious process. This often leads to inappropriate surgical debridement and antibiotic administration. Local trauma to the lesion can lead to a process of pathergy and worsening of the lesion. Here, we report on a case of cutaneous pyoderma gangrenosum affecting the dorsal hand, originally misidentified as an atypical infection with subsequent unsuccessful surgical debridement. After involvement of a multidisciplinary team, appropriate diagnosis was made, and treatment with local immunosuppressive agents achieved resolution of the lesion. (Plast Reconstr Surg Glob Open 2022;10:e4592; doi: 10.1097/GOX.0000000000004592; Published online 1 November 2022.)

Pyoderma gangrenosum (PG) is an uncommon neutrophilic dermatosis that is associated with underlying immune-mediated comorbidities, most commonly inflammatory bowel disease.1,2 The classic lesion in PG begins as a tender nodule that progresses to an ulcer with violaceous borders and surrounding erythema, the hallmark characteristic of which is pain.3 Despite a typical pattern, no definitive test exists for diagnosis, and frequently, PG is a diagnosis of exclusion.4 Due to the underlying inflammatory nature of the lesion, PG is treated with either local or systemic immunosuppressive medications. Surgical debridement is contraindicated and tends to induce pathergy, leading to worsening of the lesion.5

PG lesions affecting the hand are frequently misdiagnosed as infections. Hand lesions can be particularly challenging as many can mimic infections ranging from gout to inflammatory lesions.5 In addition, the superficial nature of tendons, joints, and neurovascular structures within the hand places them at high risk during progression of the disease process. There is limited literature describing PG affecting the hand. Case reports place an emphasis on early recognition and frequently describe pathergy after surgical debridement. Here, we present a case of PG of the hand to reillustrate the importance of early recognition.

CASE PRESENTATION

A 65-year-old right-hand dominant man with a history of diabetes but no history of autoimmune diseases, including inflammatory bowel disease (IBD), rheumatoid arthritis, or other seronegative arthropathies presented to the emergency department with a 3-week history of a painful dorsal left-hand lesion. The patient previously failed two outpatient courses of oral antibiotics without cultures. On examination, the patient had a 1 × 1 cm lesion on the dorsum of his left hand overlying the third metacarpophalangeal joint with surrounding erythema and an ulcerative center. The patient was admitted to the hospital, started on vancomycin and piperacillin/tazobactam with improvement the following morning, and discharged on oral trimethoprim/sulfamethoxazole.

The patient returned to the emergency department 1 week later with worsening pain despite antibiotic compliance. Examination was significant for spread of erythema to the proximal phalanx of the long finger, and the

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development of raised borders about the ulcerative center (Fig. 1). There was concern for atypical infection versus malignant process, and thus, the patient was brought to the operating room where an excisional biopsy and debridement were performed (Fig. 2). The patient was started on daily dressing changes with wet to dry dressings. Broad spectrum antibiotics were again administered, and infectious disease was consulted. Final cultures returned without growth. The patient was discharged on a 5-day course of amoxicillin/clavulanic acid.

The patient returned 2 weeks later with persistent pain and worsening erythema. The lesion now had a violaceous border with vesicular appearance and erythema spreading into his ring finger. The patient was again admitted and returned to the operative suite for debridement of the new ulcerative lesion. A plastic surgeon was consulted for postoperative wound management, and it was decided to allow the wound to heal by secondary intention. The patient was referred to dermatology for concern of inflammatory lesion.

At dermatology follow-up, a punch biopsy at the periphery of the lesion was taken. In addition, intraleisional Kenalog (triamcinolone) was administered without systemic therapy. The results of this second biopsy demonstrated dermal lymphocytic and neutrophilic infiltrates (Fig. 3).

The patient returned to clinic 6 weeks later with incomplete resolution of the lesion and his pain, and thus, he was started on daily topical tacrolimus 0.03%. Five months after his initial presentation, he had complete wound healing and significant improvement in pain (Fig. 4). He maintained full flexion/extension of all digits without the need for hand therapy.

DISCUSSION

PG is a rare, ulcerative neutrophilic dermatosis commonly associated with an underlying autoimmune process. There is no definitive test to confirm a diagnosis of PG, and it remains a diagnosis of exclusion. As a result,
PG is commonly misdiagnosed. Treatment guidelines rely on case reports and small retrospective studies, which indicate either systemic or local immunosuppressive therapy, with emphasis placed on prompt diagnosis. However, treatment with immunosuppressive therapy can be a difficult decision, as these patients often present with clinical concerns for infection.

The case presented above serves to illustrate several important characteristics of PG. The painful ulcer with violaceous boarders and surrounding erythema is classic for PG. In this case, delay in diagnosis led to unnecessary treatment with both antibiotics and surgical debridement. This demonstrates the pathergy response that is common for this condition, leading to worsening of the original lesion and spread to additional local sites. Early involvement of a multidisciplinary team, including plastic surgery, dermatology, infectious disease, and rheumatology, can facilitate appropriate diagnosis and treatment. Finally, the area of biopsy is of critical importance when diagnosing PG. For definitive diagnosis, the biopsy needs to include the active border of the lesion. It is possible the first biopsy in this case missed the diagnosis due to area of sample. This again emphasizes the importance of a multidisciplinary team to include a dermatopathologist.

Resolution of symptoms does not always occur, particularly with PG of the hand. In the largest case series on PG affecting the hand, Huish et al demonstrated a 100% rate of misdiagnosis, with frequent complications. This series studied seven patients with a total of 13 PG lesions. On average, each patient underwent 2.2 unnecessary procedures without clinical improvement. These surgeries included four amputations. In each case, involvement of a multidisciplinary team aided in eventual diagnosis. Case reports by Mihailidis et al and Schotanus et al chronicle similar cases of PG of the hand misdiagnosed as infection leading to surgical debridement without clinical improvement.

CONCLUSIONS

PG of the hand remains a rare condition that is both difficult to diagnose and infrequently encountered by hand surgeons. Providers must maintain an index of suspicion, particularly for patients with history of underlying immune processes, negative cultures, failure of antibiotic therapy, or a pathergic response to debridement. Prompt recognition and involvement of a multidisciplinary team allow for appropriate treatment and avoidance of unnecessary procedures.

Lucas R. Haase, MD
Louis Stokes Cleveland VA Medical Department of Orthopedic Surgery
11100 Euclid Ave
Cleveland, OH 44106
E-mail: lucas.haase@uhhospitals.org

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