Suppression of pathergy in pyoderma gangrenosum with infliximab allowing for successful tendon debridement

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Key words: debridement; pyoderma gangrenosum; tendon.

INTRODUCTION
Pyoderma gangrenosum (PG) is an inflammatory condition that causes neutrophilic infiltration of the skin and characteristic skin ulceration. More than half of patients develop PG in association with an underlying systemic disease. Its pathophysiology is complicated and not fully understood. PG can affect any part of the body, but it commonly affects the legs. The diagnosis of PG can be challenging and is often one of exclusion. PG exhibits pathergy, which is the development or worsening of a lesion at the site of trauma, and the presence of pathergy can help support the diagnosis of PG. PG can closely resemble other entities included in the differential diagnosis, such as serious skin infections, which can be found with postoperative wounds or cases of necrotizing fasciitis. Because these conditions are often managed with surgical debridement, establishing the correct diagnosis is critical. Avoidance of surgical debridement of PG is typically recommended to limit pathergy. We present a case in which worsening of a patient’s PG following surgical debridement led to the exposure of her tibialis anterior tendon, which became desiccated and superficially necrotic with subsequent functional impairment.

CASE REPORT
A 32-year-old woman was evaluated in the outpatient dermatology clinic as a second opinion for an acutely worsening ulcer over her anterior shin. The lesion was originally diagnosed as necrobiosis lipoidica and was surgically debrided. The patient had a history of rheumatoid arthritis, episcleritis, hepatosplenomegaly, and insulin-dependent diabetes. She was a smoker for 18 years. A punch biopsy showed underlying fibrin at the edge of the ulcer with proliferation of blood vessels and a few thrombi, as well as some necrosis at the base of the ulcer. Grocott methenamine silver, periodic acid-Schiff, and acid-fast bacillus stains were all negative for organisms. These findings demonstrated consistency with a healing PG ulcer.

Treatment was undertaken with cyclosporine 200 mg twice daily, clobetasol 0.05% topical ointment, and a prednisone taper with minimal improvement. After 3 weeks, she was transitioned from cyclosporine to mycophenolic acid 1500 mg twice daily and doxycycline 100 mg twice daily. The site was injected with 2.5 mL of 10-mg/mL triamcinolone.

However, at 3 weeks follow-up, she had a 10 cm × 10 cm ulcer with a violaceous undermined border and an exposed tendon at the base of the wound. Pathergic expansion of the ulcer was noted at the site of punch biopsy on the ulcer’s margin (Fig 1, A). She was admitted for concern of a secondary infection and evaluation for the presence of underlying osteomyelitis. She was treated with glucocorticoids and antibiotics intravenously. A deep vein thrombosis study, blood cultures, tissue culture (including acid-fast bacillus and fungal culture), and brucellosis testing were all negative.

Abbreviation used:
PG: pyoderma gangrenosum
Following discharge, a xeroform gauze dressing was placed on the ulcer, and she was transitioned to treatment with minocycline 100 mg orally twice daily; infliximab 5 mg/kg infused every 8 weeks was scheduled.

At follow-up, exposure of the tibialis anterior tendon persisted, and the patient noted the tendon was turning black and painful, and she complained of difficulty with ipsilateral ankle dorsiflexion (Fig 1, B). Consultation with plastic surgery noted that the tendon had desiccated due to lack of blood supply and debridement was recommended. Given the patient’s history of pathergy, concern was raised that debridement might precipitate ulcer expansion. However, due to worsening functional impairment, the patient elected for tendon debridement while on treatment with minocycline and prednisone. The dead tissue was sharply debrided over an area of 2 cm × 3 cm of the tendon only. Healthy, bleeding muscle was visualized through the tendon. Infliximab infusions began immediately following debridement.

Five days after the debridement, healthy-looking granulation tissue had developed over the tendon (Fig 1, C). The patient was followed by a dermatologist for over a year, and near complete resolution of the ulcer was observed with fully restored dorsiflexion and strength on infliximab and minocycline (Fig 1, D).

**DISCUSSION**

PG is a rare disorder with an estimated incidence of 3-10 cases/1 million people/year. Individuals of any age can be affected. PG most commonly develops in young and middle-aged adults, with an average age of onset of 40-60 years. Typically, there

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**Fig 1.** A, Pyoderma gangrenosum at admission with exposed tibialis anterior tendon. B, Edema and desiccation of the tibialis anterior tendon at follow-up. C, Granulation tissue development following debridement of the exposed tendon. D, Near resolution of the ulcer after 1 year of treatment with infliximab and minocycline.
is a rapid progression of a painful, necrolytic cutaneous ulcer with an irregular, violaceous, and undermined border.\(^7\)

Tendon exposure in a soft tissue wound is a challenging clinical scenario in the best circumstances. The added complexity of this case is the underlying cause of the ulcer and pathergy associated with trauma including surgical treatment. Viability of tendons is dependent upon blood vessels and diffusion of nutrients by synovial fluid.\(^8\) Prolonged exposure of the avascular tendon can result in desiccation. Desiccation can lead to infection, loss of tendon function, and possible rupture. Typically, debridement of the desiccated portion of the tendon is recommended, with the goal of the underlying muscle growing through the tendon and assisting with granulation tissue over the wound.\(^9\) Although pathergy following cutaneous debridement is well described, publications detailing surgical debridement of subcutaneous tissues in PG are few. PG often can expose structures deep to the dermis, and there are no standard treatment methods or recommendations for these situations.\(^2,4\) One reported case was of an untreated PG that involved exposure of the Achilles tendon and resulted in rupture.\(^2\) Another report described a multimodality approach to PG ulcers that included exposure of the extensor tendons on the dorsal hand and forearm and treatment with immunosuppressive drugs, surgical debridement, primary closure with a collagen-glycosaminoglycan biodegradable matrix, and covering with a mesh graft 14 days later. The treatment was complicated by necrosis and superinfection of the index finger, which was treated with systemic antibiotics and an unmeshed skin graft under vacuum bandage with a mesh wound contact layer.\(^10\)

In our case, the surgical debridement of the tendon did not cause pathergy or destruction of the tendon. Our patient was on an immunosuppressive regimen during and after the surgical treatment similar to the above reported case\(^10\) suggesting that immunosuppression might be essential not only for remission of the condition but prevention of pathergy caused by surgical trauma.

In most cases, debridement of ulcers suspicious for PG should be avoided to prevent pathergy; however, we were able to successfully debride an exposed, desiccated anterior tibialis tendon in a PG ulcer while the patient was on immunosuppressive treatment. Further understanding of PG is required to better predict tissue involvement and whether immunosuppression is sufficient to limit pathergy in surgical procedures.

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