

# Superficial Pyoderma Gangrenosum Treated With Infliximab: A Case Report

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*We report a case of superficial pyoderma gangrenosum (SPG) that was successfully treated with infliximab. A 22-year-old man presented with several polycyclic, purplish red plaques with some crusting and well-defined edges on the bilateral lower extremities. Histology showed superficial and deep mixed inflammatory cell infiltrate with prominent neutrophils and scarring noted as a result of multiple sinus tract formations that were characteristic of SPG. After unsuccessful results with typical treatments (ie, steroids, antibiotics, immunosuppressants), the patient was successfully treated with infliximab.*

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Superficial pyoderma gangrenosum (SPG) is a rare variant of pyoderma gangrenosum (PG)<sup>1,2</sup> that was first described by Wilson-Jones and Winkelmann.<sup>3</sup> Pyoderma gangrenosum can be classified into 4 categories: ulcerative, pustular, bullous, and vegetative. Each variant has its own distinct clinical and histologic findings.<sup>4,8</sup> The vegetative form, also known as SPG, is considered to be the most limited, superficial, and chronic variant; it also is the least aggressive form of PG with minimal to no systemic manifestations.<sup>5</sup> Superficial pyoderma gangrenosum can be differentiated from other types of PG by its clinical and histologic features as well as its prognosis.<sup>2,5,7</sup> We present a case of SPG that was resistant to numerous standard therapies but responded well to treatment with infliximab.

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## Case Report

A 22-year-old man presented with lesions on his left lower leg of 7 months' duration. He did not report any discomfort or pain associated with the lesions and had not come into contact with anything that would elicit a hypersensitivity reaction. His medical history was remarkable for focal segmental glomerulonephritis that was treated with prednisone and cyclosporine.

On clinical examination of the patient's legs, several polycyclic, purplish red plaques with some crusting and well-defined edges were noted (Figure 1A). The remainder of the skin examination was unremarkable. A biopsy from the left anterior leg showed superficial and deep mixed inflammatory cell infiltrate with prominent neutrophils, focal granuloma formation, dermal fibrosis extending into the fat, and scarring noted as a result of multiple sinus tract formations (Figures 2 and 3). These findings were consistent with SPG. Gram, Giemsa, periodic acid-Schiff, and formalin ammonium bromide stains for microorganisms were all negative. Routine blood work displayed no abnormalities.

As time progressed, the lesions continued to persist and spread further to the bilateral upper extremities and the right lower extremity. Oral prednisone was continued. Other treatments such as cyclosporine, dapsone, and doxycycline hyclate were added to wean the patient off of prednisone. These treatments yielded minimal results. The patient was treated with minocycline as well as topical and intralesional steroids, which also proved unsuccessful. Then the patient was started on etanercept subcutaneous injections (50 mg) twice weekly. This treatment proved to be more successful in clearing the lesions than the prior therapies; however, despite showing moderate improvement, the patient continued to have recurring episodic lesions. After realizing the efficacy of the etanercept injections, we placed the patient on infliximab with a dose of 5 mg/kg at 0, 2, and 6 weeks. Within 3 months the patient showed remarkable improvement with no active ulcerations (Figure 1B). Scars and some postinflammatory hyperpigmentation had healed, but

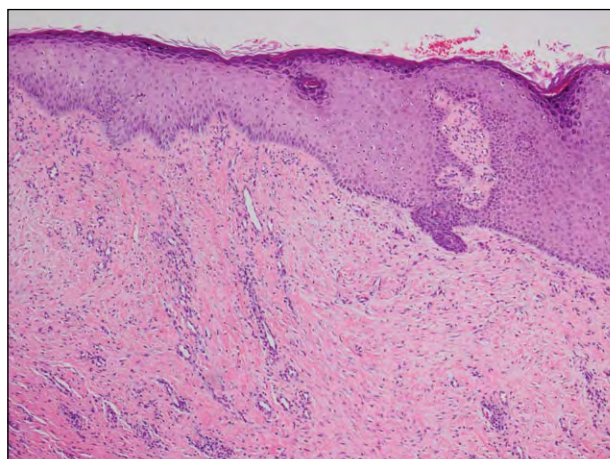


**Figure 1.** Clinical presentation of left thigh before (A) and 3 months after treatment with infliximab (B).

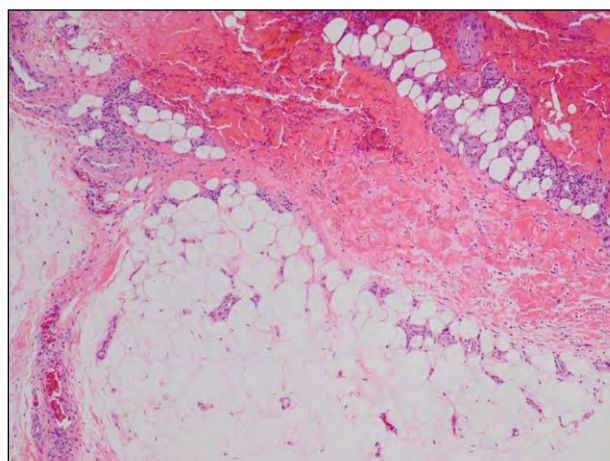
overall his condition had exponentially improved, both physically and emotionally, since beginning treatment. The patient continues to take infliximab today. The patient has been on a maintenance dose of infliximab of 5 mg/kg every 8 weeks. During this time he has not developed any new lesions and all that remains are scars from prior lesions.

### Comment

Superficial pyoderma gangrenosum clinically presents as clean, nontender, superficial ulcers with smooth edges that heal without scarring; there are no other systemic disease associations.<sup>2</sup> Lesions usually appear



**Figure 2.** Low-power view showing epidermal hyperplasia, dermal fibrosis, increased blood vessels, and chronic inflammation (H&E, original magnification  $\times 10$ ).



**Figure 3.** High-power view showing fibrosis and chronic inflammation extending into the fat (H&E, original magnification  $\times 40$ ).

on the trunk, but there have been reports of occurrences on the face, limbs, and scrotum.<sup>4,8</sup> Compared to ulcerative PG, SPG differs in that the base of the ulcers typically is nonpurulent and more superficial.<sup>5,8</sup> Ulcers from SPG also tend to enlarge slowly and commonly are painless.<sup>8</sup> The histology of SPG differs from other variants of PG with a prominence of histiocytes within the neutrophilic infiltrate,<sup>4</sup> a palisading granulomatous reaction,<sup>2</sup> the presence of giant cells<sup>4</sup> and sinus tract formations<sup>2,6</sup> in conjunction with chronic inflammatory infiltrate such as plasma cells and eosinophils,<sup>2</sup> or the presence of foreign material.<sup>2</sup> Many of these histologic features can be observed in Figures 2 and 3. It is important to note that not all of these findings will necessarily be present in every case of SPG. Additionally, unlike PG, SPG is not shown to have any associations with systemic, autoimmune, or other types of diseases.<sup>2</sup> Conversely, PG ulcers typically are deep, tender, undermined, and necrotic with

a cribriform floor and vasculitic edges. They tend to heal with scarring and can be associated with systemic diseases such as inflammatory bowel disease, rheumatoid arthritis, and lymphoreticular malignancies. The histopathology of PG includes an acute diffuse inflammatory infiltrate with neutrophils; no foreign materials or sinus tract formations are present.<sup>2</sup>

Superficial pyoderma gangrenosum has been shown to resolve with conservative management. When used alone or in combination, various topical antibiotics have been shown to be successful in treating patients with SPG.<sup>5,9</sup> Other treatments, including minocycline, dapsone, topical or intralesional steroids, and anti-inflammatory drugs, also have been shown to be successful.<sup>1,6</sup> Although there is not one defined treatment of choice, immunosuppressive medication can be avoided in most cases.<sup>1</sup> Our patient was resistant to most topical and routine treatments including cyclosporine, dapsone, and doxycycline hyclate, but his condition dramatically improved after 3 months of intravenous treatment with infliximab. Few cases of SPG treated with infliximab have been reported.<sup>9</sup>

Although the pathogenesis of PG is unknown, it seems to be caused by abnormalities in neutrophil chemotaxis and function. Since tumor necrosis factor  $\alpha$  is known to cause neutrophil activation,<sup>9</sup> we suggest that tumor necrosis factor  $\alpha$  inhibitors should be considered in resistant or widespread cases of SPG, provided the patient has no contraindications.

### REFERENCES

1. Gibson LE. Superficial granulomatous pyoderma: who are you? *J Eur Acad Dermatol Venereol.* 2002;16:97.
2. Thami GP, Kaur S, Punia RS, et al. Superficial granulomatous pyoderma: an idiopathic granulomatous cutaneous ulceration. *J Eur Acad Dermatol Venereol.* 2002;16:159-161.
3. Wilson-Jones E, Winkelmann RK. Superficial granulomatous pyoderma: a localized vegetative form of pyoderma gangrenosum. *J Am Acad Dermatol.* 1988;18:511-521.
4. Powell FC, Collins S. Pyoderma gangrenosum. *Clin Dermatol.* 2000;18:283-293.
5. Powell FC, Su WP, Perry HO. Pyoderma gangrenosum: classification and management. *J Am Acad Dermatol.* 1996;34:395-409.
6. Su WP, Davis MD, Weenig RH, et al. Pyoderma gangrenosum: clinicopathologic correlation and proposed diagnostic criteria. *Int J Dermatol.* 2004;43:790-800.
7. Wollina U. Pyoderma gangrenosum—a review. *Orphanet J Rare Dis.* 2007;2:19.
8. Ahmadi S, Powell FC. Pyoderma gangrenosum: uncommon presentations. *Clin Dermatol.* 2005;23:612-620.
9. Akhras V, Sarkany R, Walsh S, et al. Superficial granulomatous pyoderma treated preoperatively with infliximab [published online ahead of print December 15, 2009]. *Clin Exp Dermatol.* 2009;34:e183-e185.