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**Late-onset post-mammoplasty pyoderma gangrenosum treated with tobacco-pouch suture combined with oral corticosteroids**

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Dear Editor,

Pyoderma gangrenosum (PG) is a rare inflammatory skin condition belonging to the group of neutrophilic dermatoses. Classical PG, which is its most common clinical variant, typically manifests as ulcers with undermined, erythematous to violaceous edge and necrotic bed<sup>1</sup>. Lesions extend peripherally and the border often overhangs the ulceration as the inflammatory process spreads within the dermis, only secondarily causing epidermal necrosis. Recognizing PG may be challenging as it undergoes differential diagnosis with several ulcerative skin disorders and definitive laboratory and histopathological diagnostic criteria are lacking.<sup>1</sup> PG is characterized by pathergy phenomenon, which may also be induced by surgical procedures. In fact, several cases of post-surgical PG following aesthetic and reconstructive breast surgery were described in the literature, and most of them appeared a few days after surgery.<sup>2</sup>

Herein, we present a case of late-onset PG following augmentation mammoplasty successfully treated with a combined approach characterized by tobacco-pouch suture and steroid therapy.

A 43-year-old Caucasian female with an unremarkable medical history presented with an ulcer in the lateral upper external quadrant of the left breast. Three years earlier, she had undergone aesthetic augmentation mammoplasty, with silicon bilateral prostheses inserted under the pectoral major muscle.

The patient reported that the lesion initially presented as a pustular lesion which had appeared one year before, rapidly evolving to ulceration. Before coming to our attention, three skin biopsies had been performed in another department, with no definitive histological diagnosis. Swabs for bacteriological and mycological examinations had resulted negative. Systemic antibiotic therapy with penicillin, tetracycline and macrolides had been ineffective. Subsequently, a low-dose steroid therapy had been started, without any improvement.

At the admission at our Department, dermatologic examination revealed an extensive ulceration of 3 cm of maximum diameter, with necrotic bed and undermined purple borders (FIG.1 A,B).

Histopathological examination of skin biopsy taken from the edge of the lesion showed a dense neutrophilic infiltrate associated with giant foreign body multinucleate cells, some of them including fragments of birefringent exogenous material, visible to polarized light. In addition, direct immunofluorescence test was negative. Blood count, liver and renal function, serum protein electrophoresis, erythrocyte sedimentation rate and C-reactive protein were within normal ranges. Routine screening blood tests for autoimmune diseases were also negative.

Following the discovery of exogenous material, magnetic resonance imaging, requested to evaluate prosthesis integrity, resulted negative. Factitious ulcer having been suspected, an occlusive medication was applied, and the patient was instructed not to remove it. No improvement was observed after three weeks, and psychiatric evaluation ruled out the latter hypothesis. Finally, after having ruled out all the alternative diseases, a diagnosis of PG was made.

A combined medical and surgical approach was considered. As a first step, the plastic surgeon removed the lesion, reducing the defect by a tobacco-pouch suture (FIG.2A). A week after surgery new inflammatory papules and pustules appeared along the suture line (FIG.2B). This phenomenon, known as pathergy, corroborated the diagnosis of PG. Thus, high-dose corticosteroid therapy was prescribed, with rapid improvement of inflammatory lesions. Subsequently, the steroid dosage was reduced and dapsone was introduced as a steroid-sparing agent. The lesion was completely healed after five months of treatment, with a resulting scar which was smaller than the initial lesion (FIG.2C). Further improvement was obtained by application of vitamin E ointment, which made the scar softer and less atrophic.

Differential diagnosis of PG includes infectious, autoimmune, vascular and exogenous diseases<sup>1</sup>. As our patient had undergone additive mammoplasty with the insertion of sub-muscular implants, it was necessary to exclude the infectious hypothesis. PG often presents secondary to breast surgery, with median time from surgery to occurrence of cutaneous lesions of 7 days<sup>2</sup>. In our case, the time from surgery to disease onset was longer (approximately 2 years) as compared to most of the cases reported in the literature. However, other cases of late-onset PG were reported, with a time from surgical procedure to disease onset ranging from 8 months to 20 years<sup>3,4</sup>.

Before being examined at our clinic, the patient underwent a short low-dose systemic steroid therapy, without any improvement of the skin lesion: also for this reason the diagnosis of PG had been delayed, and the hypothesis of self-induced lesion had been considered. In our experience, therapy with high-dose systemic steroids usually allows rapid improvement of the lesions and represents the first-line treatment<sup>1</sup>.

Either oral prednisone (1mg/kg/day) or intravenous pulse corticosteroids have been used and the response can be seen even within 2-3 days<sup>1</sup>. Due to the well-known adverse effects of long-term corticosteroids, in our case dapsone was introduced as a corticosteroid-sparing agent until the wound was fully recovered. A combined approach of plastic surgery with skin grafts combined with immunosuppressive therapy has been proposed by other authors<sup>5</sup>. A surgical approach alone is generally not effective in PG and may trigger pathergy phenomenon. Nevertheless, in our patient, a tobacco-pouch suture of PG was beneficial resulting in appreciably quickening the healing process of the skin wound. This led to a smaller scar, with a better aesthetic result after immunosuppressive therapy.

In conclusion, such a surgical approach in association with topical or systemic treatments for PG, could lead to a better management of these difficult to treat lesions.

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### Figure legends

**FIGURE 1.** (a) Extensive ulceration with necrotic bed and undermined purple borders on the right breast; (b) magnified detail of the clinical lesion

**FIGURE 2.** (a) Pyoderma gangrenosum ulcer after reduction of the defect by means of a tobacco-pouch suture; (b) pathergy phenomenon manifesting as the appearance of papules and pustules along the suture line; (c) scarring sequelae of the ulcerative lesions after complete healing



