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CASE REPORT

Post-surgical pyoderma gangrenosum of the breast: needs for early diagnosis and right therapy

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Abstract
Post-surgical pyoderma gangrenosum (PSPG) of the breast is a rare dermatosis that worsens surgical manipulation, with a chronical relapsing course. Diagnosis is mostly clinical and made by exclusion after the failure of antibiotic therapies and surgical debridement, while the mainstay of therapy is corticosteroid-based. Here we report a case of PSPG of the breast in a young woman with breast cancer, to emphasize the needs of an early and accurate diagnosis, to guarantee the most efficacious treatment and to avoid life-threatening complications.

Keywords Pyoderma gangrenosum · Breast cancer · Breast surgery

Case report

Pyoderma gangrenosum (PG) is a rare cutaneous ulcerative disease that is part of the inflammatory neutrophilic dermatoses [1]. It is characterized by papules, pustules and painful ulceration, rapidly progressive, that does not respond to antibiotics and worsens with traumas or surgical manipulation, with a chronical relapsing course [1, 2]. Histological findings include perivascular and diffuse neutrophilic infiltrates [3]. Its pathogenesis is still poorly understood, and in many cases it is associated with underlying systemic diseases, including autoimmune diseases and solid or hematological malignancies [4]. Moreover some cases of PG after breast surgery are reported in literature [5–8]. The development of PG at a surgical site, typically within 2 weeks after surgery, is called post-surgical pyoderma gangrenosum (PSPG) [9]. Diagnosis is mainly clinical and made by exclusion after the failure of antibiotic therapies and surgical debridement [4]. The therapy of PG requires the treatment of associated concomitant disorders, and in the idiopathic disease it includes immunosuppressive drugs such as corticosteroids, that usually bring to a rapid improvement of the manifestations [10]. Recently 1 major criterion and 8 minor criteria for the diagnosis of ulcerative pyoderma gangrenosum were produced as a guideline for clinicians, allowing for fewer misdiagnoses [4].

In January 2016, a 46-year-old Caucasian female presented at our Breast Unit with a 2.5 cm palpable mass of the right breast. The core biopsy revealed an invasive ductal carcinoma grade 2, hormonal receptors positive, HER2 negative. The woman had no notable medical history, especially no history of autoimmune disorders, and was not assuming chronical therapies.

In February 2016, the patient underwent right lumpectomy with sentinel node biopsy and left breast reduction mammoplasty with a periareolar "round block" incision to improve the symmetry; no peri-operative complications were observed and the patient was discharged on postoperative day 2, with cefixima as prophylaxis. The histopathological report confirmed luminal-A like, stage I, breast cancer. Two weeks later, the patient presented at the emergency room with fever and a painful right breast with edema in the lower quadrants: amoxicilline/clavulanic acid 1 g three times a day was suggested. In few days, inflammatory papules

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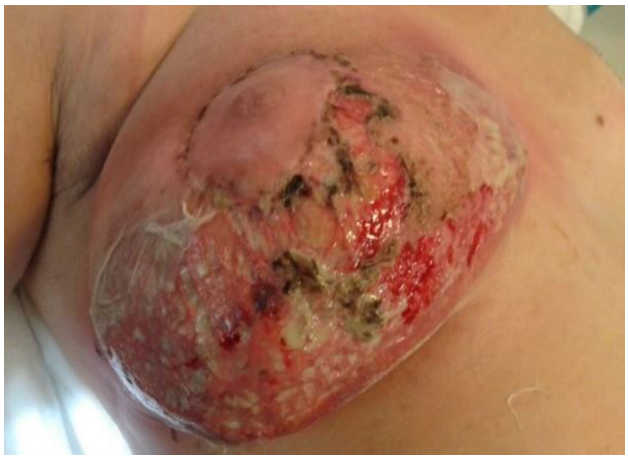

Fig. 1 Pyoderma gangrenosum of the right breast after lumpectomy for breast cancer

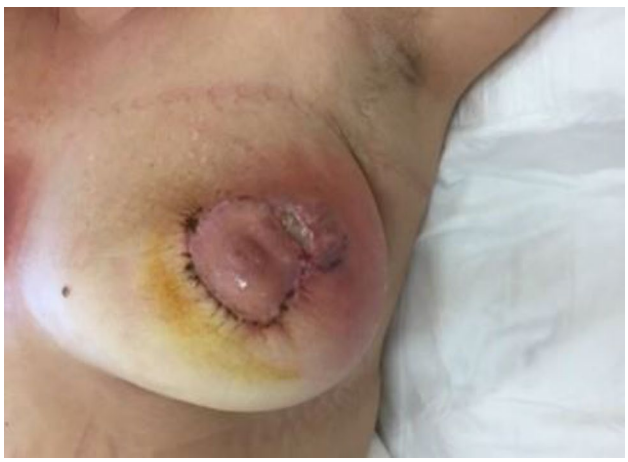

Fig. 2 PSPG spreading to the left breast, after reduction mammoplasty

and vesicles appeared on the right breast and broke down to form an erosion. Small erythematous ulcerated painful papules developed closed to the surgical scar. The lesions had necrotic eschar with purulent discharge (Fig. 1). The ulcer expanded rapidly, spreading to the other breast in 2 days' time (Fig. 2). The patient was admitted and treated with intravenous injection of antibiotics (piperacilline 4 g and tazobactam 500 mg 3 times daily in association with clarithromycine twice a day) and local wound care, without clinical improvement. In few days, the lesions got involved two-third of the skin of both breasts. The clinical impression was of sepsis, so intravenous antibiotics were changed with no improvement. A surgical removal of necrotic tissue was performed twice (Fig. 3a, b): the tissue and blood cultures remained sterile, whereas the histopathology report (March 2016) showed a diffuse neutrophilic dermatosis with vasculitis and necrosis. The clinical diagnosis of PSPG was suggested. Diagnostic criteria of ulcerative PG previously mentioned [4] were satisfied (major criterion: biopsy demonstrating neutrophilic infiltrate and minor criteria: exclusion of infection; pathergy; peripheral erythema and tenderness at ulceration site; cribriform or "wrinkled paper" scars at healed ulcer sites).

Patient started high-dose systemic steroids (methylprednisolone 1 mg/kg/day), with a swift improvement in few days. She also received 30 hyperbaric oxygen therapy (HBOT) sessions to help the lesions to heal more quickly. Adjuvant treatment with Tamoxifen was started but no complementary radiotherapy was delivered. After 6 months, the breast skin was completely healed and the patient gradually stopped steroids, without evidence of relapse (Fig. 4a–c).

Here we have reported a case of PG of the breast, developed after breast surgery in a young woman with operable breast cancer and no other underlying conditions. PG is a

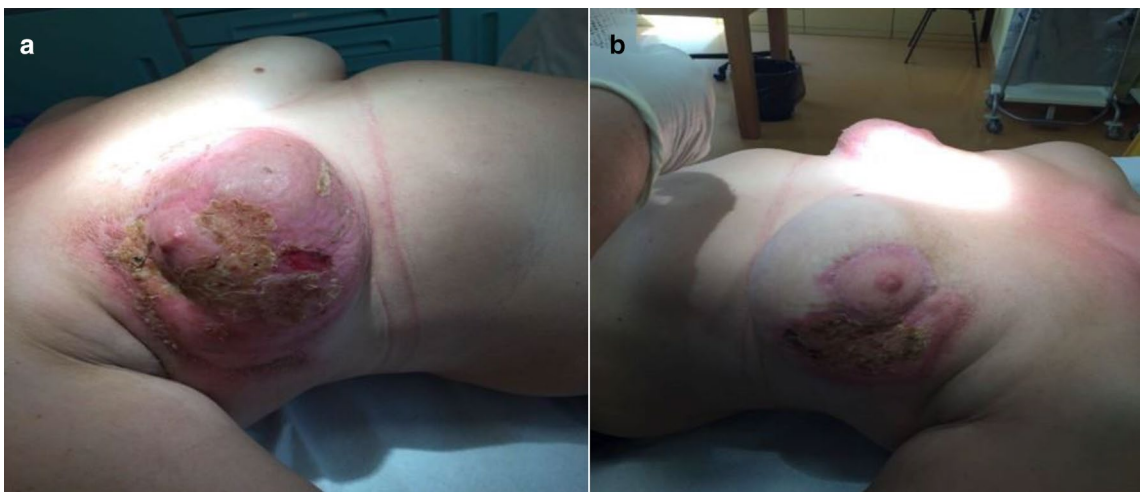

Fig. 3 Bilateral surgical debridement after antibiotics failure

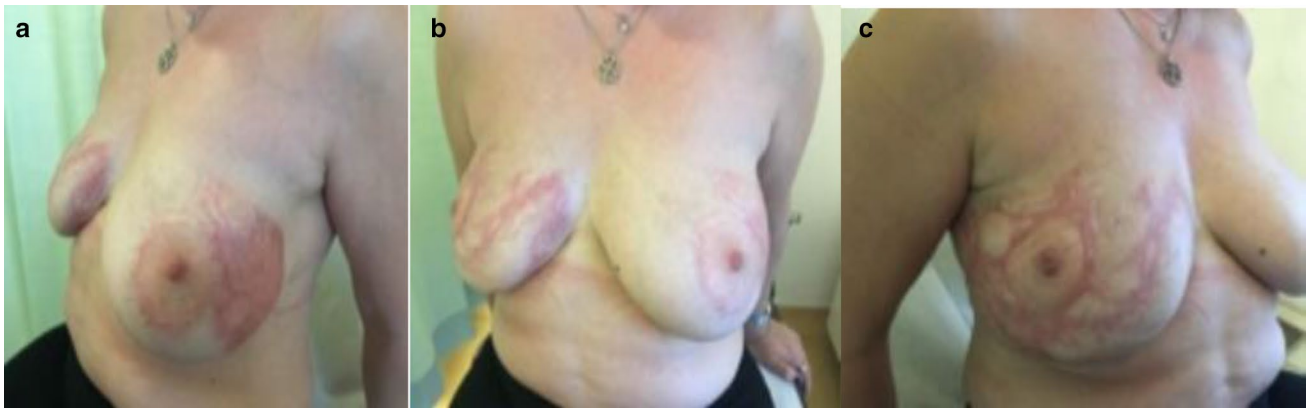

Fig. 4 Healing of breasts' skin after corticosteroids and hyperbaric oxygen therapy

rare cutaneous non-infectious ulcerative disease. The most common sites of appearance of PG are legs or lower limbs. Its pathogenesis is unknown: it is usually associated with systemic disease, but in a quarter of cases it appears as idiopathic or occurs after a trauma of the skin [11]. PG of the breast is usually precipitated by breast surgery [5–8].

PSPG can mimic infective or inflammatory causes that should be excluded in the presence of rapidly worsening wounds. There are no specific laboratory or pathological tests, but the diagnosis is usually of exclusion and guided by clinical patterns. Misdiagnosis can lead to unnecessary repeated antibiotic treatment, such as in our experience. On the contrary, rapid response to immunosuppression usually helps to confirm the diagnosis of PG. We want to stress the importance of an accurate and rapid differential diagnosis, including the search for coexisting diseases, to guarantee the most efficacious treatment, to avoid life-threating complications, to improve aesthetic results and not to delay specific oncological adjuvant therapies. Surgical management to remove necrotic tissue of PG wounds is still controversial, as it can lead to an extension of the disease instead of to promote healing, like in inflammatory processes. According to our experience, surgery should be performed after corticosteroids failure, to obtain tissue samples helpful to histological examination and to rule out other potential causes of misdiagnosis.

The role of HBOT for wound healing is well established and its use for treatment of PG has been previously described [12–14]. It may be considered to obtain a quicker healing when prolonged use of corticosteroids is contraindicated (i.e. patients suffering from diabetes, hypertension or glaucoma) or maybe even to consolidate aesthetic results.

Compliance with ethical standards

Conflict of interest All the authors declare no relevant affiliations or financial involvement with any organization or entity with a financial interest in or financial conflict with the subject matter or materials discussed in the manuscript.

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