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CASE REPORT

Pyoderma gangrenosum with severe systemic features, after augmentation mammoplasty

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Abstract

We report a rare life-threatening presentation of postsurgical pyoderma gangrenosum (PG) after augmentation mammoplasty in a 32-year-old woman. Six days after surgery, the patient presented with fever and erythema at surgical wounds. In view of a suspected postsurgical infection, the patient was first treated with antibiotics and removal of breast protheses. In spite of this treatment, in a few days the patient developed a state of shock with multiorgan failure. The antibiotic coverage was broadened several times and multiple extensive surgical debridement with resection of the mammary glands were performed but did not improve the clinical situation. Twelve days after admission, the diagnosis of PG with systemic features was suspected and clinical improvement was observed within 24 h of methylprednisolone administration. PG can mimic the cutaneous and systemic features of necrotizing wound infection of surgical site. Biopsy and culture are critical to differentiating necrotizing neutrophilic dermatosis from necrotizing infection. The diagnosis of this less common form of PG is challenging and this case highlights how the frequent misdiagnosis with infection may result in ineffective antibiotic treatment and how the unnecessary surgical debridement may prolong and exacerbate the condition.

KEYWORDS

augmentation mammoplasty, breast surgery, pathergy, postoperative pyoderma gangrenosum, postsurgical pyoderma gangrenosum, pyoderma gangrenosum, shock, systemic features

INTRODUCTION

Pyoderma gangrenosum (PG) is an inflammatory neutrophilic dermatosis with affects between 3 and 10 patients per million per year and is more common in women.[1–3]

Postoperative pyoderma gangrenosum (PPG) occurs in the early postoperative period at surgical site. It is mainly characterized by erythema, papulo-pustules and pain at surgical incisions followed by wound dehiscence. It develops rapidly into large, purulent and necrotic ulcers with violaceous and undermined borders.[4]

PG is characterized by neutrophil infiltrates in the skin. The pathophysiology remains unclear, but it may involve dysregulation of the innate and adaptative immune responses with upregulation of neutrophilic function and release of cytokines, in addition to genetic factors.[5] Operative trauma could act as a trigger, called pathergy.[1] Therefore, initial surgery and wound debridement for suspicion of postoperative infection can initiate and exacerbate the disease. Therefore, PPG can present as a more severe disease, sometimes with systemic features.[1] The most likely pathologic mechanism of the multiorgan failure in PPG is excessive expression of pro-inflammatory cytokines or systemic inflammatory response syndrome secondary to extensive skin lesions.[6–9]

PG is usually associated with systemic disease like inflammatory bowel diseases, rheumatoid arthritis and myeloproliferative disorders. In contrast to idiopathic PG, PPG has a lower association with these systemic diseases.[1,4]

The diagnosis of this less common form of PG is challenging and this case highlights how the frequent misdiagnosis with infection may prolong and exacerbate the condition.

CASE REPORT

A 32-year-old woman presented at the emergency department with pain on both breasts and fever. She had undergone a retropectoral augmentation mammoplasty by submammary fold 6 days earlier at a private clinic. The surgical wounds presented erythema and bilateral dehiscence.

The patient had a history of Crohn's disease, which had been stable for 8 years with no symptoms. Therefore, she wasn't taking any medication and hadn't consulted a gastroenterologist for the past few years. She was otherwise healthy with no other medical history.

The initial blood analyses showed a C-reactive protein (CRP) level of 100 mg/L (normal value [NV] <5 mg/L) and a white blood cell (WBC) count at 10,000/μL (NV: 3500–10,000/μL), of which 90% neutrophils. Bacterial wound infection was suspected and a treatment with high-dose antibiotics was started.

Despite this treatment, the patient had high-grade fever (>39°C) with increased leukocytosis (WBC 30,000/μL, of which 92% neutrophils). The wounds were surgically explored and cleaned, and the breast prostheses were removed. Two days after removal of the prostheses, the patient developed hemodynamic instability with hypotension, polypnea and tachycardia. Due to this deteriorated condition, the patient was admitted to the intensive care unit. Several broad-spectrum antibiotics were administered but the situation did not improve. Within a few days, the patient developed a state of shock and multiorgan failure with WBC count at 55,000/μL of which 96% neutrophils, CRP level of 280 mg/L and lactate at 28 mg/dL (NV: 5–20 mg/dL). She suffered from acute renal failure with creatinine at 1.7 mg/dL (NV < 1.2 mg/dL); disseminated intravascular coagulation with platelet count at 135,000/μL (NV: 150,000-400,000/μL), INR at 1.7 (NV: 0.85–1.30) and fibrinogen at 100 mg/dL (NV: 200–400 mg/dL); acute respiratory distress syndrome with increasing oxygen requirements; encephalopathy and shock liver with alanine aminotransferase at 70 UI/L (NV < 41 UI/L), aspartate aminotransferase at 83 UI/L (NV < 40 UI/L), total bilirubin at 3.1 mg/dL (NV < 1 mg/dL), direct bilirubin at 2.9 mg/dL. The patient required hemodynamic and ventilatory support. The wounds worsened and presented large necrotic ulcerations with purple margins, sparing both nipple-areola complexes (Figure 1a,b). The extension of necrosis required large debridement resulting in a bilateral near-total subcutaneous mastectomy with resection of the mammary glands and pectoralis major muscle. Both nipples, however, could be preserved. Blood and wound cultures remained sterile. Ten days after removal of breast prostheses, a skin biopsy was performed. Histological analysis showed a very dense, interstitial neutrophilic infiltrate in the dermis with peri-vascular involvement and deep extension (Figure 2a,b). Neutrophilic epidermal microabscesses were noted with zones of complete necrosis. Absence of infectious organisms was noted. Gastroscopy and colonoscopy were normal, excluding active inflammatory bowel disease.

Based on the history of the disease, the clinical presentation, the anatomopathological aspect of the skin biopsy and the laboratory findings, the diagnosis of postsurgical PG with systemic features was made. The patient was treated with intravenous corticosteroids (1 mg/kg/day methylprednisolone starting dose). The wounds were treated with topical 0.5% tacrolimus once a day. Within 24 h of methylprenidosolone administration, a rapid improvement of systemic symptoms and disappearance of skin lesions expansion was noted. The wounds healed in 12 weeks by secondary intention but atrophic and cribiform scars formed (Figure 1c). The oral methylprednisolone was tapered gradually and stopped after 8 weeks, and the topical tacrolimus was stopped after 12 weeks.

The dermatologists and surgeons who coauthored this case report were consulted after the acute phase of the disease for breast reconstruction and they advised a cautious conservative approach. However, 9 months later, a lipofilling procedure was attempted elsewhere. Unfortunately, a second episode of PG occurred 15 days

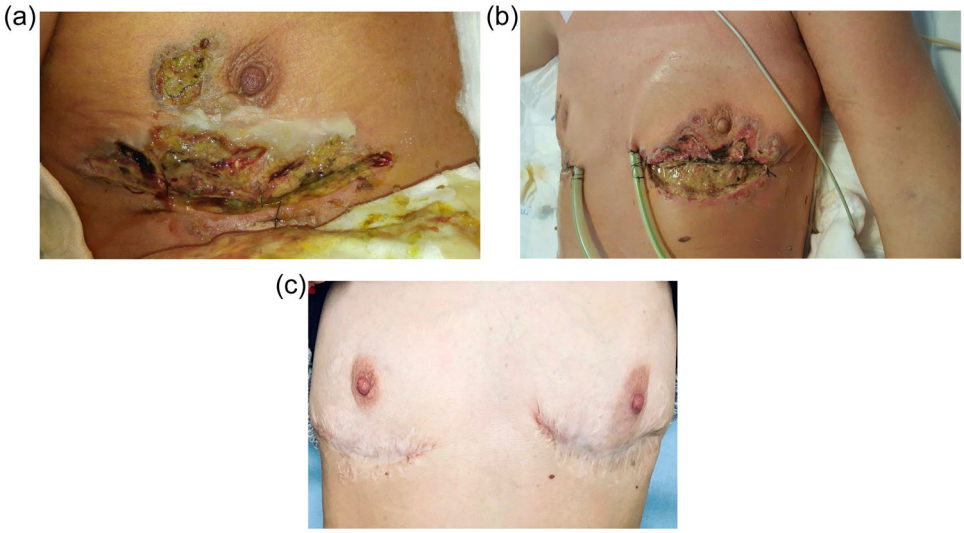

FIGURE 1 (a) Right breast on the 10th day after breast surgery and second day after removal of breast implants. The wound presented dehiscence and ulceration. (b) Left breast on the 20th day after breast surgery and 12th day after removal of breast implants. The wound presented ulceration and purple margins, sparing both nipple-areola complexes. (c) The wounds, almost 2 years after breast surgery, presented complete healing.

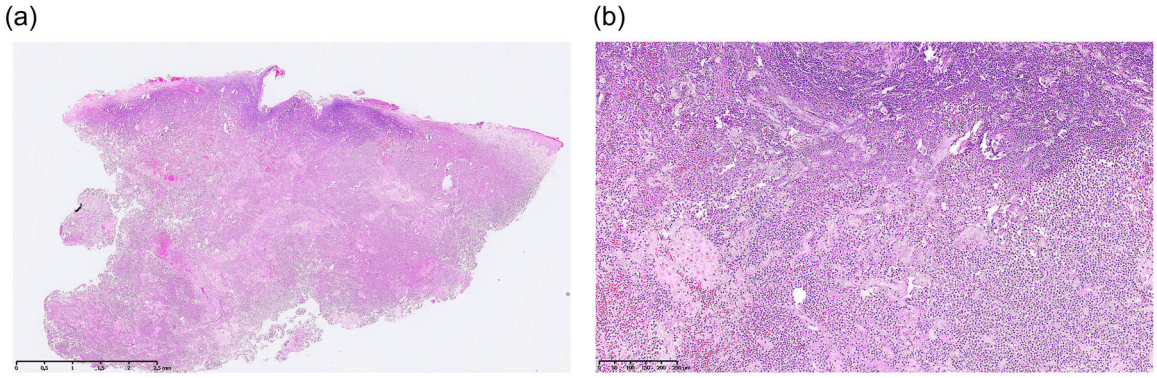

FIGURE 2 (a) Histopathological findings (hematoxylin and eosin preparation from the skin biopsy specimens) with a very dense, interstitial neutrophilic infiltrate in the dermis with peri-vascular involvement and deep extension (magnification ×1.76). (b) Dense neutrophilic infiltrate (magnification ×9.75).

after the procedure, on both thighs, used as fat donor sites (Figure 3).

DISCUSSION

The clinical course of PPG is rarely as severe as in this patient, although a few cases of acute, rapidly evolving neutrophilic dermatoses have been reported.[2,3,7,9,10] The diagnosis of PPG is challenging. It is often misdiagnosed as a postoperative infection. Ineffective antibiotics and unnecessary surgical debridement prolong and exacerbate the condition.[3,5,11] Prompt recognition of the disease and early appropriate treatment can improve patients' outcomes.

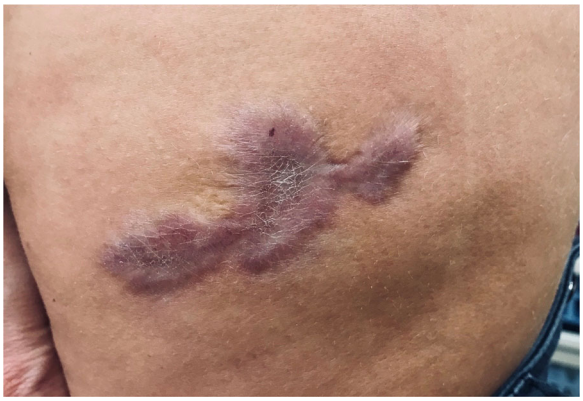

FIGURE 3 Second episode of pyoderma gangrenosum on the thigh, nine months after postoperative pyoderma gangrenosum.

In this reported case, an earlier diagnosis would have prevented an unnecessary resection of the mammary glands.

In contrast to idiopathic PG, PPG is usually less associated with systemic disease, like inflammatory bowel diseases. In our case, the patient's history of Crohn's disease is, however, probably relevant.

Patients with a history of PG may be at increased risk of recurrence, as also shown by the present observation.[1,3] If surgery is required, perioperative corticosteroid treatment may help to prevent the development of new lesions.[1,3,12–14]

AUTHOR CONTRIBUTIONS

Marine Beeckman performed the literature search. Marine Beeckman, Caroline Peeters, Maude Coyette, Halil Yildiz, Benoit Lengele, and Marie Baeck participated in data collection. Liliane Marot performed the anatomo-pathological analysis. Marine Beeckman prepared the manuscript. Marie Baeck and Benoit Lengele reviewed and approved the manuscript. Marie Baeck decided to submit the manuscript for publication. All authors approved the submitted version.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no data sets were generated or analysed during the current study.

ETHICS STATEMENT

All patients in this manuscript have given written informed consent for participation in the study and the use of their de-identified, anonymized, aggregated data and their case details (including photographs) for publication. Ethical Approval: not applicable.

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REFERENCES

1. Tolkachjov S, Fahy A, Cerci F, Wetter D, Cha S, Camilleri M. Postoperative pyoderma gangrenosum. Mayo Clin Proc. 2016;91(9):1267–79.
2. Alosaimi M, Sebzali F, Iqbal A, Rabbat C, Clase C. Pyoderma gangrenosum after insertion of a hemodialysis catheter: Koebner phenomenon, systemic inflammatory response syndrome, and a delay in diagnosis. Can J Kidney Health Dis. 2022;9:20543581221120618.
3. Zuo K, Fung E, Tredget E, Lin A. A systematic review of post-surgical pyoderma gangrenosum: identification of risk factors and proposed management strategy. J Plast Reconstr Aesthet Surg. 2015;68(3):295–303.
4. Braswell S, Kostopoulos T, Ortega-Loayza A. Pathophysiology of pyoderma gangrenosum (PG): an updated review. J Am Acad Dermatol. 2015;73(4):691–8.
5. Tuffaha S, Sarhane K, Mundinger G, Broyles J, Reddy S, Azoury S, et al. Pyoderma gangrenosum after breast surgery. diagnostic pearls and treatment recommendations based on a systematic literature review. Ann Plast Surg. 2014;77(2):e39–44.
6. Doren E, Aya-ay M. Pyoderma gangrenosum following breast reduction: treatment with topical tacrolimus and steroids. Aesthet Surg J. 2014;34(3):394–9.
7. Robbins A, Mestrallet S, Dutel-Charneux J. Un pyoderma gangrenosum post-opératoire mimant un choc septique. La Revue de médecine Interne. 2015;36:101–2.
8. Abtahi-naeini B, Bagheri F, Pourazizi M, Forozeshfard M, Saffaei A. Unusual cause of breast wound: postoperative pyoderma gangrenosum. Int Wound J. 2017;14:285–7.
9. Didan A, Donnelly A, Chua H. An unusual presentation of pyoderma gangrenosum leading to systemic inflammatory response syndrome. Case Rep Dermatol. 2017;9:146–50.
10. Sanchez I, Lowenstein S, Johnson K, Babik J, Haag C, Keller J, et al. Clinical features of neutrophilic dermatosis variants resembling necrotizing fasciitis. JAMA Brief Rep. 2019;155(1):79–84.
11. Ehrl D, Heidekrueger P, Broer P. Pyoderma gangrenosum after breast surgery: a systematic review. J Plast Reconstr Aesthet Surg. 2018;71(7):1023–32.
12. Juhász M, Maman D, Levin J, Marmur E. Pyoderma gangrenosum of the breast in a patient with a history of silicone augmentation mastopexy and suction-assisted lipectomy of the trunk. JAAD Case Rep. 2015;1(6):329–32.
13. Larcher L, Schwaiger K, Eisendle K, Ensat F, Heinrich K, di Summa P, et al. Aesthetic breast augmentation mastopexy followed by post-surgical pyoderma gangrenosum (PSPG): clinic, treatment, and review of the literature. Aesth Plast Surg. 2015;39:506–13.
14. Van Poucke S, Jorens P, Peeters R, Jacobs W, de Beeck B, Lambert J, et al. Pyoderma gangrenosum: a challenging complication of bilateral mastopexy. Int Wound J. 2004;1:207–13.

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