



A Rare Complication Following Breast Conserving Surgery: Pyoderma Gangrenosum

Glenn Costa¹, Serkan İlgün¹, David Pisani², John Agius¹

¹Department of Surgery, Mater Dei Hospital, Msida, Malta

²Department of Histopathology, Mater Dei Hospital, Msida, Malta

ABSTRACT

Pyoderma gangrenosum (PG) after breast-conserving surgery is rare, and its diagnosis is often delayed because of the similarity to wound infection and the broad differential diagnosis for PG, making it a diagnosis of exclusion. A 60-year-old woman who underwent breast conserving surgery and sentinel lymph node biopsy for invasive breast carcinoma presented with increasing erythema, fever and serosanguinous discharge in the lower outer quadrant of the right breast at the site of tumour excision on postoperative day (POD) 9. Fever persisted despite antibiotics and the patient was noted to have leucocytosis ($0.9 \times 10^9/L$), neutrophilia ($37.8 \times 10^9/L$) and elevated C-reactive protein levels ($136 \mu g/mL$) on POD 16. Microbiology and blood culture results were negative but the breast ulcer continued to expand at a rate of 1-2 cm a day. The patient underwent surgical debridement on POD 21 to rule out necrotising soft tissue infection. Persistent ulcer progression, despite debridement and antibiotics, led to clinical suspicion of PG and the patient was started on prednisolone and cyclosporin. A rapid response was seen with treatment and an optimum healing process was noted over the subsequent three-month follow-up period. Early suspicion, careful macroscopic evaluation of disease progression and appropriate use of immunosuppressive therapy are important for the management of PG. Prompt initiation of immunosuppressive therapy may avoid unnecessary treatment and aggravation of the surgical wound.

Keywords: Breast cancer; breast conserving surgery; pyoderma gangrenosum

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Key Points

- Early diagnosis of pyoderma gangrenosum (PG) is critical to avoid unnecessary treatment and aggravation of the surgical wound.
- PG of the breast although rare has been reported in the literature.
- However its onset following breast conserving surgery is very rare and may be difficult to diagnose due to its wound infection-mimicking nature.
- This case report should raise awareness about PG following breast conserving surgery as well as guide the clinician in making an appropriate and timely diagnosis to start targeted treatment.

Introduction

Pyoderma gangrenosum (PG) is a rare neutrophilic dermatosis with multiple and differing clinical presentations and associated comorbidities (1). PG is often associated with systemic diseases, such as inflammatory bowel disease, rheumatoid arthritis or haematological malignancies (2). The pathophysiology is poorly understood and is thought to involve adaptive and innate immune system dysregulation, abnormalities of neutrophil function such as chemotaxis, adhesion and trafficking, abnormal phagocytosis and genetics (3).

PG typically presents with painful lesions in different locations and with non-specific histology. This poses a clinical challenge and diagnosis is

often delayed. In the classic ulcerative variant, characterized by ulcers with inflammatory undermined borders, a broad differential diagnosis of malignancy, infection, and vasculitis needs to be considered, making PG a diagnosis of exclusion (4).

Breast PG is uncommon, with only 87 cases documented in the literature. It is most commonly associated with breast reduction surgery (38 cases, 44%) followed by augmentation mammoplasty and mastectomy with free deep inferior epigastric perforator flap (5). We present a very rare case of unilateral breast PG following breast conserving surgery in a 60-year-old woman which, to the best of our knowledge, is the first such case reported in the literature.

Corresponding Author:
Glenn Costa; glenncosta110@gmail.com

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The following case is presented in accordance with the CARE reporting checklist.

Case Report

A 60-year-old female patient with no previous co-morbidities underwent breast conserving surgery and sentinel lymph node biopsy for invasive breast carcinoma. The patient did not have any co-morbidities, either before or after the surgery. Her cancer was no special type, grade 3 (pT1c, N0). She presented to the emergency department on postoperative day 9 with increasing erythema and serosanguinous discharge in the lower outer quadrant of the right breast, at the site of tumour excision. A breast ultrasound carried out at the emergency department was suggestive of a seroma. A wound swab was taken and the patient was discharged on oral antibiotics (Ciprofloxacin and Clindamycin) with planned follow-up.

She presented one week later with recurrent febrile episodes (37.8 °C), severe tenderness and a rapidly evolving, cutaneous ulcer at the lower outer quadrant of the right breast, sparing the nipple and areola (Figure 1).

The patient was admitted for further investigations and treatment. She was noted to be febrile (Temp 38.5 °C) and tachycardic with a heart rate of 98 bpm. Blood tests revealed an inflammatory picture with leucocytosis ($0.9 \times 10^9/L$), neutrophilia ($37.8 \times 10^9/L$) and elevated C-reactive protein levels (136 µg/mL). Despite antibiotic treatment with high dose Tazocin and Metronidazole, the intermittent episodes of fever persisted and the breast ulcer continued to expand at a rate of 1-2 cm a day (Figure 2).

Microbiology and blood culture results were all negative. On the fifth day of admission the patient underwent surgical debridement to rule out necrotising soft tissue infection (Figure 3). Intraoperatively it was noted that only skin was affected and the underlying breast tissue was spared infection or necrosis.

Despite the debridement and antibiotics, the ulceration continued to progress and blood results did not improve. This led us to consider PG as part of the differential diagnosis.

A skin biopsy obtained during surgical debridement was reported as diffuse epidermal ulceration with associated gangrenous necrosis of the superficial dermis. A dense transdermal acute inflammatory infiltrate, comprised almost exclusively of neutrophil polymorphs, was evident. Associated leukocytoclastic vasculitis was also identified in places. There was no evidence of malignancy. No micro-organisms were identified histologically. These findings were supportive of the possible diagnosis of PG (Figures 4, 5).

The case was discussed with dermatology and the patient was started on oral prednisolone 60 mg daily for one week (tailed down by 10 mg every following week) and Cyclosporin 100 mg twice daily. A rapid response was noted with the steroid treatment. The patient reported reduced symptoms of pain and was no longer febrile within a matter of days. During the three-month follow-up period, a good healing process with significant improvement was evident (Figure 6).

Written informed consent was obtained from the patient for publication of this case report and accompanying images.



Figure 1. Ulcerated area with surrounding erythema on presentation



Figure 2. Evolution of ulcerated area and surrounding erythema



Figure 3. Two days following surgical debridement of non-viable skin

Discussion and Conclusion

PG is a reactive, non-infectious, inflammatory dermatosis, which falls within the spectrum of the neutrophilic dermatoses. These constitute a broad spectrum of diseases of uncertain and complex pathophysiology,

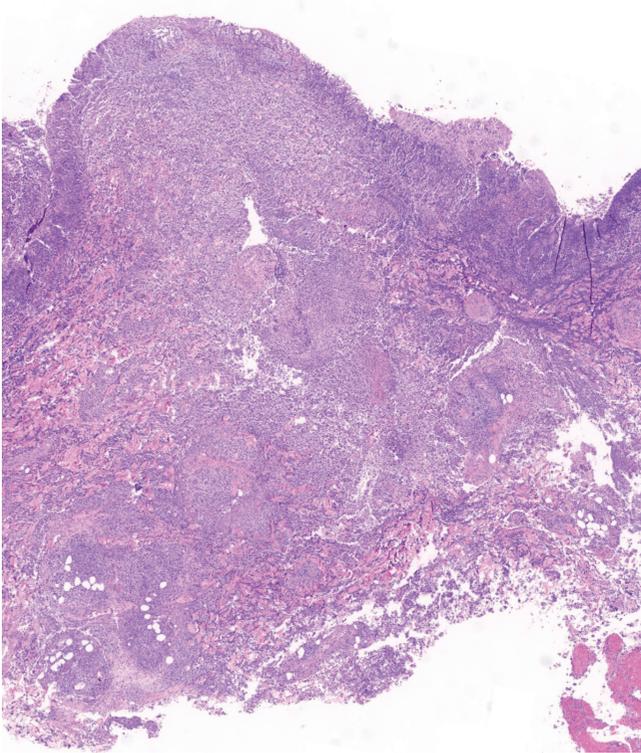


Figure 4. Skin biopsy showing diffuse epidermal ulceration with gangrenous necrosis of the superficial dermis and a dense transdermal inflammatory infiltrate (H&E, x20)

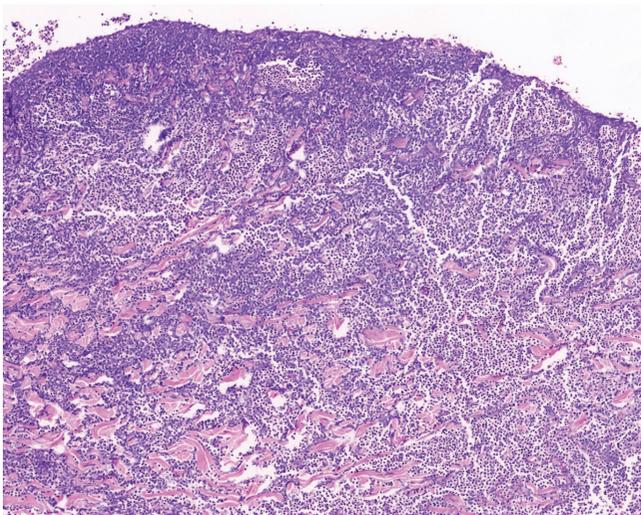


Figure 5. Diffuse epidermal ulceration with gangrenous necrosis of the upper dermis is evident. The inflammatory infiltrate is comprised almost exclusively of polymorphonuclear leukocytes. Micro-abscess formation is evident

which also includes Sweet's syndrome, neutrophilic dermatosis of the dorsal hand, neutrophilic eccrine hidradenitis and Behcet's disease. Classical PG is the most common form (85% of cases) and usually presents as an extremely painful erythematous lesion, which rapidly progresses to a blistered or necrotic ulcer. The lower legs are most frequently affected, although PG can present at anybody site (6).

Minor trauma to skin can result in exaggerated skin injury, a phenomenon known as pathergy (7). PG lesions can be easily misdiagnosed as simple non-healing ulcers and patients usually



Figure 6. Significant clinical improvement noted after three months of treatment

undergo debridement, resulting in a rapid deterioration of the condition through a pathergic response.

PG has an extensive differential diagnosis because all other causes of cutaneous ulcers should be considered. These include arterial and venous disease, haematological/immunological causes (sickle cell disease, cryoglobulinemia, anti-phospholipid syndrome), vascular occlusion, vasculitis, infections, calciphylaxis, drug-induced ulceration, primary or metastatic tumours, hypertension (Martorell ulcer) and other inflammatory disorders including cutaneous Crohn's disease (6).

PG remains a clinical and sometimes challenging diagnosis and although histology of skin biopsies can be supportive, the main value of the skin biopsy is to exclude other causes of cutaneous ulceration and to allow specimens to be sent for bacterial, mycobacterial and fungal cultures. This makes PG a diagnosis of exclusion, based on ulcerative characteristics, negative microbiological results, supportive histological findings, resistance to antibiotic and surgical therapy and improvement after steroid treatment (8).

The severity of PG influences the mode of treatment. The aim of first-line treatment is to optimise local wound care. Potent topical corticosteroids and tacrolimus ointment applied to the ulcer surface are useful and intralesional injections of corticosteroid into the erythematous active border may be considered (9).

In more severe cases, such as the case presented above, systemic therapy is required. Oral corticosteroids are the mainstay of treatment and are used to gain rapid control. Cyclosporin can be used, either alone or in combination with corticosteroids, as a steroid-sparing agent in cases where prolonged treatment is required (10). In the present case, antibiotics were initially started based on signs of inflammation and probable infection. Since the microbiology and blood culture results were negative, a therapeutic approach with corticosteroids and cyclosporin was initiated and this provided effective treatment.

PG following breast-conserving surgery is rare and is not easily diagnosed. Early suspicion, careful macroscopic evaluation of disease progression and appropriate use of immunosuppressive therapy are important for the management of PG. Prompt initiation of immunosuppressive therapy may avoid unnecessary treatment and aggravation of the surgical wound.

Informed Consent: Written informed consent was obtained from the patient for publication of this case report and accompanying images.

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Authorship Contributions

Surgical and Medical Practices: G.C., S.İ., J.A.; Concept: G.C., S.İ., J.A.; Design: G.C., S.İ., D.P., J.A.; Data Collection and/or Processing: G.C., S.İ., D.P.; Analysis or Interpretation: G.C., S.İ., D.P., J.A.; Literature Search: G.C., S.İ., D.P.; Writing: G.C., S.İ., D.P.

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