

CASE REPORT Reconstructive

Pyoderma Gangrenosum Masquerading as Necrotizing Infection after Autologous Breast Reconstruction

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Summary: Pyoderma gangrenosum (PG) is a diagnostic dilemma when it presents with a superimposed infection and previous surgery without subsequent inflammatory infection. In this setting, PG is not at the forefront of the surgeon's mind. Furthermore, the treatment for PG, systemic steroids, may cause serious morbidity if the necrotizing infection is the actual culprit. We present an autologous breast reconstruction patient with previous uncomplicated surgery and no personal history of inflammatory disease. Important clinic clues to aid the surgeon in diagnosis include irregular violaceous undermined border, purulence limited to the skin, bilateral involvement, the involvement of the abdominal wound, sparing of the mastectomy site, and relative sparing of the nipples and umbilicus. (*Plast Reconstr Surg Glob Open 2020;8:e2596; doi: 10.1097/GOX.00000000002596; Published online 10 April 2020.*)

Pyoderma gangrenosum (PG) is a diagnosis of exclusion. The incidence is extremely low at 3–10 cases per million annually.¹ Once thought to be the result of underlying streptococcal infection, PG can be associated with, but not caused by, bacteria.¹ The pathogenesis involves abnormalities in neutrophil function, underlying systemic inflammatory disease, and genetic factors. The majority of patients with PG have underlying inflammatory bowel disease, hematologic disorder, or arthritis. Pathergy, the progression of wounds after minor trauma, is reported in 20% of patients.¹

CASE

The patient is a 67-year-old woman with a history of bilateral ductal carcinoma in situ, morbid obesity, and diverticular disease. No underlying inflammatory disease has been diagnosed. She has a family history of breast cancer and a granddaughter with Crohn's disease. She had a history of multiple oncologic breast operations and

From the *Department of Plastic and Reconstructive Surgery, MedStar Georgetown University Hospital, Washington, D.C.; †Department of Plastic and Reconstructive Surgery, CHA Budang Medical Center, Seongnam, South Korea; and ‡Plastic and Reconstructive Surgery, MedStar Franklin Square Medical Center, Baltimore, Md.

Received for publication June 25, 2019; accepted October 29, 2019. Copyright © 2020 The Authors. Published by Wolters Kluwer Health, Inc. on behalf of The American Society of Plastic Surgeons. This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal. DOI: 10.1097/GOX.00000000002596 elected to undergo a bilateral mastectomy and expander placement due to breast cancer recurrence. She experienced a superficial infection that resolved with antibiotics. This procedure was otherwise uncomplicated. She then underwent bilateral muscle-sparing transverse rectus abdominis musculocutaneous flaps and was discharged on a postoperative day (POD) 3 without complications. On POD11, she presented to the clinic with oozing, malodor, and erythema of all wounds (Fig. 1). She acknowledged difficulty adhering to postoperative protocols having showered and removed her dressings preemptively. She has animals at home and reported that they had been close to her wounds.

She was admitted for IV antibiotics and wound management. She was afebrile, normotensive, and nontachycardic but had a leukocytosis of 16,700 cells/cm³ (80.2% neutrophils). She underwent superficial debridement of her medial breast wounds on POD13 after no improvement. Mucopurulent discharge limited to the dermis layer was noted. Wound cultures from admission revealed multiresistant *Enterobacter* and β -hemolytic *Streptococcus*. Meropenem and sulfamethoxazole/trimethoprim 800/160 were initiated. All other cultures were negative. She underwent repeat debridement on POD15. There was marked worsening of her medial breast wounds and extension of violaceous borders suspicious for PG. Two wound edge biopsies from the abdomen and right breast were obtained. Wound care was continued with Dakin's wet-to-dry dressings.

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Fig. 1. On presentation, the patient presents with the classic pyoderma appearance, with violaceous, tender wounds. We noted at the time mucopurulent discharge limited to the dermal layers. Upon debridement, significant pathergy with worsening of the wounds in debrided areas was noted.

Rheumatologic panel, HIV, and hepatitis were all negative. C reactive protein was 48 mg/L. The patient was started on pulse IV steroids (methylprednisolone, 1000 mg daily) for 3 days followed by prednisone 100 mg daily. She noted significant improvement immediately after steroid initiation. Remission of violaceous borders and drainage was noted by day 2 of steroids. Dressings were transitioned to the hydrogel and abdominal pads. The wound edge biopsies demonstrated benign skin with ulceration, marked acute inflammation, focal necrosis, and increased fibrosis, consistent with PG. Vacuumassisted closure (VAC; KCI, San Antonio, TX) was placed to confirm no pathergy occurred and she was discharged (Fig. 1).

She continued to improve with weekly wound care visits. Minimal devitalized, fibrinous tissue was gently debrided with curettes in office as necessary. Steroids were gradually tapered. At 2 months, her breast wounds were transitioned from VAC to collagenase ointment (Santyl, Smith and Nephew, London, UK) and moist gauze dressings (Fig. 2). At 4 months, her abdominal wound care was similarly transitioned (Fig. 3). At 10 months, all wounds were healed besides two 2×1 cm areas of the abdominal incision (Fig. 4).

DISCUSSION

PG frequently masquerades as necrotizing infection in the postoperative setting. Thus, the diagnosis of PG in the surgical patient is one of the exclusions and often delayed. Patients are treated with surgical debridement, which inadvertently exacerbates wounds. Four subtypes of PG have been described (Table 1).¹ Diagnostic



Fig. 2. Two months after the presentation, the classic violaceous borders, undermining, and mucopurulent discharge resolved with steroids and VAC therapy. Wounds have all decreased in size significantly. VAC therapy is stopped on the breasts for Santyl (Smith and Nephew, London, UK) and moist gauze, while continued on the abdomen.

criteria created by a Delphi consensus emphasize biopsy of the ulcer edge, which demonstrates neutrophilic infiltrate, pseudoepitheliomatous hyperplasia, and dermal abscess.²

Postsurgical PG (PSPG) most often occurs in breast surgery (25–39% of all cases), the majority after reduction mammoplasty (45%) or breast reconstruction (25%).^{3,4} It has also been commonly reported after



Fig. 3. Four months after the presentation, all wounds are >90% healed from the initial presentation. The patient continues Santyl (Smith and Nephew, London, UK) and moist gauze to all her wounds.



Fig. 4. Ten months after the presentation, two 2×1 cm abdominal wound remains from the initial presentation treated with gentle soap wash and packed iodoform. All other wounds are healed.

abdominal surgery.^{3,4} Associated inflammatory diseases, such as Crohn's, ulcerative colitis, rheumatoid arthritis, lymphoproliferative disorders, and hematologic malignancies, are reported in only 10–24% of PSPG cases.^{3–5} Patients may have a history of colitis associated with chemotherapy.⁶ Family history of PG is rare in PSPG (4.1%),³ although our patient had a familial history of Crohn's disease suggesting a genetic component. Several promising candidate genes have been suggested as possible triggers of Crohn's disease, but it is accepted that the etiology is likely multifactorial.

Initial signs of PSPG are erythema, wound tenderness, and dehiscence. The average time to onset is POD10 (range, 2 days to 2 months).⁵ There are 17 previous reports of PG postautologous breast reconstruction.⁵ Clues for this challenging diagnosis in the setting of autologous breast reconstruction are as follows:

- 1. irregular, violaceous, undermined border of affected wounds^{4,5,7};
- 2. purulence limited to the skin with normal appearing fat and subcutaneous tissues^{5,8};
- bilateral, often symmetric, and the involvement of breasts (88%)^{5,9};
- 4. involvement of the abdominal donor site $(86\%)^5$;
- 5. relative sparing of the mastectomy skin,⁶ except in cases of prior irradiation⁸;
- 6. relative sparing of the umbilicus and stalk^{5,6,9}; and
- 7. sparing of the nipples.^{9,10}

Table 1. Types of Pyoderma Gangrenosum

Illcerative (classic)	Tender papule, pustule, or vesicle
	• Lower extremity trunk or site of trauma
	• Expands peripherally and ulcerates centrally
	 Ulcer demonstrates a bluish or violaceous
	quality with the undermined appearance
	 Irregular expansion of the involved tissue
	• Purulent and necrotic, with depth extending
	into the subcutaneous fat or fascia
Bullous (atypical)	 Superficial variant
	Blue–gray inflammatory bullae resulting in
	superficial ulcers
	 Present on the arms and face
	 Strongly associated with a hematologic
	disorder
Pustular	• Painful pustules with surrounding erythema
	 Arise during acute exacerbation of bowel
	disease
	 May occur in the oral mucosa
Vegetative	 Localized, solitary form
	 Presents as an indolent, mildly painful,
	ulcer or plague
	 Lacks classic undermined border or
	purulent base
	1

Among reported cases of PSPG after flap reconstruction, fever and leukocytosis are present in 44% and often appear with necrotizing wounds.^{5,9} Thirty percent of patients had delayed reconstruction, where mastectomy occurred without issue.⁵ Nineteen percent of patients had a history of breast infection before the PSPG episode, as did our patient. Thirteen percent of patients presented with superimposed infection.⁵

Even with superimposed infection, the treatment of choice is high-dose systemic steroids followed by oral prednisone for 4–6 weeks.⁵ Expedient initiation of systemic corticosteroids results in a dramatic improvement in wound healing and overall condition. Debridement, occurring in 66% of reported cases, and other surgical interventions such as skin grafting often exacerbate wound progression due to pathergy.⁹ Local wound care with nontraumatic modalities, such as the hydrogel, is recommended to minimize pathergy. Alginates are useful for highly exudative wounds. However, the recurrence of PSPG may be prevented with perioperative steroids.³ For this reason, surgical interventions should only be performed after appropriate medical therapy is initiated.

Since the first successful application of VAC for PG, there has been controversy about its use, as foam changes may impart additional trauma.⁵ Several studies have shown that VAC is a useful strategy for PG compared to topical application of petroleum jelly or wet-to-dry dressings.⁵⁻⁷ VAC was highly successful for our patient. Skin grafts often fail when used in the acute setting, with concomitant PG at the donor site, but can be successful in large after stabilization of disease.³ Skin substitute use has also been described.⁵ In addition, although fear of pathergy dissuades further surgical intervention, gentle debridement is a cornerstone in our management after PG is stabilized with steroids. Removal of fibrinous materials impeding granulation can be performed via gentle curettage without disease progression. Average time to healing for PSPG after autologous reconstruction is 4 months (range, 15 days to 1 year).⁵

CONCLUSIONS

PSPG is a diagnostic challenge with potentially devastating consequences if unrecognized. The plastic surgeon must be aware of PSPG and the clinical signs that facilitate this diagnosis, as it commonly occurs in female patients after breast and abdominal surgery.

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