Postpartum pyoderma gangrenosum following lactation-induced mastitis and abscess incision

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Key words: immunosuppression; postpartum; pyoderma gangrenosum; skin grafting; surgery.

INTRODUCTION

We present the case of a 32-year-old breastfeeding woman who, at 7 weeks postpartum, developed pyoderma gangrenosum (PG) of the right breast. We will discuss how this case is novel in terms of PG management, in that early surgical intervention resulted in positive patient outcomes.

CASE REPORT

A 32-year-old breastfeeding woman presented 7 weeks postpartum with right breast pain, swelling, and erythema. The clinical diagnosis of lactational mastitis was confirmed on ultrasound, and she was commenced on intravenous flucloxacillin. Aspirated milky fluid was cultured, which grew methicillin-sensitive *Staphylococcus aureus*; thus, clindamycin was added to her treatment regimen. Her symptoms failed to improve, and 6 days later, an abscess was incised and drained. A vacuum-assisted closure dressing was placed over the wound after surgery. Over the following 3 days, she deteriorated clinically with markedly increased pain, spiking temperatures, and malaise. Linezolid and metronidazole were added to her regimen. On exam, she had an 18-cm × 10-cm deep necrotic ulcer with purulent discharge, sparing the nipple. The breast surgeon's concern was that of necrotizing fasciitis, and an immediate wider debridement was performed, removing all breast skin except for the nipple. Dermatology was then consulted. Upon examination, a large proportion of the right breast demonstrated necrotic ulceration, with rolled, inflamed, and violaceous borders (Fig 1). A diagnosis of PG was made. Scoring systems such as a PARACELSUS score of 15 supported this diagnosis.[1] The patient was commenced on intravenous methylprednisolone 1 g daily for 3 days. Her temperature normalized rapidly, and her C-reactive protein level decreased from 141 mg/L to 27 mg/L in 24 hours. The previously rolled, violaceous wound edges settled with no further progression. After 3 days, a decision was made to introduce a steroid-sparing agent to avoid potential side effects, and cyclosporine 5 mg/kg/d was commenced as a longer-term option. The right breast continued to lactate, and the very large defect now present was anticipated to pose several difficulties for the young mother. Five weeks after the initial presentation and after multidisciplinary discussion, a biodegradable temporizing matrix (NovoSorb BTM- Polynovo Biomaterials Ltd) was applied under continued dermatologist-supervised immunosuppression (Fig 2). The NovoSorb BTM is a synthetic wound dressing to assist in dermis regrowth. It is composed of 2 layers—a sealing membrane and an open-cell, porous, biodegradable

Abbreviations used:

BTM: biodegradable temporizing matrix
IL: interleukin
PG: pyoderma gangrenosum

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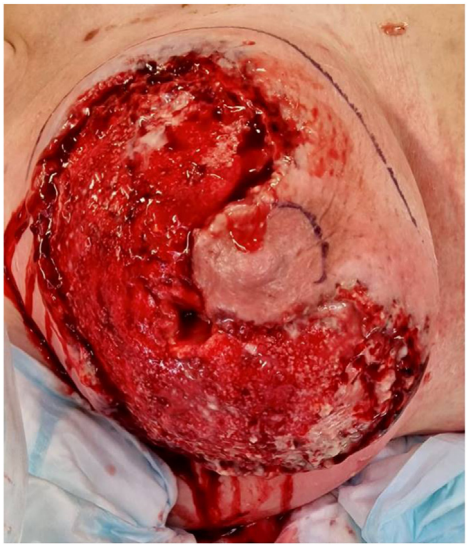

Fig 1. Active pyoderma gangrenosum of right breast.

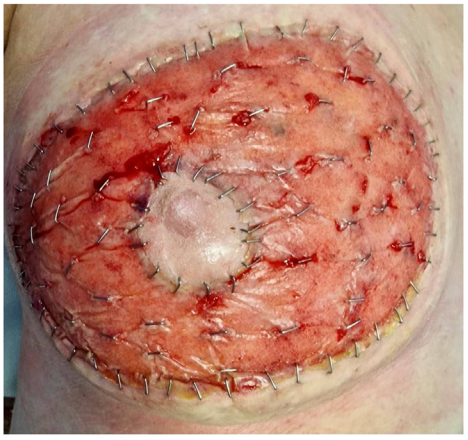

Fig 2. Pyoderma gangrenosum; right breast after matrix application.

foam—that are bound by an adhesive. The open matrix encourages cellular migration to the area with the aim of creating a vascularized dermal layer prior to grafting. The BTM was secured to the breast skin with skin clips under tension. A hole was cut in the BTM to exteriorize the nipple, and the areolar skin was everted and secured to the BTM with skin clips. The vacuum-assisted closure dressing was then reapplied. The BTM was applied with good effect. Two weeks later, a split-thickness skin graft was applied to the BTM. The skin grafting was performed successfully, with no reactivation of PG after surgery (Fig 3). Four months after grafting, the patient remains stable. Cyclosporine was weaned to stop, and the PG remained quiescent (Fig 4). A decision regarding right breast reconstruction will be considered in the coming months.

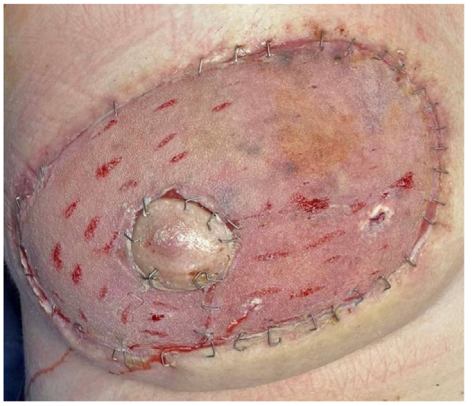

Fig 3. Pyoderma gangrenosum; right breast after skin grafting.

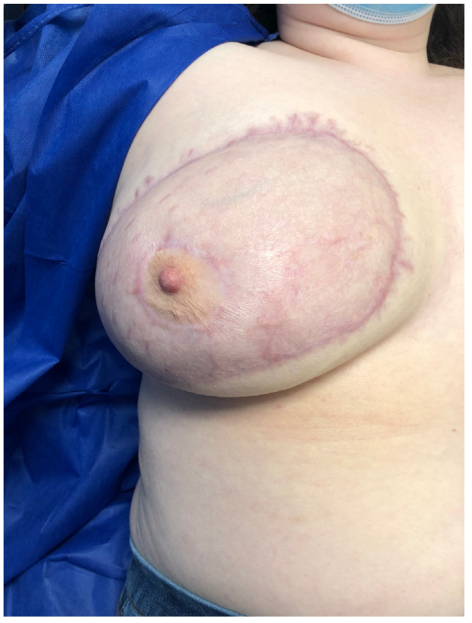

Fig 4. Pyoderma gangrenosum; current condition of the right breast.

DISCUSSION

PG presents as rapidly enlarging, painful skin ulcers with classically violaceous undermined borders and may be accompanied by fevers, general malaise, and increased inflammatory marker levels.[2] PG lesions can commonly occur in persons with underlying systemic illness, such as inflammatory bowel disease (up to 30%), hematological malignancies, and rheumatoid arthritis.[2] In our patient, the workup results for underlying conditions remain negative to date. PG is an autoinflammatory condition with involvement of the innate and adaptive immune systems, with an underlying genetic predisposition. Immune dysregulation results in abnormal neutrophil deposition and increased levels

of inflammatory mediators, including interleukin (IL) 1β, IL-8, IL-17, and tumor necrosis factor α.[2,3] A definite mechanism of association between pregnancy and PG remains unclear.[4] Pregnancy is associated with a progressive neutrophilia, which builds to a major inflammatory cascade at the initiation of labor with an upregulation of IL-6, IL-1, and tumor necrosis factor α.[4] Postpartum PG is rare and is predominantly reported at the site of cesarean section incision.[5,6] To our knowledge, PG has not previously been reported in cases of mastitis while breastfeeding is ongoing. The overall treatment of PG can be challenging. There are few standardized guidelines, and management is largely based on case reports and case series. The main goal is immunosuppression leading to ulcer clearance.[2] Ulcerated areas can have prolonged healing times despite immunosuppression.[7] Early surgical intervention in PG may risk disease reactivation.[5] However, early surgical intervention was deemed appropriate in our patient for the following reasons. The rapid systemic and local disease improvement after commencement of immunosuppression suggested that she would be a better candidate for early surgical intervention. Also, the significant ulceration that resulted from this PG coupled with the fact that the patient was a young mother with 2 small children meant that allowing the area to heal by secondary intention with immunosuppression only would have been difficult for her, exposing her to an anticipated prolonged course of cyclosporine. Finally, as PG typically heals in an atrophic, cribriform scarring pattern, the cosmetic outcome may also have been significantly less favorable without early surgical intervention. The use of a dermal matrix dressing was successful in this case. NovoSorb BTM is a novel fully synthetic matrix that is more commonly reported in burn reconstruction; however, recent retrospective analyses have shown its promise in other difficult-to-treat wound types.[8] Additionally, the efficacy and safety of skin grafting and/or negative pressure wound therapy has been reported as promising in small studies.[9,10]

Overall, this case highlights how PG should always be considered in postpartum patients presenting with rapidly progressing, nonhealing ulcers. It also shows that in certain carefully selected patients, surgical intervention, with synthetic wound matrix and grafting, can be considered alongside ongoing immunosuppression, resulting in positive patient outcomes.

Conflicts of interest

None disclosed.

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