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CASE REPORT
Breast

Pyoderma Gangrenosum after Fat Grafting in Alloplastic Breast Reconstruction: An Unusual Outcome

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Summary: Pyoderma gangrenosum (PG) is a rare and painful inflammatory skin disorder that has been recently associated with breast surgery. It is commonly mistaken for postoperative ischemia or wound infection and does not show response to antibiotics or debridement. We describe the first case of post-surgical PG (PSPG) after alloplastic breast reconstruction involving fat grafting. A 47-year-old woman underwent bilateral mastectomy and 2-stage alloplastic breast reconstruction, with fat grafting from the abdomen. Two days post-surgery, she developed bilateral erythema with tender grouped pustules that progressed rapidly into necrotic ulcerations. She did not respond to antibiotics and serial debridement. Subsequent biopsy confirmed a diagnosis of PG. She was started on steroid therapy and responded well. She was discharged on a steroid regimen, local wound care, and eventually a T-cell inhibitor. Over the next 12 months, her wounds healed without surgical intervention. PSPG has been observed in a variety of reconstructive breast surgeries, but never reported in the setting of fat grafting. As PG involves subcutaneous fat, fat grafting may accelerate and exacerbate the course of disease. Treatment for PSPG includes systemic steroid therapy or other immunomodulatory agents (or both). Surgical management remains controversial, as serial debridement and reconstruction have shown to exacerbate and stimulate disease progression. A long-term follow-up is recommended to monitor for wound healing. Delayed diagnosis of PG in breast reconstruction patients can lead to severe morbidity and disfigurement. This is first case of PSPG following fat grafting in the literature.
(Plast Reconstr Surg Glob Open 2020;8:e3223; doi: 10.1097/GOX.0000000000003223; Published online 23 November 2020.)

Pyoderma gangrenosum (PG) is a rare and painful inflammatory skin disorder described as an ulcerative wound with irregular, violaceous, raised necrotic borders. Although the etiology is unclear, it is mostly associated with systemic or autoimmune illnesses such as inflammatory bowel disease and rheumatoid arthritis. However, PG can also arise after surgery, most commonly after breast reconstruction.[1,2] Up to half of such cases are precipitated around areas of cutaneous trauma by a process called pathergy. Diagnosis of post-surgical PG (PSPG) is difficult and based mainly on clinical suspicion. Initially it may be mistaken for infection; however, infectious workups are negative, and the disease progression is refractory to antibiotics. Surgical debridement is unsuccessful and subsequent biopsies may be nonspecific, showing neutrophilic inflammation indistinguishable from other ulcerative causes. As a result, PSPG diagnosis is often delayed and becomes a diagnosis of exclusion. We present a novel case of PSPG in a breast cancer patient after a 2-stage alloplastic breast reconstruction with silicone implants and fat grafting.

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Received for publication July 27, 2020; accepted September 8, 2020.

DOI: 10.1097/GOX.0000000000003223

CASE DESCRIPTION

A 47-year-old woman with right-sided breast cancer without a history of prior PG or inflammatory bowel disease underwent bilateral mastectomy and two-stage alloplastic breast reconstruction with silicone implants and fat grafting from the abdomen to the superior poles of the breast. On post-operative day 2, she reported cyclical

Disclosure: *The authors declare no conflicts of interest or sources of funding.*

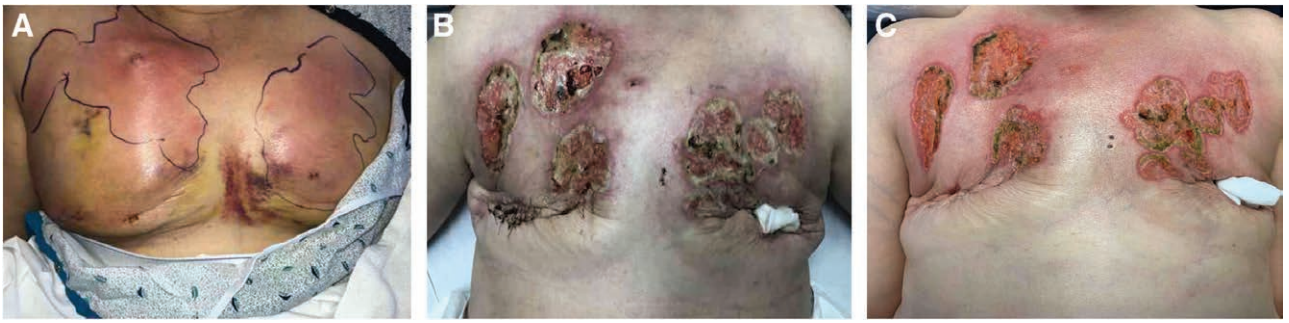

Fig. 1. Initial bilateral inflammatory changes (A) of PG in a 47-year-old woman 2 days after implantation with progression and necrosis after explantation (B), and rapid response to systemic corticosteroids after final diagnosis (C).

fevers and significant chest discomfort over the fat grafted areas. Physical examination revealed erythema superiorly on both breasts with severe pain to light palpation. She was admitted with a presumed diagnosis of cellulitis. By post-operative day 3, the erythema convalesced into tender, grouped, and draining superficial pustules (Fig. 1A). Cultures were obtained, and local wound care was initiated.

On post-operative day 4, the superficial draining pustules continued to hastily progress despite broad-spectrum antibiotic therapy with vancomycin, cefepime, and metronidazole. On examination, there were open ulcerative wound beds, with visible exposure of the left implant, which prompted urgent explantation.

Despite explantation and antibiotic therapy, the patient's wounds continued to progress (Fig. 1B). Affected tissue was sent for a histopathologic analysis, which demonstrated dense dermal neutrophilic inflammation with associated small vessel vasculitic changes consistent with pyoderma gangrenosum. She was then started on 1 g intravenous methylprednisolone daily and immediately showed significant improvement (Fig. 1C). She was discharged on 20 mg prednisone daily and 500 mg mycophenolate mofetil 500 daily. Despite significant scar formation, her wounds have continued to heal without surgical intervention over the next 12 months (Fig. 2).

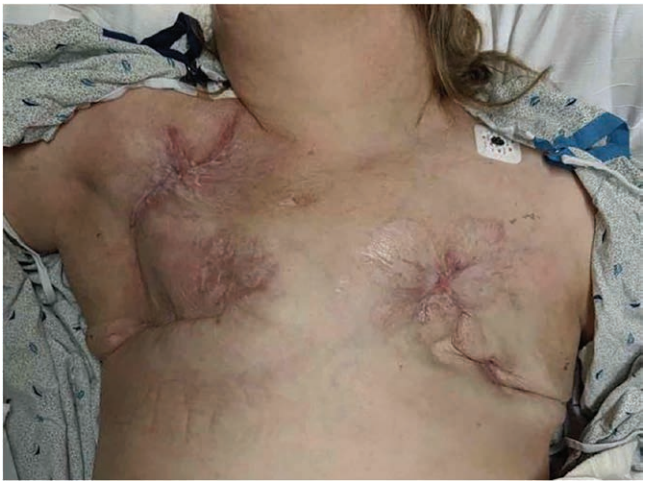

Fig. 2. Patient's recovery of PG at 6 months after initiation of steroid therapy.

DISCUSSION

Although PG was first described by Brunsting et al. in 1930,[3] only recently has the association between PG and breast surgery been acknowledged. PG can be observed in patients even without evidence of a known autoimmune or systemic disease associated with PG.[4] Furthermore, a systematic review of PSPG by Zuo et al. was unable to identify a relationship between incision size and likelihood of developing PSPG or severity of PSPG.[5]

PSPG has been observed across a variety of breast surgeries, ranging from reduction mammoplasty to deep inferior epigastric perforator flap reconstruction.[2,6] It often presents 1- to 2-weeks post-surgery,[1,5] and is often mistaken for an ischemic or infected wound. We report the first case of PSPG seen after alloplastic breast reconstruction with fat grafting from the abdomen. Autologous fat grafting for breast reconstruction has been reported to be a safe and well-tolerated technique.[7] Despite the reported safety profile of fat grafting, it is also known that PG often involves subcutaneous fat. We hypothesize that although fat grafting may not directly cause PG, the grafted fat may serve as a nidus for acceleration and exacerbation of this condition. Notably, the most significantly necrotic areas observed in our patient were areas where fat grafting was performed along the superior poles. Additionally, we find that inflammatory signs leading to PG can arise much earlier than 1 week, as our patient presented with bilateral erythema and tenderness on postoperative day 2 with significant ulceration on postoperative day 4.

There are several notable characteristics that can help differentiate PG from other differential diagnoses. PG of the breast commonly presents symmetrically after bilateral surgery, spares the area of the nipple, and does not respond to antibiotic therapy.[6] Recently, Maverakis et al. developed diagnostic criteria for PG, which consists of 1 major criteria and 8 minor criteria.[8] Our patient met the major criterion of biopsy demonstrating neutrophilic infiltrate, as well as 6 minor criteria consisting of 1) exclusion of infection, 2) pathergy, 3) history of papule, pustule, or vesicle ulcerating within four days of appearing, 4) peripheral erythema, undermining border, and tenderness at ulceration site, 5) cribriform scars at healed ulcer sites, and 6) decreased ulcer size within 1 month of initiating immunosuppressive medications. Our patient did not meet the other two minor criteria of a history of

inflammatory bowel disease or inflammatory arthritis, or multiple ulcerations with at least one on the anterior leg. Receiver operating characteristic analysis showed that 4 of 8 minor criteria had a sensitivity and specificity of 86% and 90%, respectively.

PSPG should be managed the same as conventional PG, which includes systemic steroids, or other immunosuppressive therapy such as cyclosporine or mycophenolate mofetil.[5] Intravenous immunoglobulin has also been demonstrated to be effective for long, refractory cases.[9] Surgical management for PSPG, however, remains controversial. A systematic literature review by Tuffaha et al. revealed that of the 49 reported cases of PSPG in breast surgery, 67% (n = 33) of patients underwent wound debridement.[6] Despite being well-intentioned, serial debridement can cause deep skin defects, which may require more intensive reconstructive surgery. In rare cases, groups have reported success with full thickness skin grafts for wound healing.[10] Any surgical management for PG should only be performed if the lesion is quiescent or under perioperative steroid coverage. However, there remains a concern for pathergy at donor sites especially in the setting of high doses of immunosuppressants. Long-term follow-up is important for patients who receive flaps or grafts, as late failure can occur from chronic inflammatory response.[1]

CONCLUSIONS

Delayed diagnosis of PG in breast reconstruction patients can lead to severe morbidity and disfigurement. With PSPG in the setting of fat grafting seldom reported in the literature, cognizance of the possibility and its presenting features will hopefully allow for early diagnosis and intervention.

SUMMARY

We report a 47-year-old woman who underwent bilateral total mastectomy and alloplastic breast reconstruction with silicone implants and abdominal fat grafting. Two days after grafting, the patient developed tender pustules along the superior pole of both breasts, which rapidly progressed to necrotic ulcerations, eventually diagnosed as PSPG. This is the first case reported in the literature of pyoderma gangrenosum after breast reconstruction utilizing fat grafting.

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