CASE REPORTS

A case of breast pyoderma gangrenosum after skin tag removal

E. Hayakawa*, J. Gudjonsson, T. Chang, J. Caughran

Grand Rapids Medical Education Partners, United States

Received: July 3, 2016 **Accepted:** August 16, 2016 **Online Published:** August 22, 2016
DOI: 10.5430/css.v2n4p32 **URL:** http://dx.doi.org/10.5430/css.v2n4p32

ABSTRACT

The following case report describes a 66 years old female who developed pyoderma gangrenosum (PG) on her left breast after skin tag removal. PG is an inflammatory dermatosis often confused with necrotizing infection. It is linked to breast surgery especially in those with preexisting autoimmune disorders. Knowledge of PG occurring in the breast is essential for early diagnosis and proper treatment.

Key Words: Pyoderma gangrenosum, Breast surgery, Skin/Dermatologic disease

1. INTRODUCTION

Pyoderma gangrenosum (PG) of the breast is rare and occurs in about 1 in 3 million people in the United States. The current case describes a 66 years old female who developed progressive skin ulceration after skin tag removal of the breast. Knowledge of this condition is vital in order to make an early diagnosis and minimize ineffective therapies.

2. CASE PRESENTATION

The patient is a 66 years old female who underwent an electrodessication of a single skin tag on her left breast in an outpatient dermatology office. Two weeks later, she was re-revaluated for pain and drainage from her left breast. An area was incised and drained by the dermatologist and she was placed on Bactrim. Cultures grew no organisms. Her symptoms progressed on oral antibiotics and she was admitted to the hospital three days later. At this time, she was experiencing low grade fevers, fatigue, and chills. In reviewing her past medical history, her only significant co-morbidity was hypothyroidism. Also of note, she had a negative mammogram and colonoscopy within the last year.

Breast surgery was consulted upon admission. On initial inspection, the patient's left breast had beefy red erythematous changes medially and inferiorly measuring approximately 15 cm in diameter. There was also denudement over another area approximately 6 cm with some central exudate without palpable fluctuance. Her labs showed a leukocytosis and an anemia of unknown etiology. She was initiated on broad spectrum antibiotics by infectious disease. Then silvadene with a supportive bra was applied for local wound care. Eventually, her erythema began to recede and she was discharged within 48 hours on antibiotics. All aerobic and anaerobic cultures remained negative.

Two days later, the patient presented back to the breast surgeon's office with progressive pain and drainage from her breast. She appeared diaphoretic and pale, but her vitals remained normal. Breast exam revealed a severely progressive erythema and skin loss with diffuse purulent brown drainage clinically consistent with necrotizing fasciitis. She

*Correspondence: E. Hayakawa; Email: Emiko.hayakawa@grmep.com; Address: Grand Rapids Medical Education Partners, United States.

was started on IV antibiotics and an antifungal. CT was obtained and demonstrated dermal thickening and soft tissue fat stranding without abscess.

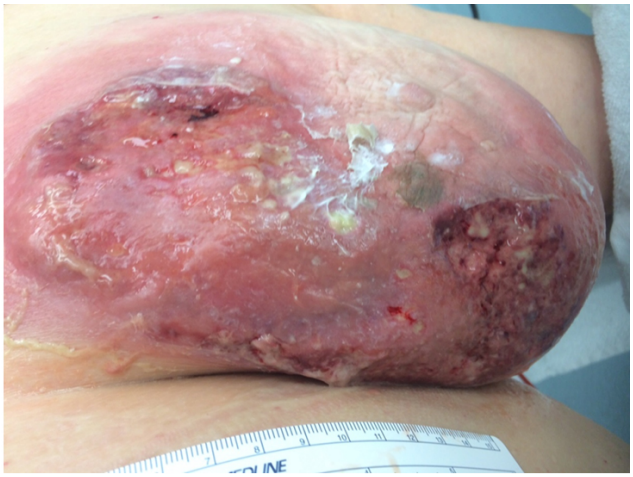

Figure 1. Left breast showing extent of PG prior to operative debridement

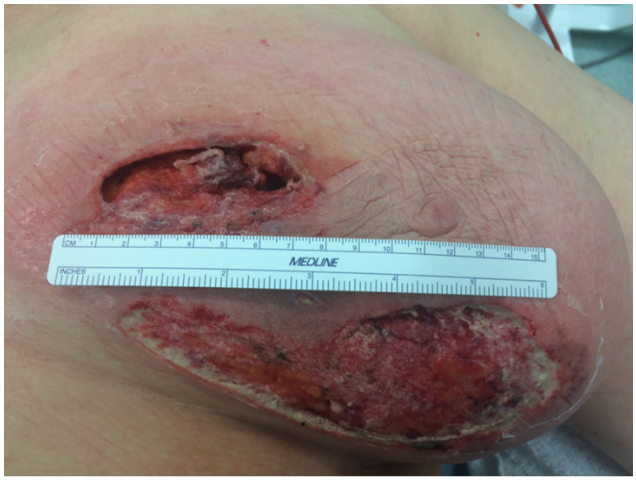

Figure 2. 48 hours after initial debridement

The patient was then taken to surgery for debridement. Operative findings revealed 20 cm of her left inferiomedial breast was affected. There were bullous changes with darkened skin edges consistent with epidermolysis and tissue necrosis with relative sparing of the breast parenchyma and nipple areolar complex (see Figure 1). All nonviable skin was debrided sharply and the wound was jet lavaged. Multiple biopsies and tissue cultures were obtained to include atypical pathogens. A wound vacuum assisted device (VAC) dressing was placed. She was taken back to the operating room two days later for VAC change which demonstrated decreased erythema and drainage (see Figure 2). Cultures from both surgeries remained negative and skin biopsy demonstrated purulent necrotizing ulceration with dense nuclear infiltrate with no signs of malignancy and negative viral stains. ANA screen, IgG, IgM, and IgA titers were all negative. However, the patient improved clinically and was sent home.

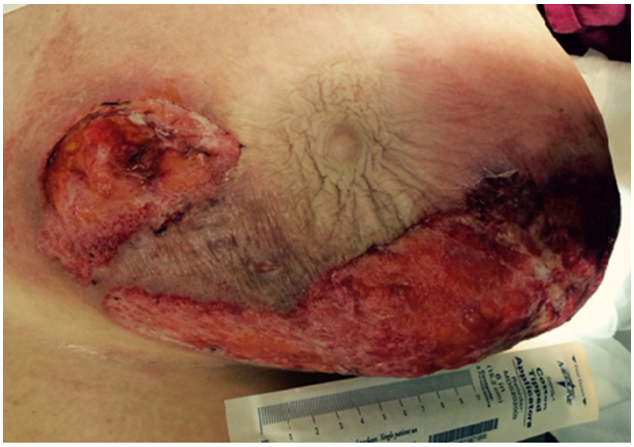

Figure 3. Left breast 7 days after debridement with progression (dusky skin edge)

The patient again returned four days later with progressive left breast pain to the office. Upon examination, she had progressive skin necrosis and was markedly tender (see Figure 3). At this point she was sent to the University of Michigan (U of M) for a second opinion. During her two week hospitalization, she was followed by a dermatologist who performed two punch biopsies that were both unremarkable. She was again restarted on broad spectrum IV antibiotics. MRI of the left breast was obtained, which showed severe skin thickening and left axillary adenopathy thought to be reactive. PG was on the differential diagnosis at the time, but felt to be unlikely given her occasional fevers, leukocytosis, and partial response to antibiotics. Her VAC dressing was at that time converted to Xeroform gauze secondary to pain. She was then discharged to a skilled nursing facility with a PICC line and continued IV antibiotics.

The patient was seen in the breast surgery office six weeks from presentation (3 weeks from initial debridement) without any local improvement (see Figure 4). At this point, it appeared that the patient's progressive skin ulceration was not infectious in nature due to repeated negative cultures and failure to improve with long term broad spectrum antibiotics. PG became the leading diagnosis based on exclusion and she was referred to Mayo Clinic for further evaluation and consideration of systemic anti-inflammatory agents given her prolonged clinical course.

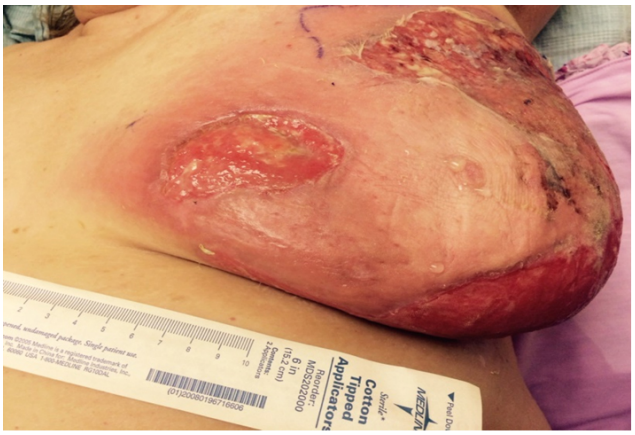

Figure 4. Appearance after returning from U of M (or 3 weeks after initial debridement)
draining vesicular/bullous changes with progression counterclockwise around nipple areolar complex

At Mayo Clinic, she completed her three week course of antibiotics. An esophagogastroduodenoscopy (EGD) was performed and duodenal biopsy did not reveal any abnormalities concerning for malignancy or autoimmune disease. A fine needle aspiration of her ipsilateral axillary node revealed inflammatory changes only. Rheumatology and hematology consultations were performed. No autoimmune or hematological diseases were found. The plan was to start oral prednisone only if the lesions worsened. She continued local Xeroform dressings and returned home (see Figure 5).

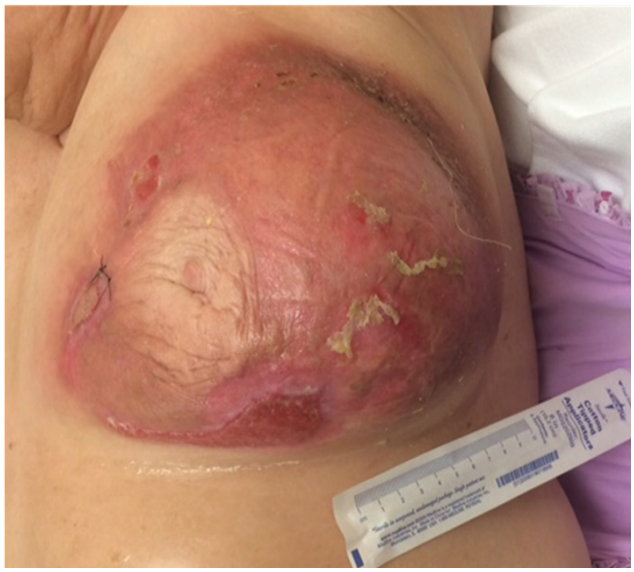

Figure 5. Left breast showing healing extent after treatment with antibiotic therapy and wound dressings

The patient then returned to see the dermatologist at the U of M following her extensive work up without much improvement. She was started on topical clobatesol 0.05% cream. After a month of treatment with the topical steroid and no additional procedures, the entire breast had healed primarily (see Figure 6).

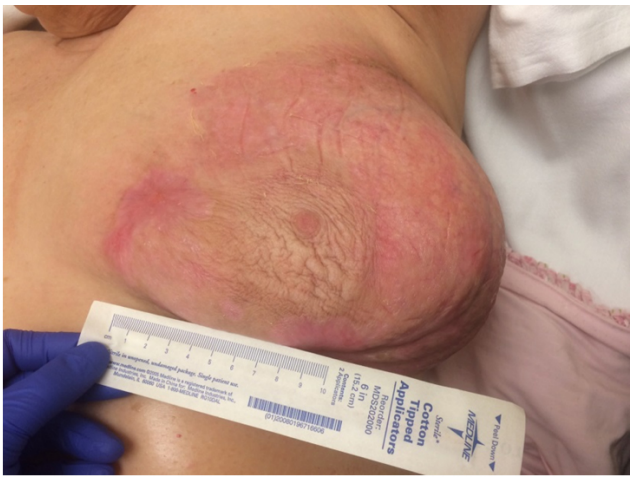

Figure 6. Left breast after topical steroid treatment

3. Discussion

PG was first mentioned in medical literature in 1930 by Brunsting et al. It is an inflammatory dermatosis of unknown origin although it has been associated with autoimmune disorders. According to various case descriptions, it starts out as pustules spreading concentrically to undermine healthy skin. It is rapidly progressive and causes painful necrolytic ulcerations. PG is a diagnosis based on exclusion and is often misdiagnosed as a necrotizing infection. The histopathology usually appears as sterile dermal neutrophilia mixed with inflammation, and lymphocytic vasculitis. PG is treated with steroids and usually has an immediate response within 48 hours. There are reports of steroids being used in intravenous, oral, and topical forms. Given the neutrophil predominate infiltrate seen on pathology; dapsone can be used in addition to steroids Treatment can also include cyclosporine A and in some cases tacrolimus.[1–14]

PG of the breast is extremely rare occurring in 3 in 1 million people in the United States. Approximately 70% of occurrences are associated with autoimmune diseases such as rheumatoid arthritis, ulcerative colitis, or hematologic diseases. The existing case reports in literature describe PG of the breast occurring after breast procedures such as reductions, reconstructions, and even biopsies. This highlights the hallmark of pathergy in the disease process. Pathergy is when trauma to tissues leads to additional necrosis and ulceration. Interestingly as the ulcerations progress, the vast majority of cases spare the nipple areolar complex. Disease onset also appears to have a latency period from the inciting event of about 6-14 days after tissue trauma. At each evaluating cen-

ter, multiple punch biopsies were obtained for analysis which may have contributed to ongoing pathergy.[3–8, 10, 13, 15, 16]

As in the present case, the patient had an initiating event with the removal of a skin tag from her breast. The disease was thought to be a necrotizing skin infection due to its appearance as well as it causing a systemic inflammatory response. It is important to note that PG can have a multisystem inflammatory reaction including leukocytosis, hypoproteinemia, and fevers as demonstrated in some case reports[3, 11] The rarity of this disease as well as the nonspecificity of its symptoms makes PG of the breast extremely difficult to diagnose. This case highlights the typical characteristics of PG of the breast including sparing of the nipple areolar complex, negative tissue biopsies, and failure to respond to antibiotics.

PG is an important condition to consider in a differential diagnosis regarding ulceration of the breast in order to avoid numerous hospitalizations, prolonged therapy, and extensive scarring.[4] Like other described cases, this patient had a delay in treatment due to her atypical clinical course with intermittent periods of improvement. Physicians should be educated about PG so that they are vigilant about post procedural care when a clinical recovery course is atypical. Patients at highest risk have inflammatory bowel disease. In some instances, PG may precede a diagnosis of systemic inflammatory disease and should lead to work up of autoimmune diseases as well as closer surveillance.[16]

4. CONCLUSIONS

PG is a rare disease of the breast making diagnosis and proper treatment extremely challenging. This disease should be included on the differential diagnosis when patients present with skin ulcerations on their breast after procedures especially those with a history of autoimmune diseases. Early recognition and treatment can minimize hospitalizations and further trauma to the breast.

CONFLICTS OF INTEREST DISCLOSURE

The authors declare they have no conflicts of interest.

REFERENCES

[1] Berry MG, Tavakkolizadeh A, Sommerlad BC. Necrotizing ulceration after breast reduction. Journal of the Royal Society of Medicine. 2003; 96(4): 186-7. PMid: 12668707. http://dx.doi.org/10.1258/jrsm.96.4.186

[2] Brunsting LA, Goeckerman WH, O'Leary PA. Pyoderma (echthyma) gangrenosumclinical and experimental observations in five cases occurring in adults. Archives of Dermatology & Syphilology. 1930; 22(22): 655-680. http://dx.doi.org/10.1001/archderm.1930.01440160053009

[3] Costa J, Monteiro D, Valença-Filipe R, et al. Pyoderma gangrenosum after breast reduction: a rare complication. Journal of Plastic Reconstructive & Aesthetic Surgery Jpras. 2013; 66(11): 336-7. PMid: 23849259. http://dx.doi.org/10.1016/j.bjps.2013.06.038

[4] Davis MDP, Alexander JL, Prawer SE. Pyoderma gangrenosum of the breasts precipitated by breast surgery. Journal of the American Academy of Dermatology. 2006; 55(2): 317-20. PMid: 16844520. http://dx.doi.org/10.1016/j.jaad.2006.02.066

[5] Dolan OM, Burrows D, Walsh M. Pyoderma gangrenosum of the breast treated with low-dose cyclosporin A. Clinical & Experimental Dermatology. 1997; 22(2): 92-5. http://dx.doi.org/10.1111/j.1365-2230.1997.tb02628.x

[6] Doren EL, Aya-Ay ML. Pyoderma gangrenosum following breast reduction: treatment with topical tacrolimus and steroids. Aesthetic Surgery Journal. 2014; 34(3): 394-9. PMid: 24448967. http://dx.doi.org/10.1177/1090820X13520448

[7] Gul U, Soylu S, CAkmak S, et al. Successful treatment of pyoderma gangrenosum with cyclosporin-A: case report. Turkiye Klinikleri Dermatoloji Dergisi. 2009.

[8] Gulyas K, Kimble FW. Atypical pyoderma gangrenosum after breast reduction. Aesthetic Plastic Surgery. 2003; 27(27): 328-31. PMid: 15058560. http://dx.doi.org/10.1007/s00266-003-3017-y

[9] Horner B, El-Muttardi N, Mercer D. Pyoderma gangrenosum complicating bilateral breast reduction. British Journal of Plastic Surgery. 2004; 57(7): 679-81. PMid: 15380703. http://dx.doi.org/10.1016/j.bjps.2004.03.004

[10] Leppard WM, Reynolds MF, Schimpf DK, et al. Pyoderma gangrenosum of the breast after bilateral simple mastectomies for ductal carcinoma in situ. American Surgeon. 2011; 77(7): 144-6.

[11] Mansur AT, Dgoktay B. Pyoderma gangrenosum on the breast: a case presentation and review of the published work. Journal of Dermatology. 2010; 37(1): 107-10. PMid: 20175832. http://dx.doi.org/10.1111/j.1346-8138.2009.00756.x

[12] Momeni A, Satterwhite T, Rd EJ. Postsurgical pyoderma gangrenosum after autologous breast reconstruction: case report and review of the literature. Annals of Plastic Surgery. 2015; 74(3): 284-8. PMid: 24557050. http://dx.doi.org/10.1097/SAP.0b013e318296b7ae

[13] Reddy R, Favreau T, Stokes T, et al. Pyoderma gangrenosum following breast reconstructive surgery: a case report of treatment with immunosuppression and adjunctive xenogeneic matrix scaffolds. Journal of Drugs in Dermatology. 2011; 10(5): 545-7. PMid: 21533303.

[14] Swinson BD, Morrison CM, Sinclair JS. Pyoderma gangrensum–a complication of breast biopsy. Ulster Medical Journal. 2002; 71(71): 66-7. PMid: 12137170.

[15] Duke G, Al SA, Husain A, et al. Pyoderma gangrenosum: a rare cause of breast ulceration. Ochsner Journal. 2012; 12(2): 155-8. PMid: 22778682.

[16] Gateley CA, Foster ME. Pyoderma gangrenosum of the breast. British Journal of Clinical Practice. 1990; 44(12): 713-4. PMid: 2102206.