



Pyoderma gangrenosum localized on the breast

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ARTICLE INFO

ABSTRACT

Article History

Received 08 / 08 / 2014

Accepted 08 / 11 / 2014

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Keywords:

Breast
Inflammatory bowel disease
Pyoderma gangrenosum
Ulcer

Breast ulceration is usually associated with breast cancer. However, it is important to know other factors that may be involved in its etiology. In this case report, we presented a very rare cause of pyoderma gangrenosum (PG) in the breast of a female patient without any prior history of breast tissue trauma or surgical intervention. More than one-half of PG cases develop in association with an underlying systemic condition including inflammatory bowel disease (IBD), hematologic disorder and arthritis. In contrast to other extraintestinal manifestations, cutaneous and ocular disorders occur at equal frequency in both Crohn's disease and ulcerative colitis. PG has been detected in 0.75% of IBD patients. It usually is related to the activity of colitis. The classic lesion begins as erythematous pustules or nodules. The patients with ulcerative PG have had an associated disease such as IBD, arthritis, monoclonal gamopathy, and internal malignancy. Lesions may be single or multiple. It can be resolved by treatment of the underlying colitis. For mild localized cases, topical corticosteroid or topical tacrolimus should be considered as the first choice. Severe cases can require systemic glucocorticoids, immunosuppressants or anti-TNF therapy. This case differs from others reported in the literature because in addition to breast, hand and foot lesions consistent with pyoderma gangrenosum were also present in this patient.

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1. Introduction

Pyoderma gangrenosum (PG) is a rare neutrophilic dermatosis which is known as an inflammatory and ulcerative disorder of the skin. It is usually related to the activity of colitis. The classic lesion begins as erythematous pustules or nodules. PG may present with bullous, vegetative, peristomal and extracutaneous lesions (Tromm et al., 2001). Lesions may be single or multiple. It can resolve with treatment of the underlying colitis. Severe cases can require systemic glucocorticoids, immunosuppressants or anti-TNF therapy. Here, we present a case of pyoderma gangrenosum with both breast and hand/foot involvement.

2. Case

A 43-year-old female patient without any history of systemic disorder presented with painful lesions on her right breast,

right thumb and left ankle which first appeared ten days ago as small, erythematous pustules (breast and ankle) and a bulla (thumb); she was subsequently hospitalized and investigated when her lesions became larger in size. Additionally, she described travelling joint pain and watery, non-bloody diarrhea three or four times per day which occurred also at nighttime. Previously, the patient had been started on prednisolone 75 mg/day ten months ago by the dermatology department for lesions that had developed on her right breast which had been pathologically confirmed to be consistent with PG; the therapy had been tapered and discontinued nine months later when lesions had regressed. Two weeks after cessation of prednisolone therapy, diarrhea had started and aforementioned skin lesions had developed one month later. She had no trauma or history of surgical intervention to the breast.



Fig. 1. Medially localized, partly ulcerated pustular lesion with yellow discharge and scar appearance from the previous ulcer on the right breast and bullous lesion on the right thumb (before treatment).

She did not have a history of systemic illness and smoking. During physical examination, a medially localized, partly ulcerated pustular lesion (6x4 cm in size) with yellow discharge and scar appearance from the previous ulcer were observed in the right breast as well as a bullous lesion on the right thumb and pustular lesions surrounded by erythema on the medial malleolar region of the left foot (Fig. 1 and 2).



Fig. 2. A pustular lesion surrounded by erythema and painful swelling on the medial malleolar region of the left ankle (before treatment).

Her left ankle was tender, painful and warm to touch. Examination of the other breast or both axillae did not reveal any pathological abnormalities. Abdominal examination results were normal. Hepatitis B surface antigen (HBsAg), hepatitis C virus antibody (Anti HCV), human immunodeficiency virus antibody (Anti HIV), antinuclear antibody (ANA), romatoid factor (RF), venereal disease research laboratory (VDRL) test, syphilis indirect hemagglutination (IHA),

p-antineutrophil cytoplasmic antibody (p-ANCA) and c-antineutrophil cytoplasmic antibody (c-ANCA) were all negative. Biochemical parameters (erythrocyte sedimentation rate: 117 mm/h; C-reactive protein: 10.8 mg/dL; white blood cells: 10.600 K/ μ L; haemoglobin: 12.40 g/dL and platelets: 394.000 K/ μ L) were in normal range except for albumin which was 2.7 mg/dL. Breast ultrasonography did not yield any pathological finding. Punch biopsies of the lesions were not pathologically specific but were compatible with the clinical diagnosis of pyoderma gangrenosum (Fig. 3). There was no growth in the cultures of the superficial layer obtained from the lesions. Colonoscopy revealed ulcerated lesions with erythematous surrounding which extended from the proximal of the sigmoid colon up to caecum with occasional normal mucosa appearance. Attempts to access to terminal ileum failed. Biopsy specimens were obtained from the adjacent site of the lesions. While pathology report established non-specific colitis, clinical diagnosis was indeterminate colitis. The patient was started on treatment with local creams, methyl prednisolone (60 mg/day), cyclosporin (3 mg/kg) and mesalazine (pentasa) (4x1 g/day). Two months later, ulcers healed, lesions crusted (Fig. 4 and 5), diarrhea resolved and joint pain improved.

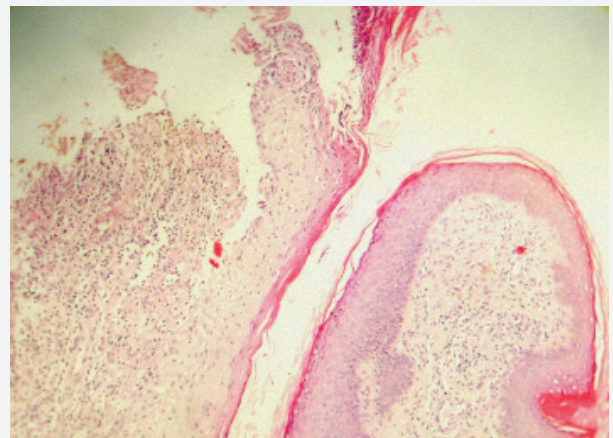


Fig. 3. Microscopic appearance of the punch biopsy material obtained from the right breast: Ulceration also involving the follicle and inflammatory granulation tissue are observed. This appearance supports the diagnosis of pyoderma gangrenosum in conjunction with the clinical features of the patient (H-E x 100).

3. Discussion

Pyoderma gangrenosum occurs rarely and its annual incidence has been estimated to be 3-10 cases per million (Ruocco et al., 2009). PG affects people in the age group of 40-60 years. It occurs more frequently among women (Ruocco et al., 2009; Binus et al., 2011). More than one-half of PG cases develop in association with an underlying systemic condition including inflammatory bowel disease (IBD), hematologic disorder and arthritis (Bennett et al., 2000). In contrast to other extraintestinal manifestations, cutaneous and ocular disorders occur at equal frequency in both Crohn's disease and ulcerative colitis. PG has been detected in 0.75% of IBD patients (Farhi et al., 2008).

Although PG is uncommon, it has serious consequences. Lesions initially occur as solitary or multiple papules or pustules. They occur mostly in the legs but may be seen in any



Fig. 4. Posttreatment healing of the right breast with visible cicatrice

part of the body. The frequency of pyoderma gangrenosum cases with breast involvement is very low (Duke et al., 2012) and the majority of them has a history of surgery or pathergy phenomenon. What differentiates the presented case from others reported in literature is the absence of such history and presence of concurrent lesions on both the breast and extremities. PG is related to the activity of IBD in 50% of the cases. For mild localized cases, topical corticosteroid or topical tacrolimus is preferred as the first choice. Also, dressings for the lesion, leg elevation and rest are recommended. Systemic treatment is indicated for patients with more generalized involvement. In this case, systemic glucocorticoids were preferred initially. Cyclosporin may or may not be added to this treatment regimen (Callen, 1998; Ahronowitz et al., 2012).

For our patient, cyclosporin was used in combination with glucocorticoid and resulted in treatment success. Infliximab has been reported to be effective for patients with both PG and Crohn's disease (Brooklyn et al., 2006).

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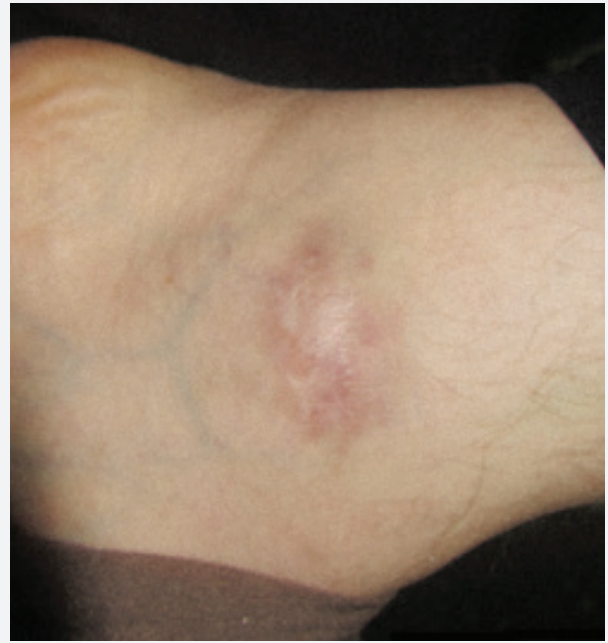


Fig. 5. Posttreatment healing of the left ankle with cicatrice.

As suggested by the post-treatment images of the lesions shown, permanent scarring and the risk of spreading underscore the need to rapidly diagnose and treat the condition. Also, in our patient, parallel occurrence of the activation of the underlying disease and development of the lesions is remarkable. Another striking feature of this case is the relapse of the condition about one month after discontinuation of steroid therapy. PG should be considered when establishing a differential diagnosis of breast ulcers. Additionally, the presented case further corroborates the need to perform a colonoscopic examination as the initial procedure in order to avoid a delay in the diagnosis when PG is detected.