CASE REPORT

DOI: 10.5336/caserep.2017-58570

Pyoderma Gangrenosum Developing After Total Colectomy Due to Ulcerative Colitis

Seda YILMAZ,[a]
Kadir GİŞİ,[b]
Bülent KANTARÇEKEN[b]

Departments of
[a]Internal Medicine,
[b]Gastroenterology,
Kahramanmaraş Sütçü İmam University
Faculty of Medicine,
Kahramanmaraş

Received: 20.10.2017
Received in revised form: 12.02.2018
Accepted: 13.02.2018
Available online: 17.09.2018

Correspondence:
Kadir GİŞİ
Kahramanmaraş Sütçü İmam University
Faculty of Medicine,
Department of Gastroenterology,
Kahramanmaraş, TURKEY
kadirgisi@gmail.com

Copyright © 2018 by Türkiye Klinikleri

ABSTRACT Pyoderma gangrenosum (PG) is a rare inflammatory cutaneous disorder starting with painfulnodules and pustules that enlarge to form cutaneous ulcers. PG, is one of the neutrophilic dermatosis. Pyoderma gangrenosum has been shown in a rate of 1.4% of ulcerative colitis (UC) and 5% of Crohn's disease (CD). Without Ulcerative colitis activity; PG has been shown together with Ulcerative colitis. The patient was admitted to clinic with swelling of left ankle and periostomal flowing lesion on his back for one week. Seven months ago, he/she was operated emergently because of free air on abdominal computerized tomography while detecting for bloody diarrhea lasting for one month. He was diagnosed as ulcerative colitis and near total colectomy was applied. His biopsy from skin was compatible with pyoderma gangrenosum. In conclusion, ulcerative colitis patients with total colectomy should be followed for extraintestinal findings (complications), too.

Keywords: Pyoderma gangrenosum; colectomy; colitis, ulcerative

Pyoderma gangrenosum (PG) is rarely seen; destructive, inflammatory skin disease characterized by painful nodules or pustules that progress to large and extended ulcerations.[1] In 50% of the patients with PG, there is an underlying disease. These include inflammatory bowel disease, arthritis, leukemia, hepatitis, primary biliary cirrhosis, intestinal malignancies and monoclonal gammopathy. In the patients with ulcerative colitis (UC), the presence of PG is independent from the activity of the disease. However, rarely, activation of UC may lead to PG development. In this article, we present an UC case with arthritis and the lesions consistent with PG around ileostomy and on his right back, which have been developed approximately 7 months after total colectomy.

CASE REPORT

34-years-old male patient who has been diagnosed with UC and who have undergone nearly total colectomy, was presented with the complaints of running lesion in his right back and swelling on his left ankle, which have been lasting for the last 1 week. Seven months ago, he was operated emergently because of free air on abdominal computerized tomography while detecting for bloody diarrhea. Computerized tomography showed normal

terminal ileum, perforation of cecum, totally necrotized right and transvers colon and partly necrosis in the left colon. Pathologic result of the patient, who had multiple colectomies, was consistent with ulcerative colitis. For the patient in whom arthritis was detected in the right ankle during the post-operative follow-up, sulphasalazine was recommended. However, patient discontinued the drugs. In his control, all rectal mucosa was hyperemic and edematous, consistently with the activation. His blood pressure was 120/80 mmHg, pulse was 80/min, fever was 39°C, his general status was moderate, he had ileostomia and abdominal midline incision scarring and he had arthritis in the left ankle (Figure 1). In his right back, pustular and occasionally ulcerated purulent lesions, the largest of which measures 3x4 cm and ulcerated lesions around ileostomia were detected (Figure 2). His laboratory investigations showed the following results: C-reactive protein: 391 mg/dl; sedimentation rate: 81 mm/hr, white blood cell: 17.4/mm^3; hemoglobin: 11.5 g/dl. Hepatitis marker, VDRL, HIV serology and immunological markers were all negative. The patient was examined by orthopedic clinic for his arthritis on the left ankle. Cutaneous and subcutaneous edema in the ankle and Achilles tendinitis were detected; drainage was performed. His cultures did not show any growth. The patient with fever was examined by the department of infectious diseases and he was initiated on antibiotic therapy. The patient was examined by the rheumatology clinic for Behçet's disease, pathergy test was performed; the result was negative. Pathological result of the skin lesion included ulcer in the epidermis, extensive infiltration of leukocytes with polymorphic nuclei in the ulcerated area, few lymphocytes and histiocytes and neutrophils in the epidermis of the surrounding tissue. Some findings consistent with pyoderma gangrenosum were detected. The patient was started 1 mg/kg systemic corticosteroid and mupirocin-containing cream for the lesions. Regression of lesion and fever during the clinical monitorization, his acute phase reactants decreased. Informed consent was obtained from the patient.

DISCUSSION

Most common extraintestinal sign of the inflammatory bowel disease is arthritis. Arthritis is present in 2-20% of the patients with UC and Crohn's disease.[2] Peripheral arthritis is more commonly seen in the patients with colon involvement and in those with severe intestinal involvement.[1] Brunsting et al., who have firstly described rapidly progressing and painful, necrotic and irregular skin ulcerations in 5 patients in 1930 and they called it as Pyoderma gangrenosum.[3] They are non-infectious ulcerated cutaneous lesions with unknown pathogenesis. There are the theories that it is a vasculitis or a cell-mediated reactive process. In its histopathology, there is sterile abscess accompanied

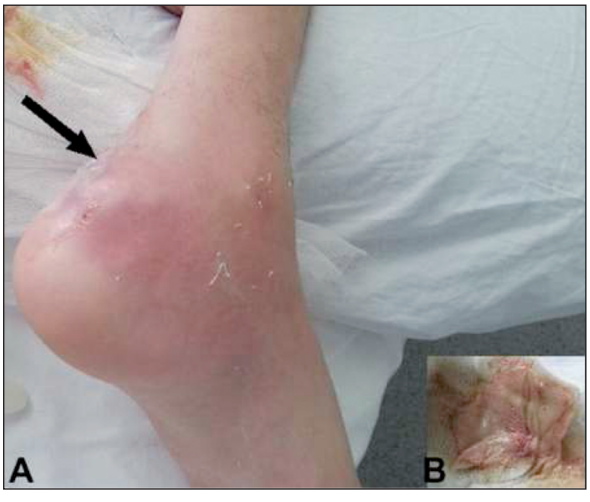

FIGURE 1: A. Arthritis in left ankle and B. Purulant material drained from the lesion.

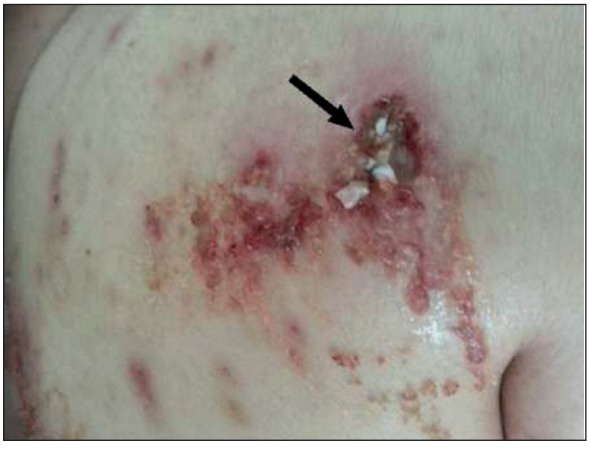

FIGURE 2: Pustuler, purulant lesion on the right scapular area of the patient.

by marked neutrophil infiltration.[4] Sterile abscess results from venous and capillary thrombosis, hemorrhage, necrosis and extensive cell infiltration. PG has four different clinical types, which include ulcerative, pustule, bullous and vegetative. Ulcerative PG appears to be an irregular, necrotic and mucopurulent ulcer and is associated with inflammatory bowel disease.[5] Classification is done by clinical and histological status, progression rate, related diseases and therapeutic modalities.[1] In our patient, the lesions were considered as ulcerative PG due to their appearance and association with UC. When PG is concomitant with ulcerative colitis, the skin lesions generally appears several months and several years after the onset of the colitis regardless of the clinical course of the disease. Skin lesions are rarely seen before the intestinal signs. They may also appear after proctocolectomy. There was a case report in which PG appearing as an extraintestinal complication of the ulcerative colitis could also occur during the activation of the colonic disease.[6] Our patient had no bloody diarrhea but the result of the biopsy obtained from rectum during his visits approximately 1 month before was consistent with activation. In the study performed by Abela et al., it was reported that PG typically occurs in the trunk and in the extremities, with a probability rate of 95%.[7] However, head and neck were less commonly defined as atypical areas. In our patient, it was detected around the back and ileostomia. In the treatment of PG, medical treatment of the underlying inflammatory bowel disease may provide regression in the skin lesions. Occasionally, the resection of the ulcerated bowel is accompanied by PG regression. However, as the response cannot be estimated, as seen in other extraintestinal signs, the treatment does not include intestinal surgery. As it is a rapidly progressing disease that, although rarely, may result in death due to sepsis and fluid loss, it requires aggressive therapy. For the treatment of inflammatory bowel disease accompanied by PG, the first therapies of choice include IV, PO and topical corticosteroids. In our patient diagnosed with UC, who underwent nearly total colectomy, skin lesions and arthritis were observed. Lesion regression was achieved using intermediate-dose systemic corticosteroid, antibiotic therapy and local therapy and he had no fever.

Consequently, the patients diagnosed with UC, who underwent total colectomy, should also be closely monitored for extraintestinal signs.

Source of Finance

During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.

Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Seda Yılmaz;* ***Design:*** *Kadir Gişi;* ***Control/Supervision:*** *Bülent Kantarçeken;* ***Literature Review:*** *Seda Yılmaz;* ***Writing the Article:*** *Seda Yılmaz.*

REFERENCES

1. Wolff K, Stingl G. Pyoderma gangrenosum. In: Fitzpatrick TB, Freedberg IM, Eisen AZ, Wolff K, Austen KF, Goldsmith LA, et al, eds. Fitzpatrick's Dermatology in General Medicine. 6th ed. New York: McGraw-Hill Book Company; 2003. p.969-76.
2. Bakelants E, van der Hilst J, Corluy L, Achten R, Gyssens I, Messiaen P. The diagnostic tangle of pyoderma gangrenosum: a case report and review of the literature. Neth J Med 2014;72(10):541-4.
3. Powell FC, Su WP, Perry HO. Pyoderma gangrenosum: classification and management. J Am Acad Dermatol 1996;34(3):395-409.
4. Brunsting LA, Underwood LJ. Pyoderma vegetans in association with chronic ulcerative colitis. Arch Derm Syphilol 1949;60(2):161-72.
5. Weismann K, Graham RM. Systemic disease and the skin. In: Champion RH, Burton JL, Burns T, Breathnach S, eds. Rook's Textbook of Dermatology. 6th ed. Oxford: Blackwell Science Limited; 1998. p.2721-3.
6. Ahmadi S, Powell FC. Pyoderma gangrenosum: uncommon presentations. Clin Dermatol 2005;23(6):612-20.
7. Tasdelen-Fisgin N, Aydin F, Tanyel E, Çandır N, Sünbül M, Bektaş A, et al. [Pyoderma gangrenosum]. Klimik Dergisi 2006;19(2):82-4.