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PYODERMA GANGRENOSUM TRIGGERED BY SURGICAL PROCEDURES IN PATIENTS WITH UNDERLYING SYSTEMIC DISEASES

Tomoko Hiraiwa[1], Hirotoshi Furukawa[2], Toshiyuki Yamamoto[1]

[1]*Department of Dermatology, Fukushima Medical University, Fukushima, Japan*
[2]*Department of Dermatology, Hoshi General Hospital, Fukushima, Japan*

Corresponding author: Dr. Tomoko Hiraiwa *tmk429313@yahoo.co.jp*

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Introduction

Pyoderma gangrenosum (PG) is a disease characterized by refractory, sterile, deep ulcers, predominantly in the extremities, occasionally in association with several systemic diseases. We herein describe three cases of PG, which were triggered by iatrogenic or surgical procedures in patients with acute myeloid leukemia, Takayasu's arteritis (TA), and ulcerative colitis, respectively.

Case Report

Case 1

A 68-year-old female was diagnosed with acute myeloid leukemia, and hospitalized for chemotherapy. A central venous catheter was inserted into her chest. A few days later, erythema appeared at the site of insertion, which spread rapidly with redness and swelling. Initial treatment with antibiotics was ineffective, and she was referred to our department. Physical examination revealed a well-circumscribed, large deep ulceration with ring-shaped, elevated edematous borders (Fig. 1), and a reddish granulated surface. Skin biopsy from the edge of the ulcer showed dense neutrophilic infiltration in the dermis as well as the epidermis. Vasculitis was not noted. Laboratory tests showed increased levels of C-reactive protein (CRP; 6.5 mg/dl), anemia, leucopenia and thrombocytopenia due to chemotherapy. Bacterial culture of the ulcer was negative. We therefore diagnosed the patient with pyoderma gangrenosum triggered by catheter insertion. She was successfully treated with oral prednisolone (30 mg/day).

Case 2

A 28-year-old Japanese man was seen in a hospital complaining of fever and diarrhea lasting 5 to 6 days. Laboratory findings showed leukocytosis in the peripheral blood (25.47×103/μl) and an increase of CRP (30.47 mg/dl). He was admitted to our hospital and treated with antibacterial agents for suspected infection, which was ineffective. A few days later, contrast enhanced CT results showed that there was a circumferential thickening of the vessel wall from the ascending aorta to the aortic arch, a part of the descending aorta, brachiocephalic artery, and left common carotid artery. He was then diagnosed with TA, and treatment with steroid pulse therapy and anticoagulant therapy with heparin was begun. During admission, he had complained of an abscess formation and painful subcutaneous induration on his left arm at the site where a drip was inserted (Fig. 2). A puncture and drainage resulted in deep ulceration. Bacterial culture was proved aseptic.

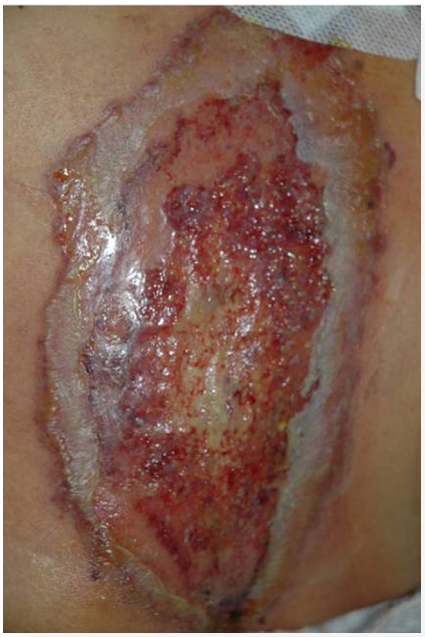

Figure 1. Pyoderma gangrenosum lesion showing large ulceration surrounded with edematous borders and erythema.

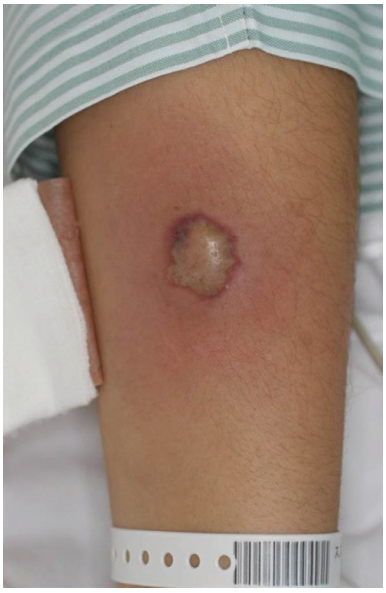

Figure 2. Large abscess formation at the site of intravenous infusion on the forearm.

Case 3

A 75-year-old male was suffering from ulcerative colitis for 2 years. He got operation for left inguinal hernia, at this time he was treated with prednisolone (15 mg per day). Three weeks later, the operation scar was ulcerated and got enlarged. Laboratory examination showed increased levels of white blood cells (34,000/μl) and CRP (14.3 mg/dl). Bacterial cultures were negative. Physical examination showed a deep ulceration covered with necrotic tissues, with erythematous borders on the left lower abdomen (Fig. 3). Histological examination revealed a number of neutrophilic infiltration in the dermis. He was successfully treated with hydrocortisone sodium succinate pulse therapy (500 mg/day for 3 days).

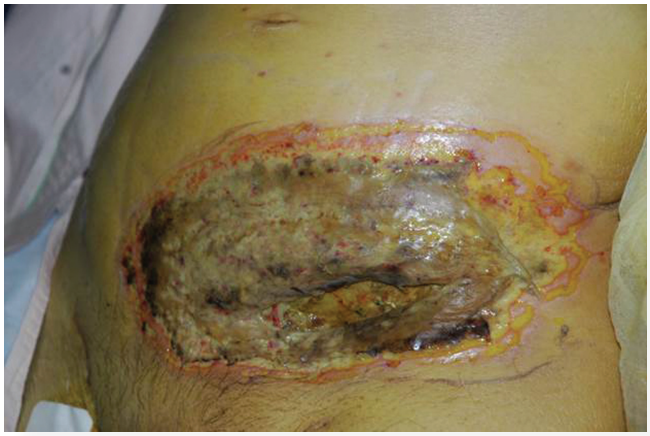

Figure 3. Deep ulceration covered with necrotic tissues bordered by edematous ring.

Discussion

There are several reports of PG occurring at percutaneous surgical sites, after procedures such as breast surgery, pacemaker implantation, splenectomy, hysterectomy, endoscopic tube insertion, cholecystectomy, appendectomy, and cesarean delivery [1]. In the present study, Case 1 was suffering from leukemia. He underwent central vein catheter insertion, which rapidly developed into PG. Similar cases have been reported which were triggered by injection of interferon [2] or tattoo placement [3] in patients with leukemia. Occasionally, PG is the initial presentation prior to the onset of leukemia, however the pathogenic link between PG and leukemia remains poorly understood. Case 2 had TA, who developed a large abscess at the infusion site of an intravenous drip in the forearm that resulted in deep ulceration after puncture. TA is characterized by stenosis or occlusion affecting mainly the aorta and its branches in young women. Several kinds of cutaneous manifestations are occasionally seen in association with TA, with representative lesions such as erythema nodosum and PG. To date, the association of PG and TA has not been frequently reported. PG occurring in patients with TA usually involves the upper limbs, followed by the scalp, face, neck, trunk, buttocks, and pubic region, in addition to the lower limbs [4]. Inflammatory cytokines, such as TNF-α, are considered to play an important role in the pathogenesis of TA. Recent studies have shown that TNF-α targeting therapies are effective for both TA [5] and PG [6], suggesting possible pathogenic similarities between these disorders. Case 3 developed PG following surgical operation of hernia. Cases of postoperative PG following hernia operation have been reported [7, 8]. Such phenomena are called pathergy, which means hyper-reactivity of the skin in response to even minor trauma. Pathergy can be seen in about 20% of cases [9]. The mechanism is still unknown, however, an aberrant immune response to minor trauma, defective cell-mediated immunity, aberrant integrin oscillations on neutrophils and abnormal neutrophil tracking, have been speculated. Because the majority of patients with PG have systemic disorders, development of PG triggered by surgical operation or iatrogenic procedures should be widely recognized.

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