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LETTER TO THE EDITOR

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Peristomal pyoderma gangrenosum in patients who underwent colectomy and colostomy for incurable inflammatory bowel disease

Dear Editor,

Peristomal pyoderma gangrenosum (PPG) is a subtype of PG and begins with painful tender pustular lesions that form fistulous tracts or ulcerations spreading outwards. The etiology of PG is not well understood, but it is considered to be an aberrant immune response characterized by histopathological neutrophilic infiltrate. Most frequently, PG is associated with rheumatoid arthritis, inflammatory bowel disease including Crohn's disease (CD) and ulcerative colitis (UC), and hematological disorders.[1–4] Antitumor necrosis factor (TNF) therapy is widely used to treat inflammatory bowel disease, rheumatologic disease, and even psoriasis.

In case 1, a 30-year-old woman had been diagnosed with CD, which is a potentially devastating condition difficult to manage. She had undergone hemicolectomy and colostomy prior to visiting us. After surgery, ulcerations with hypergranulation appeared around the stoma and rapidly spread (Figure 1A). We diagnosed the condition with PPG associated with CD. She was inadequately managed by oral administration of corticosteroids and cyclosporine. As her condition gradually worsened, she was subsequently administered a supplementary 40 mg adalimumab every other week. Within several weeks of admission to our hospital, the cutaneous lesions rapidly improved, and the ulcerations diminished in size. Within three months of starting supplementary adalimumab treatment, there was a dramatic improvement in PPG (Figure 1B). During the 12 month follow-up period after she started adalimumab, there were no reports of symptom recurrence or any other adverse effects.

In case 2, a 60-year-old woman had a history of severe UC. She underwent total colectomy and ileal colostomy due to the difficulty in managing the CD. After the surgery, ulceration with redness and swelling appeared around the ileostomy site and spread rapidly (Figure 1C). We diagnosed her with PPG associated with UC. Although her PPG was being treated with oral corticosteroids and cyclosporine, the peristomal ulcerations worsened slightly. She was subsequently administered 40 mg adalimumab every other week which resulted in marked improvement in PPG (Figure 1D). PPG did not flare up again, and no side effects were observed.

Peristomal pyoderma gangrenosum is characterized by painful ulcerations with hypergranulation occurring in the area surrounding an abdominal stoma. We reported two cases of PPG in Japanese patients who underwent colectomy and colostomy for incurable inflammatory bowel disease. In a previous larger clinical study, only 12 patients out of 1295 consecutive patients who demonstrated stoma formation had associated PPG. Zuo *et al.*[5] mentioned that approximately 25% of cases are caused by incidental or iatrogenic trauma, with 14% of cases of postsurgical PPG precipitated by abdominal surgery. We suggest that surgical damage could be a trigger to induce PPG under aberrant immune conditions such as incurable inflammatory bowel disease.

In the present cases, adalimumab was effective in treating PPG with complete healing and no recurrence. Adalimumab is the first recombinant human IgG1 antibody that binds specifically to TNFα and blocks its interaction with TNF receptors on the cell surface. Adalimumab resulted in marked improvement in PPG, suggesting a key role of some immune response including TNFα in the pathogenesis of PPG.

CONFLICTS OF INTEREST

The authors declare no conflict of interest.

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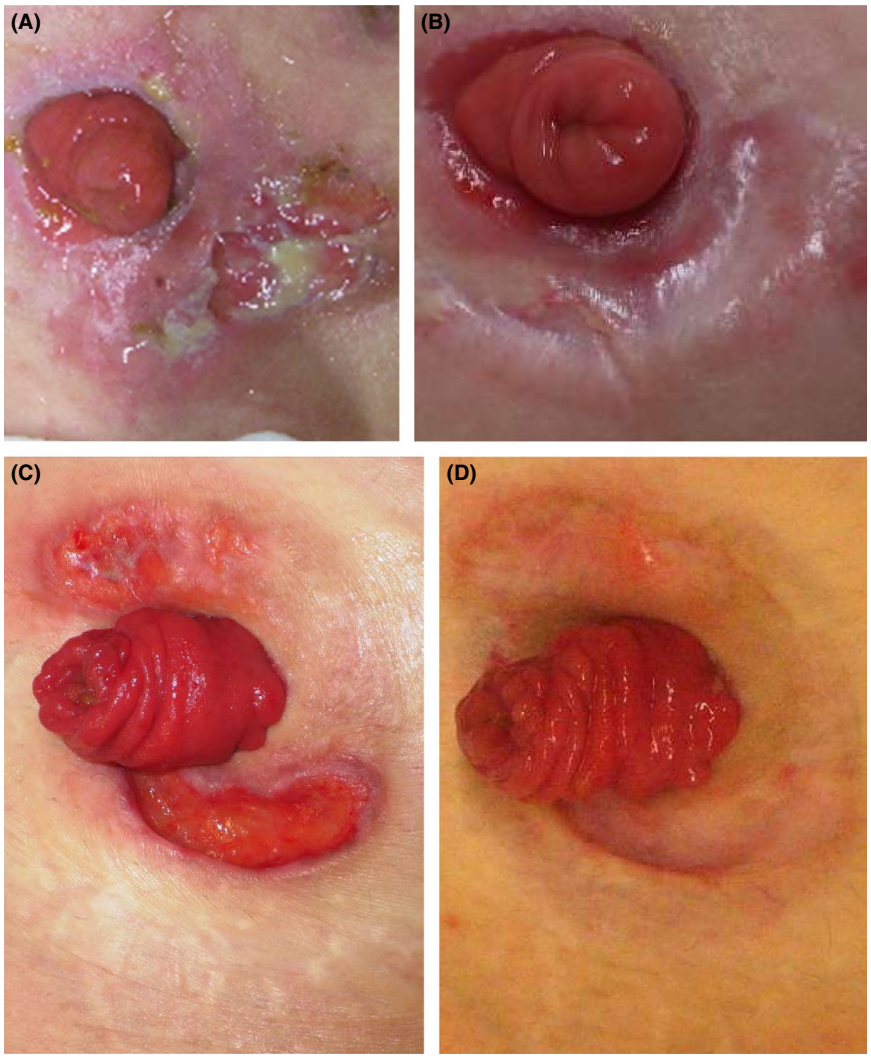

FIGURE 1 A, Case 1. Physical examination revealed ulcerations with granulation tissue, elevated borders, and perilesional erythema. B, Case 1. The patient was treated with adalimumab and PPG improved. C, Case 2. Physical examination revealed a well-circumscribed ulceration with ring-shaped, elevated, edematous borders and a reddish, granulated surface. D, Case 2. PPG successfully treated with adalimumab treatment

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