Case Report

Report of a Rare Case of Nasal Mucosa Pyoderma Vegetans in a Patient with Ulcerative Colitis

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ABSTRACT

Some dermatologic manifestations are common in ulcerative colitis (UC). Herein, we present a 36-year-old woman with ulcerative colitis and uncommon nasal mucosa pyoderma vegetans. The patient presented to our hospital with symptoms of active colitis and a concomitant 3×4×5 cm dermato-mucosal lesion in her left nasal lumen. After surgery of the mucosal lesion, the treatment for her active colitis was initiated with intravenous infliximab and oral asacol. After a 1-year follow-up, no sign of recurrence favoring mucosal lesion was noted and symptoms of ulcerative colitis were managed properly.

KEYWORDS
Ulcerative colitis; Pyoderma vegetans; Infliximab

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INTRODUCTION

Dermatological manifestations are common in inflammatory bowel diseases (IBD). Patients with IBD should be periodically examined regarding their dermatologic conditions.[1-3] In a previous report from Iran, the prevalence of dermatological manifestations was reported to be 5.9% in patients with IBD with a higher rate in Crohn's disease (7.29%) compared with patients with ulcerative colitis (4.07%). They were more common in women (52%) than in men (48%).[2] One of the rare dermatological manifestations in patients with IBD is pyoderma gangrenosum.[1-8] which is more common in patients with ulcerative colitis.[2] Since pyoderma gangrenosum is a rare occurrence, its explicit prevalence is unknown, but generally it has been estimated to occur in 3-10 million patients annually.[4] In Iran, the prevalence of pyoderma gangrenosum in patients with ulcerative colitis has been reported to be 1.4%.[2] The diagnosis of pyoderma gangrenosum is based on physical examination, and examining the lesions regarding its type, number, size, and location as well as associated symptoms of ulcerative colitis.[4] Sometimes pyoderma gangrenosum is very painful for the patient.[5] Most pustular lesions in patients with IBD should be considered as pyoderma gangrenosum variants and be treated accordingly.[7] Even though histopathologic examination is not deemed diagnostic for pyoderma gangrenosum, skin biopsy should be performed to rule out other conditions simulating pyoderma gangrenosum.[4,8] Pyoderma vegetans is a sign of IBD,[6] but rarely occurs in Iranian patients.[2] In case of facing

pyoderma vegetans in a patient without significant medical history, his/her digestive tract should be examined thoroughly to rule out IBD.[6] The mean age for the onset of dermatological manifestations in Iranian patients with IBD is 31 years.[2]

CASE REPORT

The patient is a known case of ulcerative colitis limited to her left colon for 15 years under irregular treatments. She was receiving unknown herbal medications for a long time and discontinued her standard treatments in the past three years. She had intermittently developed skin lesions diagnosed as pyoderma gangrenosum in her shoulder, thigh, and genital areas during the past 4 years (Figure 1). She applied herbal medications for the skin lesions with inconsequential benefits.

Six months before admission to our center, she developed a small mucosal lesion in her nose. She was referred to Shariati Hospital Gastrointestinal Clinic because of the rapid growth of the lesions and unresponsiveness to local treatments used by her dermatologist. She reported defecation with mild bleeding three times daily. She had no fever or abdominal pain. On physical examination, her vital signs were within the normal range. The only remarkable findings were old skin scars on her extremities and genital area and a lobular dermatomucosal lesion measured 3×4×5 cm in her left nasal lumen (Figure 2).

Laboratory assays showed white blood count (WBC)=6,300/mm^3, hemoglobin=10.6gr/dlit, platelet count=405,000/mm^3, ESR=61 mm 1st hr, CRP=37mg/L with normal stool smear and culture. During the preceding 6 months, the dermatologist who referred the patient had injected corticosteroids into the lesion several times. Oral cyclosporine followed by dapsone was administered by the dermatologist with no significant effect on her nasal lesion. Since the lesion was large and patient complained of respiratory difficulty, surgical intervention was done and the lesion was excised completely. Histopathological examination was indicative of pyoderma vegetans (Figure 3).

Meanwhile, colonoscopy was done which confirmed pancolitis on both macroscopic and microscopic views. No evidence was detected in favor of cytomegalovirus or clostridium difficile on laboratory or pathological examination. After initial evaluation and ruling out latent infections such as tuberculosis, intravenous infliximab (300 mg) was started on weeks 0, 2, and 6 and then every 8 weeks. Simultaneously oral asacol (4.8 grams per day) was started and she underwent regular monitoring every 8 weeks until week 48 of injection. After 2 months, the clinical symptoms improved significantly and in a 1-year follow-up no recurrence of the nasal lesion was found. The macroscopic and microscopic signs of ulcerative colitis were also improved.

DISCUSSION

In case of simultaneous pyoderma gangrenosum and ulcerative colitis, treatments are delivered directly targeting the skin lesions as well as systemic agents to control ulcerative colitis.[1,4,7,8] Topical therapeutics include highly potent steroids, calcineurin inhibitors (tacrolimus, pimecrolimus), human platelet releasing growth factor, and intralesional injection of cyclosporine.[4] Sometimes, antibiotics are required to control allied skin infection.[6,8] Systemic treatment for pyoderma gangrenosum involves cyclosporine (3-5 mg/kg/day) which is effective for extensive pyoderma gangrenosum and is usually administered with steroids. In most cases, cyclosporine could be applied instead of steroids. Dapsone (maximum dose of 200 mg/day) is effective in mild cases. Thalidomide in conjunction with steroids has also been advocated. When the patient does not respond to routine treatments, infliximab which is a chimeric anti-tumor necrosis factor alpha antibody is an appropriate option. However, it is mandatory that the patient be evaluated regarding latent/active infections such as tuberculosis and be monitored during treatment with infliximab in terms of appearance of infective signs.[4] Despite various treatments reported in the literature, an exact treatment guideline does not exist for pyoderma gangrenosum.[6] In the presented patient, in view of the presence of active colitis (evident in colonoscopy) and mucosal lesion of pyoderma vegetans in

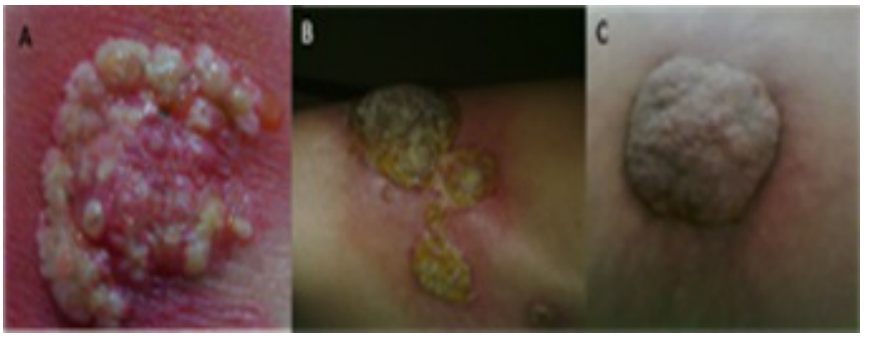

Fig. 1: **(A) Pyoderma gangrenosum on the right thigh, (B) Pyoderma gangrenosum on the shoulder, (C) Pyoderma gangrenosum on the genital area**

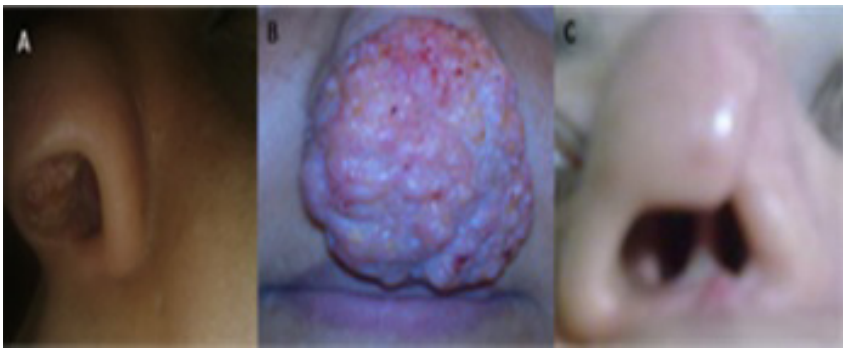

Fig. 2: **(A) The dermato-mucosal lesion in her nose in the first month, (B) The dermato-mucosal nasal lesion in month 6, (C) Scar of the skin lesion after surgery**

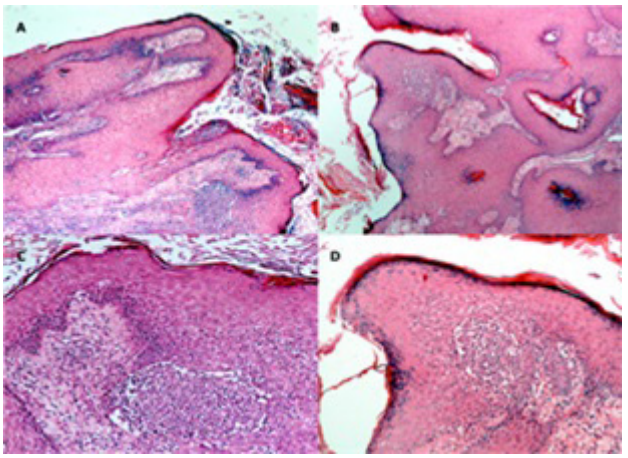

Fig. 3: **(A, B) Microscopic features of the verrucous plaques are acanthotis and papillomatous hyperplasia of epidermis along with multiple micro-abscesses (pustules) in dermis (H&E stain, original magnification 60X), (C,D) Higher power view of the lesion shows acanthotic papillomatous epidermis and subepidermal micro-abscess (pustule) (H&E stain, original magnification 150X).**

her nose associated with other dermatologic manifestations of pyoderma gangrenosum, intravenous infliximab (300 mg, 5 mg/kg) with oral asacol (4.8 gr per day) was initiated. After 2 months, complete resolution of symptoms as well as improvement in macroscopic and microscopic views in colonoscopy was observed. In a one-year follow-up, no signs in favor of recurrence of nasal lesion were noted. It seems that infliximab was effective not only for her ulcerative colitis signs, but also on extra-intestinal manifestation (i.e., pyoderma vegetans).

CONFLICT OF INTEREST

The authors declare no conflict of interest related to this work.

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