Pyodermatitis vegetans responding to rifampicin and clindamycin

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INTRODUCTION

Pyodermatitis vegetans, or pyoderma vegetans (PDV), is a rare inflammatory cutaneous disease characterized by well-circumscribed, exudative vegetating plaques with elevated margins affecting the face, scalp, intertriginous regions, and, less often, the trunk and distal extremities.[1-3] PDV is sometimes present in association with oral lesions termed, *pyostomatitis vegetans* (PSV).[2,3] PDV is known to be correlated with gastrointestinal disorders, particularly inflammatory bowel disease (IBD).[4-6] Other diseases reported in association with PDV include T-cell lymphoma, chronic myeloid leukemia, liver diseases, malnutrition, HIV infection, and primary immunodeficiency disorders.[5] Multiple topical and systemic medications have been described to treat PDV with varying efficacies.[6] We report on a case of PDV in a patient with Crohn's disease treated with a combination of rifampicin and clindamycin with good results.

CASE REPORT

A 35-year-old man known to have Crohn's disease for 13 years presented to the dermatology clinic with purulent skin lesions that had started 2 months previously. The patient had undergone a total colectomy 3 years earlier due to his recalcitrant Crohn's disease that was resistant to multiple therapies, including infliximab and adalimumab. He was not taking any medications at the time of presentation.

Skin examination found multiple well-defined erythematous and vegetative plaques with dark brown, raised borders and overlying yellow crusts along with multiple hypopigmented scars over the trunk (Fig 1), axillae, inguinal folds, and lower limbs. No mucosal, scalp, or nail lesions were observed.

Abbreviations used:

HS:	hidradenitis suppurativa
IBD:	inflammatory bowel disease
PDV:	pyodermatitis vegetans
PSV:	pyostomatitis vegetans

Histopathologic studies found extensive neutrophilic infiltrate in the stratum corneum. The epidermis showed psoriasiform hyperplasia with scattered neutrophils. The papillary dermis exhibited edema and dilated blood vessels. The reticular dermis showed infiltration of a mix of inflammatory cells, including many neutrophils, scattered lymphocytes, and eosinophils, over a background of fibrosis and granulation tissue (Fig 2). Direct immunofluorescence studies were negative. Bacterial culture found a growth of *Staphylococcus aureus*. Fungal and mycobacterial cultures were negative. Results of basic blood tests including complete blood count, hemoglobin level, liver function, and renal profile were unremarkable.

Our initial impression was an atypical hidradenitis suppurativa (HS) or PDV. Therefore, the patient was treated with a combination of clindamycin and rifampicin, at a dose of 300 mg, twice a day each. Three weeks into the treatment course, the diagnosis of PDV was supported by the histopathology results, which showed neutrophilic infiltration and areas of granulation tissue in the dermis. However, the patient was already showing a good response to the combined antibiotic treatment. After completing 6 weeks of clindamycin and rifampicin treatment, all lesions had regressed and were completely asymptomatic (Fig 3). The patient remained disease free for the next 3 months. However, the lesions

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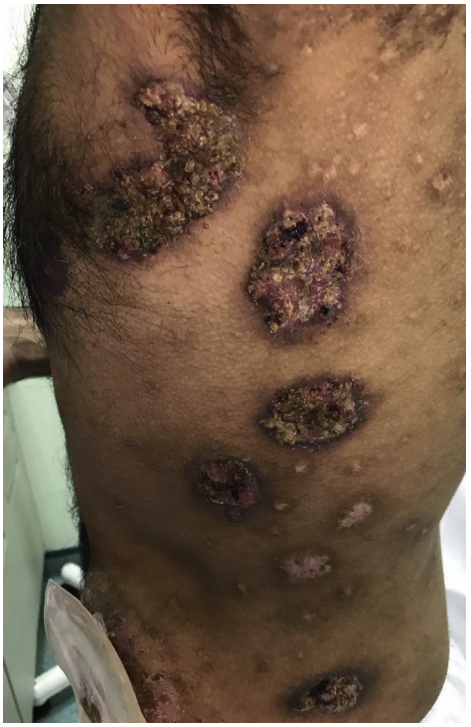

Fig 1. PDV. Vegetating plaques over the left trunk.

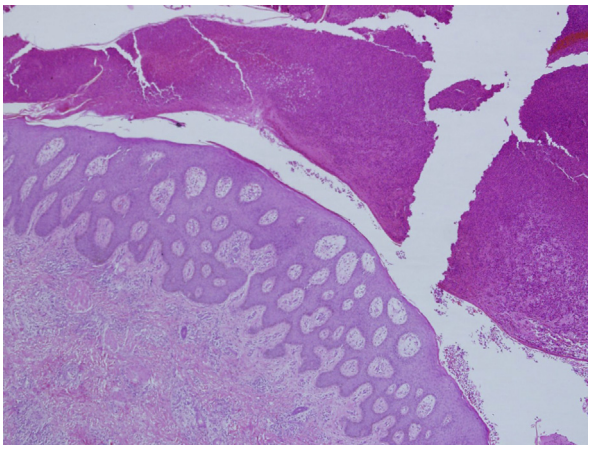

Fig 2. Histopathologic examination of the vegetative plaque shows neutrophilic infiltration in the stratum corneum, psoriasiform hyperplasia with neutrophils in the epidermis, papillary dermis edema, and fibrosis with mixed inflammatory cell infiltrate in the reticular dermis. (Hematoxylin-eosin stain; original magnification: ×40.)

recurred 4 months later (Fig 4), and a treatment with 0.5 mg/kg of prednisone daily rapidly controlled the disease. The patient is currently in a clinical remission on a daily dose of 10 mg of prednisone.

DISCUSSION

PDV is a rare inflammatory neutrophilic dermatosis that was initially described in 1898 by Hallopeau.[7] Previous reports have indicated that PDV is frequently associated with IBD in 70% of cases, and up to 100% in a recent cohort study.[1-4,6,8] PDV commonly affects adults in the third decade of life with a male/female ratio of 3:1.[3,9] The etiology of PDV is still unknown. However, variable immunologic and microbial factors have been proposed.[8]

Cutaneous lesions are characterized by erythematous vesiculopustules that coalesce to form exudative vegetating plaques commonly involving the scalp, axilla, and groin.[6,10] Other areas reported to

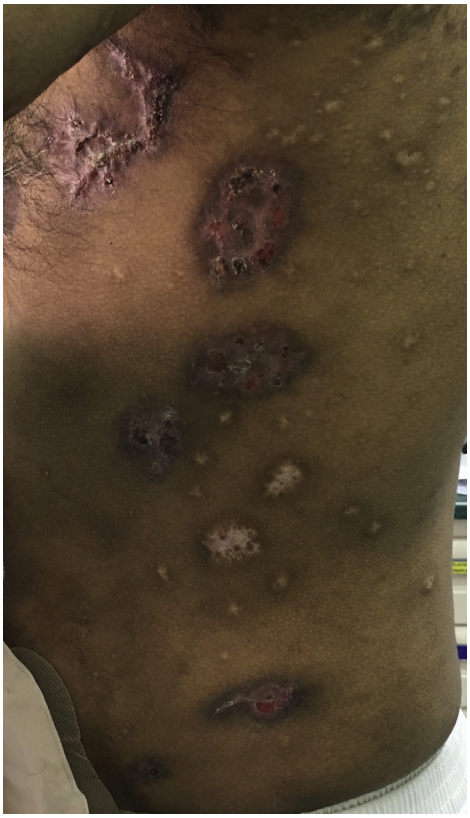

Fig 3. PDV. Resolution of the lesions after 6 weeks of treatment with oral clindamycin and rifampicin.

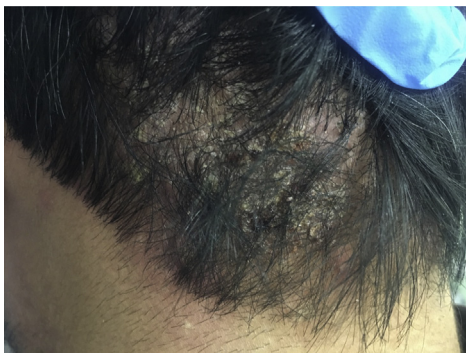

Fig 4. PDV. Recurrence of the lesions over the scalp after 4 months off treatment.

be affected include the face, trunk, and extremities.[1-3] HS has also been reported in association with IBD.[11] In our case, the initial observation of the lesions in intertriginous folds raised the possibility of atypical HS. However, the histologic findings were not supportive of this diagnosis.

The histopathologic features of PDV include epidermal hyperplasia containing large amounts of neutrophils and eosinophils. Acanthosis, acantholysis, and suprabasilar clefts can also be observed with PDV. In the dermis, dense inflammatory infiltrates with various inflammatory cells, including eosinophils, neutrophils, lymphocytes, and plasmocytes, are observed. Immunofluorescence studies usually show negative results.[3,12] Fungal, viral, bacterial, and mycobacterial cultures are typically negative, but a secondary bacterial infection with at least a single pathogenic bacteria, such as *S aureus*, β-hemolytic streptococci, *Pseudomonas aeruginosa,* or *Escherichia coli* anaerobes, is usually identified.[1] In our case, the observed growth of *S aureus* is most likely caused by a secondary bacterial infection.

Multiple previous studies have considered PDV as a clinical marker for IBD activity and recommended that a total/subtotal colectomy is a therapeutic option for complete remission of symptoms.[3,6,13] However, in 2015, Uzunçakmak et al[9] reported on a case of PDV developed shortly after a total colectomy and discontinuation of systemic corticosteroid.[9] Here we report a first presentation of PDV 3 years after total colectomy in a patient with Crohn's disease.

Previous studies have demonstrated that treating PDV is primarily accomplished by controlling the underlying IBD. To date, there is no standardized treatment for PDV. A variety of pharmacologic therapies have been suggested. Topical or systemic corticosteroids were recommended as a first-line therapy. Other reported therapeutic agents include dapsone, sulfasalazine, tacrolimus, azathioprine, cyclosporine, isotretinoin, infliximab, and methotrexate.[13] For patients with positive bacterial cultures, appropriate antibiotics were added as a conjunctive therapy. In spite of the morphologic similarities of PDV to HS, the combined antibiotic therapy of rifampicin and clindamycin is not a treatment that is suggested for PDV. Here we report a good response of PDV to a combination of oral clindamycin and rifampicin.

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