Pyoderma gangrenosum with an underlying ulcerative colitis associated with bone tuberculosis

Milind A. Patvekar, Neha C. Virmani

Department of Dermatology, Dr. D.Y. Patil Medical College and Hospital, Pimpri, Pune, India

ABSTRACT

Pyoderma gangrenosum is a rare noninfective neutrophilic dermatosis, characterized by progressive painful ulceration. It is frequently associated with systemic disorders like inflammatory bowel disease, rheumatoid arthritis and myeloproliferative diseases. However, its association with infectious diseases in particular with tuberculosis is extremely rare. Diagnosis is based on the history of an underlying disease, a typical clinical presentation, histopathology and exclusion of other diseases leading to ulcerations of similar appearance. Immunosuppression with corticosteroids remains the mainstay of treatment. We report a case of a 49-year-old male with long-standing ulcerative colitis, associated with tuberculosis of hip, who presented with nonhealing ulcers over the lower extremity.

Key words: Pyoderma gangrenosum, tuberculosis, ulcerative colitis

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Address for correspondence:
Dr. Milind Patvekar, Queens Tower, Flat B-204, Udyog Nagar, Opp. Lokmanya Hospital, Chinchwad, Pune – 411 033, Maharashtra, India.
E-mail: drmilind1212@rediffmail.com

INTRODUCTION

Pyoderma gangrenosum (PG) is a rare noninfectious neutrophilic dermatosis with distinct clinical features. Often associated with other underlying systemic diseases, it presents with rapidly progressive painful ulceration with undermined edges and violaceous border.[1-3] Extracutaneous involvement is reported to occur in bone and lungs.[2] These lesions usually require aggressive therapy in the form of immunosuppression and they heal with a characteristic cribriform scar. We report a case of PG in a patient of long-standing ulcerative colitis, associated with tuberculosis of the hip joint.

CASE REPORT

A 49-year-old male patient, a known case of ulcerative colitis on prophylactic sulfasalazine since 10 years, presented to us with two nontraumatic, painful, expanding ulcers; one each over anterior and anterolateral aspect of the left leg since 2 months. These lesions started as small follicular pustules that evolved into ulcers involving most of the pretibial region. Two months preceding the onset of these lesions, he also had an ongoing history of pain in the right hip which worsened on exertion.

Two years back he had similar lesions, one over the dorsal aspect of right feet and two over the back that resolved with cribriform scarring. During this period he had severe left-sided hip pain with complaints of burning micturition and increased urinary frequency, and was diagnosed with disseminated tuberculosis as documented in the MRI and urinary PCR reports. He was, thereby, put on antituberculous therapy (ATT), DOTS Category 1 which he abruptly stopped after 2 months of intensive phase. His physical condition was uneventful, until the preceding 2 months of his presentation to us.

Examination revealed two ulcers of sizes 10 x 8 cm and 3 x 2 cm, each with a purulent base and violaceous, undermined edges over the anterior and anterolateral aspect of left lower leg [Figures 1 and 2]. Investigations showed an elevated ESR, with a negative Mantoux test. Owing to his past history and present complaint, magnetic resonance imaging of bilateral hip was done that revealed altered signal around the left sacroiliac joint [Figure 3] and a small collection adjacent to right ischial tuberosity with altered signal within [Figure 4], suggestive of an infective etiology. Urinary PCR was negative for *Mycobacterium tuberculosis*. X-ray lumbosacral spine appeared normal and test for HLA B27 was

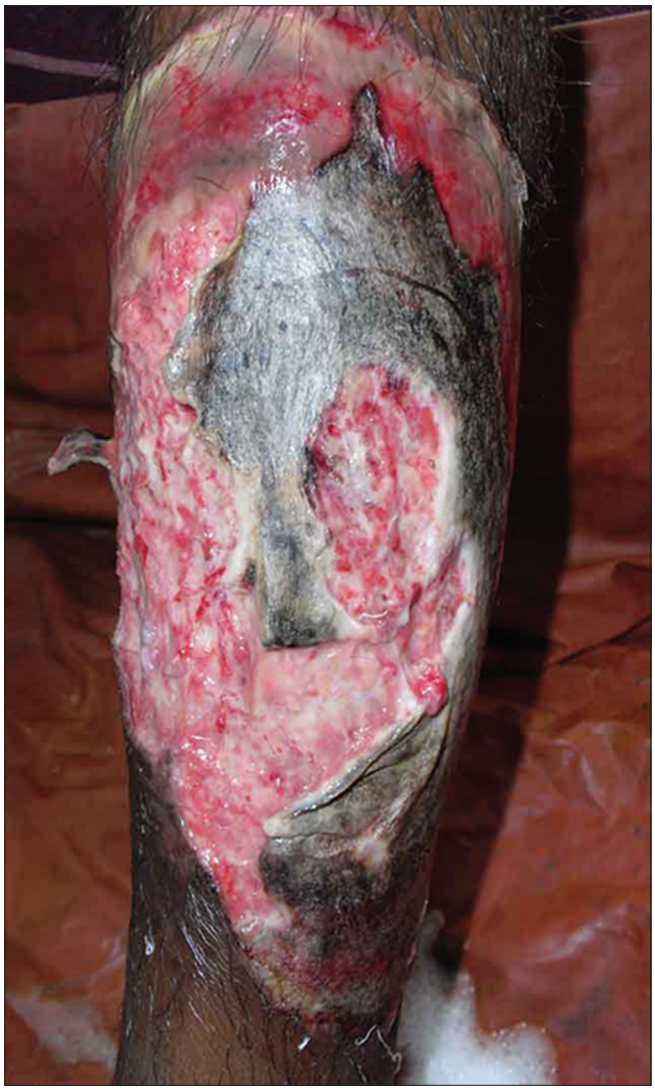

Figure 1: Ulcer measuring 10x8 cm with purulent exudate and undermined edge with overlying necrotic skin over left pretibial region

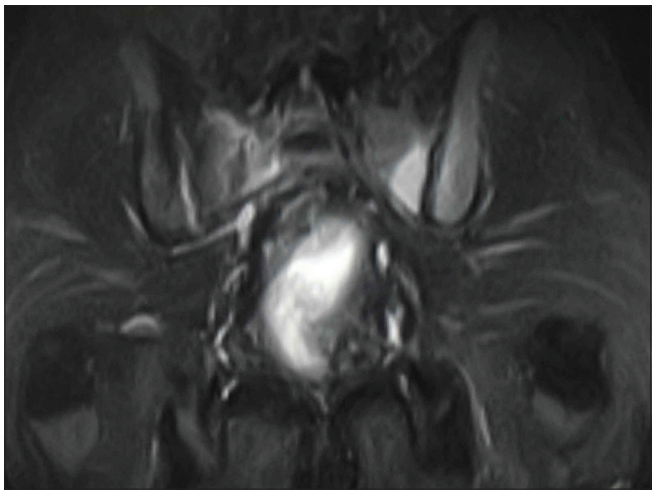

Figure 3: MRI showing altered signal around left sacroiliac joint

negative. Cytological examination of the biopsy taken from the edge of the ulcer revealed normal epidermis with dermis

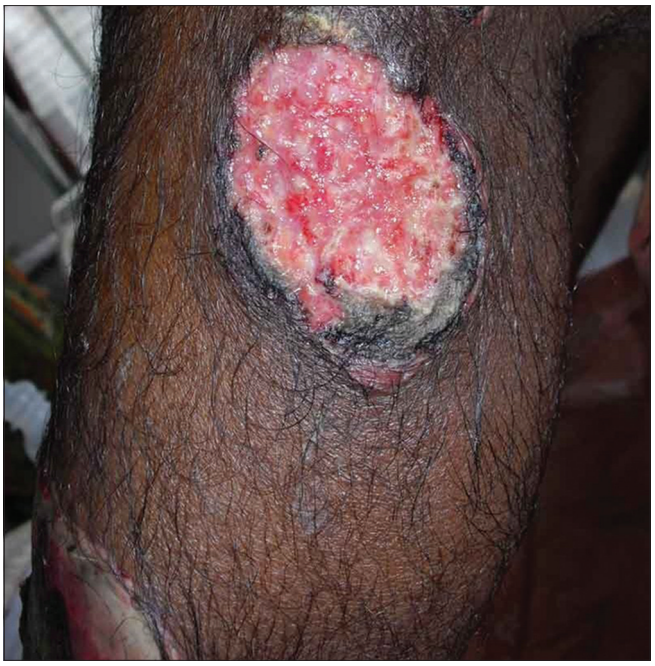

Figure 2: Ulcer showing hypergranulation tissue with ragged edge over anterolateral aspect of left lower leg

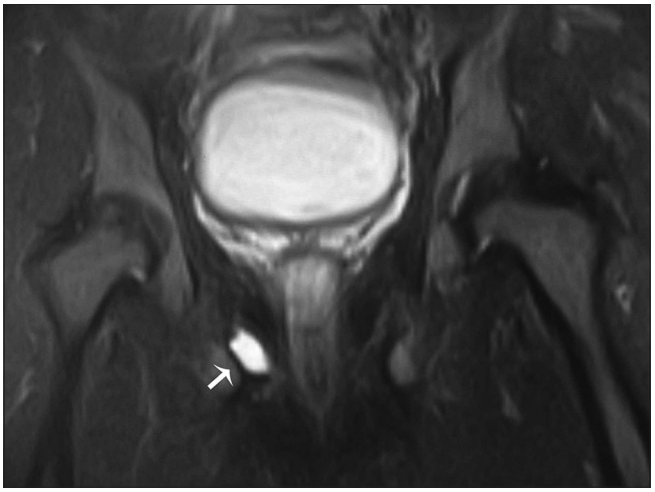

Figure 4: MRI showing small fluid collection adjacent to right ischial tuberosity suggesting an infective etiology

showing perivascular lymphocytic infiltration with endothelial swelling and focal neutrophilic abscess [Figures 5 and 6], consistent with pyoderma gangrenosum. Patient was promptly started on ATT, DOTS Category 2 owing to his defaulter status, with resultant improvement in his lesions. After 1 month of starting the ATT he subsequently received two pulses of dexamethasone cyclophosphamide with intervening low-dose cyclophosphamide. Daily dressings were done until complete healing occurred.

DISCUSSION

Pyoderma gangrenosum (PG) also known as phagedena

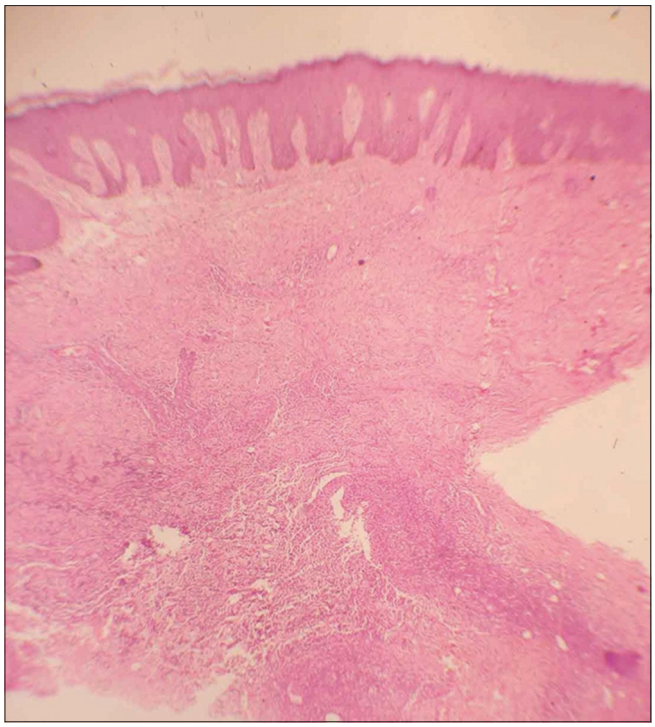

Figure 5: Histopathology; showing normal epidermis with dermal neutrophilic infiltrate. (H and E, ×100)

geometrica, dermatitis gangrenosa and phagedenic pyoderma is a rare neutrophilic dermatoses[3] with distinctive clinical features and a frequent association with systemic disease.[2,3] It was first described and named by Brunsting, Goeckerman and O'Leary in 1930. They believed that streptococcal infection was a significant component leading to secondary cutaneous gangrene and hence the current nomenclature.[3,4] Although the cause remains obscure, most probably an immunological anomaly of the hyperergic reaction type has been implicated.[5] PG most commonly occurs in adults aged 30-50, with a higher predilection for lower extremities and trunk. Several variants have been described; ulcerative, pustular, bullous, vegetative, vulvar and peristomal.[1,3,4] These often start as a small papule or collection of papules, which break to form small ulcers with a "cat's paw" appearance that coalesce to form a single ulcer with central necrosis[2] and have sharply demarcated undermined edges with violaceous border.[1-3,5-7] Lesions are less than 10 cm in size, but can be very large. They tend to endure, lasting months to years and heal with an atrophic cribriform scar. Associated symptoms include fever, malaise, myalgia and arthralgia. Extracutaneous involvement is reported in bone and lungs with neutrophilic infiltrate. Pathergy occurs in 25-50% of cases-lesions develop at the site of minor trauma.[3,5]

PG is known to be associated with diseases like inflammatory bowel disease (which may precede it, follow it, or occur simultaneously), arthritis, monoclonal gammopathy, myeloproliferative disorders, acne conglobata, diabetes mellitus, malignancies and a host of other conditions like

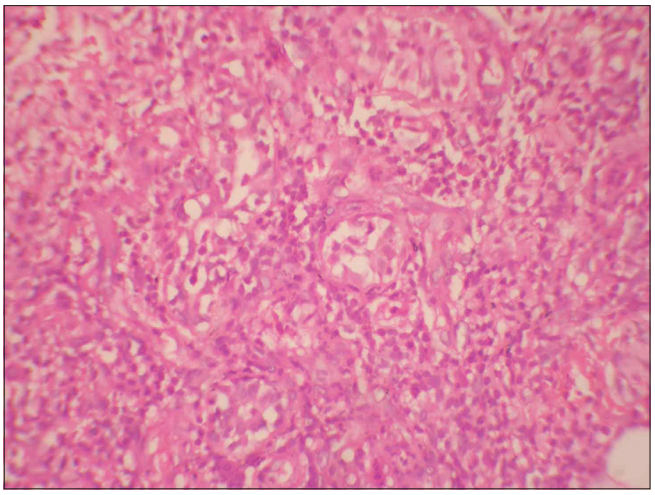

Figure 6: Histopathology view of dermis showing focal neutrophilic abscess. (H and E, ×400)

hidradenitis suppurativa, sarcoidosis and Takayasu's arteritis. Its association with HIV and other neutrophilic dermatosis has also been documented.[3-6]

To date less than half a dozen cases of pyoderma gangrenosum associated with different forms of tuberculosis have been reported.[7-9] Our case of PG with bone tuberculosis, further strengthens the association of the two conditions, as was evident by the clinical improvement in the lesions following anti tuberculous therapy and the development of lesions, at two different instances with an underlying tuberculous pathology.

Diagnosis depends mainly on recognition of the evolving clinical features and history of underlying disease because the histopathological changes are not specific. Long-term immunosuppression, often with high doses of corticosteroids or low doses of cyclosporine, with local care remains the cornerstone for treatment.[1,2,3,6] Successful treatment of the underlying disease results in almost complete remission of pyoderma gangrenosum. Recently, good outcomes have been reported for treatments with anti-tumor necrosis factor α, and infliximab.[5]

To conclude, this case of PG in a known case of ulcerative colitis is being reported for its rare association with tuberculosis.

REFERENCES

1. Naik CL, Singh G, Kumar L, Lokanatha K. Bullous pyoderma gangrenosum associated with ulcerative colitis. Indian J Dermatol Venereol Leprol 2008;74:68-70.
2. Brooklyn T, Dunnill G, Probert C. Diagnosis and treatment of pyoderma gangrenosum. BMJ 2006;333:181-4.
3. Bhat RM. Management of pyoderma gangrenosum-an update. Indian J Dermatol Venereol Leprol 2004;70:329-35.
4. Paramkusam G, Meduri V, Gangeshetty N. Pyoderma gangrenosum with oral involvement: Case report and review of the literature. Int J Oral Sci 2010;2:111-6.

5. Inan I, Myers PO, Braun R, Hagen ME, Morel P. Pyoderma gangrenosum after totally implanted central venous access device insertion. World J Surg Oncol 2008;6:31.
6. Thornton JR, Teague RH, Low-Beer TS, Read AE. Pyoderma gangrenosum and ulcerative colitis. Gut 1980;21:247-8.
7. Shirin H, Shirin K, Pouneh D, Mohammad SR. Lymph node tuberculosis associated with pyoderma gangrenosum. Infect Med 2005;25:76-9. Available from: http://www.medscape.com/viewarticle/499297. [Last accessed on 2011 Oct 17].
8. Antón Botella F, Peña Somovilla JL, Salcedo Aguilar J, Turón Alcaine JM, Ruiz Valverde A, Campo Hernandez JM. Pyoderma gangrenosum associated with testicular tuberculosis. An Med Interna 1989;6:549.
9. Kim NY, Choi JY, Lee KH, Shin JW. Pyoderma gangrenosum in a patient with colonic tuberculosis. Am J Gastroenterol 1994;89:1257-9.

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