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Recalcitrant Pyoderma Gangrenosum- Like Atypical Mycobacterial Infection Successfully Treated with Combination of First and Second Line Anti Tubercular Drugs

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Abstract

A 65-year-old male presented in the outpatient department with multiple painful ulcers and pustules on the whole trunk. The total duration of disease was 14-months. He was provisionally diagnosed as a case of pyoderma gangrenosum. He was initially treated with oral steroid but the condition worsens. The biopsy of the lesion was done which showed caseating granuloma. He was treated as a case of cutaneous scrofuloderma but no response was seen in two months. Then culture for mycobacterium was send and result was positive for atypical mycobacteria. The treatment direction was shifted towards the management of atypical mycobacterial infection with the addition of two second line drugs in first line antitubercular drugs and then he cured completely in 9-months.

Keywords: Pyoderma Gangrenosum; Atypical Mycobacteria; Antitubercular Drugs

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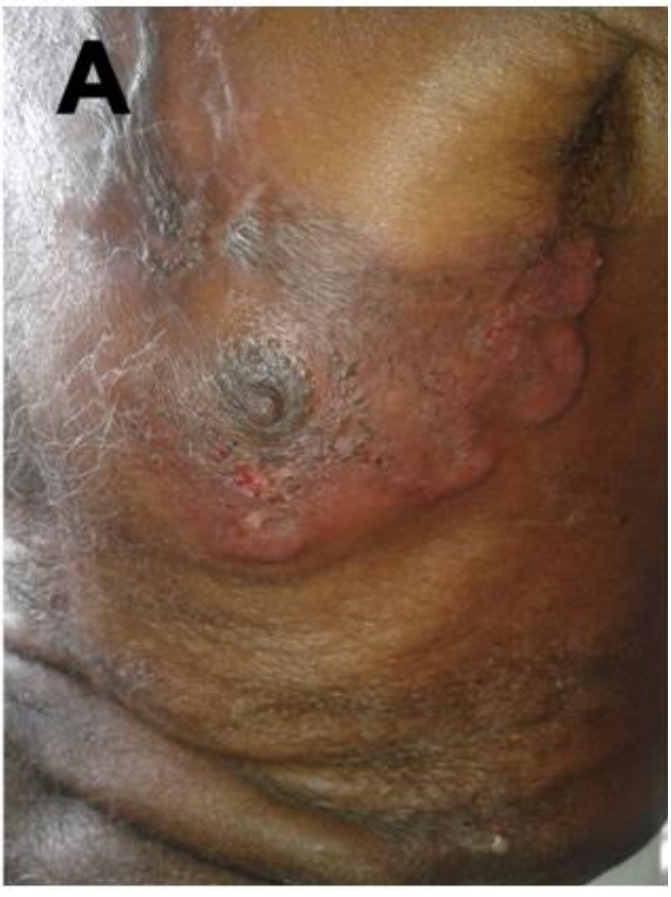
Case Report

A 65-year-old male presented with 14-month duration of very painful, purulant ulcers on the whole trunk (Figure 1). Ulcers started as furuncle-like lesions. He also had infiltrated erythematous plaques and nodules. He was very lean and thin but with no fever, no fatigue and no systemic symptoms.

There was past history of pulmonary tuberculosis 6-year back for which he took complete treatment of antitubercular drugs. On the basis of history and examination following differential diagnosis of the case was made pyoderma gangrenosum, tuberculous scrofuloderma, erythema nodosum leprosum, actinomycosis, cutaneous T-cell lymphoma and cutaneous B-cell lymphoma.

Figure1: A. Tender erythematous plaque on chest.



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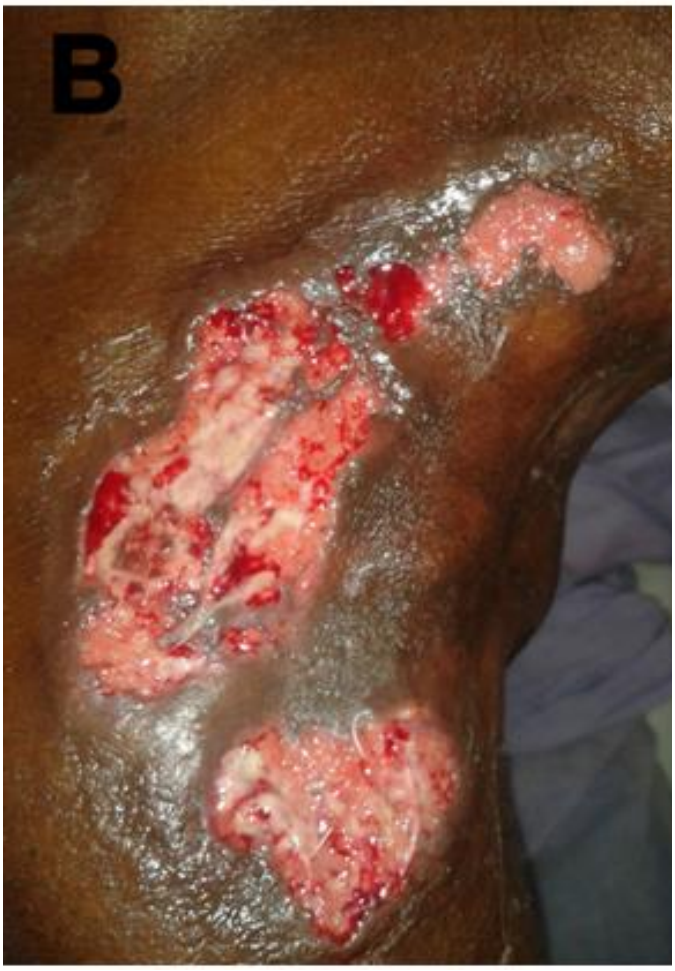
B. Pyoderma gangrenosum like tender ulcerative plaque.

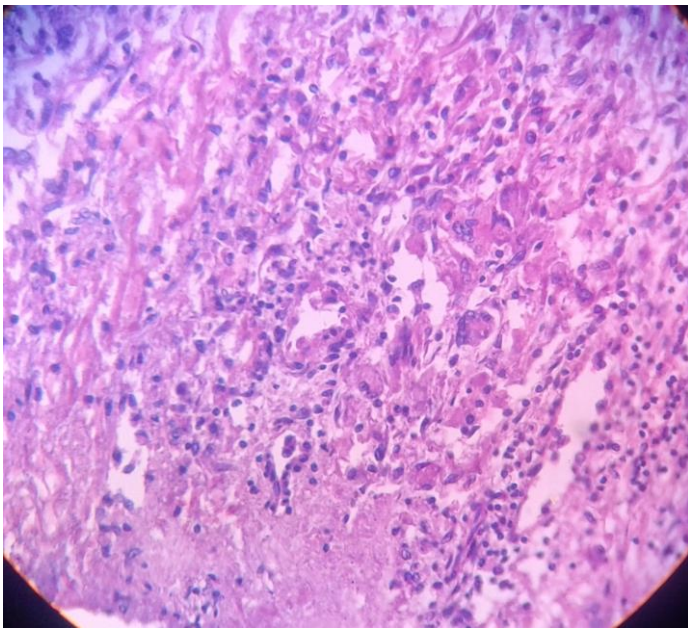


The routine blood investigation findings of patient were within normal range but there was moderate lymhocytosis (54%). Serological tests for HIV, HBV and HCV were negative. X-ray chest showed mild fibrosis in right upper lobe. Ultrasonography of whole abdomen was normal. Pathergy test was negative.

A biopsy from the active margin of the lesion was done that revealed a mixed granulomatous dermal infiltrate. A higher power showed epithelioid cell tubercles with mixed cell infiltrate of lymphocytes and neutrophils (Figure 2). Lymphocytes were normal in morpholgy with no atypia. Fite stain of that smear showed no acid fast bacilli. Slit-skin smears from ear lobule and from lesion were negative for Acid-Fast Bacilli (AFB). CD4/CD8 ratio was normal.

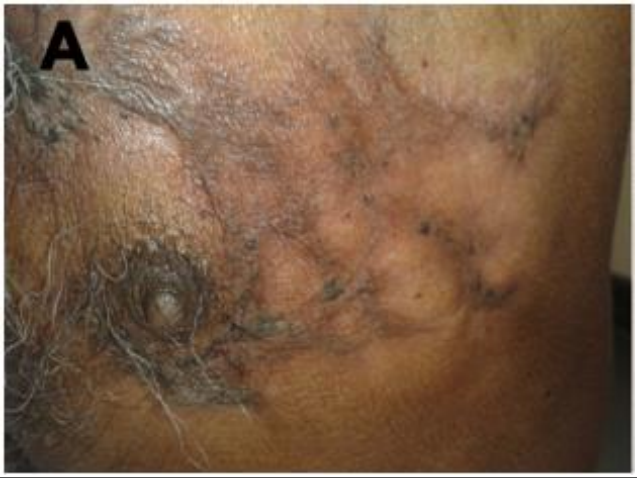
This case was initially treated as a case of pyoderma gangrenosum on clinical ground and oral steroid was given for 15-days but when condition worsen then biopsy and other tests were done. On the basis of investigation findings a provisional diagnosis of cutaneous scrofuloderma was made. The combination of all the five drugs of first line antitubercular regimen (rifampicin+isoniazide+ethambutol+pyrizin-amide+streptomycin) were given to the patient for two months but there was no response and lesions were continuously appeared. Then cultures for fungi, mycobacteria, and Actinomycosis were done. The culture report was positive for atypical mycobacterial infection. Tissue PCR for mycobacterium tuberculosis was negative.

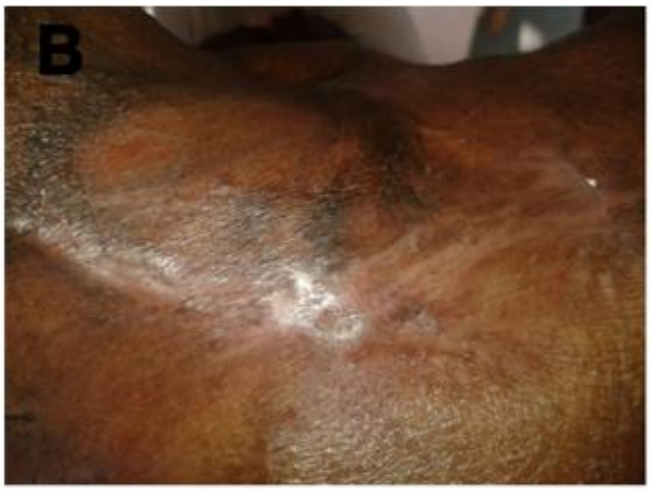
Figure2: Histopathology of the lesion in 40X view (H&E stain) showing caseating tuberculoid granuloma with Langhan's giant cells.



Now on the basis of culture report ciprofloxacin was added with the antitubercular drugs and given to the patient but still there was no improvement in next one month. In the last, combination of minocyclin, ciprofloxacin and five antitubercular drugs were started under the supervision and improvement was seen within three weeks of the treatment. The ulcers were healed in five months (Figure 3). Azithromycin and minocyclin were given as maintenance for next one month. Total duration of the treatment was 9-months. There was no recurrence of disease in two month follow-up.

Figure3: A and B: Scars of the lesions after complete treatment





On the basis of clinical features, histopathological findings, microbiological findings and also the therapeutic response to specific drug regimen; the final diagnosis of recalcitrant pyoderma gangrenosum-like atypical mycobacterial infection was made.

Discussion

Atypical mycobacteria are acid-fast bacilli other than M.tuberculosis that cause pulmonary, cutaneous or systemic disease [1]. These are found in water, wet soil, house dust, dairy products, cold-blooded animals, vegetation and human feces [2, 3]. Atypical mycobacteria are accidentally transmitted by inhalation, ingestion or percutaneous penetration, which can result in pulmonary, lymph node or skin diseases [4]. They occur more frequently in immunocompromised hosts and in AIDS patients [5, 6]. Predisposing factors for infection include trauma, surgery, chronic underlying illness, organ transplantation, drug abuse and tattoos. Infection originates from the ground, natural or stored water [7]. The entry is by inoculation, spreading from a deep focus or by blood dissemination [4, 8].

The cutaneous manifestations of atypical mycobacterial infection are varied. It presents like pyoderma gangrenosum-like lesions to ecthyma gangrenosum-like lesions or abscesses, discharging sinuses, panniculitis-like to warty plaques like lesions [3]. Atypical mycobacterial infections are frequently misdiagnosed due to non-specific clinical presentation with very low level of suspicion and improper culture techniques [1].

Treatment of atypical mycobacterial infection with first line antitubercular drugs is highly unsatisfactory as there is resistance with these drugs [9]. Our case was unique in terms of resistant to first line drugs for atypical mycobacteria and showed response after addition of two second line drugs and posed a treatment dilemma.

Learning Point

1. Pyoderma gangrenosum-like atypical mycobacterial infection is rarest entity.
2. Diagnosis of atypical mycobateria is very difficult, only culture can help but most of the time it is the diagnosis of exclusion.
3. Treatment of such case is highly dissappointing if not take carefully. As resistance was common with first line antitubercular drugs so think about one or two adjuvent drugs for complete clearance of the disease.
4. Mantenance treatment is required for one to two months with follow-ups.

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