Case Reports

Multiple pulmonary nodules in association with pyoderma gangrenosum

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This report describes a patient with extensive pyoderma gangrenosum in whom there were co-existent lung abnormalities. The patient's X-ray showed peripherally sited multiple pulmonary lesions bilaterally. A lung biopsy showed chronic non-specific inflammatory changes with neutrophil and lymphocyte infiltration which were similar to the skin lesions. This case was diagnosed as multiple aseptic nodules in pyoderma gangrenosum. The pulmonary infiltrative shadows were controlled only with prednisolone treatment. Steroid therapy is considered to be the first choice to control pulmonary lesions of this disease.

Introduction

Pyoderma gangrenosum is a chronic inflammatory cutaneous disease of unknown aetiology. Although it is basically considered to be a dermatological disease, its clinical appearance seems to be systemic in some aspects. For example, pyoderma gangrenosum is often associated with chronic inflammatory bowel disease, rheumatoid arthritis, gammopathies or multiple myeloma. However, few cases with pulmonary involvement have been reported. The present case report describes a case of pyoderma gangrenosum with multiple aseptic pulmonary lesions which responded to corticosteroid treatment.

Case Report

A 50-year-old male office worker was admitted to the authors' dermatological ward in November 1993. He complained of painful skin ulcerations on both legs (Plate 1). Histological examination showed inflammatory changes with neutrophil and lymphocyte infiltration, without angitis or granuloma. These macroscopic and microscopic findings demonstrated typical features of pyoderma gangrenosum. Steroid treatment was started and the eruptions gradually improved. He was discharged in March 1994 with a maintenance dose of prednisolone (7.5 mg day⁻¹).

Disease remission continued until September 1994, when he started to complain of cough and slight chest pain. Chest X-ray showed bilateral infiltrative shadows accompanied by pleural effusion and pleural thickening on the right side (Plate 2). He was treated with oral antibiotics but the shadows did not change, so he was referred to the authors' department on 7 November 1994.

The patient was afebrile. There was a dull area on percussion and breath sounds were slightly weak at the right basal area, but crackles were not audible. Laboratory findings revealed leukocytosis (white blood cells 13,200 mm⁻³; 75% neutrophils, no eosinophilia), haemoglobin 13.4 g dl⁻¹, platelet count 461,000 mm⁻³, erythrocyte sedimentation rate 30 mm h⁻¹, C-reactive protein 2.1 mg dl⁻¹, IgG 2530 mg dl⁻¹, IgA 418 mg dl⁻¹, IgM 197 mg dl⁻¹ (normal range of IgG: 885–1822 mg dl⁻¹, IgA:
Ulceration of the leg is a typical manifestation of pyoderma gangrenosum.

96–393 mg dl\(^{-1}\), IgM: 44–295 mg dl\(^{-1}\)). Other biochemical blood tests were normal. Rheumatoid factor was negative and both cytoplasmic antineutrophil cytoplasmic autoantibody (C-ANCA) and perinuclear cytoplasmic antineutrophil cytoplasmic autoantibody (P-ANCA) were negative. Tumour markers were negative. Repeated sputum cultures, including those for tuberculosis, were negative. Tomography showed bilateral peripherally located multiple nodular lesions (Plate 3). A lung biopsy performed from the right middle lobe bronchus showed chronic nonspecific inflammatory changes with neutrophil and lymphocyte infiltration which were similar to the skin lesions, but there was no evidence of bronchiolitis obliterans, granuloma, vasculitis or malignancy. Bronchial washing fluid culture was negative. Percutaneous aspiration by thoracentesis was performed, and fluid cultures including anaerobic culture and culture for tuberculosis were negative. Cytologic examination of the lavage fluid showed no malignant cells, only neutrophils and lymphocytes. After examination, antibiotic therapy with clindamycin (1200 mg day\(^{-1}\)) and isepamicin (400 mg day\(^{-1}\)) was given intravenously for 10 days, but there was no pulmonary improvement. This case was diagnosed as multiple, aseptic nodules in pyoderma gangrenosum. Oral prednisolone was increased to 40 mg day\(^{-1}\), and the infiltrative shadows rapidly decreased over a few weeks. The patient was discharged on 25 November 1994 and prednisolone was tapered to 10 mg day\(^{-1}\). During the subsequent 10 months, no relapse has been recognized.
Discussion

Pulmonary involvement of pyoderma gangrenosum is uncommon and has been seldom reported (1). Previously reported histological findings of pulmonary lesions are similar to those of the present case (2-4). The lung abscesses were aseptic (2-4). In the other cases, pulmonary lesions showed a single unilateral opacity on chest X-ray. The present patient had an unusual form of pyoderma gangrenosum with multiple pulmonary involvement which has not been reported previously.

Pyoderma gangrenosum is characterized by an aseptic inflammatory cutaneous disease of unknown aetiology. It is associated with basic immunological disorders like Crohn’s disease, ulcerative colitis, rheumatoid arthritis, vasculitis, lymphomas, leukaemias, dysproteinemias or multiple myeloma (5). There may be auto-immune factors in the development of the disease process. The previous reported cases with pulmonary lesions also had basic diseases, ulcerative colitis and rheumatoid arthritis. This case showed increase of immunoglobulins, but there were no clinical complications of other immunological disorders (6). Sarcoidosis or vasculitis like Wegener’s granulomatosis (7) was ruled out by the clinical and histological findings.

In this case, the response to steroid therapy was dramatic, as in previously reported cases. Steroid therapy is considered to be the first choice to control pulmonary lesions of pyoderma gangrenosum.

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References