An unusual cause of lung mass in a young female

Sir,

Hereby presenting a 17-year-old girl who was referred with complaints of cough and expectoration lasting for 1 month. She was diagnosed to have non-Hodgkins lymphoma (NHL) 1 year back, for which she underwent chemotherapy and was in remission. Patient had no past history of asthma, atopy, pneumonia, or any chronic lung disease. Routine hematological investigations including a peripheral smear were within limits, apart from an elevated erythrocyte sedimentation rate (ESR) of 62 mm/h. Chest X-ray showed right upper lobe and left lower lobe opacity [Figure 1]. Sputum for ordinary culture sensitivity showed no growth and sputum for acid fast bacilli by Ziehl-Neelsen stain was negative. Sputum for fungal culture revealed heavy growth of *Candida tropicalis* consistently over three consecutive times. Parenteral antifungals were initiated with amphotericin B which was given for 14 days followed by oral itraconazole. She improved symptomatically following therapy, but chest X-ray continued to show persistent opacities. In view of these persistent opacities, bronchoscopy was done. Bronchial lavage was negative for acid fast bacilli by Ziehl-Neelsen stain and bronchial lavage pyogenic culture and fungal culture yielded no growth. Bronchial lavage cytology, brush cytology, and bronchoscopic biopsy were negative for malignancy. Patient then underwent computed tomography (CT)-guided fine needle aspiration cytology (FNAC) and biopsy of the lesion. Histopathology specimen showed fragments of lung tissue with areas of coagulative necrosis with fibroblastic proliferation, ill-defined granulomas, and chronic inflammatory infiltrates consisting of lymphocytes, eosinophils, few neutrophils, and plasma cells. Impression was that of a necrotizing granulomatous inflammation with eosinophils suggestive of fungal bronchocentric granulomatosis [Figure 2]. Patient was started on oral steroids at 1 mg/kg body weight for a month with significant radiological clearance [Figure 3].

Bronchocentric granulomatosis is a rare idiopathic condition, defined by Liebow in pure pathological terms as necrotizing granulomatosis centered on peripheral conducting airways.[1] Many authors restrict the term bronchocentric granulomatosis to asthmatic patients, in whom this lesion should be considered a manifestation of allergic bronchopulmonary aspergillosis (ABPA) and about 30% of reported cases of bronchocentric granulomatosis have been associated with ABPA.[2] It may be due to a variety of other noninfectious and infectious causes as well.[3-5] Noninfectious causes predominantly are rheumatoid arthritis, ankylosing spondylitis, chronic granulomatous diseases, glomerulonephritis, scleritis, diabetes insipidus, red cell aplasia, bronchogenic carcinoma, Wegener's granulomatosis, following heart and bone marrow transplantation. In all cases of bronchocentric granulomatosis infectious etiology has to be thoroughly investigated and any remote infective foci must be ruled out.

Radiographic appearance of Bronchocentric granulomatosis has consistently been divided into two main patterns: Lobar consolidation or masses/nodules. Consolidation could be lobar and segmental with atelectasis which may also be fleeting or migratory over a period of time. It is one of unusual causes for nonresolving pneumonia. The mass lesions are another pattern of presentation of BCG and these lesions are unilateral in

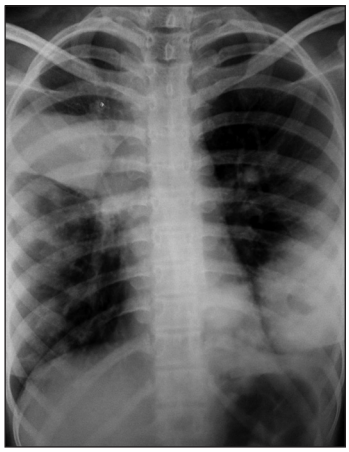

Figure 1: Chest X-ray showed right upper lobe and left lower lobe opacity

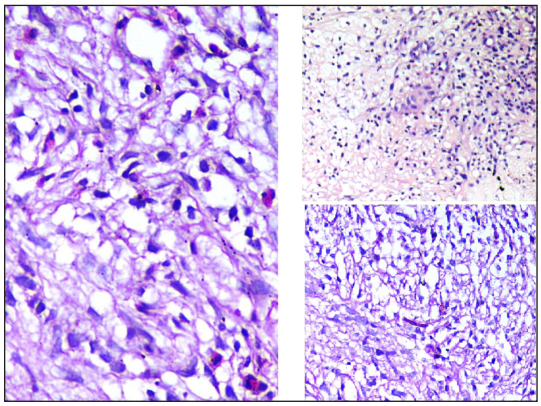

Figure 2: Histopathology showing ill-defined granulomas and chronic inflammatory infiltrates consisting of lymphocytes, eosinophils, few neutrophils, and plasma cells

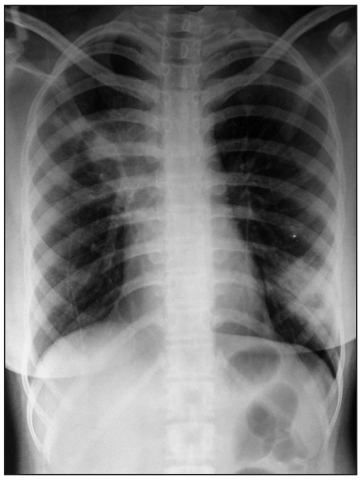

Figure 3: Chest X-ray after 1 month of steroid therapy which shows regression of lesions

the majority of cases and have only occasionally been reported to show cavitation.[6,7] Histopathology of BCG exhibits foci of prominence of palisading histiocytes with intraluminal exudate and eosinophilia. Fungal hyphae can be identified in most of these cases as also evidences in our subject and are located within the bronchocentric granulomas as well as the impacted mucus.[8] Confirmatory diagnosis is by biopsy from the lesion and a definitive diagnosis of bronchocentric granulomatosis can only be made by histopathological examination.

Corticosteroid therapy has in general been quite effective treatment resulting in radiological clearing and subsidence of clinical symptoms in many patients. The duration of therapy must be monitored on the basis of clinical and radiologic response. In our patient an antifungal agent, itraconazole, was used in addition to corticosteroids as reports have suggested that antifungal agents such as itraconazole are beneficial in the treatment of ABPA.[9] We were also concerned about the risk of invasive fungal infection in this patient in view of her immunodeficiency state in combination with glucocorticoid therapy.

The coexistence of these two rare entities of *Candida tropicalis* and BCG in an NHL patient with post chemotherapy status is unlikely to be coincidental and it raises the question of a broader link between abnormal phagocyte functioning and the development of hypersensitivity reactions. Persistent lung shadows in patients having underlying malignancy are generally attributed to progressive disease, metastasis, or opportunistic infections. In our case an infectious etiology of *Candida tropicalis* (Candida non-albicans) could be isolated by culture repeatedly for three times which is extremely rare as most often the fungi isolated is Aspergillus.

Rarity of this case was the association of an unusual pulmonary mycosis in the form of *Candida tropicalis* with brochocentric granulomatosis in an NHL patient, which is not previously reported. This case is an eye opener pointing towards other rarer but often overlooked causes for persistent lung opacities in an immunocompromised host.

Sandeep Satsangi, Vishak Acharya[1], Hema Kini[2], Anupama KV

Junior resident, Department of Internal Medicine,
[1]Associate professor, Department of Pulmonary Medicine,
[2]Professor and Head of Department of Pathology, Kasturba Medical College and Hospital, Mangalore, Karnataka, India
E-mail: Sandeep satsangi:agamthesmartest@gmail.com

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