

A YOUNG LADY WITH VANISHING LUNG SHADOWS

*Johnson Samuel MD, FRCP
Balamugesh Thangakunam, MD, DM*

Basildon University Hospital, United Kingdom

This case was presented at the Great Cases Session at the American Thoracic Society International Conference, May 16, 2010 in New Orleans, LA.

Abstract

A previously well 20-year-old young lady who presented with nonspecific right-sided chest pain was found to have a rounded shadow on chest X-ray. Investigations to rule out malignancy revealed multiple lung masses. Initial blood tests and percutaneous image guided biopsy were inconclusive. Surgical lung biopsy revealed features suggestive of Bronchocentric Granulomatosis. Her lung shadows spontaneously resolved and there was no evidence of symptomatic or radiological recurrence on follow up for five years. Bronchocentric Granulomatosis is a rare condition particularly in non-asthmatic individuals and should be considered in the lesser-known differential diagnosis of benign lung shadows.

Case presentation

History and Examination

A 20-year-old lady presented to the Emergency department with right-sided pleuritic chest pain after watching a football match in the local pub. A Chest radiograph showed a 1cm oval opacity in the periphery of the left upper zone. She was treated with a course of antibiotics and a repeat chest radiograph two weeks later showed an increase in the size of the opacity (Fig 1).

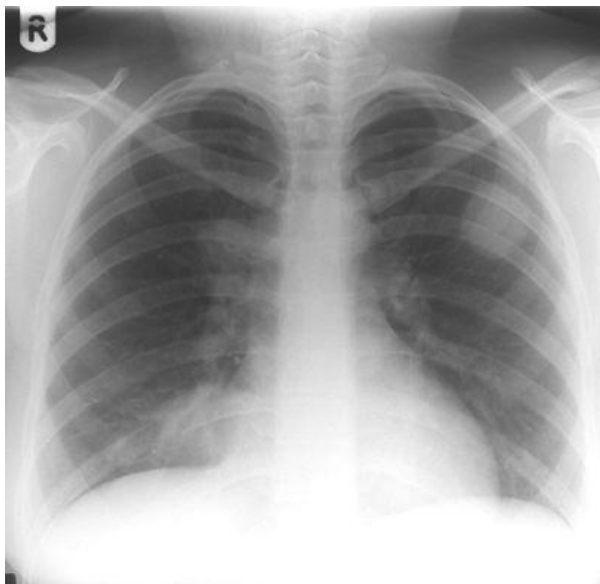


Fig 1: Chest radiograph showing a rounded opacity in left upper zone.

She was then referred to the respiratory clinic for evaluation. She was a non-smoker with no previous medical history of note. There was no history of recent respiratory infections. Clinical examination including breast examination was entirely normal.

Investigations

Haemoglobin was 11.2 gm/dl, white cell count $7.0 \times 10^9/L$, platelet count $255 \times 10^9/L$ and erythrocyte sedimentation rate 22mm/hr. Differential white cell count was normal with absolute eosinophil count $0.1 \times 10^9/L$. Serum angiotensin converting enzyme was 36 IU/L. Serum compliment levels were normal and tumour markers were negative. Human chorionic gonadotrophin level was not elevated. Rheumatoid factor was 38 IU/ml, autoantibodies and Human Immunodeficiency Virus tests were negative. Mantoux test was non-reactive.

Computed Tomography (CT) showed three opacities, two of which were on the right side (Fig 2).

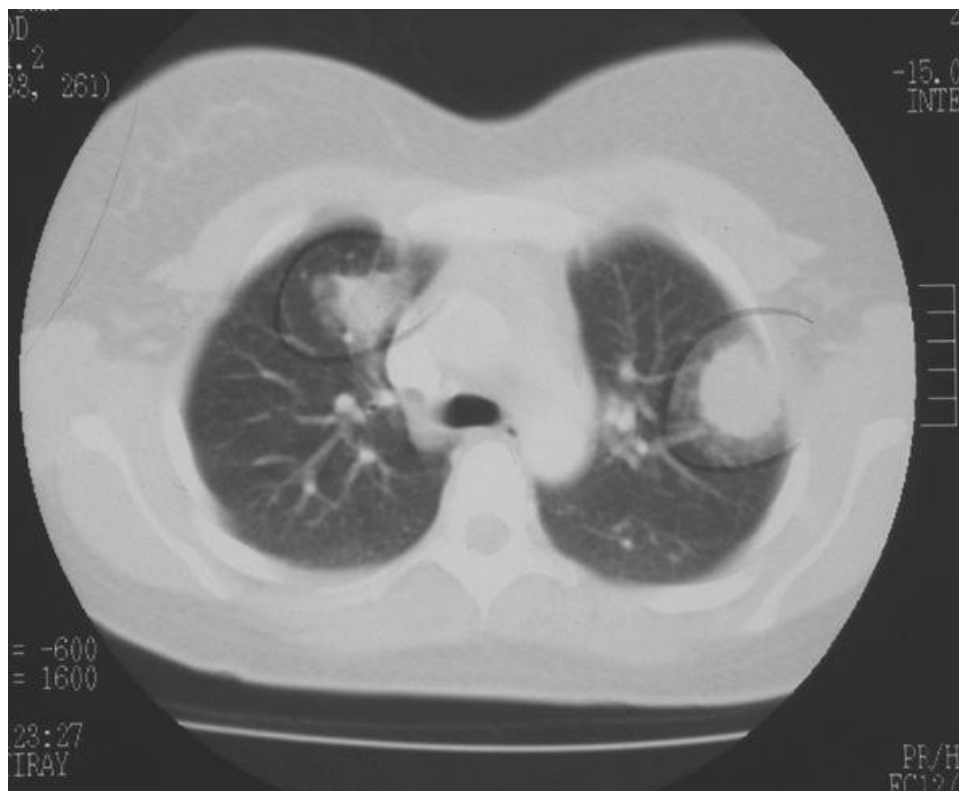


Fig 2: Computerised Tomography scan of chest showing bilateral lung opacities

A CT guided biopsy showed necrotizing chronic granulomatous inflammation with no evidence of malignancy. There were many eosinophils seen in the biopsy sample (Fig 3).

As the diagnosis was still uncertain and as there was the suspicion of metastatic malignancy of uncertain origin, she underwent a video assisted thoracoscopic biopsy (VATS biopsy). Histopathology of the nodule revealed abundant necrosis, surrounded by a rim of chronically inflamed fibrous tissue with focal areas of discontinuous elastic lamina (Fig 4).

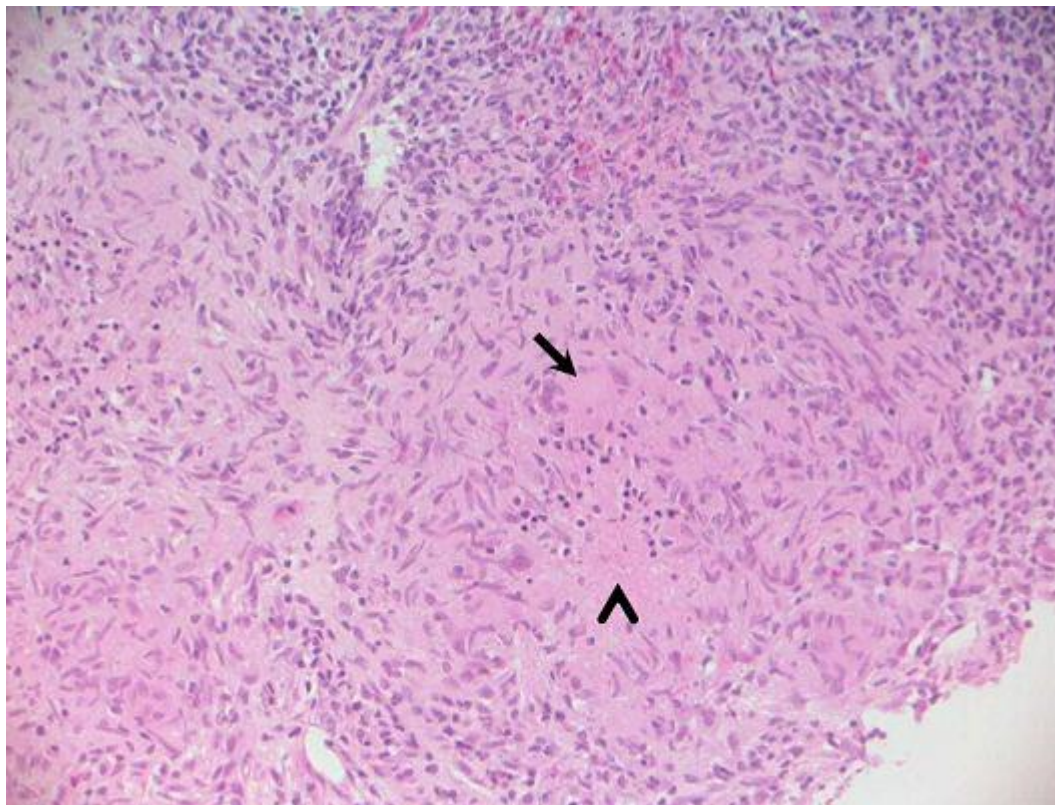


Fig 4: Video assisted thoracoscopic biopsy showing focal necrotizing granulomatous inflammation. Arrowhead indicated the central necrosis. Short arrow indicated the multi-nucleated giant cell (High power view – formalin fixed, haemotoxylin and eosin staining).

Mycobacterial and fungal cultures were negative. There were no features of vasculitis or malignancy. Based on the histopathology and the clinical features, a diagnosis of Bronchocentric Granulomatosis was made.

Follow up

The patient was asymptomatic throughout she did not receive treatment. On follow up, after initial increase in size, the nodules started regressing and serial chest X-ray's showed complete resolution of the lung lesions in four months. She has since been followed up in the respiratory clinic for five years and there was no radiological evidence of recurrence. Her serial spirometry during follow up was normal. She remains asymptomatic and has joined the police force leading an active lifestyle.

Discussion

Bronchocentric granulomatosis is a necrotizing granulomatous inflammation of the small to medium sized bronchi and bronchioles (1). Approximately half the patients are asthmatics and among these patients, the disease appears to be a hypersensitivity reaction to inhaled allergens particularly *Aspergillus fumigatus*. (2,3). In the non-asthmatic group, the causative agent is usually not identified. Similar appearances have been described in patients with Pulmonary Echinococcosis, Wegener's Granulomatosis and Rheumatoid Arthritis (4). Occasionally, it may show histological features indistinguishable from tuberculosis.

Asthmatic patients, who are usually in the age group 20-40 years, present with worsening wheeze, dyspnoea, cough, fever and occasionally haemoptysis. Peripheral blood may show eosinophilia and *Aspergillus fumigatus* is commonly grown in sputum culture. Non-asthmatic patients are often asymptomatic or may present with an acute febrile illness. Radiologically, Bronchocentric Granulomatosis can present as mass lesions, alveolar infiltrates, pneumonic consolidation or reticulo-nodular opacities (5). As this can mimic malignancy as in our case, a surgical lung biopsy may be required to rule out vasculitis or malignancy. There are no definite diagnostic criteria and diagnosis is based on clinical, radiological and pathological correlation. Some patients may need corticosteroids for rapid clinical and radiological resolution while others recover without treatment. Surgical resections have been done to firmly establish the diagnosis given the concern about malignancy. However, when there are no alarming symptoms or signs, it may be appropriate to monitor progress or consider medical treatment.

Bronchocentric Granulomatosis though rare is a reassuringly benign and usually self-limiting condition, which can present with alarming radiological signs. It should be considered in the asthmatic and asymptomatic patient when other sinister causes are ruled out by definitive investigations, which may include a surgical biopsy.

Acknowledgement:

Dr. Nazar Alsanjari, Pathology department, Basildon University Hospital, Basildon. For kindly providing the photomicrographs of the biopsy specimens.

References:

1. Liebow AA. Pulmonary angiitis and granulomatosis. American Review of Respiratory Disease 1973;108:1-18.
2. Sulavik SB. Bronchocentric granulomatosis and allergic bronchopulmonary aspergillosis. Clin Chest Med. 1988;9:609-21.
3. Koss MN, Robinson RG, Hochholzer L. Bronchocentric granulomatosis. Hum Pathol. 1981;12:632-8.
4. Bonafede RP, Benatar SR. Bronchocentric granulomatosis and rheumatoid arthritis. Br J Dis Chest. 1987;81(2):197-201.

5. Ward S, Heyneman LE, Flint JD, Leung AN, Kazerooni EA, Miller NL. Bronchocentric granulomatosis: computed tomographic findings in five patients. Clin Radiol. 2000;55:296-300.

Corresponding author:
Balamugesh Thangakunam,
Speciality doctor,
Respiratory services,
Basildon University Hospital,
Basildon, Essex, United Kingdom.
SS16 5NL.
E mail: drbalamugesh@yahoo.com