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Case Report

Peribronchial inflammatory myofibroblastic tumour: a case report

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ABSTRACT

Inflammatory Myofibroblastic Tumour (IMT) is a rare neoplasm of mesenchymal origin, most commonly seen in the lungs of children and adolescents, but it can occur in older persons also. IMT also called inflammatory pseudotumor accounts for less than 1% of all lung tumours. Approximately half of the patients are asymptomatic. However, the patients with symptoms show cough, haemoptysis, dyspnoea and chest pain. Biopsy by thoracotomy or video assisted thoracoscopic surgery is often necessary to confirm the diagnosis. In this case report, we discuss IMT in a 56-year-old male, who presented with cough and fever of one and a half months duration.

Keywords: Inflammatory myofibroblastic tumour, Inflammatory pseudotumour, Haemoptysis, Dyspnoea, Fibrosarcoma, Bronchoscopy, Histiocytoma, Xanthogranuloma, Plasmocytoma

INTRODUCTION

In majority of the chronic inflammatory lesions in Pulmonology, the real culprit turns out to be Tuberculosis. But in fact, the differential diagnoses of such lesions are very vast. Inflammatory pseudotumours (IPT) or Inflammatory Myofibroblastic Tumour (IMT) of the lung are rare and were first described in the lung in 1939.[1] Although, IPT are regarded as inflammatory or reactive lesions rather than neoplasms, they may have features such as local invasion or recurrence, distant metastases, and cytogenetic clonal changes.[2] IPT has been described by various names in the literature such as plasma cell granuloma (heart and lung), inflammatory myofibrohistiocytic proliferation, histiocytoma, xanthoma, fibroxanthoma, fibrous xanthoma, xanthogranuloma, xanthomatous pseudotumor, plasma cell-histiocytoma complex (lung), plasmocytoma, solitary mast cell granulomas, and inflammatory fibrosarcoma (urinary bladder).[3,4]

In 1954, Umiker and Iverson[5] coined the term "inflammatory pseudotumour" due to its clinical and radiological features of the lesion which mimic those of malignant tumours. IPT accounts for less than 1% of all lung tumours with no gender or race preponderance.[2] Approximately half of the patients are asymptomatic, whereas 26-56% of the patients have symptoms including cough, haemoptysis, dyspnoea and chest pain. Therefore, the clinician should be aware of the entity called IMT for the favour of adequate treatment. In this case report, we discuss IMT in a 56-year-old male, who presented with cough and fever of one and a half months duration.

CASE REPORT

A 56-year-old male presented with history of, cough, which was scantily productive with mucopurulent sputum, occasional low grade intermittent fever and loss of weight of one and a half months duration. He had one episode of streaky haemoptysis. History of contact with tuberculosis patients or family history was not available

or he was not on any regular medications in the past. He was a manual labourer with heavy smoking (smoking score 400) and an alcoholic. Clinical examination was uneventful. Haemogram revealed neutrophilic leukocytosis (total WBC count 18800/µL with 72.7% neutrophils) with a raised Erythrocyte Sedimentation Rate (ESR) (132 mm/hour). Other blood parameters were within normal limits. Chest X-ray revealed an ill-defined homogenous opacity in the right upper zone (Figure 1).

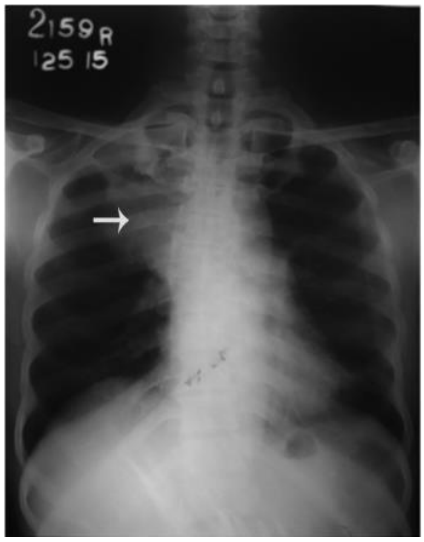

Figure 1: Chest X-ray with an ill-defined homogenous opacity in the right upper zone (Arrow head).

Computed Tomography (CT) thorax showed a contrast enhancing necrotic mass of 7.5 x 4.5 x 8.5 cm in size in the right upper lobe, encasing the right upper lobe bronchus which favoured bronchogenic carcinoma (Figure 2A and 2B).

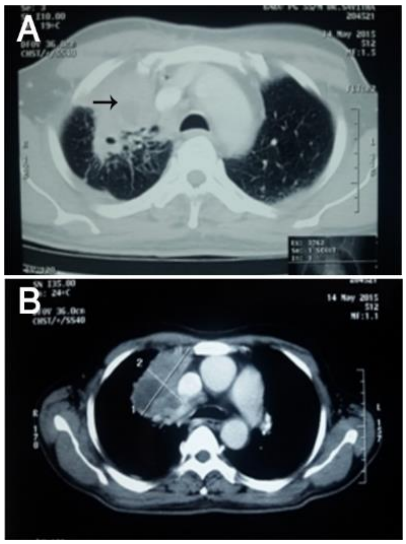

Figure 2: A) and B) Computed tomography of thorax showed a contrast enhancing necrotic mass of 7.5 x 4.5 x 8.5 cm in size in the right upper lobe, encasing the right upper lobe bronchus (Arrow head).

Empirical broad spectrum antibiotics (Ceftriaxone-Sulbactam and Metronidazole) and supportive therapy was given. Patient showed only partial clinical improvement with the treatment. After 72 hours of starting antibiotics, a repeat haemogram showed decreased total WBC count of 13500/µL and ESR dropped to 110 mm/hour. Patient became afebrile. Sputum AFB, culture and cytology was negative. Since, the clinical improvement was inadequate; bronchoscopy was done which showed an extra luminal compression of right upper lobe bronchus with unhealthy mucosa. Mucoid secretions were oozing from anterior segment orifice, with no intraluminal growth. Bronchial washings and brushings were taken. An endobronchial biopsy was also taken from the site of extraluminal compression. Since there was no definite intraluminal growth, CT guided Fine-Needle Aspiration Cytology (FNAC) and biopsy were also done. After completing seven days of antibiotics, the patient was discharged with an advice to review. Bacterial culture of bronchial washings, AFB smears and cytology of bronchial washings and brushings were negative. Endobronchial biopsy showed chronic inflammatory cells. During the review, he had become afebrile with minimal persisting cough. Constitutional symptoms had not fully resolved. The lesion did not show significant resolution in the repeated chest X-ray. Repeat CT guided FNAC and biopsy were done. These too showed chronic inflammatory infiltrates. He was referred to the Department of Thoracic Surgery, where resection of the right upper lobe with sampling of mediastinal lymph nodes was done (Figure 3A). The histopathology report of the lobectomy specimen was peribronchial inflammatory myofibroblastic tumour with lipoid pneumonia (Figure 3B).

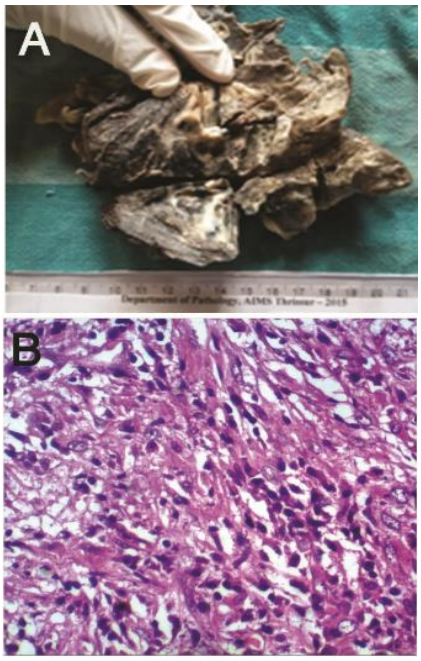

Figure 3: A) Gross appearance of resected specimen B) Photomicrograph showing the myofibroblastic proliferation & plasma cell infiltration (H&E; x400).

There were no features of malignant neoplasm. The lymph node samples showed reactive changes only. The patient improved symptomatically. He got discharged and is on follow-up.

DISCUSSION

IPT is usually considered to be a benign tumour, principally occurring in younger individuals. They are the most common isolated primary tumour of the lung in children younger than 16 years.[6] More than half of patients are less than 40 years of age. IMT is a rare neoplasm of mesenchymal origin, most commonly seen in the lungs of children and adolescents, but it can occur in older persons also.[7] The cause of IPT is unknown. Some causes are trauma and surgical inflammation, immune-autoimmune condition, and low-grade fibrosarcoma with inflammatory cells.[3,8] Approximately, half of the patients are asymptomatic, whereas ~40% of the patients have symptoms including cough, haemoptysis, dyspnoea and chest pain.[9] Our patient had cough and haemoptysis with mild fever.

The radiographs of IPT usually appears as a single peripheral, well-defined, lobulated mass with lower lobe predominance.[10] IPT can also be FDG-avid on PET/CT images.[11] The differential diagnosis of IPT presenting as a solitary pulmonary nodule is wide and biopsy is usually required to make an accurate diagnosis.[3] Radiographic images and invasive diagnostic procedures, including bronchoscopy and percutaneous fine needle aspiration biopsy, are considered insufficient for diagnosis. Therefore, open lung biopsy or videothoracoscopic resection is often necessary.[12] In our case, bronchoscopic biopsy was done once and CT guided biopsy was done twice but the results were inconclusive. The final diagnosis was revealed only after surgical excision.

Macroscopically, inflammatory pseudotumours are well circumscribed, un-encapsulated, firm, usually yellow-white masses containing variable inflammation, haemorrhage, calcification, and rarely cavitation. Microscopically, the lesions consist of variable mixtures of fibroblasts and granulation tissue, fibrous tissue, and inflammatory cells including lymphocytes, histiocytes, giant cells, macrophages, neutrophils, eosinophils and typically large numbers of plasma cells.[1] Immunohistochemistry has demonstrated the polyclonal nature of plasma cells with immunoglobulin G predominance.[2]

Based on cellular types and main histological properties, Matsubara et al. categorized inflammatory pseudotumour into three groups: A) Organized pneumonia formed by gradual healing of the intraalveolar exudation (44%), B) Fibrous histiocytoma formed by storiform proliferation of plasmocyte and lymphocyte aggregates (44%), and C) lymphoplasmocytic type formed by the aggregation of both plasmocyte and lymphocytes (12%).[13]

The treatment usually is surgery with radical excision. IPT can be locally aggressive with invasion of bronchi, the mediastinum, chest wall, and diaphragm, and surgical removal of adjacent structures may be necessary.[14] Radiation and corticosteroids have been used to treat patients who cannot undergo surgery but do not have proven survival benefit compared with surgery. The risk of relapse is low with radical excision, but relapse and distal metastasis occur many years after treatment. Sarcomatous degeneration has also been found in IPT, so careful surveillance is crucial. The prognosis of these rare tumours is excellent after complete surgical excision. There is a low incidence of recurrence with long-term follow-up after complete removal of the mass. Patients with recurrent disease should undergo re-resection.[1] IPT most commonly recurs locally and rarely metastasizes to distant sites.

CONCLUSION

Although IMT is a rare condition, the clinician has to keep such uncommon differential diagnoses also in mind whenever he is confronted with pulmonary lesions simulating malignancy. Small biopsy specimens usually do not give a definite diagnosis. Surgical excision is diagnostic as well as therapeutic. Complete surgical resection leads to excellent survival and prevents recurrence.

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