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Pulmonary hyalinizing granuloma-a rare case report of pulmonary nodule

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Abstract: Pulmonary hyalinising granuloma is rare, non-infectious fibrosclerosing lesions of the lung which can mimic with metastatic disease. It was first described in literature by Engleman et al in the year 1977. Pulmonary hyalinizing granuloma presents as multiple, mostly bilateral, recurrent pulmonary nodules that affect patients of both genders with equal frequency. Its etiology and pathogenesis is yet to be well defined. We report the case of a 57 year old woman presented with worsening of dry cough & dyspnoea for last 11 months. Her CT scan shows multiple pulmonary nodules, CT guided FNAC shows non-specific inflammatory cells. Biopsy of pulmonary lesion is consistent with pulmonary hyalinizing granuloma(PHG).
Keywords: Pulmonary hyalinizing granuloma,pulmonary nodule.

INTRODUCTION

Pulmonary hyalinizing granuloma (PHG) is rare fibrosclerotic inflammatory lung disease. Their etiology is unknown, but they may be caused by an exaggerated immune response to an unknown antigen [1]. It was first published by Engleman et al in 1977 [2] .Clinically pulmonary hyalinizing granuloma can be asymptomatic(25% of cases) or patient may present with dry cough, chest pain, haemoptysis, dyspnoea, fever, fatigue, pharyngitis, sinusitis[2]. Extra pulmonary involvement (kidney, larynx,skin) [3,4] occurs in some cases. It is commonly occur in adult between 19 and 77 years (mean age being 44 years) [5], there is no sex or racial predilection [6]. Exact etiology of this condition is still remain unclear, although an exaggerated immune response to the antigenic stimuli by infection (fungus, tuberculous bacilli) or autoimmune diseases has been suggested [7,8]. Chest radiography and computed tomography will reveal single or multiple well-defined nodules. The prognosis for patients with PHG is generally excellent with no significant impact on longevity [9].

REPORT OF CASE

55 year old woman came to cardiothoracic department of R G Kar Medical College & Hospital with history of worsening of dry cough & mild dyspnoea for last 11 months, on examination patient was lean & thin afebrile and her vitals were within normal ranges. She was housewife and there was no history of hemoptysis, chest pain, addiction history or occupational exposure. Her routine blood analysis, urine & sputum findings were normal. Her erythrocyte sedimentation rate & C reactive protein were within normal range. Among past medical history she had cough, hemoptysis, and respiratory distress 2 years ago and she was diagnosed as pulmonary tuberculosis by her physician and she completed 7 months courses of anti tubercular drugs. There was no history of pulmonary tuberculosis contact or family history of bronchial asthma or any autoimmune disorders. Her breath sounds over right side of lung was diminished in intensity and CT scan shows multiple mass with irregular borders located in the middle and lower lobe of right lung, (Fig 1). No mediastinal or cervical lymphadenoathy were noted. Pulmonary function test showed mild obstructive ventilatory defect and her anti nuclear antibody was negative. Her bronchoalveolar fluid (BAL) was negative for acid fast bacilli and malignant cells. CT guided FNAC from the mass show acute on chronic inflammatory cells (Fig 2). An open lung biopsy of mass showed interweaving collagen bands with chronic inflammatory cells in between them, few staghorn shaped blood vessels and foci of calcification were noted (Fig 3). Spindle cells are arranged in short fascicles with few of them shows epithelioid like arrangement (Fig 4). Initially our diagnosis was solitary fibrous tumor but immunohistochemistry for CD34 showed negative result (Fig 5).Section was negative for mycobacteria or any fungus. Among special stain it showed positivity for reticulin, masson trichome stain and negativity for PAS, congo red. These features are highly suggestive for pulmonary hylinizing granuloma.

Fig-1: CT scan shows multiple irregular mass.

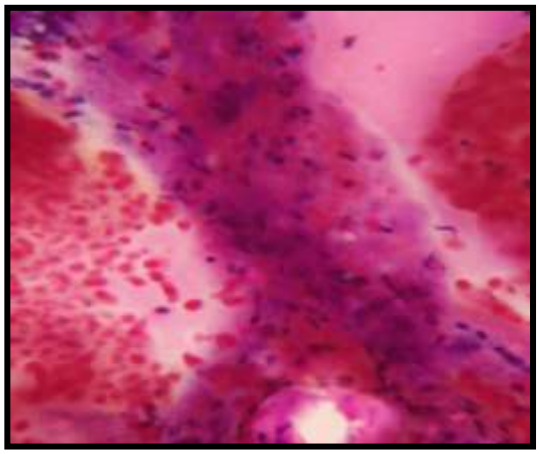

Fig-2: CT guided FNAC shows acute on chronic inflammatory cells.

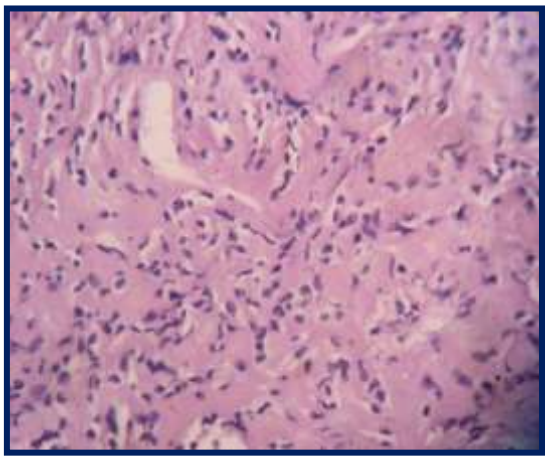

Fig-3: (H & E X 40) studied section shows band of collagen with chronic inflammatory cells and few staghorn shaped blood vessels.

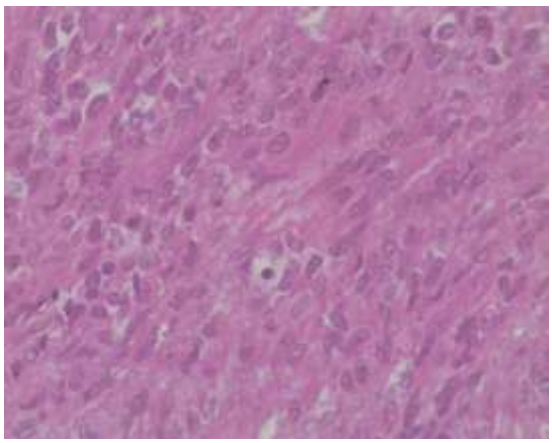

Fig-4: (H & E X 40) Spindle cells are arranged in fascicles with few of them show epithelioid like pattern.

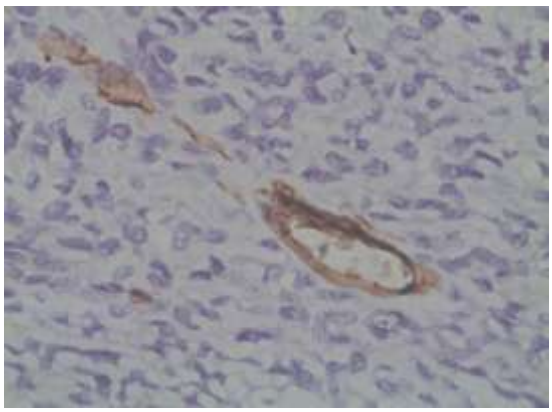

Fig-5: IHC with CD34 shows negative result.

DISCUSSION

Pulmonary hyalinizing granuloma is a rare fibrosing nodular disease of lung that manifest as bilateral, multiple and recurrent pulmonary nodule affect mainly adult with no sex predilection [9]. Many patients are asymptomatic and few of them presents with non specific symptoms such as- cough, fever, dyspnoea, chest pain. The etiology of PHG is unclear; there have been reports of patients who were previously exposed to mycobacteria, aspergillosis, histoplasma prior to contracting PHG [10]. Many patients present with immunologic abnormalities such as elevated level of antinuclear antibody, anti rheumatoid factor, positive antiglobulin test.

Sclerosing mediastinitis, retroperitoneal fibrosis, rheumatoid arthritis, posterior uveitis, Sjogren's syndrome, hemolytic anemia, and other diseases are often associated with PHG [11]. Association of PHG with many lymphoproliferative diseases such as- lymphoma, castleman's disease have been described [12,13]. Many patients have elevated serum IgG4 and elevated tissue IgG4-positive plasma cells in the PHG [19].

Chest radiography and CT scan show solitary or often multiple randomly distributed unilateral and

bilateral pulmonary nodules with well defined border. Lesions which are without calcification are typically focal and the calcified mass are more often bilateral & multiple [14].Cavitation was reported in few cases. Nodular size range from few millimeters to 15 cms (average size 2 cms) [13]. Lymphadenopathy is typically absent in this cases [5].

The importance of PHG is that it is included in more common differential diagnosis such as tuberculosis, sacoidosis, histoplasmosis, plasma cell granuloma, wegner's granulomatosis [14,15]. Other important differential diagnoses are solitary fibrous tumor which have similar clinical and radiological findings and metastatic carcinoma (mucinous adenocarcinoma, thyroid carcinoma, choriocarcinoma) [14,16] .

F-18 fluordeoxyglucose positron emission tomography (FDG-PET/CT) can reveal increased metabolic activity in PHG lesions [17]; however accurate diagnosis can only be made with histopathological examinations [11]. Prognosis of PHG is generally excellent, single lesion can be cured by surgery and successful resolution of multiple nodules by administration of glucocorticoids have been described [5,13,18].

CONCLUSION

Pulmonary hyalinising granuloma, a usually benign condition, should be kept in mind when encountered with patients presenting with nonspecific chest symptoms and bilateral pulmonary nodules on chest radiographs.

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