SCLEROSING HAEMANGIOMA OF LUNG PRESENTING AS SPONTANEOUS HAEMOTHORAX

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Introduction

Sclerosing haemangioma was first described by Leibow and Hubbell [1] in 1956. This is a rare differential diagnosis of pulmonary nodule, which is much commoner in young females. Initially it was thought to be of endothelial origin but some recent electron microscopic studies have demonstrated surfactant apoprotein, their origin being suspected from type II pneumocytes. We present here a young lady of 37 years who presented with sudden spontaneous haemothorax and left lower lobe atelectasis due to nodular mass in perihilar area in lower lobe with absorption atelectasis. Diagnosis was made by histopathological studies after removal of the mass on surgery. We have not come across any case report of this lesion presenting as sudden massive spontaneous haemothorax and major lobar atelectasis.

Case Report

A 37 year old lady Mrs RB presented to a peripheral hospital with 2 days symptoms of left sided acute onset chest pain and low grade fever. Only relevant history was of her having suffered from left sided pneumonia 2 years prior to the present illness. At that time she was treated with parenteral antibiotics for 14 days. She was told that recovery was slower than expected but no other history or the chest radiograph of that time was available. At hospitalization she was found to have pallor (Hb-9gm%). There was no clubbing, cyanosis, peripheral lymphadenopathy or any other significant general examination finding. Her temperature was 99.8°F, with a pulse rate of 120/min regular. There was mild dyspnoea. Examination of the respiratory system revealed fullness on the left side of the chest, dullness on percussion in the left lower hemithorax with absence of breath sounds, and an area of bronchial breath sounds just above the dullness in left interscapular region. The chest radiograph revealed a homogenous opacity in the left mid and lower zones completely obliterating the left costophrenic angle (Fig 1).

A diagnosis of left sided pleural effusion was made and on aspiration 700 ml of frankly haemorrhagic fluid was aspirated. Haematocrit of fluid was 37%. The physician made a diagnosis of

haemothorax. Antibiotics were started and one unit of blood was transfused. Next day she was transferred to our hospital for further management. She continued to have left sided pain and on and off low-grade fever. She had pallor and respiratory system examination was as before, except for added sign of a localized wheeze in left interscapular area. Investigations revealed an Hb - 9.8gm%, ESR-02 mm, TLC - 9200/mm³ (P67, L28, E05, M02), platelet count was 1,55,000/cu mm, other coagulation profile including BT, CT, PT and PTTK was normal. Serum bilirubin was slightly raised 2.1 mg% with an indirect Van den Berg's test. Chest radiograph had not shown any significant change. Ultrasonography of the chest showed moderate amount of pleural fluid. Liver and spleen were normal. A CT scan of the thorax showed, a mass occupying the perihilar area of the left lower hemithorax with collapse of the left lower lobe and a minimal amount of pleural effusion (Fig-2). A MRI of the thorax showed it to be a non-vascular structure with no significant attachments to the mediastinum. Fibreoptic bronchoscopy showed external compression of left lower lobe bronchus. There was no intrabronchial lesion seen. FNAC of the mass lesion was performed under CT guidance, which revealed a bloody aspirate consisting mainly of alveolar macrophages, pneumocytes and neutrophils consistent with an inflammatory picture. Since the first FNAC did not provide a clear picture, FNAC was repeated which again showed the same findings. There were no malignant cells seen. Since a definitive diag-

Fig. 1 : Chest radiograph showing evidence of significant pleural fluid (left)

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Fig. 2 : Computed tomography of thorax showing the mass lesion (arrow), atelectatic left lower lobe and pleural fluid

nosis was not reached, she was taken up for open thoracotomy on the 7th day after admission. It revealed a 5x5 cm encapsulated looking cystic mass at the anterior end of oblique fissure which was adherent to the mediastinal and the diaphragmatic pleura. On cut section of the mass there was clotted blood and fimbrial looking tissue with adhesions between lung and parietes (Fig-3). The left lower lobe expanded well after the mass was excised. Histopathology of the mass was suggestive of a diagnosis of sclerosing haemangioma. It is possible that this mass had caused an obstructive pneumonia two years before as seen from the history, however in absence of any previous X-ray this cannot be definitely proved.

Discussion

Liebow & Hubbell [1] originally described sclerosing haemangioma in 1956. Katzenstein et al [2] in 1982 described 51 cases and noted that most patients were in the age group of 15-69 years with average age of 42 years, 84% of the patients were women and 78% were asymptomatic at the time of detection. They were generally discovered accidentally as rounded solitary pulmonary nodule on routine chest radiographs most of them being in the lower lung fields. Some patients had presented with vague chest pain and haemoptysis. Keengwa et al [3] in 1982 described 14 cases from Hong Kong, all in females and 7 were asymptomatic, detected as a coin lesion accidentally and others had cough or minimal chest pain and haemoptysis. Most were located near pleura. They rarely metastasize as only one case of Katzenstein had lesions in hilar nodes. These tumours are generally circumscribed and can be shelled out from the adjacent lung parenchyma. Initially an endothelial origin for the tumour was proposed (Liebow et al), but electron microscopic studies have now demonstrated surfactant apoprotein, now believed to be arising from the Type II pneumocyte [4-6]. Some of them have named them as pneumocytomas [3]. In the past, sclerosing

Fig. 3 : Microphotograph (x 40) of the lesion showing tubulo-papillary structures lined by low cuboidal epithelium. Numerous hyalinised blood vessels, inflammatory cells, a few fibroblasts and rounded histiocytes seen. Areas of haemorrhage and scattered hemosiderin pigment in stroma. Sclerosing haemangioma

haemangioma has been confused with plasma cell granuloma (histiocytoma), which is also called fibroxanthoma because of the fibrosclerotic tissue, cholesterol spaces and fat filled macrophages [7]. Degenerative changes occur in both tumours, the difference is in the absence of plasma cells in the former. Sclerosing haemangioma may involve adjacent lymph nodes but do not metastasize. As a rule conservative resection is generally curative and recurrences are unusual [2]. As already described in most of the published reports these cases have been asymptomatic radiographic detections or in some minimal cough or haemoptysis. We did not find any case presenting as massive spontaneous haemothorax and also atelectasis of a lobe due to external pressure as was in this case.

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