Case report

Pleural haemangioma: A rare cause of recurrent pleural effusion

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A middle aged female presented with recurrent unilateral pleural effusion. Thoracoscopy revealed a vascular tumor in the apical region of pleural cavity arising from the chest wall. Biopsy from the tumor showed features of pleural hemangioma. She was successfully managed by surgical excision of the tumor. The case is being presented because of its rarity.

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1. Introduction

Haemangioma is a rare congenital benign lesion and is thought to be associated with an imbalance of proangiogenic factors and angiogenesis inhibitors [1,2]. A haemangioma is a benign and usually self-involuting tumor (swelling or growth) of the endothelial cells that line blood vessels, and is characterized by increased number of normal or abnormal vessels filled with blood. Pleural hemangioma as a cause of recurrent pleural effusion has been reported rarely. We present case of a middle-aged female with recurrent pleural effusion who was diagnosed with benign pleural haemangioma on thoracoscopic biopsy.

1.1. Case summary

A 45 year old non-smoker female presented with complaints of dyspnea, dry cough and left sided chest pain for the past one and a half years. She gave history of recurrent re-filling and multiple thoracentesis over this period. There was no past history of tuberculosis. There was no history of fever, joint pains and weight loss. There was no history suggestive of chronic use of any medications. She was housewife by occupation. On examination, her vital signs were normal. Respiratory system examination showed findings suggestive of left sided bulging with decreased movements of left side, stony dull note on percussion and decreased vocal resonance suggesting left sided effusion. Her chest radiograph was suggestive of massive left pleural effusion with left upper zone rounded opacity (Fig 1). Pleural fluid aspiration was done which was straw colored and had a cell count of 150 cells/cubic mm, 60% polymorphonuclear cells and 40% mononuclear cells, glucose-131 mg/dl, protein-4400 mg/dl, ADA-7.72U/L, LDH-98U/L. Gram's and ZN stains were negative for bacteria and acid fast bacilli. Cytology for malignant cells was negative. Computed tomography revealed a well defined heterogeneously enhancing mass along the posterior chest wall in left upper hemithorax with massive left sided effusion (Fig 2) MRI was done to rule out chest wall origin or invasion and it suggested a well defined lesion in the D4, D5 paravertebral region? neurogenic tumor with massive left side pleural effusion with left lower lobe collapse.

Thoracoscopy was done using a rigid thoracoscope (Karl Storz). A large tumor arising from the posterior chest wall of the apical region was seen. The tumor was highly vascular and bled on touch. (Fig. 3) Biopsy from the tumor was taken with electrocautery assistance to prevent excessive bleeding. Biopsy was suggestive of pleural hemangioma. (Fig. 4).

Patient was referred for surgical management. Left posterolateral thoracotomy with excision of the tumor was done and intercostal drain was placed. The drain was removed after 4 days of surgery when any fluid ceased to drain through it. There was no re-accumulation of pleural fluid on follow-up chest skiagram after 14 days (Fig. 5). Patient is in our regular follow up and she is absolutely normal.

2. Discussion

Haemangioma is a rare congenital benign lesion and is thought to be associated with an imbalance of proangiogenic factors and angiogenesis inhibitors [1,2]. Hemangiomas constitute 7% of all benign tumours [3]. Hemangiomas are generally seen in liver, bone, soft tissue and lung [4]. Pulmonary hemangiomas of lung, chest
wall (rib or muscle) and mediastinum have been reported [5–9] but pleural hemangioma has been reported rarely [4,10]. Majority of such hemangiomas are discovered incidentally or when spontaneous rupture results in hemorrhage and hemorrhagic pleural effusion [10]. We are presenting a patient of left sided pleural hemangioma, which presented with recurrent massive and straw pleural effusion.

Approximately 80–90% of haemangiomas develop before the age of 30 [7] but our patient presented at the age of 45 years.

Diagnosis of haemangioma depends primarily on imaging and pathological examinations. The most widely used imaging studies include computed tomography and MRI [9,11]. The previous reported case of pleural hemangioma was diagnosed on exploration.

Fig. 1. Chest X ray of patient showing left sided effusion with left upper zone rounded opacity with intercostal drain in situ.

Fig. 2. Chest computed tomogram showing heterogeneously enhancing mass lesion along posterior chest wall in left upper lobe and left sided moderate pleural effusion.

Fig. 3. Thoracosopic appearance of a large vascular tumor arising from posterior chest wall.

Fig. 4. Histopathological slide showing hemangioma.
In our case, the diagnosis was achieved on thoracoscopic biopsy. The treatment of haemangioma should be individualized, depending on the location of the tumour mass, the depth of its infiltration, and the age and cosmetic requirements of the patient. Comprehensive treatment strategies are recommended, including dry ice cryotherapy, radiotherapy, steroid treatment, sclerosing agent injection, vascular ligation, vascular embolism and surgical excision [12,13].

We managed our patient with surgical excision of the tumor. To conclude, in patients presenting with recurrent pleural effusion, possibility of benign tumors like hemangioma should be kept in mind and investigated. Thoracoscopy can be a useful and easy tool to confirm etiology of such benign tumors.

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