Hemorrhagic Pleural Effusion Due to Pleural Hemangioma

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Abstract
We report a rare case of isolated pleural hemangioma presenting as recurrent, haemorrhagic pleural effusion. Chest radiograph and computed tomography of chest was suggestive of left apical tumour. Diagnosis of hemangioma was revealed only after thoracotomy and excision of the tumour mass.

INTRODUCTION
Hemangiomas are neoplasms characterized by an increase in number of normal or abnormal vessels. It may be difficult to distinguish them from arterio-venous malformations or hamartomas. Majority of hemangiomas are superficial lesions often on the head and neck, but may occur internally with nearly one-third in the liver and very rarely as pleuropulmonary hemangiomas. When pleural hemangiomatosis is diffuse it may be associated with congestive cardiac failure and consumptive coagulopathy. Hemangiomas constitute 7% of all benign tumours.

CASE REPORT
A 55-year old male farmer, non-smoker presented with symptoms of dry cough and dyspnoea on exertion of one-month duration. On general examination the vital parameters were normal. Respiratory system examination revealed increased volume of the left hemithorax with markedly reduced breath sounds. Cardiovascular, abdominal and neurological examinations were unremarkable. Chest radiograph revealed a massive left sided pleural effusion (Fig. 1). Hematological investigations revealed hemoglobin of 15.2 gram% with total white blood cell count of 8500 per cu mm, 85% polymorphs, 7% lymphocytes, 3% eosinophils and 5% monocytes. Other biochemical parameters were within normal limits. Pleural fluid was hemorrhagic with proteins of 2.8 gram%, red blood cells of 28,500 per cu mm, white blood cells of 30 per cu mm. There were no pyogenic or acid-fast bacilli seen on microscopy. Pleural fluid adenosine deaminase was elevated to 73.6 IU/L (N< 40 IU/L). Pleural fluid did not show presence of malignant cells. Pleural biopsy revealed only fibrin with occasional clusters of mesothelial cells. The ultrasonography of the abdomen was normal. An intercostal tube was inserted in left 5th intercostal space in the anterior axillary line. Chest radiograph following the intercostals tube drainage revealed a homogeneous opacity in the left upper zone (Fig. 2). On computed tomography (CT) of the thorax using contrast, malignant lesion in left apical region was suspected (Fig. 3). Fibreoptic bronchoscopy was normal. Transbronchial lung biopsy as well as CT guided fine needle aspiration cytology (FNAC) of the mass was inconclusive. Hence, as a diagnostic and a therapeutic option and patient was subjected to thoracotomy. It revealed an extraparenchymal pleural tumour, which was excised and sent for histopathological analysis. On histopathology, the parietal pleura showed marked thickening with proliferating fibroblasts, capillaries and marked mixed inflammatory cells consisting of polymorphs.
and lymphocytes. Section from the tumour mass showed fibro-muscular capsule and the tumour consisted of numerous vascular channels of varying sizes with ill-defined muscle layer in their walls. Few of the vessels revealed proliferating endothelial cells obliterating their lumen. These vascular spaces were separated by loose fibromyxoid and fibro-fatty stroma with sparse inflammatory cells with foci of hemorrhage and calcification. Thus, a diagnosis of pleural hemangioma was made.

DISCUSSION

Pleuropulmonary hemangiomas may be congenital or may be as a part of Von-Hippel Lindau disease (capillary hemangiomatosis). The epidemiology is rare and the pathogenesis is unknown. The age of presentation has a wide range with cases reported from two months to 76 years. However maximum cases occur in young adults before the age of thirty-five years. Majority of such hemangiomas are discovered incidentally or when spontaneous rupture results in hemorrhage and hemorrhagic pleural effusion as seen in our case. They may present with non-specific complaints like cough, chest pain and dyspnoea or symptoms due to compression of adjacent structures like superior vena cava or neurological symptoms secondary to intraspinal extension. Calcification on plain radiograph is reported only in 10% of cases. They can present as diagnostic dilemmas as in our case and may be evident only after exploration. These tumours are usually well encapsulated and heterogeneous in appearance. Only a few cases show homogenous density as in this case. On gross examination, they appear as bluish red masses, which are soft, spongy and may be pulsatile in situ. Mass of blood vessels may be capillary, arterial, venous, or mixed with or without thrombosis and recanalisation.

Pleuropulmonary hemangiomas are curable with resection or embolisation. Corticosteroids and radiation therapy have been reported to shrink hemangiomas. Cyclophosphamide can be used in cases that do not respond to prednisolone or radiation therapy. Pleuropulmonary hemangiomas, which are benign tumours, are different from sclerosing pulmonary hemangiomas which present with cough chest pain or hemoptysis found predominantly in young females and have a malignant potential.

To conclude, tumours of benign etiology should always be kept in mind while treating hemorrhagic pleural effusion.

REFERENCES

3. Davis JM. Benign blood vascular tumours of the mediastinum.

**Announcement**

Nominations are invited to fill the vacancy for the post of Governing Body Member created due to the resignation of Dr. Shashank R Joshi, who has been nominated as Editor of the Journal of Association of Physicians of India (JAPI). Those members who are interested and have continuous membership of API for one year are eligible. The application should be duly proposed and seconded by two valid members of API and reach the Hon. General Secretary, Association of Physicians of India, Laud Mansion, 3rd Floor, MK Road, Mumbai - 400 004 on or before **30th June, 2003**.

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