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"CASE REPORT OF PLEURAL CAPILLARY HEMANGIOMA: A RARE CAUSE OF RECURRENT UNILATERAL HEMORRHAGIC PLEURAL EFFUSION WITH BONY EROSION, COUNTERFEITING AS A MALIGNANT PLEURAL MASS – FIRST CASE REPORT ON PLEURAL CAPILLARY HAEMANGIOMA CAUSING RIB EROSION"

Pathology

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ABSTRACT

Background: A non-smoker male who was apparently normal before the present illness and without any significant past medical and surgical history presented with sudden onset dyspnoea, chest pain and recurrent haemorrhagic pleural effusion. Imaging was suggestive of malignant neoplasm of pleura, but on histopathological examination of pleural mass, capillary haemangioma was diagnosed. Capillary hemangioma is an extremely rare benign neoplasm of pleura that clinically and radiologically mimics malignant neoplasm of thorax. However, preoperative biopsy procedure may be catastrophic to the patient due to risk of life threatening bleeding episodes. So, pleural haemangioma should always be kept in the differential diagnosis of recurrent unilateral haemorrhagic pleural effusion.

KEYWORDS

Non Smoker, Pleural Effusion, Capillary Hemangioma, Malignant Neoplasm of Thorax.

INTRODUCTION

Pleural tumors are small but significant subset among thoracic malignancies. They constitute 0.3% to 3.5% of thoracic tumours. (1) These are divided into benign and malignant pleural neoplasms. Mesothelioma, metastasis, hematolymphoid and mesenchymal neoplasms constitutes the main differential diagnosis pleural neoplasms. (2) Malignant mesothelioma accounts for more than 90% of primary pleural tumours. Of the remaining 10%, approximately5% are constituted by Solitary Fibrous tumours of pleura and remaining 5% includes a variety of less common primary pleural tumours, such as lipomas, lymphomas, thymomas, melanomas, vascular neoplasms, sarcomas and other rare neoplasms.(1,3)

Here, we present a case of pleural capillary hemangioma, an extremely rare benign mesenchymal tumor of pleura, counterfeiting as a malignant pleural mass clinically and radiologically. It is the first case report on pleural capillary haemangioma causing rib erosion.

CASE SUMMARY

A 46 years old male who was apparently alright 4 months back, presented with complaints of chest pain, cough and dyspnea for 4 months which was more during walking. The symptoms increased in severity over the past 12 days. He had undergone repeated thoracocentesis for recurrent hemorrhagic pleural effusion during this period. The patient had no history of fever and weight loss. There was no past history of tuberculosis, other chronic illnesses and use of long-term medications. On physical examination, his vital signs were within normal limit. On respiratory examination, he had reduced chest movements on left side with dull note on left side chest on percussion suggestive of left pleural effusion. Contrast Enhanced Computerized Tomography chest scan showed heterogeneously enhancing pleural based nodular lesion with irregular borders and areas of necrosis along the posterior pleura of left side measuring 38 x 30 x 30 mm causing left 7th rib erosion with moderate to severe left side pleural effusion, suggestive of neoplastic mass. Mediastinal lymphadenopathy was also noted in CT scan. Pleural fluid examination showed a cell count of 300 cells/cubic mm, 25% polymorphonuclear cells, 70% lymphocytes and 5% other cells, total protein- 5.1 g/dl, ADA-9.7 U/L. Gram's and ZN stains were negative for bacteria and acid fast bacilli. Pleural fluid cytology showed signet ring like cells and scattered atypical cells. However, ultrasonography guided biopsy of pleural lesion was done which was suggestive of a benign angiomatous lesion, most likely capillary haemangioma. Immunohistochemical studies were performed, showing positivity for CD31 and factor 8, and a Ki67 proliferation index <5%, yielding a definitive diagnosis on pathology of pleural capillary haemangioma.

The patient then undergone left posterolateral thoracotomy and excision of pleural mass along with excision of segment of 7th rib. Final diagnosis on histopathological examination of resected specimen was suggestive of Capillary haemangioma with proliferating neoplastic vascular channels abutting the bone in excised rib sections. After surgical management, there was no re- accumulation of pleural fluid on follow-up chest skiagram. Patient is in our regular follow up and he is absolutely normal.

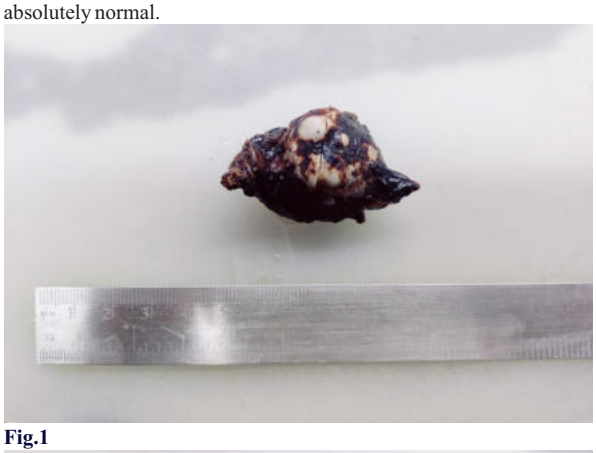

Fig.1

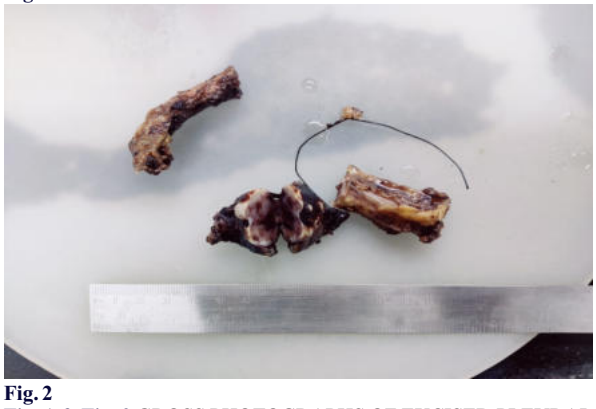

Fig. 2

Fig. 1 & Fig. 2 GROSS PHOTOGRAPHS OF EXCISED PLEURAL SOFT TISSUE MASS AND SEGMENT OF 7TH RIB

Fig. 3 Cut Surface Of The Pleural Lesion

DISCUSSION

The benign mesenchymal tumors of mediastinum ranges from the most common lipoma to extremely rare pleural hemangioma. (4) Haemangiomas constitute 7% of all benign tumours. (5) They result from an imbalance of proangiogenic factors and angiogenesis inhibitors, characterized by increased number of normal or abnormal vessels filled with blood due to proliferation of endothelial cells lining the blood vessels. (6,7). Haemangiomas are generally seen in skin, liver, bone, soft tissue and lung. Hemangiomas of thorax (lung, chest wall, ribs and mediastinum) have been reported but pleural haemangiomas are extremely rare reported cases. (3, 8-13). Most hemangiomas are observed in young adults, before the age of 35 years (5), however in our case report, patient is 46 years old. Pleural haemangioma may be unifocal or multifocal. (14) They are usually found by chance or after spontaneous rupture causing bleeding or haemorrhagic pleural effusion. Fifty percent of pleural hemangiomas are asymptomatic and diagnosed incidentally on imaging. Rest present as chest pain, cough, dyspnea (due to compression of adjacent organs). Rarely, it presents as spontaneous hemothorax due to rupture of tumor, hemorrhagic pleural effusion and dysphagia, superior venacava syndrome and neurological complications from intra spinal tumor extension. Pleural hemangioma is a rare cause of adjacent bony erosion due to pressure effect. (15) In our case, presence of features such as recurrent haemorrhagic pleural effusion, irregular mass with areas of necrosis, bony erosion, mediastinal lymphadenopathy on chest CT scan and atypical cells on pleural fluid cytology were inferring the lesion as malignant but USG guided biopsy with IHC study led to the diagnosis of pleural capillary haemangioma, however it could lead to life-threatening bleeding in our patient.

Diagnosis of pleural haemangioma depends on radiological and histopathological examination. An extrapulmonary mass with benign features and typical centripetal enhancement pattern on contrast CT should raise a suspicion of pleural haemangioma. (16) When imaging features are suggestive of pleural haemangioma, preoperative biopsy is not recommended as it may lead to torrential bleeding. Histopathological and immunohistochemical examination of excisional biopsy is gold standard. (17)

It is important to differentiate pleural haemangioma from other uncommon tumours that may present as primary lesions of the mediastinum. The conditions that may be confused with pleural haemangiomas include lymphangiomas, sclerosing haemangioma, and haemangioendothelioma. Lymphangioma may be distinguished from haemangioma by the presence of ectatic lymphatics filled with chyle and a prominent inflammatory interstitial lymphoid infiltrate distributed haphazardly within the collagenous stroma. Epithelioid haemangioendothelioma, a recently described variant of vascular neoplasm of low or borderline malignant potential, are the most important differential diagnosis of pleural haemangioma. Epithelioid haemangioendothelioma can grow in a solid pattern similar to capillary haemangiomas; however, the cells are characterized by abundant eosinophilic cytoplasm with prominent cytoplasmic vacuolization and occasional abortive intracytoplasmic lumen formation. The lack of the characteristic cytoplasmic vacuolization, cellular atypia, and abundant eosinophilic cytoplasm distinguishes capillary haemangiomas from these tumours. (18)

The most usual approaches for management are surgical resection (as in our patient), cryotherapy, embolization or vascular ligation, especially when the pleural haemangioma is unifocal. Drug therapy including corticosteroids and cyclophosphamide have also been used in multifocal pleural haemangiomas. For lesions that are symptomatic or compromising vital structures, surgical excision remains the treatment of choice.(14,18) It is unusual for haemangiomas to relapse. (19) Our patient also has not had any recurrence of pleural effusion since surgery.

CONCLUSIONS

Capillary haemangioma in the pleura is extremely rare. For recurrent haemorrhagic unilateral pleural effusion, the possibility of benign tumour such as haemangioma should be considered. Imaging is an important method for diagnosis to avoid catastrophic bleeding during preoperative procedure of biopsy. Awareness of this possibility is important to reduce the time-to-diagnose and proper management, though rarity in its incidence in pleura. For localized haemangioma, surgical resection is the most common approach. While for multifocal pleural capillary haemangioma, individualized and multimodal treatment approach may be necessary, including dry ice cryotherapy, sclerosing agent injection, radiotherapy, vascular embolism, surgical excision and drugs therapy including corticosteroids. Immunosuppressive agents may also be potential therapeutic drugs. (20) Despite the rarity of these lesions, awareness of their histopathological features in the mediastinum is of importance to avert the possibility of incorrect diagnosis. Hence, reporting of our case may be helpful to improve the clinical and therapeutic strategy.

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