

PLEURAL HAEMANGIOMA – A CAUSE OF RECURRENT PLEURAL EFFUSION : CASE REPORT

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Dr. Ashish Deshmukh, primarily evaluated the patient, did pleuroscopy and biopsy.

Dr. Ananad Deodhar performed surgical excision of the mass.

Dr. Dinesh Kulkarni processed the surgically removed specimen and did histopathology report.

All authors have collected references, wrote and corrected manuscript and approved the same.

ABSTRACT

A middle aged female presented with recurrent left sided pleural effusion. Pleuroscopy revealed a well circumscribed pleural mass. Biopsy from that mass revealed features of capillary haemangioma. She was successfully treated by surgical excision of the pleural mass. The case is being reported for the extreme rarity of haemangioma in pleura.

KEY WORDS

Recurrent pleural effusion, unilateral, pleuroscopy, pleural mass, capillary haemangioma

INTRODUCTION

Haemangioma is a benign tumour which can occur externally on skin, mucus membrane and also in internal organs, arises from endothelial cells and is characterized by increased number of normal or abnormal vessels filled with blood. Pleural haemangioma as a cause of recurrent pleural effusion is unusual. (1) Pleural hemangioma is a rare and its incidence being observed in patients mostly below 35 years of age with no gender predilection. It's pathophysiology is associated with an imbalance of proangiogenic factors and angiogenesis inhibitors (2) They may be asymptomatic or can present as incidental findings or with mild symptoms of cough, chest pain. If it presents as a solitary nodule on imaging, it may mimic carcinoma in adults. The diagnostic dilemma with pleural effusions resolves after the explorative CECT or thoracoscopy. Radiologically, these tumours are well-defined, variably dense and occasionally cystic masses. However, histopathology of the lesion remains gold standard for confirmation. CD 4 positivity for endothelial cells is detected on immunohistochemistry. Pleural haemangiomas are not to be confused with sclerosing pulmonary hemangiomas which have a malignant potential. Management of pleural haemangioma depends on it's location, size, depth of infiltration, age of the patient. Various non-surgical options available are radiotherapy, dry ice cryotherapy, steroid treatment, sclerosing agent injection. Vascular ligation, vascular embolization, and ultimately surgical excision are considered depending on the above-mentioned factors. Imaging also plays a role in follow-up to rule out any residual tumour or recurrence. (3)

We present case of a middle-aged female having recurrent pleural effusion with pleural mass and diagnosed as capillary haemangioma on pleuroscopic biopsy as well as excised mass.

CASE REPORT

A 43 year old female clinically came with complaints of breathlessness, cough and left (Lt) sided chest pain for about two years. She had history of hospitalization three times for recurrent (Lt) sided pleural effusion. She didn't have history of fever, joint pains, weight loss or hemoptysis. She was a house wife from rural area and was neither a smoker nor alcoholic. She didn't have past history of tuberculosis. On examination her vitals were normal and chest examination revealed features of (Lt) sided pleural effusion and confirmed on x-ray chest. Pleural fluid aspiration was done and sent for cytology examination. The pleural fluid was straw coloured with total cell count of 173 cells/cmm with 62% neutrophils and 38% lymphocytes. Gram stain didn't reveal organisms, ZN stain was negative for acid fast bacilli. It was negative for malignant cells. Fluid biochemical parameters revealed glucose 130 mg%, proteins 2.8 gm%, Adenosine Deaminase (ADA) 8.3 U/L. She was referred for pleuroscopy which revealed a large mass at apical region and biopsy was taken (Fig 1), it was highly vascular and electrocautery was done to avoid bleeding. The biopsy showed features of capillary hemangioma. Surgical excision was planned. After proper fitness and under general anasesthesia, left posterolateral thoracotomy was done and the tumour mass was excised. Intercostal drain was removed on fourth postoperative day. There was no refilling of pleural cavity. She is on regular follow up and asymptomatic.

The mass was well circumscribed, grayish, 4x3x3 cm, soft to firm (Fig. 2). Cut surface was grayish with haemorrhages. Sections from the mass revealed tumour mass composed of plenty of capillaries of variable sizes, lined by endothelium and RBCs in their lumina (Fig. 3) and fibrous stroma and was reported as capillary haemangioma

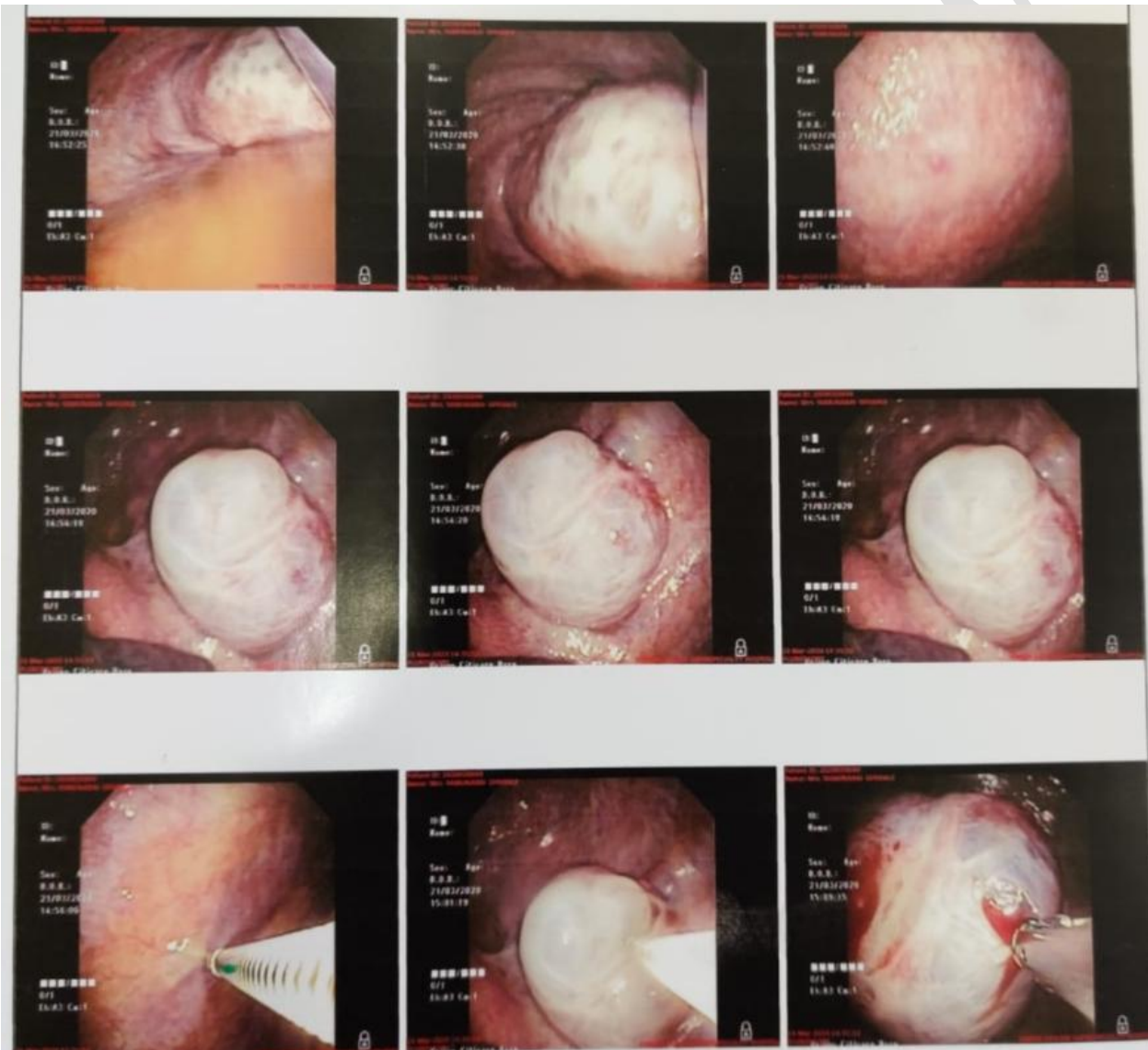


Fig.1 Pleuroscopy showing a well circumscribed mass on pleural surface and biopsy taken .



Fig. 2 shows gross appearance of excised pleural mass

UNDER PEER REVIEW

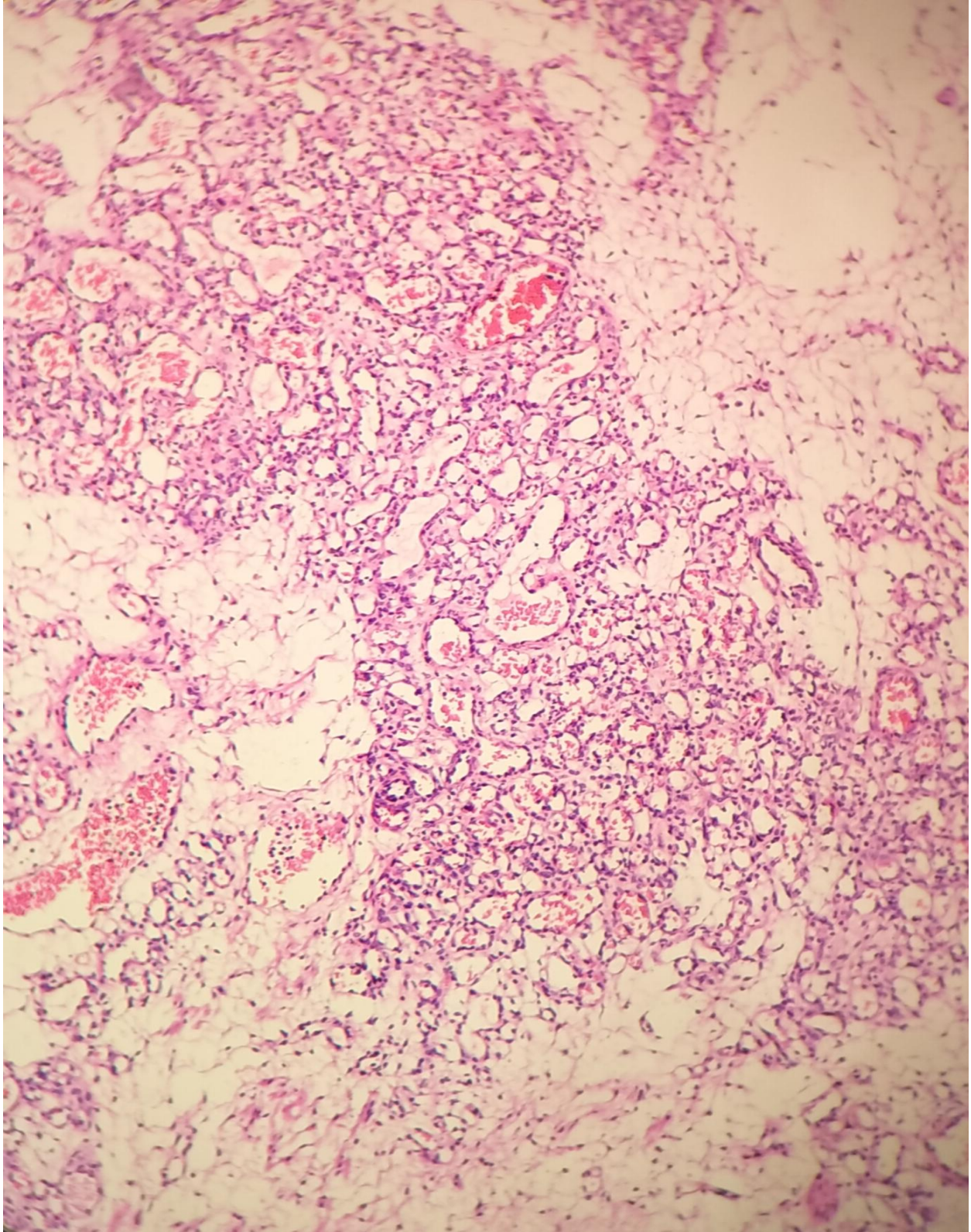


Fig. 3 from microscopic section from mass showing plenty of small thin capillaries lined by endothelium and containing RBCs in their lumina (H & E 10x X 10x)

DISCUSSION

Haemangioma is a rare benign neoplasm and can present since birth. They constitute about 7% of all benign neoplasms. It can occur externally on skin and also be seen in internal organs.(2) Capillary hemangiomas are observed in skin, mucus membrane whereas cavernous hemangiomas are generally seen in liver, bone, soft tissue and lung. Hemangiomas of lung, chest wall (rib or muscle) and mediastinum have been reported, but pleural hemangiomas are quite rare. Majority of pleural haemangiomas are discovered incidentally or when they rupture and result in hemorrhagic pleural effusion. Our patient of pleural hemangioma presented clinically with recurrent (Lt) sided pleural effusion. Approximately 80 - 90% of haemangiomas develop before the age of 30 but our patient presented at the age of 43 years. Diagnosis of haemangioma depends primarily on imaging and pathological examinations. The most widely used imaging studies include computed tomography and MRI. The previous reported case of pleural hemangioma was diagnosed on exploration. In our case, the diagnosis was made on pleuroscopic biopsy of mass and histological confirmation. As haemangioma was never our differential diagnosis. Diagnostic biopsy was done, fortunately there was little bleeding. 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. We managed our patient with surgical excision of the tumor.

The take home message is (1) An extrapulmonary mass with benign features and typical centripetal enhancement pattern on contrast CT should raise a suspicion of pleural hemangioma to be added in the differential diagnoses.

- (2) Awareness of this possibility is important to reduce the time to diagnose and proper management, though rare in its incidence in this location.
- (3) When imaging features are suggestive of pleural hemangioma, preoperative biopsy is not recommended as it may lead to torrential bleeding.(4)

CONCLUSION

We recommend, in patients presenting with recurrent pleural effusion, possibility of benign tumors like hemangioma to be kept in mind and investigated. **Pleuroscopy** can be a useful and easy tool to confirm etiology of recurrent effusion. This is a case of capillary hemangioma located in the pleura, an extremely rare location, accompanied by recurrent pleural effusion, an unexpected presentation in this type of tumor.

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