Case Report

Mediastinal Hemangioma Mimicking as Bronchial NET

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Abstract

We present a rare case of mediastinal capillary hemangioma in a 54-year-old female. She presented with back pain in the left suprascapular region, and the chest radiograph revealed left pleural effusion. On further workup with high-resolution computed tomography (CT) chest, a hypervascular pleural-based neoplastic lesion in the left upper hemithorax with gross left pleural effusion and subtotal collapse of the left lung was identified. ^{18}F-fluorodeoxyglucose positron emission tomography/CT was suggestive of a weakly metabolic well-defined pleural-based soft tissue lesion in the left upper hemithorax along the mediastinal side. Neuroendocrine tumor was suspected. ^{68}Ga-DOTATATE PET/CT was advised, which showed intense uptake in the lesion. The mass was resected completely. Histopathological examination established the final diagnosis as benign vascular tumor consistent with a capillary hemangioma.

Keywords: *^{68}Ga-DOTATATE positron emission tomography/computed tomography, capillary hemangioma, mediastinal carcinoid, mediastinal mass, neuroendocrine tumor, pleural effusion*

Introduction

Somatostatin receptor (SSTR) scintigraphy is an established modality for imaging well-differentiated neuroendocrine tumors (NETs). It is known that nonmalignant inflammatory diseases (e.g., tuberculosis) may also accumulate SSTR analogs. Mediastinal hemangioma showing intense SSTR expression and mimicking NET has not been reported earlier to the best of our knowledge. We report a case of mediastinal capillary hemangioma manifesting as pleural effusion, successfully managed by surgical excision.

Case Report

A 54-year-old female presented to the outpatient department with the complaints of the left upper back pain for 4 months, initially relieved on conservative management. On recurrent and nonrelieving symptoms, she underwent a chest X-ray. Chest X-ray revealed large left-sided pleural effusion, for which pleural tapping was done. On needle thoracocentesis, straw-colored fluid was aspirated. Evaluation of pleural fluid cytology was negative for malignant cells. Single-phase contrast-enhanced computed tomography (CT) revealed a hypervascular pleural-based neoplastic lesion in the left hemithorax with gross left pleural effusion and subtotal collapse of the left lung. Noncontrast ^{18}F-fluorodeoxyglucose positron emission tomography/CT (^{18}F-FDG PET-CT) was suggestive of a weakly metabolically active well-defined soft tissue nodular lesion in the left upper hemithorax along the mediastinal side, and NET was suspected [Figure 1].

^{68}Ga-DOTATATE PET/CT was advised, which showed intensely increased SSTR expression in a well-defined heterogeneously enhancing pleural-based soft tissue nodular lesion in the left upper hemithorax, with areas of necrosis within [Figure 2]. It measured 4.3 cm × 3.6 cm in the maximum transverse dimensions and 5 cm in superoinferior extent, primary NET was suspected and histopathological correlation was advised.

The patient was planned for surgical excision and underwent thoracotomy with complete excision of the lesion with decortication and mediastinal lymph node excision (level 5 and level 9). Postoperative recovery period was uneventful. Postoperative chest radiograph showed minimal left pneumothorax.

Gross pathological examination of the tumor showed well-circumscribed lobulated neoplasm. Microscopic examination

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revealed many capillary sized, dilated, and congested thin-walled vessels lined by a single layer of endothelial cells and few large caliber arterial blood vessels. Many vessels showed fibrin thrombi. There was no evidence of significant atypia or mitotic activity. The stroma was edematous and showed areas of hemorrhage and occasional hemosiderin-laden macrophages. Immunohistochemistry was performed; endothelial cells expressed CD34, FLI-1, and Erythroblast transformation-specific [ETS]-related gene (ERG). The Mib 1 labeling index was 8%–10%. These findings were suggestive of benign vascular tumor consistent with a capillary hemangioma [Figure 3].

The patient was followed up in the outpatient department and was doing well.

Discussion

Hemangiomas are benign vascular tumors resulting from abnormal growth of blood vessels. They are proliferation of blood vessels of varying diameters with thin and uniform fibromuscular walls and lined by a single layer of benign cuboidal epithelium. They have varying amounts of stromal elements (e.g., fibrous tissue, fat, and myxoid elements).

Mediastinal hemangiomas are rare tumors, constituting <0.5% of all the mediastinal masses.[1,2] They share common histological subtypes with other vascular tumors in the body. These could be capillary, cavernous, venous, arteriovenous malformation, angiofibroma, angiolipoma, glomus tumor, hemangioendothelioma, and hemangiopericytoma. True hemangiomas are benign, but these should be differentiated from potentially malignant vascular tumors such as hemangioendothelioma and hemangiopericytoma via histological examination. Overall capillary and cavernous types account for >90% of the cases.[3]

These typically occur in young patients (<35 years accounting for 75% of the cases).[2] Approximately 50% of the cases are asymptomatic and detected incidentally, while others present with nonspecific complaints of cough, chest pain, fever, dyspnea (as a result of compression of adjacent structures), respiratory distress, hemoptysis, and superior vena cava syndrome.[1,4,5] They can be classified further based on their location, into anterior (most common), middle or posterior mediastinum.[6]

Typical CT findings include heterogeneously enhancing, hypervascular, well-circumscribed mass, with occasional case reports of infiltrative appearance. Multiple punctuate calcifications and phleboliths can be seen in 21% and 7% of cases, respectively. A study by McAdams *et al*. showed four distinct patterns of heterogeneous attenuation: central, mixed central and peripheral, peripheral, and nonspecific increased attenuation.[7] Chest CT is useful in assessing the extent of the lesion before planning for surgical intervention. Magnetic resonance imaging could be useful; however, no preoperative investigations are diagnostic.

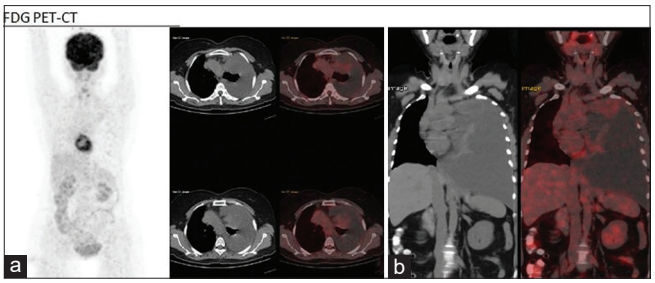

Figure 1: ^{18}F-fluorodeoxyglucose positron emission tomography/computed tomography maximum intensity projection and axial (a) and coronal (b) showing a weakly metabolically active pleural-based soft tissue mass in the left hemithorax

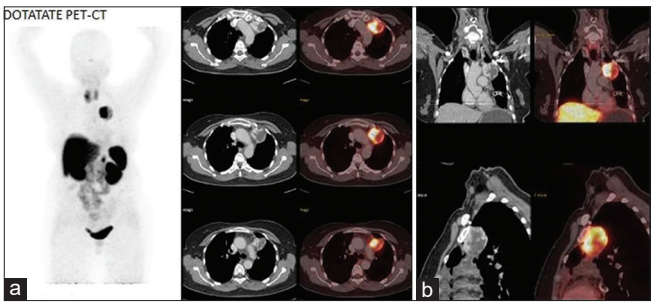

Figure 2: ^{68}Ga-DOTATATE positron emission tomography/computed tomography maximum intensity projection and axial (a) and coronal and sagittal (b) showing intense somatostatin receptor expression in a well-defined heterogeneously enhancing pleural-based soft tissue nodular lesion in the left upper hemithorax, with areas of necrosis within

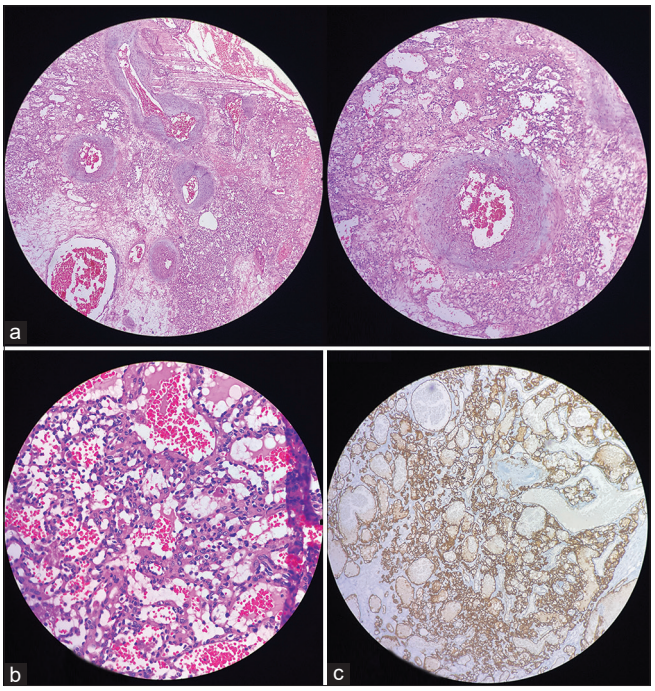

Figure 3: Microscopy images scanner view. (a) Admixture of varying sized vessels, high-power view. (b) Anastomosing capillary sized vascular channels and immunohistochemistry. (c) Endothelial cells staining positively with CD34

Other differentials such as Castleman disease, paraganglioma, vascular malformation, ectopic parathyroid

adenoma, and hypervascular metastasis should be considered when hypervascular anterior mediastinal masses are encountered.[6]

Transthoracic needle aspiration biopsy is reported to be of little diagnostic use.[8] Surgical excision is the recommended choice for diagnostic and therapeutic purpose. However, there is a potential complication of substantial blood loss. According to Cohen *et al*., subtotal resection is a therapeutic option for infiltrative cases, without increased risk of local recurrence, hemorrhagic morbidity, malignant transformation, or becoming symptomatic.[1] Other treatment modalities including radiotherapy and venovenous extracorporeal membrane oxygenation have been reported in critical cases.[5,9]

^{68}Ga-DOTATATE PET/CT is an established method for the evaluation of NETs. It is used for SSTR imaging, which is overexpressed in various tumors, such as NETs (carcinoid, insulinoma, and pheochromocytoma), meningioma, pituitary adenoma, Merkel cell carcinoma, and medullary thyroid carcinoma. There are very few benign differentials for uptake of SSTR analogs. Low-grade uptake is known in reactive nodes, prostatitis, sarcoidosis, and other inflammatory conditions, as activated macrophages and lymphocytes express SSTRs on their surface.[10] There are few case reports demonstrating somatostatin receptor expression in vertebral, intraosseous, primary nodal hemangiomas, in which the exact mechanism of uptake is unclear.[11-15] ^{68}Ga-DOTATATE uptake in bone hemangiomas is well-known; however, its uptake in a mediastinal capillary hemangioma is worth knowing as it might change the clinician's perspective.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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