Radiology Case Reports 15 (2020) 2519–2521

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Case Report

Multiple pulmonary myofibromas mimicking metastatic disease in an adult patient

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ARTICLE INFO

Article history:
Received 13 September 2020
Revised 23 September 2020
Accepted 25 September 2020

Keywords:
Myofibroma
Spindle cell neoplasm
Pulmonary nodules

ABSTRACT

We report a case of multiple bilateral pulmonary myofibromas in an asymptomatic 76-year-old man, who was referred to our institution for investiagtion of incidentally discovered pulmonary nodules, orginially suspected to represent pulmonary metastases. Myofibromas are unusual benign neoplasms, infrequent in adults, and rarely affecting the lungs. Pulmonary neurofibromas can mimic lung metastases and their diagnosis requires histopathological assessment.

Introduction

Myofibromas, also referred to as myofibroblastic tumors and inflammatory pseudotumors, are rare benign neoplasms composed of contractile myoid cells arranged around thin-walled branching hemangiopericytoma-like blood vessels. Immunohistochemistry is usually used to differentiate myofibromas from other spindle-cell tumors [1].

Case presentation

A 76-year-old male nonsmoker was referred to our institution for investigation of bilateral pulmonary nodules incidentally discovered on routine chest radiograph. Reportedly, the patient has had a transthoracic needle biopsy, with histopathological report from another institution describing a spindle cell neoplasm. A contrast enhanced chest computed tomography (CT) was performed, demonstrating several bilateral 2- 15 mm well-defined, solid pulmonary nodules with peripheral enhancement (Fig. 1). Subsequent positron emission tomography showed hypermetabolic activity in the largest nodule in the right lower lobe, with SUV of 4.9 (Fig. 2). CT-guided transthoracic needle biopsy of this hypermetabolic nodule was performed next. Histopathological report of the biopsy sample described clusters of cells in delicate fibrovascular stroma, small nuclei with little pleomorphism and no mitotic activity, and absence of necrosis, compatible with myofibroma.

Surgical excision of the largest nodule was done next. Histopathological findings of the excised lesion confirmed benign pericystic neoplasm with muscle differentiation, and immunohistochemical profile: HMB45, ERG, TTF1, Napsin, CD56, chromogranin, synaptophysin, P40, CK 5/6, S100, AE1/AE3, CK

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https://doi.org/10.1016/j.radcr.2020.09.044

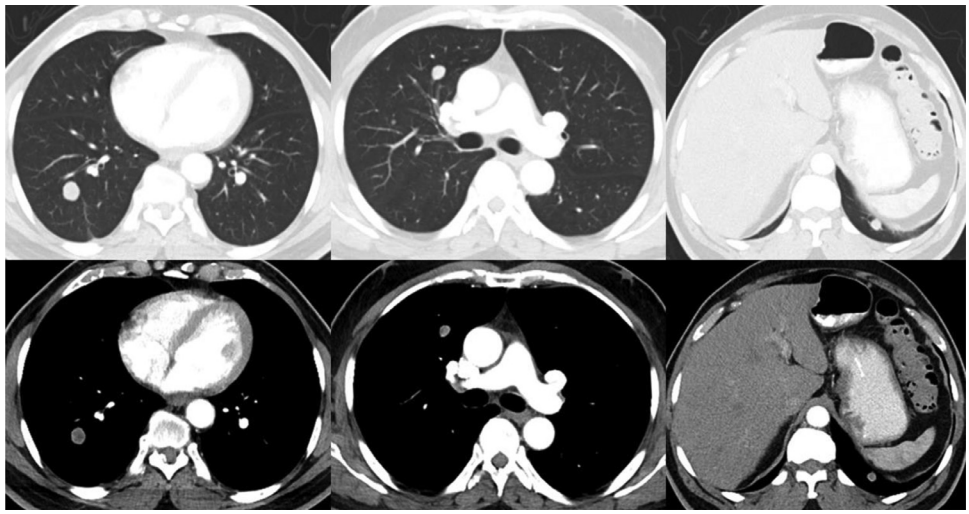

Fig. 1 – Contrast enhanced chest CT in lung and mediastinal windows shows multiple bilateral well-defined pulmonary nodules with peripheral enhancement.

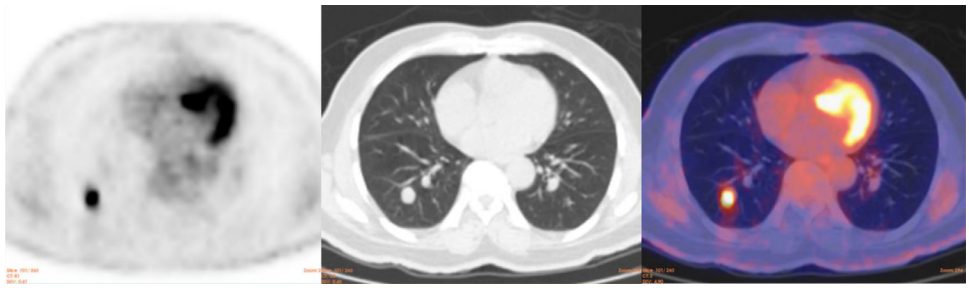

Fig. 2 – PET-CT of the thorax showing a hypermetabolic right lower lobe nodule with SUV of 4.9. The rest of the bilateral pulmonary nodules were not hypermetabolic.

8/18, CD34 – negative; caldesmon, vimentin, CD 99 – positive (diffuse, strong); SMA – positive (focal, moderate); Ki67 – less than 5%; consistent with myofibroma.

Discussion

Etiology of myofibromas is unknown, however, few reported cases suggest a familial pattern [2]. Myofibromas are more frequent in newborns and children under 2 years of age (88%-90% of the cases), and are rarely found in adults [2,3]. Myofibromas are subdivided in solitary and multicentric forms, that is, "myofibromatosis" [4]. They usually involve cutaneous and subcutaneous tissues [1]. Less common locations include bones and internal organs: lung, heart, gastrointestinal tract, and central nervous system [5–8].

Common presentation of myofibromas on ultrasound is a well-defined thick-wall cystic lesion. On CT, myofibromas usually present as well-defined soft tissue nodules with central low attenuation, due to necrosis, and peripheral enhancement [1,4,9,10]. Magnetic resonance imaging appearance of myofibromas consists of low signal on T1-weighted imaging, high or low signal of the center on T2-weighted imaging, and peripheral enhancement after gadolinium administration [4]. On positron emission tomography, myofibromas are often hypermetabolic [11].

Diagnosis of myofibroma requires either biopsy or surgical excision with immuno-histochemical evaluation, which usually reveals positivity for vimentin, muscle-specific actin and myoglobin, and negativity for S100 protein, cluster of differentiation (CD) 68, and desmin, thus excluding tumors of neural, histiocytic, and smooth muscle origin [9].

There is no recommended treatment for this entity. However, most myofibromas regress spontaneously, especially if there is no visceral involvement [1,4].

In conclusion, pulmonary myofibromatosis, although rare, should be considered as a differential diagnosis in patients with suspected pulmonary metastases without known malignancy.

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