Case Report

Asymptomatic localized pleural amyloidosis mimicking malignant pleural mesothelioma: report of a case

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Abstract: We herein report an asymptomatic 65-year-old male with localized pleural amyloidosis mimicking malignant pleural mesothelioma. He had a history of exposure to asbestos and was admitted for investigation of an abnormal pleural thickness detected by chest radiography. Positron emission tomography showed elevation of standardized uptake value corresponding to the pleural thickness. Partial pleurectomy including the tumor was performed for the purpose of diagnosis and local disease control. The pathological examination showed that the tumor was pleural amyloidosis. The tumor was diagnosed as localized primary amyloidosis, because serum monoclonal protein concentration did not increase. Pleural amyloidosis should be considered as a differential diagnosis from pleural mesothelioma.

Keywords: Pleural amyloidosis; pleural mesothelioma; partial pleurectomy

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Introduction

Amyloidosis is characterized by the extracellular deposition of an amyloid substance (1), and it is classified into systemic and localized types. In primary systemic amyloidosis, peripheral nerves, heart, skin, muscle, and less frequently, the pulmonary tract may be involved. Although the presence of amyloid deposits in the lung parenchyma has been sometimes reported in the literature, pleural amyloidosis is extremely rare (2).

Case presentation

A pleural thickness was found on a routine chest X-ray of an asymptomatic 65-year-old man. He had no previous medical history. He had 80 pack-year smoking habit. His occupation was in the oil stove repair, and he had history of exposure to asbestos.

His physical examinations revealed that all values were within the normal limits, pulmonary function tests were within the normal limits. Laboratory examinations revealed: complete blood count and serological data including the C-reactive protein level remained within the normal limits. His serum levels of carcinoembryonic antigen (CEA) and cytokeratin 19 fragment (CYFRA) were 4.8 ng/mL (normal range, 0–5 ng/mL) and 2.5 U/mL (normal range, 0–3.5 U/mL), respectively.

Chest computed tomography (CT) revealed that the pleural thickness, plaque, and tumor were diffusely located in the bilateral thoracic cavity (*Figure 1A*). The intrapulmonary tumor was not found. ^{18}F-fluorodeoxyglucose-positron emission tomography (FDG-PET) showed that the maximum standardized uptake value (SUV_{max}) of this left sided pleural tumor was 4.84, and other lesions revealed a lower uptake (*Figure 1B*), and therefore malignancy could be suspected. No hilar and mediastinal lymphadenopathy, distant metastasis, or abnormal findings of other organs were found CT and positron emission tomography-computed tomography (PET-CT). The thoracotomy surgery was performed for the purpose of diagnosis and local disease control.

A left posterolateral thoracotomy at the fifth intercostal space was performed, Partial visceral pleurectomy including

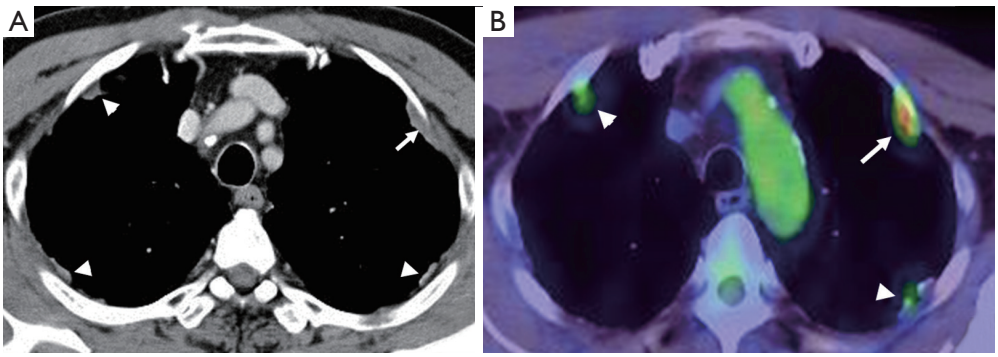

Figure 1 Chest computed tomography (CT) and positron emission tomography (PET) images of this case. (A) Enhanced chest CT scan revealed that the pleural tumor (white arrow) in the left thoracic cavity and pleural thickness and plaque (white arrow head) were diffusely located in the bilateral thoracic cavity; (B) ^{18}F-fluorodeoxyglucose-positron emission tomography (FDG-PET) showed that the maximum standardized uptake value (SUVmax) of this left sided pleural tumor was 4.84 (white arrow), and pleural thickness and plaque revealed a lower uptake (white arrow head).

Figure 2 Thoracotomy findings (top, lateral side; left, cranial side) (3). Left-sided posterolateral thoracotomy at the 5th intercostal space with extrapleural dissection, showed that the mass and pleural lesion were not found on the parietal pleura of the chest wall. Masses were palpated on the visceral pleural surface of left upper lobe and left S6. The tumors on left upper lobe were resected by pleural decortication.
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the mass was carried, whereas the mass were not found on the parietal pleura of the chest wall. The visceral pleurectomy for the left upper lobe of the lung, where two masses were palpated on the visceral pleural surface was completed (*Figure 2*) (3). Intraoperative frozen-section analysis of the specimen showed no malignancy. The other tumor was present on the left S6 and removed by lung parenchyma wedge resection using a stapler. Defect visceral pleura was repaired with polyglycolic acid sheet and fibrin glue. Prolonged pulmonary air leakage was not found, and postoperative course was uneventful.

The cut surface of pleural mass revealed a hyalinizing structure and fibrillary thickened pleura with calcification (*Figure 3A*). Histopathologically, the lesion mainly comprised amyloid deposition showing Congo red stain was positive in the pleural tissue, chronic inflammatory cell infiltrate and multinucleated giant cell were found (*Figure 3B,C*). The stainability of Congo red stain was not lost by potassium permanganate treatment, the lesion was pathologically confirmed to be amyloidosis, excluding the amyloid protein A (AA) type. The appearance of serum monoclonal protein (M protein) was not found, there are no significant clinical symptom of multiple myeloma. Therefore, primary immunoglobulin light chain (AL) type amyloidosis of pleura was suggested. No symptoms of other organ amyloidosis have occurred 1 year after surgery.

Discussion

Amyloidosis is characterized by the deposition of an amyloid substance (1). There are four major categories primary or immunoglobulin AL disease, secondary or AA disease, hereditary or mutant transthyretin (ATTR) disease, and dialysis-associated or β_2-microglobulin (β_2M) disease (4). AL disease arises from the deposition of monoclonal κ or λ immunoglobulin light chain, in these cases, there are two sub-type of disease: systemic and localized. Amyloid is deposited in several organs and leads to organs failure in systemic disease. In this case, clinical course, family history, blood test and pathological examination showed primary localized pleural AL type amyloidosis.

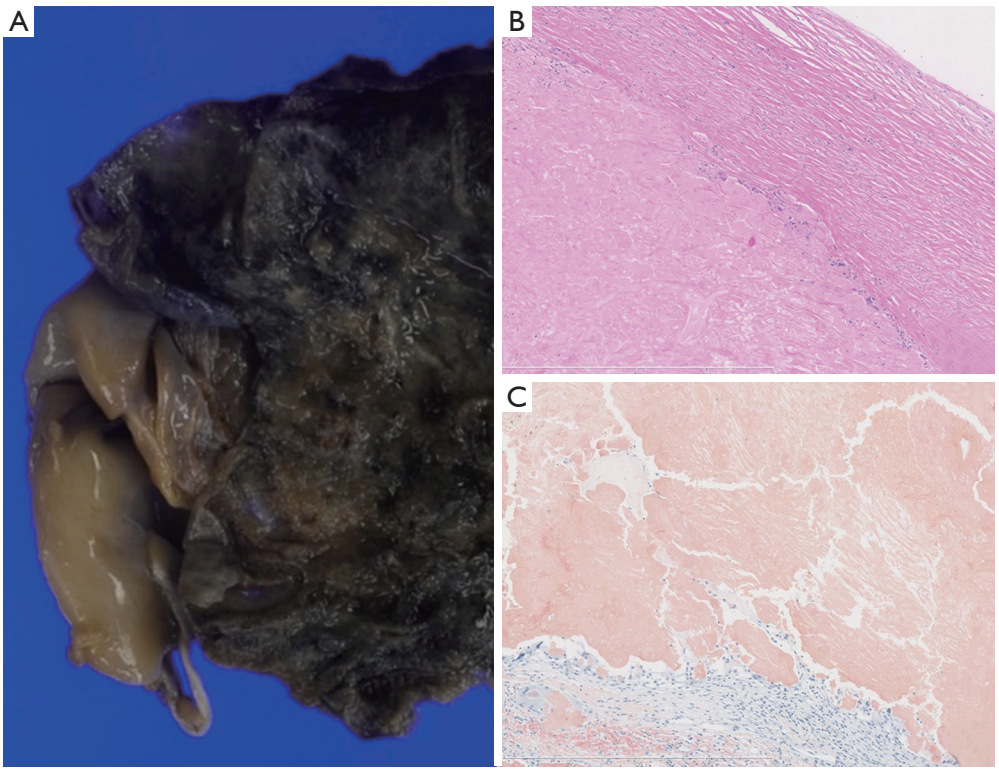

Figure 3 Resected specimen and pathological findings. (A) The surface of pleural mass of resected specimen, revealed a hyalinizing structure and fibrillary thickened pleura with calcification and osteogenesis; (B) histopathological examination showing hyalinizing structure deposition in the parietal pleura (hematoxylin and eosin; original magnification, ×10); (C) the deposition in the pleura was stained with Congo red stains, and chronic inflammatory cell infiltrate were found (Congo red; original magnification, ×10).

Berk *et al.* (4) reported that localized amyloid deposits arise from a small number of plasma cells surrounding the lesion, and the most commonly involved sites were the skin, urethra, and urinary bladder, eye and lung parenchyma. Localized pulmonary amyloidosis has been reported on many occasions (4), but pleural amyloidosis is rare (2). The cases of pleural amyloidosis accompanied with pleural effusion was previously reported (5,6), they had symptom of dyspnea etc. Therefore asymptomatic localized pleural amyloidosis like this patient is extremely rare. Adams *et al.* reported that the most important diagnosis of pleural amyloid includes mesothelioma, either a sarcomatoid or the unusual desmoplastic variant, and solitary fibrous tumor of the pleura (5).

The case with the detection of a pulmonary amyloid lesion by FDG-PET was reported, the uptake of FDG could be related to the abnormal production of immunoglobulins by plasma cells (7). This false-positive findings of PET-CT makes it difficult to diagnose as pleural amyloidosis in distinguishing malignancy.

In conclusion, we experienced a case of localized pleural amyloidosis mimicking pleural mesothelioma clinically and radiologically. In the future, the detection of pleural lesion is supposed to increase with prevalence of CT screening. And false-negative diseases like as this case outside of malignant pleural dissemination and pleural mesothelioma may be increasing by prevalence of PET-CT. Localized pleural amyloidosis should be considered as one of the differential diagnosis for a pleural lesion.

Acknowledgements

None.

Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

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