An Uncommon Etiology for Pleural Effusion with Rib Lesion in a 78-year-old Woman: A Case Report

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Case report

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Abstract

Background: Pleural effusion with rib lesion is generally considered as metastasis of thoracic carcinoma or rib tuberculosis and tuberculous pleuritis. Myelomatous pleural effusion with rib lesion is rare and poses a great diagnostic challenge. We described an easily-misdiagnosed extrapulmonary disease with concomitant pulmonary and rib manifestations, bilateral pleural effusion with lytic rib lesion in multiple myeloma, in order to raise awareness of this disease and avoid misdiagnose.

Case presentation: A 78-year-old woman was referred to the thoracic department with chest distress, breathlessness, poor appetite and weight loss for 3 months. Chest computed tomography demonstrated bilateral pleural effusion and lytic lesion on the left 5th rib. Whole-body bone scan showed the intense uptake lesions on the sixth, seventh vertebrae and a central defect with the peripheral uptake of the "donut"-like change sign on the left 5th rib. Percutaneous CT-guided core biopsy for the rib lesion was performed and the diagnosis was multiple myeloma. Combined chemotherapy and supportive treatment were prescribed. She did not respond to the therapy and died 3 months later due to pleural cavity infection and cardiopulmonary failure.

Conclusion: Some uncommon extrapulmonary diseases with concomitant pulmonary and rib manifestations should be reminded in order to avoid misdiagnose. Extramedullary multiple myeloma can affect any tissue and is an aggressive entity with poor prognosis. Percutaneous CT-guided core biopsy for the rib lesion is of higher positive rate than pleural fluid cytology and more minimally invasive than pleural biopsy via thoracoscope or rib excision biopsy.

Background

Pleural effusion with rib lesion is generally considered as metastasis of thoracic carcinoma or rib tuberculosis and tuberculous pleuritis. In patients not receiving active immunosuppressive therapy, some underlying hematologic diseases can inflict the chest(1). Multiple myeloma (MM) is the second most common hematological malignancy affecting mainly the elderly. Extramedullary multiple myeloma (EM) can affect any area of tissue including the breast, bone, lung, or pleura and is an aggressive, mostly resistant entity with poor prognosis. The incidence of EM varies largely among studies, possibly due to different imaging technique used (2). Myelomatous pleural effusion (MPE) is uncommon with 6% of patients with MM (3). MPE with rib lesion is rare and poses a great diagnostic challenge. We reported a unique case of symptomatic bilateral pleural effusion with lytic rib lesion in MM. Then percutaneous CT-guided core biopsy for the rib lesion was performed and the diagnosis was established. This method was of higher positive rate than pleural fluid cytology and more minimally invasive than pleural biopsy via thoracoscope or rib excision biopsy.

Case Presentation

A 78-year-old woman with a history of hypertension was referred to the thoracic department with chest distress, breathlessness, poor appetite and weight loss for 3 months. She had family history of lung cancer for her brother. Physical examination revealed tachypnea and weak breath sounds on both lungs. There was palpable, tender, swollen 5th rib anteriorly at the level of left nipple. Chest computed tomography (CT) demonstrated bilateral pleural effusion and lytic lesion on the left 5th rib whose cortex was destroyed (Fig. 1a). Whole-body bone scan was performed 3 hours after the injection of 20 mCi ^{99m}Tc -MDP and showed the intense uptake lesions on the sixth, seventh vertebrae and a central defect with the peripheral uptake of the "donut"-like change sign on the left 5th rib, suggesting multiple, metastatic lesions (Fig. 1b). Laboratory findings revealed normal erythrocyte sedimentation rate, C-reactive protein, blood cell analysis, electrolyte, tumor markers, procalcitonin, TB-PCR, globulin, A/G ratio, IgA and IgG. The total protein was 54 g/L (normal 58–80 g/L), albumin 30 g/L (normal 38–60 g/L), serum creatinine 158 $\mu\text{mol/L}$ (normal 44–88 $\mu\text{mol/L}$), β_2 microglobulin 6.92 mg/L (normal 0.8–3.0 mg/L) and IgM 0.36 g/L (normal 0.63–2.77 g/L). Bence-Jones protein in urine and serum protein electrophoresis was negative. Thoracentesis revealed hemorrhagic effusion (positive Rivalta test and red blood cells). A pleural catheter was subsequently inserted into left pleural cavity. Pleural fluid tests for pyogenic bacteria and acid-fast staining were negative. Biomarkers such as adenosine deaminase, interferon- γ , and interleukin-27 were negative. Repetitive cytological examination found scattered cells with heterogeneous nuclear which were difficult to distinguish with adenocarcinoma. Bronchoscopy showed no endobronchial pathology. So, percutaneous CT-guided core biopsy using a 13-gauge core needle under local anesthesia was performed for the left 5th rib lesion (Fig. 1c). Puncture pathology for rib lesion revealed abundant immature plasma cells (Fig. 1d). The immunohistochemistry of CD19, CD38 and CD138 were positive. A diagnosis of multiple myeloma (MM) was made. Subsequent bone marrow aspiration, immunofixation electrophoresis and cytogenetics confirmed as λ type, ISS-II stage and 46,XX[20]. The surgery for rib resection was halted considering the poor prognosis and the patient underwent combined chemotherapy (bortezomib and dexamethasone) and supportive treatment. She did not respond to the therapy and died 3 months later due to pleural cavity infection and cardiopulmonary failure.

Discussion And Conclusions

Concomitant pulmonary and rib lesions usually are considered as metastases. Some uncommon extrapulmonary diseases with concomitant pulmonary and rib manifestations should be reminded in order to avoid misdiagnose. Lymphoma is the most common type to involve the chest wall and ribs and accounts for about 0.3–1.0% of extranodal lymphoma. It often appears as a large extraosseous soft-tissue mass, with relative preservation of the underlying rib cortex. Up to 10% of malignant pleural effusions are due to lymphoma(4). Multiple myeloma (MM) is the second most common hematological malignancy affecting mainly the elderly. The median age at presentation is approximately 70 years. The clonal plasma cells infiltration of bone marrow and presence of monoclonal immunoglobulin in serum and/or urine are its characteristics. Extramedullary multiple myeloma (EM) can affect any area of tissue including the breast, bone, lung, or pleura and is an aggressive, mostly resistant entity with poor

prognosis. Myelomatous pleural effusion (MPE) is uncommon with 6% of patients with MM (3). It is usually related to secondary causes such as congestive heart failure, chronic renal failure, pulmonary infection, serious hypoalbuminemia and thrombosis. MPE with rib lesion is rarer and poses a great diagnostic challenge in need of better strategic approaches (5). Laboratory findings may reveal abnormal globulin, IgA, IgG, A/G ratio, or Bence-Jones protein; however, these are often insensitive and lagging like this case. Diagnostic procedures for MPE have been pleural fluid cytology or pleural biopsy via thoracoscope, as well as bone marrow biopsy. Cytological examination for malignant plasma cells in the pleural fluid has a limited diagnostic sensitivity. Despite guidance from NICE which positioned whole-body MRI as the first-line diagnostic imaging test for suspected myeloma (6), it is currently performed at only a few centers due to limited MRI capacity(7). 99 m-Tc MDP bone scan is a sensitive but insufficient method for detecting bone lesions in MM (8) and may show the "donut"-like change due to the lytic center. Percutaneous CT-guided core biopsy for the rib lesion was of higher positive rate than pleural fluid cytology.

Rib destruction has many etiologies and is more likely to be caused by metastatic rather than primary bone tumors. Metastatic rib lesions commonly include the lung, breast, thyroid and kidney. Specific radiological findings, such as ground-glass patterns in fibrous dysplasia, trabecular patterns in hemangioma, calcification in osteochondroma and fluid levels in aneurysmal bone cyst, usually are benign bone tumors(9). Solitary plasmacytoma of the rib should be considered when confronted with a lytic rib tumor. It is often resectable with good prognosis (10). On the contrary, MPE with rib lesion is characterized by an aggressive clinical course with a low possibility of survival exceeding 6 months and the surgery is contraindicated (11).Traditional chemotherapy is less effective in MPE than in MM without PE. Researchers must develop more effective and durable treatments for MM patients with PE.

Some uncommon extrapulmonary diseases with concomitant pulmonary and rib manifestations should be reminded in order to avoid misdiagnose. Extramedullary multiple myeloma can affect any area of tissue and is an aggressive entity with poor prognosis. Percutaneous CT-guided core biopsy for the rib lesion is of higher positive rate than pleural fluid cytology and more minimally invasive than pleural biopsy via thoracoscope or rib excision biopsy.

Abbreviations

Pleural effusion (PE)

Multiple myeloma (MM)

Myelomatous pleural effusion (MPE)

Extramedullary multiple myeloma (EM)

Computed tomography (CT)

Declarations

Ethics approval and consent to participate:

We have had the ethics approval and consent of the Shanxi Medical University ethics committee.

Consent for publication:

Written consent to publish this information had been obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editors-in-Chief of this journal.

Availability of data and materials:

The datasets generated and/or analysed during the current study are not publicly available till publication but are available from the corresponding author on reasonable request.

Competing interests:

The authors declare that they have no competing interests.

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Authors' contributions:

XQ R, YP W, and J J analyzed and interpreted the patient's data and the medical treatment. DA Q performed the biopsy procedure of the rib lesion, and was a major contributor in writing the manuscript. All authors read and approved the final manuscript.

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Figures

Figure 1

The imaging for a 78-year-old woman. a, chest CT showed bilateral pleural effusion and lytic left 5th rib lesion. b, whole-body bone scan showed the intense uptake lesions on the sixth, seventh vertebrae and a central defect with the peripheral uptake of the "donut"-like change sign on the left 5th rib. c, percutaneous CT-guided core biopsy of the left 5th rib lesion. d, puncture pathology for rib lesion revealing abundant immature plasma cells (Hematoxylin and eosin,×200).

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