Case Report

Multiple myeloma with pleural involvement

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ABSTRACT
This study presents the differential diagnosis of metastatic tumors of the pleura vs. primary pleural malignancies in a male patient whose diagnosis was multiple myeloma with pleural involvement and confirmed by bone marrow aspiration and pleural biopsy. Computed tomography (CT) manifestations of this case were retrospectively analyzed, and compared with those of primary pleural malignancies. The CT manifestations of this case mainly involved bilateral multiple pleural nodules with irregular thickening. These nodules were also associated with rib damage and lung metastases, manifestations characteristic of metastatic tumors. The presence of a primary pleural malignancy correlated with patients’ clinical history and imaging data, and involved unilateral pleural involvement, and to a lesser extent, mediastinal pleural involvement and pleural effusions. A pleural biopsy can establish a definitive diagnosis. Therefore, multiple myeloma with pleural involvement can be differentiated from primary pleural malignancies by using a combination of imaging data and clinical laboratory tests.

KEY WORDS: Multiple myeloma, Metastatic tumors, Computed tomography (CT).

INTRODUCTION

Multiple myeloma is the most common type of malignant plasma cell disease. It involves uncontrolled proliferation and extensive infiltration of malignant plasma cells, and the deposition of monoclonal immunoglobulins. As a result, a variety of adverse reactions are induced which include extensive bone destruction, recurrent infections, anemia, hypercalcemia, hyperviscosity syndrome, and renal dysfunction.¹ The incidence of multiple myeloma is estimated to be 2-3/100,000, with a male/female ratio of 1.6:1. This disease mostly affects patients over the age of 40; however, multiple myeloma with pleural involvement is a very rare clinical entity. Here, we report a case of pleural involvement associated with multiple myeloma, and a differential diagnosis of metastatic tumors of the pleura vs. primary pleural malignancy.

CASE REPORT
A 36-year-old male patient presented with chest pain, convulsive pain in the posterior left hip, and...
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radiating pain in the left lateral leg. The patient reported no history of trauma, hyperpyrexia, afternoon fatigue, or low fever. On examination, no redness or swelling in the left hip joint was noted, and no pathological reflexes were elicited. The patient had no family history of related diseases.

Laboratory tests detected a white blood cell count of $31.9 \times 10^9$, 83.1% neutrophils, a platelet count of $412 \times 10^9/L$, serum alkaline phosphatase levels of 121.5 U/L, serum levels of (kappa light chain (KAP) and lambda light chain (LAM) of immunoglobulin) to be 1.57 g/L and 4.69 g/L, respectively, $\alpha_2$ microglobulin levels of 5.7 mg/L, and urine levels of KAP and LAM to be 55.3 and 3570 mg/24h, respectively.

Abdominal ultrasonography detected splenomegaly, and a chest computed tomography (CT) scan (Fig-1A, B) revealed localized, low density areas in the bilateral clavicles and sternum. Within these areas, high density stripes were visible. Bone destruction was also observed in the bilateral ribs associated with surrounding soft tissue thickening and multiple inhomogeneous nodular thickening of bilateral pleurae (Fig-1C, D). Magnetic resonance imaging (MRI) examination detected inhomogeneous L2-5 vertebral bodies with hypointense signals on both T1 and T2 weighted images (WI), which were most pronounced in fat-suppressed images. In addition, abnormal signal intensity in an irregular lamellar shape was notable in the bilateral acetabulum, pubis, ischium, and femoral head and neck regions. The irregular shape was also hypointense on T1WI images, and slightly hyperintense on fat-suppressed T2WI.

In a whole-body bone scan, increased radioisotope uptake was observed in the L4 vertebral body, the bilateral femoral head, and the right acetabulum, as well as in the skull, humerus, and bilateral ribs.

Bone marrow aspiration was performed (Fig-1E), and an increased number of toxic particles were observed among the granulocytes. Blood smear tests also detected the presence of immature granulocytes and atypical lymphocytes (10%), while the proportion of plasmablasts and proplasmacytes was significantly increased. Levels of albumin were 51.3%, and the $\alpha_1$, $\alpha_2$, $\beta$, and $\gamma$ globulin levels were 4.6%, 13.4%, 11.9%,

Figure-1: Representative images of the chest CT scans, bone marrow aspiration, and left pleural biopsy performed. Panels A and B present axial chest CT images. Bone damage to multiple ribs bilaterally, surrounding soft tissue thickening and multiple inhomogeneous nodular thickening of bilateral pleural effusions are observed. Panels C and D represent multi-planar reconstruction images of chest CT scans that detect nodular pleural thickening. Panel E represents the blood smear test of a bone marrow aspiration that was performed. An increased number of toxic particles are detected in the granulocytes, along with the presence of immature granulocytes, 10% atypical lymphocytes, and a significantly higher proportion of plasmablasts and proplasmacytes. Panel F is a representative image of a left pleural biopsy that was performed. The results are consistent with the presence of plasma cell tumor (myeloma) involving the pleura (H/E staining; 20× magnification).
and 18.7%, respectively. To obtain a biopsy specimen, an area of thickened left pleura that was not associated with rib damage was selected. Analysis of the resulting specimen suggested a diagnosis of plasma cell tumor (myeloma) involving the pleura (Fig-1F).

For treatment, a chemotherapy regimen including melphalan, prednisone, and thalidomide (MPT) was administered for six courses. Upon completion of this regimen, the patient was in generally good condition. Subsequently, a bone marrow aspiration assay detected plasma cells at 3%; therefore, a chemotherapy regimen including vincristine, adriamycin, and dexamethasone (VAD) was administered. Following this regimen, the general status of the patient was also good, and the extent of the pleural involvement was lessened.

**DISCUSSION**

A retrospective analysis of the present case identified prominent manifestations of pleural lesions and multiple changes in bone substance. Thus, a diagnosis of primary pleural malignancy with multiple bone metastases was initially formed. However, upon further analysis, additional manifestations were noted that were inconsistent with the original diagnosis.

Primary pleural malignancy most commonly presents as malignant pleural mesothelioma. Patients with this disease often have had long-term exposure to asbestos, or a history of viral infection. The lesions present are usually unilateral, and manifest as extensive pleural thickening of the affected side with hump-like nodules. The lesions can arise in the lower chest, and are often associated with a narrowed thorax and pleural effusion. Furthermore, the lesions can typically be detected on an enhanced scan. Additional changes characteristic of malignant pleural mesothelioma include a fixed mediastinum, copious pleural effusions, and irregular pleural thickening. Distant metastasis in these cases is less common. In the present case, the patient had no history of exposure to asbestos or viral infection. Moreover, the patient presented with multiple changes in bilateral pleural effusions, with an absence of any narrowing of the thorax or pleural effusion. Common manifestations of other primary pleural malignancies such as liposarcoma, fibrosarcoma, and rhabdomyosarcoma, include extensive pleural thickening, often more than 1cm, and mediastinal pleural involvement. Diffuse or irregular pleural thickening can be detected by CT, and is a strong indicator that pleural malignancies are present. However, CT data lack specificity. The present case did not show mediastinal pleural involvement, yet exhibited bilateral nodular pleural thickening without diffuse changes. These results suggest that the patient did not exhibit the typical manifestations associated with pleural malignancies.

Thirdly, primary pleural malignancies are generally not associated with increase in serum levels of KAP, LAM, and β2 microglobulin, or increased levels of KAP and LAM in urine as observed in the present case. Therefore, these clinical data further confirm that a diagnosis of primary pleural malignancy could be ruled out.

Since the primary manifestations of the present case included multiple bilateral pleural nodules and damage to multiple bones, indicative of the presence of metastases, the patient could have been diagnosed with multiple pleural and bone metastases. However, elevated serum levels of KAP, LAM, and urine levels of KAP and LAM were not consistent with this diagnosis. Furthermore, the possibility that the pleural lesions were metastases and the rib changes were primary was an additional consideration. Therefore, by performing a pleural biopsy, the nature of the lesions could be confirmed.

Although metastasis from multiple myeloma is very rarely observed, multiple myeloma with pleural involvement was an alternative diagnosis in the present case, especially since pleural changes had been detected. Therefore, this case serves to demonstrate the importance of performing a comprehensive assessment of clinical manifestations, imaging findings, and laboratory tests, in order to improve diagnostic accuracy.

**REFERENCES**


**Authors Contribution:**

Liang Chen performed the clinical procedure. Liang Chen and Jia-Huan Wang are both the manuscript writing. Liang Chen Jia-Huan Wang Wan-An Zhu Chun-Lai Dai Xiao-Yan Sun and Yu-Xue Bai are both the assisted in the procedure and performed post operative follow up.