

Primary Osteosarcoma of the Lung: A Case Report

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We report a rare case of primary osteosarcoma of the lung. A 73-year-old Japanese man with a productive cough and hemoptum was referred to us for further evaluation of a huge cavitating mass in the left upper lobe, shown on a radiograph of his chest. The result of a tumor biopsy, via fiberoptic bronchoscope, raised a strong suspicion of sarcoma. Therefore a left upper lobectomy was performed without any adjuvant therapy. The tumor, which measured 72×70×62 mm, was well-defined, whitish-yellow in color and soft in consistency. Histological examination of the tumor showed a dense proliferation of spindle cells and the presence of many collagen fibers. Eosinophilic osteoid, with no epithelial structures, were noted in the stroma. Immunohistochemically, the tumor cells were positive for mesenchymal, but negative for epithelial markers. These pathological features suggested the tumor was an osteosarcoma. A general inspection of other organs did not reveal any more tumorous lesions, therefore, the final diagnosis of the tumor was primary osteosarcoma of the lung. (Ann Thorac Cardiovasc Surg 2006; 12: 126–8)

Key words: osteosarcoma, lung, rapid progression

Introduction

Osteosarcoma usually arises from skeletal lesions in the extremities, chest, abdominal wall, head and neck, but, occasionally from the parenchymatous organs. Osteosarcoma primarily arising from the lung parenchyma has rarely been reported in the past. In this paper, we report a case of primary osteosarcoma of the lung which occurred in a 73-year-old Japanese man who died with rapid metastases after surgery.

Case Report

A 73-year-old Japanese man was referred to our hospital with a productive cough and hemoptum over the pe-

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riod of a month. A chest radiograph revealed a huge cavitating mass shadow in the left upper lung field (Fig. 1). A computed tomography (CT) of his chest showed a well-defined cavitating mass in the left upper lobe (measuring approximately 70×65 mm) which was enhanced heterogeneously by the injection of contrast material (Fig. 2).

A transbronchial biopsy was carried out through a fiberoptic bronchoscope. Pathological examination of the specimen revealed undifferentiated sarcomatous neoplastic change in the form of small, uniformly round cells. Therefore, the tumor was suspected of being a sarcoma. Further systemic inspection, by ⁶⁷Ga scintigraph and ^{99m}Tc scintigraph, was made in order to locate possible further primary lesions, however, none were found, hence we concluded that the tumor in the lung was a primary sarcoma.

Adjuvant therapy was not given at the patient's request. The tumor was completely resected with left upper lobectomy of the lung. Radical lymph node dissection of hilar and mediastinal nodes revealed no involvement of the tumor. The tumor (measuring 72×70×62 mm) showed a well-defined margin and made expansive growth to adjacent structures without apparent invasion.

The cut surface of the tumor was whitish-yellow in



Fig. 1. Routine chest radiograph revealed a well-defined huge cavitating mass in the left upper lung field.

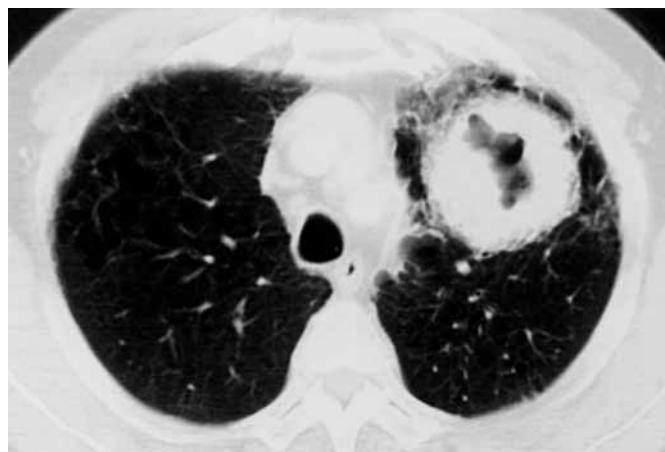


Fig. 2. A computed tomography of his chest showed a well-defined cavitating mass in the left upper lobe of the lung.

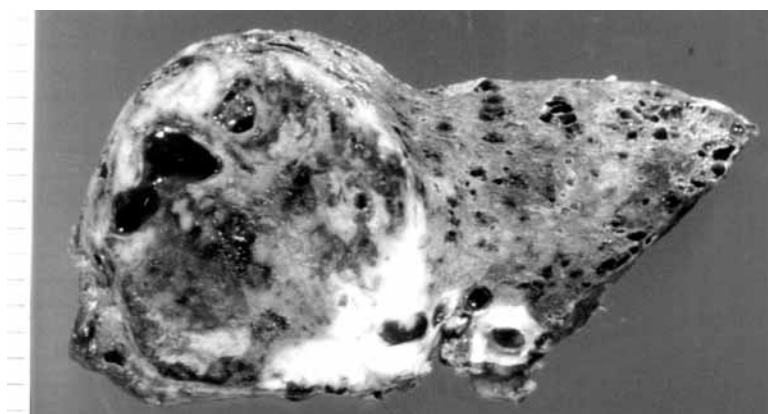


Fig. 3. A cut section of the gross specimen. The tumor which measured 72×70×62 mm, was whitish-yellow in color and ligeneous in consistency.

color and soft in consistency. Macroscopically, it revealed necrosis, hemorrhage and cystic change, but calcification was not observed (Fig. 3).

Microscopically, sporadic intracapillary growth of the tumor was evident. The tumor was characterized by a dense proliferation of atypical spindle cells and fusiform cells. Polykaryocytes, like osteoclast, were noted in some areas. Abundant collagen fibers and eosinophilic osteoid were noted in the stroma, but epithelial appearance was not evident in the tumor (Fig. 4). Immunohistochemical analyses revealed that the tumor cells were diffusely positive for vimentin and osteonectin, but negative for osteopontin, desmin, myoglobin, CD34 and S-100 protein. Epithelial markers, such as broad-spectrum keratin (AE1/AE3), epithelial membrane antigen (EMA) and keratin CAM5.2, were not present. Given these pathological findings, the tumor was considered to be an os-

teosarcoma.

The patient rejected any adjuvant chemotherapy after surgery and, subsequently, multiple subcutaneous tumors located mainly on the scalp were evident at the 6 month follow-up. An incisional biopsy of the tumors confirmed metastases of the original osteosarcoma. Further examination revealed the emergence of multiple, new metastatic lesions in the lung, liver, brain and bone within a short period. They showed rapid growth and the patient, receiving only the best supportive care, died 7 months after the operation. A post mortem autopsy was not carried out.

Discussion

Osteosarcomas develop most frequently from skeletal lesions in the extremities, chest, abdominal wall, head and neck, though occasionally they can be found in the

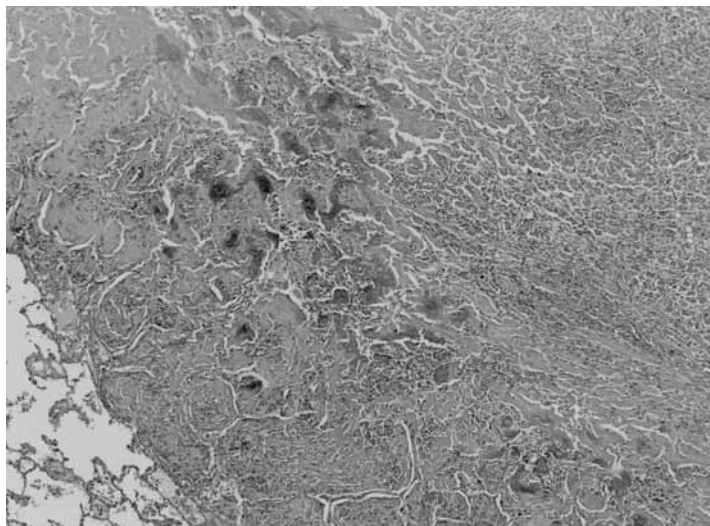


Fig. 4. Histopathological appearance of the tumor showing a dense proliferation of atypical spindle cells, osteoclast and eosinophilic osteoid and the presence of many collagen fibers with no epithelial structures in the stroma. ($\times 40$ H&E)

parenchymatous organs.¹⁾ Extraskelatal osteosarcomas are rare, accounting for about 1% of all sarcomas. The lung is the most extraordinary site for them to arise. To our knowledge, only 10 cases of primary osteosarcoma of the lung have ever been reported, including our case.^{2,3)}

The average onset age of extraskelatal osteosarcoma is the sixth decade of life, in contrast to skeletal osteosarcoma that has peak incidence in the second decade of life. The incidence in both sexes is equal. Most cases of osteosarcoma in the lung take the form of a huge lung mass on diagnosis²⁾ and show a rapid progression. Recurrence after resection occurs in more than half of the patients. The lung is the most common site of recurrence (>80%) followed by the liver, lymph nodes and bones. Skin metastasis of extraskelatal osteosarcoma, as shown by scalp metastases in our case, is exceedingly infrequent.⁴⁾ Postoperatively, most local recurrences and distant metastases occur within 3 years. The prognosis is poor, with a 5-year survival rate of 37%.⁵⁾ Wide excisions and radical resections in the initial operation are thought to be effective for local control. Resection of the metastatic lesions can occasionally achieve a good outcome.^{1,5)} Aggressive treatment with chemotherapy and radiotherapy has been reported to be useful in some literature,¹⁾ but the effects of adjuvant therapy are unproven because of the scarcity of this type of tumor.

In order to diagnose a tumor in the lung as primary osteosarcoma, some differentials must be excluded. Firstly, the possibility that it might be a metastatic osteosarcoma from another primary site because the lungs

are the predominant site of metastasis.⁵⁾ Secondly, carcinosarcoma of the lung, characterised by the presence of carcinomatous foci in sarcoma tissue is also possible. In our case, a preoperative general examination did not reveal any lesions, except for the pulmonary tumor and the diagnosis of osteosarcoma of the lung was based on analysis of the multi-sliced sections of the resected specimen with immunohistochemical analysis. Mesenchymal origin was confirmed by a positive reaction to mesenchymal markers and epithelial origin was excluded by a negative reaction to epithelial markers. Therefore, we finally diagnosed this pulmonary tumor as a primary osteosarcoma of the lung.

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