Giant-Cell Bone Tumour With Pulmonary Metastases: A Case Report*

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Abstract

Giant-cell tumour of bone (GCTB) which represents 5% of all primary bone tumours is encountered in patients between 20 - 40 years of age. Bones adjacent to the knee, the distal end of the radius and occasionally the sacrum or pelvis are the usual sites for these tumours.

GCTB was discovered accidentally in 1987 in a 30-year-old male patient who had an operation due to a traumatic right forearm fracture. Neither pulmonary nor other organ involvement had been described at that time. In 1990, the patient admitted to our hospital due to shortness of breath with abnormal chest X-ray. 10x11cm. solid mass and pleural effusion in the right hemithorax were seen at the thorax computed tomography. Transthoracic fine needle aspiration biopsy was reported as a pulmonary metastases of giant-cell bone tumour. Chemotherapy was applied, but there was not complete response to the chemotherapy. In 1995, the patient was operated second time on the right radius because of local recurrence. Till 1998, he had no any respiratory system complaints. Then the patient had sudden onset of dyspnoea and chest pain. He died due to pulmonary embolism in the third day of hospitalisation. Giant-cell bone tumour is generally classified as a benign tumour. In 1957, for the first time, Lasser and Tetewsky reported pulmonary metastases although there was no histopathologically malignant transformation. Because pulmonary metastases of GCTB is rare and he had long and comfortable life with his pulmonary metastases, we wanted to present our patient.

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Introduction

Metastases from a primary neoplasm are generally regarded as biological evidence of malignancy. Giant-cell tumour of bone (GCTB), classified as a benign tumour, may rarely lead to metastases in the lung, but still has a good prognosis (1). The primary lesion and the lung metastases usually have the same histological features of a benign tumour (2).

Approximately 55 cases of pulmonary metastases from GCTB have been reported until 1999 (2,3). The aim of this presentation is to contribute to the accumulated experience related to GCTB with pulmonary metastases.

Case Report

A 41-year-old male patient had been operated on his broken right forearm caused by trauma in 1987. During the operation, the tumour on the distal part of the right radius had been acci-

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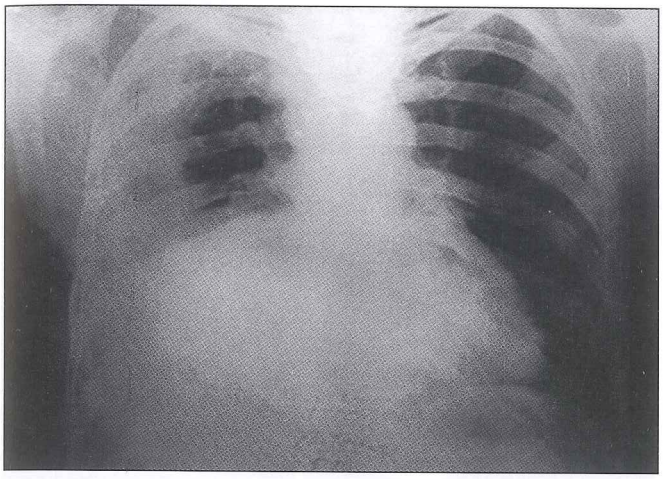


Figure 1. *Chest X-ray of the patient three years after the first diagnosis of GCTB*

dentally noticed and the biopsy had revealed GCTB. There were no metastases at the time of the diagnosis neither in the lungs nor pertaining to other systems.

In 1990, the patient was admitted to the hospital suffering from shortness of breath. His chest X-ray showed a homogenous density in the right hemithorax which extended to the 3rd anterior rib (Figure 1). Massive pleural effusion and a 10x11 cm solid mass which had cystic necrosis areas were seen on his computed thorax tomography (CT). The mass originated from the right middle lob and reached the lower lobe and could not be distinguished from mediastinal vasculature. Thoracentesis and pleural biopsy were performed. No evidence of malignancy was found in pleural fluid and pleural biopsy sampling. Flexible fiber optic bronchoscopy was performed and again there were no pathological signs indicating malignancy. Both the pleural fluid and the bronchial lavage fluid from the right middle lobe were evaluated as class II. Then, transthoracic fine needle aspiration biopsy was performed and the specimen was diagnosed as a pulmonary metastastatic lesion of GCTB. Because there was mediastinal invasion, no surgical intervention was attempted. Adriamycin was given in three weeks intervals for four cycles. Although the pleural effusion disappeared, no change occurred in the solid mass with this treatment. The follow-up of the patient was continued by periodic visits.

In 1995, eight years subsequent to the initial diagnosis, the patient, due to a local recurrence of GCTB although his clinical performance was excellent, underwent a second operation. At this time, his thorax CT was evaluated as stable.

The patient, who had had no complaints until 1998, was readmitted to our emergency department suffering from sudden onset of dyspnea, right pleural pain and hemoptysis. His chest X-ray showed a homogenously dense area in the lower 2/3 of the right hemithorax (Figure 2). Sinusal tachycardia was noted in the ECG tracings and hypoxemia and hypocapnia in arterial blood gas analyses. He was hospitalized for suspicion of pulmonary embolism and given anticoagulation therapy. His thorax computed tomography, performed a week before admission to the hospital, was again evaluated as stable (Figure 3,4). Ventilation/perfusion scintigraphy performed in 48 hours was reported to show a high probability for pulmonary embolism. The patient died on his third day of hospitalisation, 11 years after the first diagnosis of GCTB .

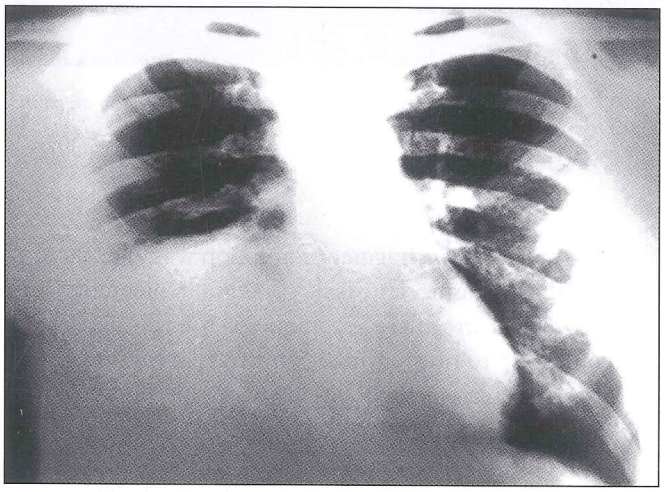


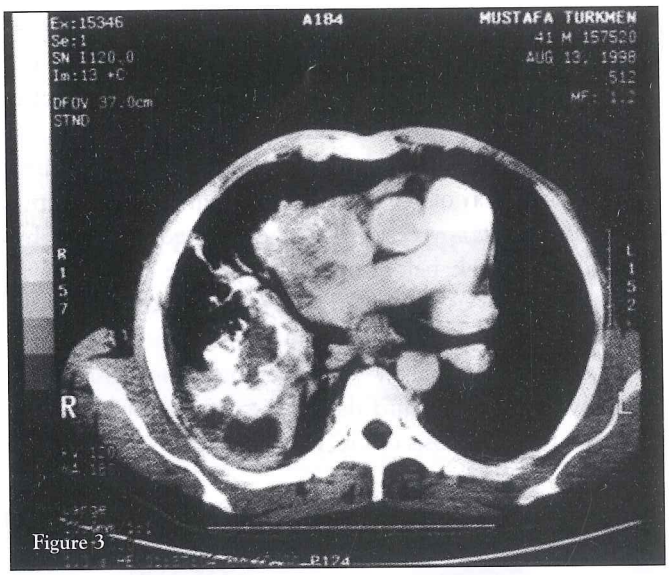
Figure 2. *Chest X-ray of the patient eleven years after the first diagnosis of GCTB*

Discussion

In 1940, Jaffe et al. described the potential for metastases without undergoing sarcomatous transformation in benign GCTB (2). Gresen et al. found only one case with lung metastases among 195 cases followed in the Mayo Clinic between 1910 and 1969; and reported that there was a malignant transformation in this case (4). In 1957, Stargardter and Cooperman reported a patient with giant-cell tumour of the femur in whom metastatic bone and pulmonary lesions developed five years after amputation for local recurrence (5).

The incidence of lung metastases from GCTB ranges from 1% to 9%. Because of its rarity, very little is known about the long term outcome, the risk factors for metastases and the best treatment for the pulmonary lesions (2,6,7).

Usually mononuclear stromal cells, vacuolar changes and granulation are noted in the primary and metastatic lesions and both have the same histopathological appearance. The



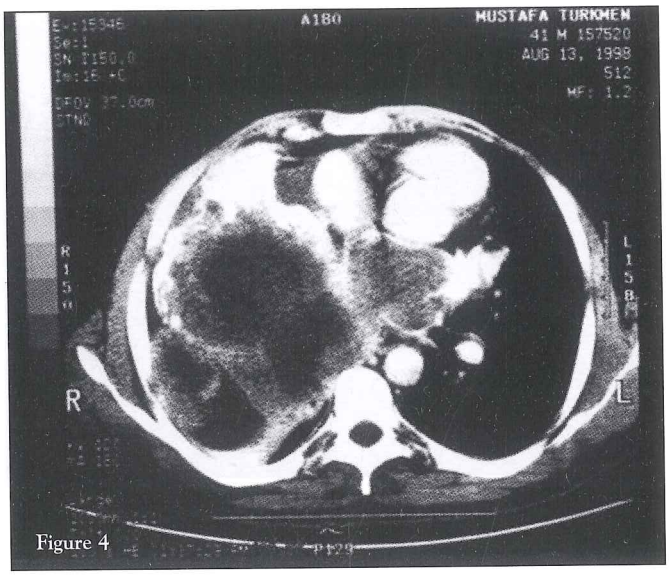


Figure 3-4. *Thorax CT of the patient eleven years after the first diagnosis of GCTB*

absence of true tissue fragment formation would exclude an epithelial neoplasm. The lack of cellular pleomorphism, atypia, nuclear irregularities, spindle cell forms and necrosis would help to rule out malignant transformation as well as other sarcomatous processes (1).

Distal radius, distal femur and sacrum are the most comman sites of the tumour, respectively (2,6). The mean interval between the primary diagnosis and the onset of lung metastases has been reported to be between 2.9 and 4.1 years in larger series (2,7). The primary tumour site was the distal radius and the interval between the initial diagnosis and the onset of the lung metastases was 3 years in our patient. The mean interval between the primary diagnosis and local recurrence is reported as 2.9 years (ranging from 3 months to 23 years) (2). In our patient, local recurrence occurred 8 years after the primary tumour was diagnosed and 5 years after lung metastases, contrary to expectations.

In larger series it has been reported that the patients' age or gender is not a risk factor for metastatic disease (2). It is estimated that there is a seven-fold increase in the risk of developing lung metastases after local recurrence of GCTB (7). Furthermore, in larger series, lung metastases were seen in most of the patients with giant-cell tumour of the radius. This suggests that a primary tumour at this site may be a higher risk for lung metastases. Vascular invasion and surgical manipulation are the other risk factors for metastases. The reported incidence of subsequent metastatic lesions in the lungs after surgical resection alone ranges from 33% to 46% (2).

Although resection of pulmonary metastatic lesions is thought to be necessary, Kares et al. made the diagnosis via fine needle aspiration biopsy and we did the same (1).

The majority of the lung metastases are solitary pulmonary nodules and the most successful therapy is complete resection by thoracotomy. Radiation has been used without apparent success and chemotherapy has only occasionaly proved to be successful. Although spontaneous regression of metastatic giant-cell tumour in the lung has been reported, apparently no prior tissue diagnosis had been established in that reported case (1-3). Surgical intervention was not attempted in our case due to mediastinal invasion and the size of the mass.

A pulmonary metastasis from GCTB does not always implicate a bad prognosis, but has been reported as cause of death in 16% to 25% of the patients. However, the overall survival rate of GCBT patients with lung metastases is much higher than that for other metastasized lung tumours (3). The mortality rates recorded for patients with GCTB metastasing to the lungs range from 0% to 37%. The mean follow-up period is 11.9 years and eight years survival rates range from 14% to 23%. The patients have a more favorable prognosis than was originally thought (2). Our case also survived comfortably for nearly 11 years.

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