Unusual Cause of Spontaneous Bilateral Pneumothorax Secondary to Osteosarcoma Metastasis

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Abstract
Spontaneous pneumothorax is an emergency situation and early diagnosis and management is very important. Pneumothorax is rarely the first manifestation of lung metastasis. Spontaneous pneumothoraces have been reported in some osteosarcoma patients. We hereby report a case of a bilateral spontaneous pneumothorax, revealing metastasis of an osteosarcoma.

Introduction
Simultaneous bilateral spontaneous pneumothorax is a very rare condition, mainly seen in patients with underlying lung disease.¹ Metastatic causes of pneumothorax are rare but must be considered in patients with a history of neoplastic disease. Pneumothorax may be associated with several types of lung tumors: primary tumors, bullous or excavated metastasis, carcinomatous lymphangitis. Primary lung tumors are often excised. In contrast, only 4% of lung metastases are excised, whatever the primary tumor. The most frequently involved tumors are derived from epithelial tissue, genital tumors, and sarcoma. Only 1% of spontaneous pneumothorax cases are due to lung metastasis.

Case Report
A 22 yrs old male patient admitted through emergency department with sudden onset of dyspnoea. He had no history of chest trauma, palpitation, syncope or leg oedema. The patient was normotensive and not known to be diabetic. He was non-smoker and non-addicted. He had swelling above left knee joint associated with pain for 15 days, for which he was under treatment by a local doctor who diagnosed it as septic arthritis of left knee joint but no improvement was noted after conservative management, and then the patient developed sudden onset of respiratory distress for which he was admitted to our institution.

On examination, pulse rate was 84 bpm, regular; respiratory rate 34/min, BP-114/76 mmHg, central cyanosis was present. Chest examination showed tympanic percussion note and diminished vesicular breath sound bilaterally. Other systemic physical examination was normal. Chest x-ray (PA) revealed bilateral pneumothorax. Patient was put on intercostals tube drain on right side initially and then stabilized and further investigation done. CT thorax (Figure 1) also showed similar finding i.e. bilateral pneumothorax without any obvious pulmonary nodule or subpleural lesion. X ray of the left knee joint showed expansile lesion at lower end of femur with periosteal elevation and adjacent soft tissue swelling. MRI of the left knee joint revealed heterogeneous mass lesion in metaphyseal region of left femur with soft tissue extension –possibly osteosarcoma. Punch biopsy from the lesion revealed osteosarcoma by H/P examination (Figure 2). Patient was treated with three cycles of neo-adjuvant chemotherapy with inj. cisplatin and doxorubicin followed by above knee amputation and on follow up and receiving chemotherapy is till date asymptomatic.

Discussion
Several mechanisms of spontaneous pneumothorax in malignancy have been suggested: (1) Constitution of a fistula between parenchyma and pleura due to necrosis of a subpleural tumoral nodule, secondary to vascular lesion or chemotherapy; (2) Partial bronchial obstruction by a valve tumor, with alveolar distension, dehiscence of alveolar walls, and passage of air in interlobular septa to the pleura, forming blebs that can break; (3) Tumor emboli with infarction and necrosis; and (4) Tumoral infiltration of the wall of a preexisting benign cavity and rupture into the pleural space.²

Isolated cases and small series of pneumothorax due to lung metastasis have been described in children and young adult who are frequently
associated with bone sarcoma.\textsuperscript{3} Chemotherapy like doxorubicin for osteosarcoma treatment leads to tumor necrosis 1 to 8 days after initiation of treatment thereby increasing the risk of pneumothorax. But, before initiation of chemotherapy, spontaneous pneumothorax as a complication of malignancy is rare (1%) and usually unilateral. The lung is the first site of dissemination of osteosarcoma. At first presentation 10% of osteosarcoma patients show pulmonary metastasis. Lung metastasis of osteosarcoma is usually multiple and bilateral, but is unique in 35% of cases: 49% are parenchymatous and 51% are subpleural, 5% of the latter being complicated by pneumothorax. Lung metastases of osteosarcoma are often calcified, more rarely bullous. These bullous metastatic lesions can progress to full nodules.

We are presenting a relatively rare early presentation of spontaneous pneumothorax which is bilateral rather than unilateral. Though in one study on 32 patients sensitivity of detection of lung metastasis was 57% by chest x-ray and 88% by chest computed tomography but we cannot detect any nodule or cavitary lesion. Ferrari and colleagues showed that all patients died within 40 months without surgical resection of lung lesion whereas after surgical treatment the 10 year survival rate was 30%.\textsuperscript{4}

**Conclusion**

Our case presented at emergency with bilateral spontaneous pneumothorax which is very rare. Repeated spiral CT thorax is necessary to detect pulmonary lesion. We must watch for recurrence of pneumothorax in future from undetectable pulmonary nodule.

**References**