Case report

Spontaneous pneumothorax from radiographically occult metastatic sarcoma

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Abstract

We present two cases of spontaneous pneumothorax secondary to metastatic synovial cell sarcoma. Radiographic techniques initially did not suggest lung nodules in either patients; thoracoscopy allowed sequential bilateral detection of small pulmonary metastases in one case and confirmed the diagnosis during treatment of recurrent pneumothorax in the second case. This report underlines that pneumothorax may be the first clinical manifestation of metastatic sarcoma and can occur even if current imaging techniques are not indicative of metastatic disease. In the latter instance, thoracoscopic exploration has to be envisaged in order to rule out the possibility of metastatic lung involvement. © 1997 Elsevier Science B.V.

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1. Introduction

Spontaneous pneumothorax (SP) secondary to malignant pulmonary neoplasm, although well documented, is uncommon [1,3,7,10]. It is estimated that less than 1% of all SP are tumor-associated [1,10]. Metastatic osteogenic or soft tissue sarcoma are most commonly associated with pneumothorax, particularly in the setting of administration of cytotoxic chemotherapy [4,5] or radiation [8]. In the majority of these previously reported cases, the patients either had known metastatic pulmonary disease or the diagnosis was rapidly suggested by nodules detected by imaging modalities.

Before the advent of thoracic computed tomography (CT) scans, five cases of pneumothorax secondary to radiologically occult pulmonary metastases and without recent chemotherapy were described [1,9]; during the last 15 years, only one additional report has been published [2,4]. Recently, we observed two young patients, both with known synovial cell sarcoma, in which pneumothorax suggested pulmonary metastatic disease. Despite negative CT scans, thoracoscopy allowed sequential bilateral detection of a small pulmonary metastases in one case and confirmed the diagnosis after 1 year in the second case.

2. Case reports

2.1. Case 1

A 39-year-old man developed a right SP 3 years after excision of a malignant synovial cell sarcoma from his left shoulder. There were no significant symptoms or radiographic evidence of local recurrence or metastatic disease. The pneumothorax was initially treated by tube thoracostomy but recurred within a few weeks. Thoracoscopic exploration and apical talc poudrage were
performed at another institute. Neither nodules nor bullous lung tissue were noted on chest X-ray, CT scan, or during thoracoscopy.

The patient was admitted to our clinic 1 year later for treatment of second recurrence of the right-side pneumothorax. A CT scan suggested a small right pneumothorax and three peripheral nodules 0.5–2 cm in diameter, one each in the upper, middle and lower lobe. Thoracoscopic excision of one nodule located in the middle lobe close to the fissure confirmed the presence of metastatic synovial cell sarcoma. No bullous alterations were found this time either. The remaining two lesions were excised by thoracotomy.

2.2. Case 2

A 35-year-old man underwent excision of a synovial cell sarcoma of the thumb and developed a left SP 1 year later. Chest X-ray did not suggest possible metastatic disease; CT scan was not performed at that time. Thoracoscopy revealed a small peripheral nodule in the upper lobe, which was histologically consistent with metastatic synovial cell sarcoma. A second metastasis located in the posterior-basal segment of the left lower lobe was excised by thoracotomy at a later date.

Actually the patient developed an isolated new mass on the left diaphragm at the site of the former excision of pulmonary metastases highly suspect for local recurrence. Thoracic CT scan revealed a right pneumothorax without evidence of other nodules. To exclude right-side metastatic disease, thoracoscopy was performed. A 2-mm metastasis was detected at the surface of the right lower lobe (Fig. 1). Histology of the resected small nodule showed tumor infiltration of the visceral pleura probably responsible for the pulmonary air leakage.

3. Discussion

Metastatic soft tissue sarcoma is often exclusively located in the lung. The appearance of SP in metastatic lung disease, however, is rare and most commonly described in advanced disease or during cytotoxic therapy [1,3–5,7,10]. In a series of 1143 patients with SP seen at the Mayo Clinic between 1953 and 1973, only 10 were attributed to malignant pulmonary neoplasms and in only two of these 10 cases did SP precede the development of demonstrable pulmonary nodules on chest X-ray [1]. In more recent studies reported after introduction of CT scan and magnetic resonance imaging (MRI), one further case of untreated radiographically occult tumor causing SP has been described [2]: after local excision of a scalp angiosarcoma, bilateral pneumothoraces appeared, attributed pulmonary micrometastasis, detected during post mortem examination of the lung. In two additional reports of pneu-
Pneumothoraces associated with radiographically undetectable pulmonary metastasis, recurrent and often bilateral pneumothoraces occurred during and after systemic chemotherapy of patients with osteogenic sarcoma [6,8]. In both of our cases, SP was the first indicator of pulmonary metastases. While in one of our patients, the first thoracoscopic exploration failed to detect metastases, 1 year later nodules appeared on CT scan at the time of the second recurrence of SP and the suspicion of pulmonary metastases was confirmed by thoracoscopic excision biopsy. In the second case, thoracoscopy was able to detect small peripheral metastases on both sides—on the left side despite negative chest X-ray and 3 years later on the right side despite negative CT scan findings.

Several theories regarding possible mechanisms of pleural disruption in metastatic or primary lung disease have been put forward [10]. Spontaneous rupture of necrotic tumor tissue or of oncological treatment may create bronchopleural fistulae. Alternatively, intermittent bronchiolar obstruction by nodules at the lung periphery may lead to subpleural blebs [10]. Histological examination of the nodules excised in our second case revealed neither tumor necrosis nor blebs. It is possible that tumor invasion and disruption of the visceral pleura and of peripheral bronchioles might have produced slow air leakage resulting in a small and clinically silent pneumothorax.

If a patient with a known malignant neoplasm, especially sarcoma, develops a pneumothorax, pulmonary metastatic disease has to be considered. The first line of approach in all patients consists of a chest CT scan. As shown in this report, however, small subpleural pulmonary metastases causing minor air leaks might be detected by careful thoracoscopic exploration while the CT scan is negative. Some authors recommend clinical observation of such patients with potential radiographically occult metastases for 60 days to provide evidence of pulmonary nodules [1]. However, we believe that thoracoscopic exploration should be attempted in every case of SP in a patient with a history of malignancy in order to prove metastatic involvement of the lung. Being a useful preliminary diagnostic procedure, videothoracoscopy should not be considered as an adequate method to treat pulmonary metastatic disease. If immediate radical resection of all lesions is intended, thorough manual palpation of the lung by means of bilateral open exploration cannot be replaced by video-assisted thoroscopic surgery (VATS) techniques.

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References