CASE REPORT

Lingual Carcinoma Metastases Presenting as Spontaneous Pneumothorax

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Spontaneous pneumothorax is a rare manifestation of primary lung cancer or metastasis. We report a 39-year-old man with well-differentiated squamous cell carcinoma of the tongue and cervical lymph node metastases. He developed lung metastases and spontaneous pneumothorax 22 months later after intra-arterial infusion chemotherapy. The patient was managed with partial lung resection under thoracotomy. The pneumothorax resolved completely after the operation. Histological examination demonstrated metastatic squamous cell carcinoma, which had led to a bronchopleural fistula with subsequent induction of pneumothorax. The patient recovered uneventfully and continued to receive adjuvant chemotherapy in the oncology surgery outpatient department. Unfortunately, the tumors of the tongue and cervical lymph nodes progressively enlarged despite treatment. Eventually, the patient died of respiratory failure 5 months later. In most of the previously reported cases, pulmonary metastases associated with spontaneous pneumothorax usually originate from osteogenic or soft-tissue sarcomas. Although rare, pulmonary metastasis should be considered in the etiology of spontaneous pneumothorax. Despite advanced disease, surgical treatment may be feasible. [*J Formos Med Assoc* 2009;108(9):736–738]

Key Words: fistula, pneumothorax, thoracoscopy

Case Report

A 39-year-old man was diagnosed in June 2004 with squamous cell carcinoma of the tongue with cervical lymph node metastases. He did not have pneumothorax or pulmonary metastases at the time of diagnosis. Histologically, the tumor was a well-differentiated squamous cell carcinoma. Adjuvant therapy was intra-arterial infusion chemotherapy with methotrexate (50 mg/week) and mitomycin C (2 mg/week) in the oncology surgery outpatient department. However, the tumors of the tongue and cervical lymph nodes progressively enlarged despite treatment.

In April 2006, the patient was readmitted with sudden chest pain, and radiography revealed left pneumothorax. Despite chest tube air drainage, the air leakage persisted after 10 days. Computed tomography showed a perforated cystic nodule near the surface of the left upper lobe, and left pneumothorax (Figure 1A). There was also a small amount of pleural effusion, but the cytological examination of thoracentesis showed no malignant cells. As the lesion was considered to be a metastatic tumor and the possible cause of pneumothorax, thoracotomy was performed. It revealed a 5-mm nodular perforation into the pleural cavity (Figure 1B). Partial resection was

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Received: September 13, 2007
Revised: January 21, 2008
Accepted: March 14, 2008

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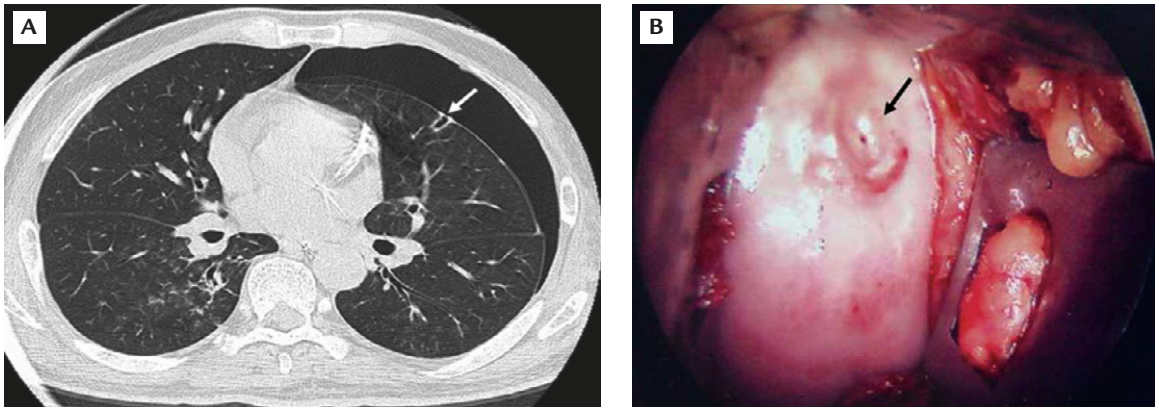

Figure 1. (A) Thoracic axial computed tomography shows a perforated cystic mass (white arrow) located in the left upper lobe near the pleura. (B) An approximately 5-mm perforated nodule (black arrow) was detected during thoracoscopic exploration of the left upper lobe.

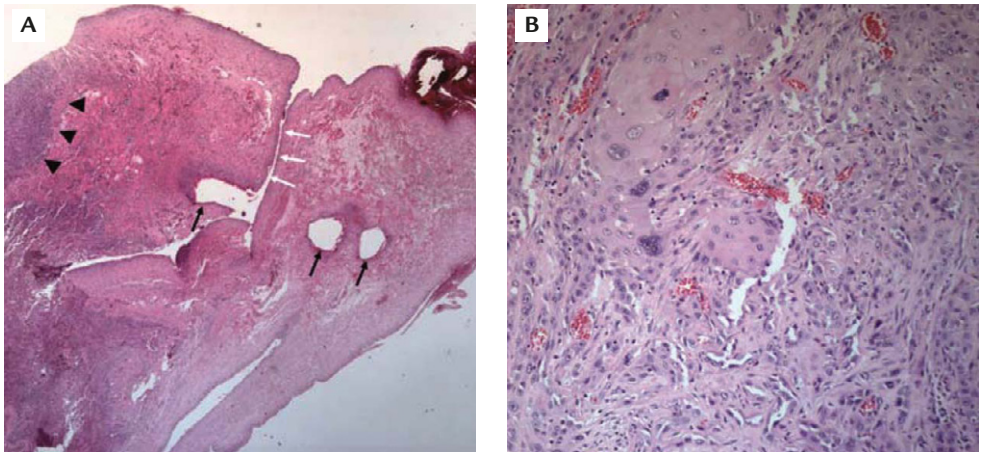

Figure 2. (A) A bronchopleural fistula (white arrows) was lined by granulation tissue without conspicuous invasive tumor in the underlying collapsed lung. Several subpleural bullae (black arrows) were seen. There was a tumor (black arrowheads) in the thickened pleura adjacent to the fistula [hematoxylin & eosin (H&E); original magnification, 2×]. (B) Pathology revealed well-differentiated squamous cell carcinoma with focal keratinization. Several giant cancer cells were seen (H&E; original magnification, 20×).

carried out. Histological examination demonstrated dissemination of well-differentiated squamous cell carcinoma (Figure 2) at the periphery of the lung and the adjacent thick pleura. There was a bronchopleural fistula lined by granulation tissue. No conspicuous invasive tumor was noted in the bronchopleural fistula or in the underlying collapsed lung. The alveoli in the periphery of the tumor were enlarged. These findings indicated that the perforating lesion was a metastasis from the previous lingual carcinoma. The patient recovered uneventfully and continued to receive adjuvant therapy in the oncology surgery outpatient department. Unfortunately, the tumors of the tongue and cervical lymph nodes progressively enlarged despite treatment. The patient died of respiratory failure 5 months later.

Discussion

Spontaneous pneumothorax secondary to malignant pulmonary neoplasm, although well-documented, is uncommon.[1–4] It is estimated that < 1% of all cases of spontaneous pneumothorax are tumor-associated.[1,4] Metastatic osteogenic or soft-tissue sarcomas are associated most commonly with pneumothorax, particularly in the setting of administration of cytotoxic chemotherapy or radiotherapy.[5] Although metastases from

squamous cell carcinoma of the head and neck usually occur in the lung, spontaneous pneumothorax in such cases is extremely rare.[6]

It is well known that malignant tumors can cause secondary pneumothorax, and the following mechanisms have been suggested to explain how this occurs. First, spontaneous rupture of a necrotic tumor or rupture as a result of oncological treatment causes communication between the bronchus and the pleural cavity;[4,7] second, tumor nodules at the periphery of the lung cause a ball-valve action, which over-distends the lung to form subpleural bullae, which leads to rupture and bronchopleural fistula;[4] and third, in rare cases, the tumor spreads to the pleura itself.[2]

In our case, although examination of the resected specimen of the left lung revealed tumor dissemination at the periphery of the lung and the adjacent thick pleura, neither necrosis of the tumor and lung tissue nor any tumor invasion of the bronchopleural fistula was demonstrated. The alveoli at the periphery of the tumor were enlarged and the visceral pleura had ruptured at the periphery, which led us to suspect that the pneumothorax was caused by the ball-valve action of tumor pressure on the bronchioles.

The optimal management of pulmonary malignancies with spontaneous pneumothorax has yet to be determined. Some patients have been managed successfully with conservative measures, including bed-rest, closed-system air drainage, and pleurodesis, but without resection, the pneumothorax tends to recur. Fenlon et al[8] stated that pneumothorax that results from bronchopleural fistula as a complication of chemotherapy may prove resistant to conventional treatment, and thus requires thoracostomy and pleurectomy. Our patient had intractable air leakage, and resection was indicated because of suspected pulmonary metastasis. Surgical removal of such metastasis can improve respiratory function rapidly and may have a positive bearing on the patient's overall prognosis.

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