Bilateral spontaneous pneumothorax in an osteosarcoma patient with pulmonary metastases: A case report

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Abstract. Spontaneous pneumothorax is a rare complication of chemotherapy in the treatment of lung neoplasms. It is relatively common in osteosarcoma lung metastases patients following chemotherapy, but the cause is still unknown. Here, we report a case of bilateral spontaneous pneumothorax occurring in a 40-year-old female in whom lung metastases from osteosarcoma were treated with combination chemotherapy. The patient had osteosarcoma on the right side of the mandible, and developed bilateral spontaneous pneumothorax. Through contrasting chest computed tomography, pneumothorax was identified at the site of the pulmonary metastases and occurred following the formation of bullae. The pneumothorax and bullae were produced after the patient received chemotherapy. In October 2014, the patient succumbed to the disease due to hemorrhage of the primary osteosarcoma lesions.

Introduction

Spontaneous pneumothorax in patients with osteosarcoma is a well-known phenomenon. It occurs as a complication of pulmonary metastases of osteosarcoma or during chemotherapy. Pneumothorax is a clinical emergency (1,2). The incidence rate of the pneumothorax in osteosarcoma patients is extremely low; spontaneous pneumothorax occurs in <2% of patients with pulmonary metastases (3). Pneumothorax affects respiratory function and endangers patient life by interfering with the treatment of osteosarcoma. Subsequently, pneumothorax significantly decreases quality of life and increases the risk of mortality in osteosarcoma patients (4). It reported that <10% of sarcoma patients with spontaneous pneumothorax survive for >2 years following the initial diagnosis of pneumothorax (5). Surgical resection is considered the best treatment for this complication (3). Here, we present a case of bilateral spontaneous pneumothorax with pulmonary metastases. We observed that pneumothorax developed after the formation of bullae through chest computed tomography (CT). In addition, all of these events occurred after the patient had received chemotherapy. Written informed consent was obtained from the patient's family.

Case report

A 40-year-old female presented with right lower lip numbness in 2007 and a mass was identified on the right mandibular area in April 2008. CT revealed right mandibular body bone destruction and a soft tissue mass. Following resection of the right mandible, osteosarcoma was revealed by biopsy. The right mandible osteosarcoma relapsed in February 2009. Following surgery, the patient was treated with four cycles of dacarbazine + ifosfamide + epirubicin. However, the osteosarcoma relapsed once more in June 2013. In September of the same year bilateral pulmonary metastases was confirmed by CT. The patient was then treated with one cycle of adriamycin (ADM) and cisplatin (DDP). Chest CT revealed left liquid pneumothorax on 12 December 2013. Following tube drainage therapy, the left liquid pneumothorax was cured and another cycle of ADM + DDP was administered. The patient suffered from severe anemia and thus, chemotherapy could not be applied regularly (every 3 weeks). Notably, the occurrence of pneumothorax was observed during the long intervals between chemotherapy treatments. We compared the chest CT results taken on 27 May 2014 (Fig. 1) with those taken on 14 July 2014 (Fig. 2). It was observed that bullae were present on the site of the pulmonary metastases. When pneumothorax occurred, the bullae disappeared. We hypothesize that the pneumothorax occurred due to the rupture of the bullae. In October 2014, the patient succumbed to the disease due to hemorrhage of the primary osteosarcoma lesions.

Discussion

In the present case, we identified that pulmonary metastases cause the formation of bullae. When the bullae ruptured, pneumothorax occurred in our osteosarcoma patient. We also consider that chemotherapy could play a significant role in delaying the onset of pneumothorax. Effective chemotherapy may prevent the growth of the tumor, and delay the appearance of the bullae.

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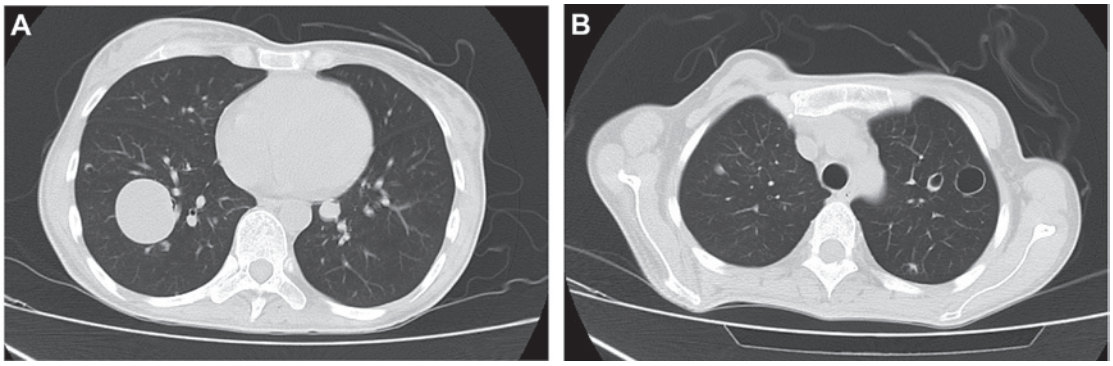

Figure 1. Chest computed tomography image from 27 May 2014. (A) Bullae on left ventricule; (B) Bullae on bifurcation of trachea.

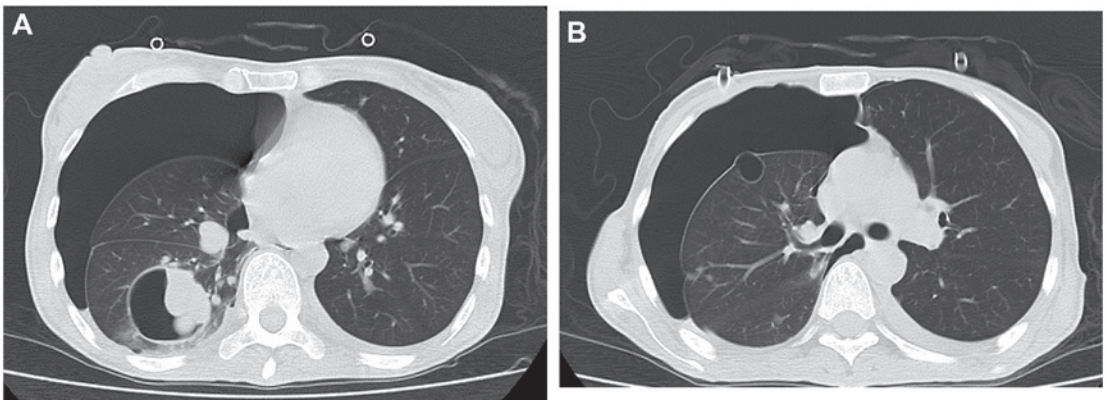

Figure 2. Chest computed tomography image from 14 July 2014. (A) Bullae on left ventricule; (B) Bullae on bifurcation of trachea.

The association of spontaneous pneumothorax with osteosarcoma is well known (1,6,7). Patients with osteosarcoma that have pulmonary metastases have a higher risk of spontaneous pneumothorax than patients with pulmonary metastatic carcinoma, but the reason for this remains unclear (8). It is also unclear whether there is any correlation between pneumothorax and lung metastasis (9). Regardless of whether patients with osteosarcoma have pulmonary metastases, those who receive chemotherapy have a higher risk of spontaneous pneumothorax than those who do not (8). It has been hypothesized that the pneumothorax was the result of ruptures of the necrotic subpleural micrometastasis in patients treated with chemotherapy (2). However, Smevik and Klepp reported that the bullous change caused the metastatic pulmonary disease, and that the peripheral tumor produced partial bronchial obstruction and caused a ball-valve phenomenon (8). This case provided us with radiographic evidence of this phenomenon.

Through studying this patient, we identified an association between the bullae on the site of the pulmonary metastasis and the pneumothorax. By comparing the chest CT film before and after the occurrence of pneumothorax, we identified that the bullae near to the pleura had disappeared when the pneumothorax occurred and thus, we hypothesize that the rupture of the bullae may have caused pneumothorax to occur. Although the pathogenesis of spontaneous pneumothorax is not clear, we conclude that the metastases under the pleura and the enlargement of the bullae are necessary for the phenomenon. However, the reason why the ball-valve phenomenon occurred in pulmonary metastases in osteosarcoma while it is rarely observed in other lung tumors is unknown. The answer should be elucidated following further research on the molecular biology in the future.

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