A Case of Metastatic Osteosarcoma Developed Pneumothorax after Chemotherapy

Masahiko KANAMORI1,2, Masanori NOBUKIYO2, Kayo SUZUKI2
Shigeharu NOGAMI3, Taketoshi YASUDA3, Tomoatsu KIMURA3

Abstract
A case of osteosarcoma arising in the left humerus of an 18-year-old man associated with the congenital deficiency of the talus is presented. He developed spontaneous pneumothorax after the combination chemotherapy of Adriamycin (ADM) and cisplatinum (CDDP). The patient managed with tube thoracostomy in each event, and recovered. The mechanism of spontaneous pneumothorax is not completely understood, but it will be appropriate to consider these findings in relation to the rapid regression of the pulmonary metastatic lesion following chemotherapy.

Key words: osteosarcoma, pneumothorax, lung metastasis

Introduction
Spontaneous pneumothorax is a rare manifestation of metastatic lung cancers and described in advanced diseases or during cytotoxic chemotherapy which is manifested by sudden onset of dyspnea. The cause or mechanism of spontaneous pneumothorax has been unknown, as well as the association with site of metastases or type of cancers. The side effects of chemotherapeutic drugs have been also reported rarely1).

A case of osteosarcoma arising in the left humerus of an 18-year-old man associated with the congenital deficiency of the talus is presented. He developed spontaneous pneumothorax after the combination chemotherapy of Adriamycin (ADM) and cisplatinum (CDDP). The patient managed with tube thoracostomy and each lung was expanded, and recovered.

Case report
An 18-year-old man presented with left shoulder pain. The family indicated the swelling of his shoulder, which had been slowly enlarging. The patient had visited another hospital, and was pointed out the abnormal shadow of the left proximal humerus on the radiographs. Physical examination revealed tenderness over the left shoulder, which was swelling hard. Moreover, a small nodule (1 × 2 cm) was palpable in the axillary region. Radiographs revealed an osteosclerotic lesion of the proximal humerus (Fig. 1). A chest radiogram showed multiple metastatic lung lesions at that time (Fig. 2). He underwent an open biopsy of the bone and resection of a swelling lymph node. Histologic examination of the biopsied specimen of the left humerus and a metastatic lymph node showed a conventional osteoblastic osteosarcoma.

After biopsy, therefore, systemic ADM and CDDP were given. At first, ADM (30 mg + 40 mg) and CDDP (100 mg) were given. The patient felt just nausea without other systemic complication for 1 week after chemotherapy. The chemotherapy was effective for the lung metastasis (Fig. 3). Three weeks later, therefore, he received the chemotherapy of ADM (50 mg + 40 mg) and CDDP (130 mg) again. After nine days of 2nd chemotherapy, however, he developed a right-side spontaneous pneumothorax. We could diagnose only chest radiograph as a routine examination (Fig. 4), because the patient did not complain due to mental retardation based on a congenital deficiency of the talus. The patient was managed with right-side tube thoracostomy and the lung was expanded. Moreover, 12th days of 2nd chemotherapy (3 days

1Department of Human Science (1), Faculty of Medicine, University of Toyama
2Department of Orthopaedic Surgery, Faculty of Medicine, University of Toyama
after the event of right-side pneumothorax, he developed the left-side spontaneous pneumothorax again (Fig. 5). The patient managed with left-side tube thoracostomy and the lung was expanded again. After that, however, bilateral lung metastases were developed and progressed despite additional BCD (Bleomycin, Cyclophosphamide, Dactinomycin) chemotherapy. The patient died due to respiratory failure after 4 months from the initial therapy.

Discussion

Osteosarcoma might occur as a sporadic event rather than a related occurrence in patients with the congenital deficiency of corps callosum. To date, there has been no report of osteosarcoma associated with congenital deficiency of corps callosum. A delay in the diagnosis of pneumothorax occurred due to the mental retardation in this case.

Pneumothorax occurring as a complication of the anti-tumor effects of cytotoxic chemotherapy has been reported in occasional cases of a variety of tumors. There appears to be an increased incidence of spontaneous pneumothorax in patients with metastatic osteogenic sarcoma compared to those with other tumors metastatic to lungs21. In patients with osteosarcoma, pneumothorax was reportedly observed spontaneously, or as a result of rapid regression of a tumor following chemotherapy20. ADM and CDDP
have been incorporated in the treatment of osteogenic sarcoma since the 1970s. But the pathophysiology of side effects is not completely understood, and further investigation of the pharmacokinetics of ADM and CDDP is needed. But it will be appropriate to consider these findings in relation to the rapid regression of the pulmonary lesion following chemotherapy. In this report, we described a rare complication in patients with osteosarcoma.

**Conclusion**

This report underlines that spontaneous pneumothorax can occur as the rare manifestation of lung metastasis of the osteosarcoma. The mechanism of spontaneous pneumothorax is not completely understood, but it will be appropriate to consider these findings in relation to the rapid regression of the pulmonary lesion following chemotherapy.

**REFERENCES**

