CASE REPORT

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Spontaneous pneumothorax without any detectable pulmonary metastases in a patient with osteosarcoma

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Abstract An 11-year-old girl with osteosarcoma in the left distal femur, developed unilateral spontaneous pneumothorax. Pneumothorax was found at the initial presentation, but chest CT failed to reveal pulmonary metastases, bullae or blebs.

Résumé Une fillette de 11 ans présentant un ostéosarcome de la région distale du fémur, chez qui est apparu un pneumothorax spontané. Le pneumothorax a été constaté à la première visite, mais la tomographie par reconstruction d'image n'a permis de révéler ni métastases pulmonaires, ni bulles pulmonaires ou sous-pleurales.

Introduction

Spontaneous pneumothorax in patients with osteosarcoma is a well-known phenomena. It occurs either as a complication of pulmonary metastases of osteosarcoma or during chemotherapy. Here, we present a case of unilateral spontaneous pneumothorax without any detectable pulmonary metastases in a patient with osteosarcoma.

Case report

An 11-year-old girl with no prior medical history complained of left knee pain for 6 weeks and right-side chest pain for 10 days. She was 151 cm tall, weighed 29 kg, and had not yet experienced menses. Radiographs of her left knee revealed osteolysis in the distal femur (Fig. 1).

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Chest X-rays showed right pneumothorax but CT (slices: 5 mm) failed to reveal any pulmonary metastases, bullae or blebs (Fig. 2). An open biopsy of the left femur showed osteosarcoma. The patient required 15 days of tube drainage. She was then treated with two cycles of methotrexate and two cycles of doxorubicin and CDDP. There was no recurrence of pneumothorax during chemotherapy. 4 months after the initial presentation, she underwent a prosthetic replacement of the left distal femur. However, the postoperative chest CT (performed 6 months after the initial presentation), revealed two pulmonary metastatic nodules in the ipsilateral side (Fig. 3). These were excised using thoracoscopic procedures. She is now receiving subsequent repeated chemotherapy.

Discussion

In this case, we excluded idiopathic spontaneous pneumothorax and pneumothorax associated with ectopic endometriosis due to the clinical and radiographic findings.

The association of spontaneous pneumothorax with osteosarcoma is well known [2, 6]. Patients with metastatic osteosarcoma in the lung have a higher risk of spontaneous pneumothorax than patients with metastatic carcinoma, but the reason for this remains unclear [4]. Whether or not patients with osteosarcoma have pulmonary metastases, those who receive chemotherapy have a higher risk of spontaneous pneumothorax than those who do not [4]. It has been hypothesized that the pneumothorax was the result of ruptures of the necrotic subpleural micrometastasis in patients treated with chemotherapy [1]. However, Sarno et al. reported that the bullous change heralded the metastatic pulmonary disease, and that the peripheral tumor produced partial bronchial obstruction and acted as ball-valve phenomenon [3].

Our case, with the patient being found to have pneumothorax at the initial presentation, was rare. She was not receiving chemotherapy, and had no detectable metastatic pulmonary diseases, bullae or blebs on CT. We found only few such cases in the English literature [5].

Fig. 1 Anteroposterior plain radiograph showing diffuse osteolysis of the left distal femur. Biopsy revealed osteosarcoma

Fig. 2 Chest computed tomographic image showing the right pneumothorax at the initial presentation

Fig. 3 Chest computed tomographic image obtained 6 months after the initial presentation showing pulmonary metastases (arrowhead)

In our case, we also consider that a subpleural or pleural micrometastasis may have played an important role in the occurrence of pneumothorax, because the patient later developed metastases in the ipsilateral lung.

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