Case Report

Epidural Pneumatosis Associated with Pneumomediastinum Occurring at Rest

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We report a rare case of epidural pneumatosis associated with pneumomediastinum that occurred at rest. A 17-year-old male had spontaneously felt shortness of breath while attending class during the day. Chest and neck CT revealed interstitial free air collection along the right pulmonary vascular sheath. It was speculated that some cough or strain-induced increase in intra-alveolar pressure in the alveoli along the right upper lobe caused free air leaks from the ruptured alveoli, which accumulated along the bronchovascular tissue sheath and moved into the mediastinum and subcutaneous area, especially in the right neck, right anterior chest wall, and right axillary region. CT also demonstrated subcutaneous emphysema extended into the epidural space through the intervertebral foramen. He exhibited a benign self-limited course. (Ann Thorac Cardiovasc Surg 2009; 15: 38–41)

Key words: pneumomediastinum, mediastinal emphysema, epidural pneumatosis, pneumorrhachis, aerorachia

Introduction

Pneumomediastinum (mediastinal emphysema) and subcutaneous emphysema are well-known complications of barotrauma and occur in up to 10% of cases of blunt chest trauma,[1)] but they rarely occur at rest. Epidural pneumatosis (pneumorrhachis, aerorachia) is a complication associated with trauma, but it does not often occur with pneumomediastinum. Herein we report a case of epidural pneumatosis caused by a pneumomediastinum that occurred while the subject was in a seated position during high-school class. CT revealed a radiolucent line along the pulmonary artery in the right upper lobe, suggesting that air entering the pulmonary interstitial space passes to the mediastinum through leakage via bronchovascular layers. Furthermore, the air route of the epidural space from the pneumomediastinum was recognized on chest and neck CT.

Case Report

A 17-year-old healthy youth, body weight 60 kg (132 lb), body height 177 cm (5 ft 9 in), and body mass index 19.2, had spontaneously felt a shortness of breath while attending class during the day. He was sitting and studying at that time. Since the sensation did not improve, he visited the hospital that night. He was admitted with a pneumomediastinum, conspicuous subcutaneous emphysema, and complaints of neck discomfort and dyspneic sensation as a result of swelling in the neck. He had no history of bronchial asthma, violent cough, or vomiting; he had no apparent medical history of any kind.

On admission, the patient's body temperature was 38.0°C. A physical examination revealed widespread subcutaneous emphysema involving both sides of the

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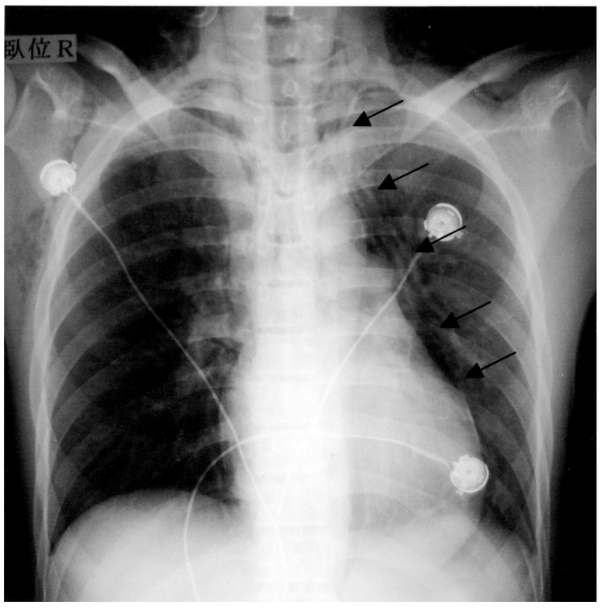

Fig. 1. Chest radiograph showing air (arrows) along the left border of the mediastinum and subcutaneous emphysema.

neck, right anterior chest wall, and right axillary region. Dysphonia was not observed. Dyspneic sensation was felt because of his neck swelling, and pulse oxymetry revealed his SpO_2 as 98% (room air). His respiratory rate was 15 breaths per minute. Continuous oxygen supplementation was started, and his SpO_2 became 100%. He had no neurological findings.

A laboratory examination showed white blood cell count (WBC) 12,700/mm^3 (normal range: 4,000–8,500) and C-reactive protein (CRP) 0.01 mg/dL (normally <0.50). An administration of antibiotics (cefazolin sodium) was started because of the elevated fever and WBC count.

Chest radiography showed the presence of mediastinal and subcutaneous emphysema (Fig. 1). Pneumothorax was not observed. A chest and neck CT scan revealed air in the mediastinum to the level of the diaphragm; in the cervical, right anterior chest wall; in the right axillary subcutaneous region; and in the spinal canal (Fig. 2). The niveau formation in the emphysematous area was not observed. CT also showed linear air collections arising within the lung and dissection along the right pulmonary artery (A^1a) (Fig. 3). The air collections extended toward the hilum and were responsible for the pneumomediastinum. Moreover, CT revealed subcutaneous emphysema that extended into the epidural space through the intervertebral foramen at the level of C7 and Th1 (Fig. 4).

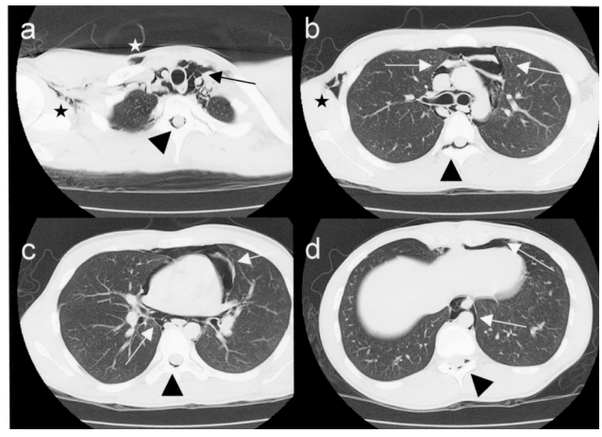

Fig. 2. Chest CT revealed mediastinal (arrows), subcutaneous (stars), and epidural (arrowheads) pneumatosis.

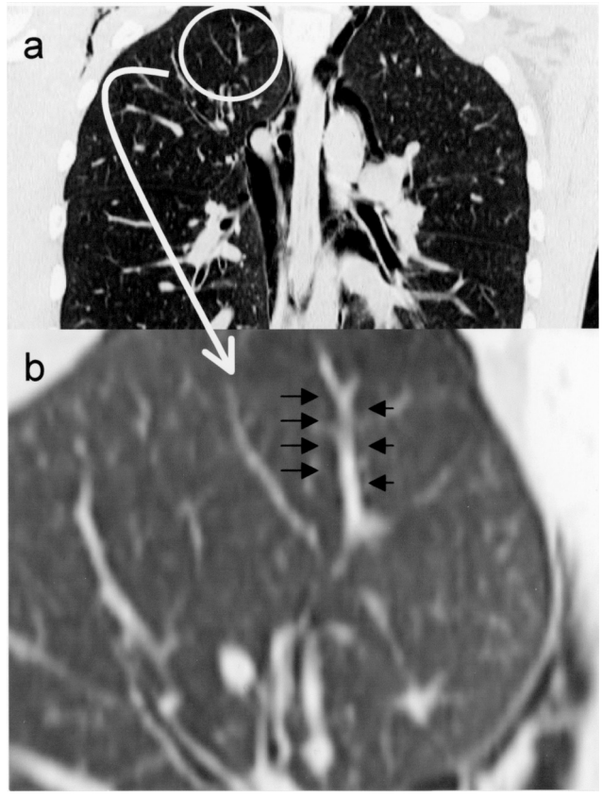

Fig. 3. CT revealed interstitial free air collections (in the circle, arrows) along the right pulmonary artery (A^1a).
This finding disappeared by the third hospital day.

The patient remained at rest and was treated conservatively, and the dyspnea disappeared. One day later, because the WBC count decreased to 8,200/mm^3, but the CRP increased to 0.68 mg/dL, the administration of

antibiotics was continued. Two days later, the subcutaneous emphysema of the neck and anterior chest had decreased. Three days later, the WBC count (4,700/mm^3) and CRP (0.42 mg/dL) normalized, and the antibiotics administration was discontinued. The need for oxygen supplementation disappeared completely, and the patient's SpO_2 was maintained in the 96% to 98% range. A resolution of the pneumomediastinum and subcutaneous emphysema was observed on CT, and the patient was discharged to his home after 7 days.

Discussion

Pneumomediastinum and subcutaneous emphysema are well-known complications of barotrauma. A possible explanatory mechanism for pneumomediastinum is high bronchioalveolar pressure resulting in air leakage into the pulmonary perivascular interstitium. On the other hand, spontaneous (atraumatic) pneumomediastinum, defined as the presence of extraluminal gas in the mediastinal space with no clear traumatic cause, has been reported in association with asthma, violent coughing, vomiting, sports, and excessive shouting.[2] When alveolar pressure increases temporarily, air entering the pulmonary interstitial space passes to the mediastinum via leakage through the bronchovascular layers.[2] The air dissects the paths of least resistance into the mediastinum to the fascial planes of the neck. This pathophysiological process was first described by Macklin and is called the Macklin effect.[3] As with our case, spontaneous pneumomediastinum occurs without an apparent precipitating factor or disease, usually in young people, and resolves within approximately 7 days.[4] However, in this case there was no trauma, asthma, vomiting, or past history. Although we could not be certain why this patient had pneumomediastinum, we speculated that some cough or strain-induced increase in intra-alveolar pressure in the alveoli along the right upper lobe, free air leaks from the ruptured alveoli, the air accumulates along the bronchovascular tissue sheath and then moves into the mediastinum and subcutaneous area, especially in the right neck, right anterior chest wall, and right axillary region. Our CT evidence supports this hypothesis.

Epidural pneumatosis accounts for the visualization of air in the spinal epidural space. Trauma of the skull or spine, epidural abscess, epidural anesthesia, and unsuccessful lumbar puncture may cause epidural pneumatosis.[2] However, epidural pneumatosis can occur in association with pneumomediastinum. The presence of air in the epidural space has been reported in association with pneumomediastinum in a few cases.[2] There are no fascial barriers between the posterior mediastinum and the retropharyngeal and epidural spaces; thus air can diffuse freely to the epidural space via the neural foramen and produce epidural pneumatosis.[5] Typically, air enters from the posterior aspect because of a relatively lower resistance in the loose connective tissue compared to that in the rich anterior vascular network.[6] In our case, the route from the subcutaneous emphysema to the epidural space was clearly demonstrated on the CT. Epidural air related to pneumomediastinum is a benign situation, and no further investigation or treatment is necessary.[2,4,6] Our patient showed a benign self-limited course.

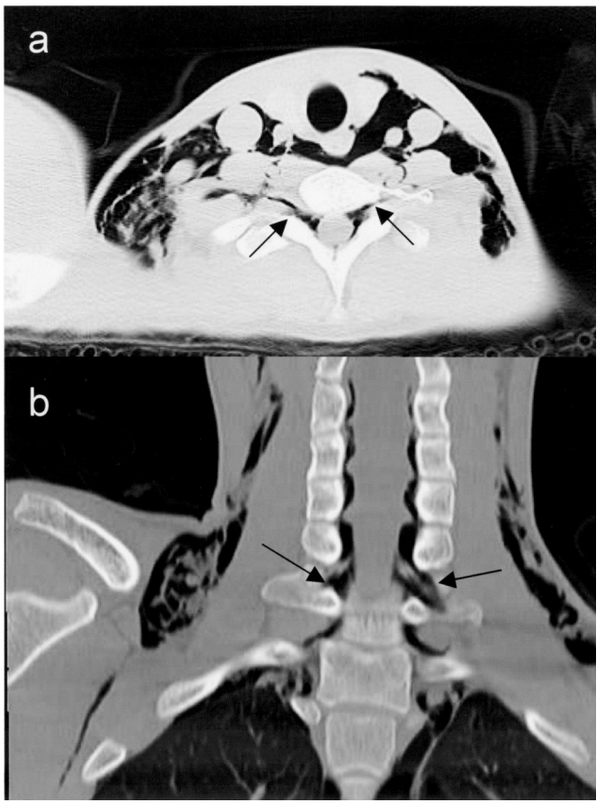

Fig. 4. Neck CT at the level of C7 and Th1.
Subcutaneous emphysema extended into the epidural space through the intervertebral foramen (arrows).

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reveal the Macklin effect. *AJR Am J Roentgenol* 1999; **172**: 129–30.
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