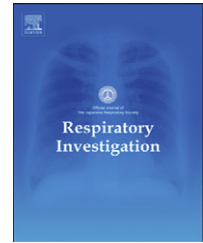




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Case report

Two cases of spontaneous epidural emphysema during asthmatic attack

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ABSTRACT

Two cases of spontaneous epidural emphysema that occurred during asthmatic attacks in a 13-year-old and a 15-year-old are reported here. Epidural emphysema was diagnosed in both cases by using computed tomography (CT), and in 1 case by using magnetic resonance imaging (MRI). Neither patient had neurological findings. Both patients were discharged with no respiratory difficulties. It is generally believed that a diagnosis of epidural emphysema can only be made on CT. In this report, MRI was used to make the diagnosis of subdural emphysema, and it demonstrated that the air was localized within the epidural fat.

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1. Introduction

Epidural emphysema, or pneumatosis, is a known complication of severe trauma involving the vertebrae [1–3]. However, epidural emphysema during an asthmatic attack is extremely rare. It is difficult to identify the area of emphysema on plain X-rays due to their poor discrimination ability; however, epidural emphysema can be easily seen on computed tomography (CT) [4]. In this report of 2 cases, epidural emphysema

was identified on CT in both cases and on magnetic resonance imaging (MRI) in 1 case.

1.1. Case 1

A 13-year-old male with a history of childhood bronchial asthma visited a nearby hospital due to cough, wheezing, and laryngeal involvement. He had no history of trauma, and there was no family history of pneumothorax. He was diagnosed with bronchial asthma and prescribed fluticasone. Because his symptoms did not improve, he visited another clinic. At that clinic, subcutaneous emphysema was noted, and the patient was referred to our hospital.

On physical examination, crepitus was noted in the neck and back. There was no wheezing in either lung field.

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The patient had no neurological symptoms, and the neurological examination was negative.

All laboratory tests were all within normal limits. On chest X-ray, subcutaneous emphysema was noted in the neck and bilateral axilla, while mediastinal emphysema was noted in the right hilar region and along the descending aorta. On CT, there was subcutaneous emphysema in the neck and bilateral

axilla, mediastinal emphysema, and epidural emphysema within the cervical and thoracic spinal canal (Fig. 1). No emphysematous change or cystic change was observed in the bilateral lung fields. Since the patient's symptoms disappeared on admission, he was observed without additional treatment.

1.2. Case 2

A 15-year-old male with a history of childhood bronchial asthma visited a nearby hospital complaining of cough and wheezing. As with case 1, he had no history of trauma, and there was no family history of pneumothorax. He had noted wheezing while playing table tennis. He was treated with corticosteroids and a bronchodilator at the hospital, but his symptoms worsened, and he was referred to our hospital.

On physical examination, crepitus was noted from the left precordial area to the neck, and loud wheezing was heard in bilateral lung fields. The patient had no neurological symptoms, and the neurological examination was negative.

In laboratory testing, the serum C-reactive protein was 1.2 mg/dL, and serum IgE was 1,653 mg/dL. On chest X-ray, subcutaneous emphysema was noted in the neck and left precordial area. On CT, there was subcutaneous emphysema in the neck and precordial area, mediastinal emphysema, and epidural

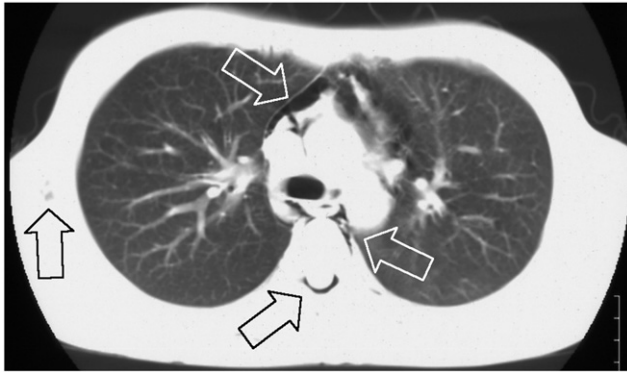


Fig. 1 – Computed tomography shows subcutaneous emphysema in the right axilla, mediastinal emphysema in the anterior mediastinum, and epidural emphysema (arrows).

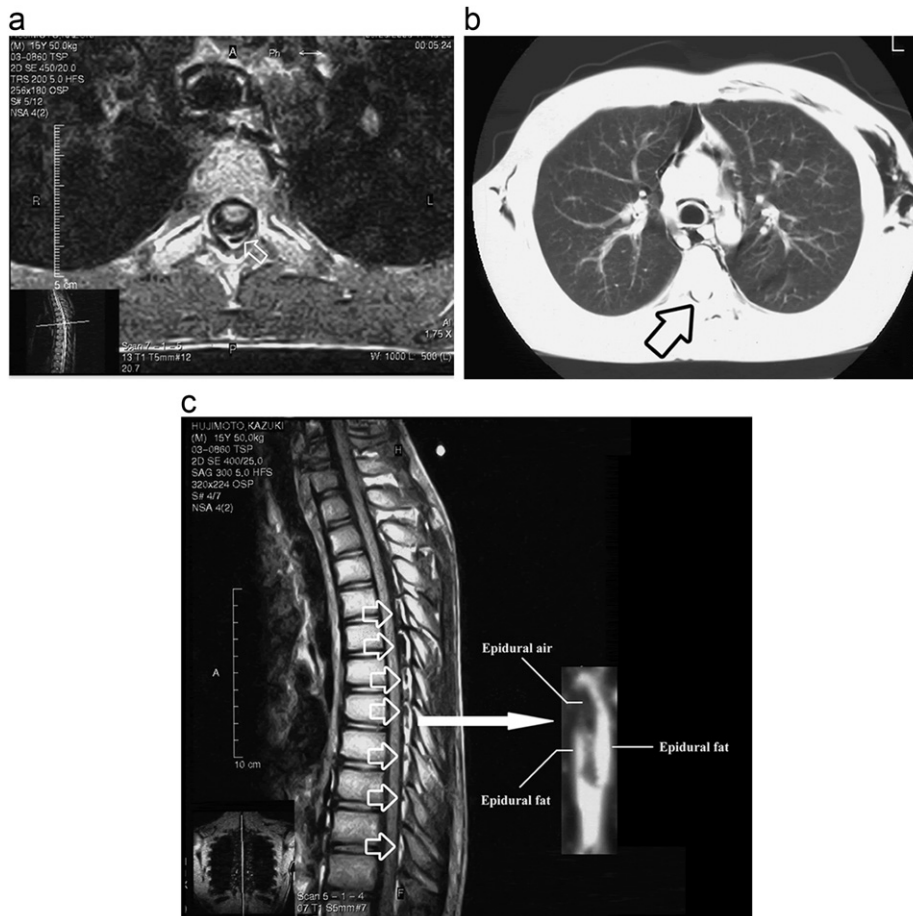


Fig. 2 – (a) T1-weighted MRI shows a no-signal area interposed between the epidural fat from T5 to T11 in the axial view (arrows). This figure shows the T7 level. (b) CT of the T7 level shows that the no-signal area is air (arrows). (c) In the sagittal view of T1-weighted MRI, the no signal area is surrounded by epidural fat (arrows).

emphysema within the thoracic spinal canal. No emphysematous change or cystic change was observed in bilateral lung fields. MRI was done to examine for spinal cord compression. T1-weighted MRI showed a no-signal area from T5 to T11 in the axial view (Fig. 2). CT of the same level showed that the no-signal area was air (Fig. 2b). In the sagittal view of T1-weighted MRI, this air was surrounded by epidural fat (Fig. 2c). The patient was treated with corticosteroids and a bronchodilator, and he was discharged after 9 days of hospitalization.

2. Discussion

Epidural emphysema, also called pneumatosis, results in air in the spinal epidural space [5]. Epidural emphysema has been reported in association with spontaneous pneumomediastinum [6], primary spontaneous pneumothorax [7], asthmatic attacks [8–12], violent coughing [4,9], forceful vomiting [13], strenuous exercise [14], Marfan's syndrome [15], and anorexia nervosa [16], as well as during sleep [17], and following trauma [2]. The average age of patients with epidural emphysema is under 20 years, because these young patients have friable connective tissue. The cause of epidural emphysema is unknown; however, it is interesting that pyrexia was noted in 11 of 22 reported cases, including the present 2 cases.

A possible mechanism by which epidural emphysema occurs involves hyperinflation, which causes rupture of the alveolar walls, after which air leaks into the perivascular sheaths and progresses to the hilum of the lung and the posterior mediastinum. Eventually, air passes through the intervertebral foramina into the epidural space [10].

It is generally thought that a diagnosis of epidural emphysema can only be made on CT [8]. The reason for this is that the images of mediastinal emphysema, subcutaneous emphysema, and subdural emphysema are superimposed on chest X-ray [18]. It is interesting to note that there was a linear lucency along the posterior border of the spinal canal in 1 reported epidural emphysema case [19]. In the present report, MRI was used to make the diagnosis and to determine the location of subdural emphysema in case 2. On MRI, the air was seen as a no-signal area. Furthermore, the air was located within the epidural fat on MRI. This report is the first to show that epidural emphysema is present within epidural fat. Although CT is superior for detecting epidural emphysema, because of radiation exposure, MRI is the preferred method for younger patients.

In case 2, the spin-echo method was used. However, to detect epidural emphysema, the gradient-echo method may be more sensitive than the spin-echo method [20].

Regarding neurologic findings, there were no neurological symptoms in the previous epidural emphysema cases [5,7–10,12,15], and the present 2 cases also had no neurological findings.

Treatment of epidural emphysema during an attack of bronchial asthma involves removal of the cause [11], giving a bronchodilator to reduce airway pressure, corticosteroids or antibiotics to suppress inflammation, accelerating clearance of secretions, providing oxygenation, and maintaining an intravenous drip.

Epidural emphysema during an asthmatic attack may appear to be an index of bronchial asthma severity [9,11,12]; however, it is known to be a benign complication because the entrapped air is often absorbed into the tissues within 7 days, as was observed in the present cases [11,17]. However, follow-up of this rarely seen entity is still important because serious neurological complications may occur in some cases [7]. In fact, the patient in 1 case died of intractable meningitis [21]. Although MRI was not useful for treatment in the present cases, it might be useful in severe cases, such as epidural emphysema caused by epidural abscess and trauma.

Conflict of interest

Authors have no potential conflict of interest.

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