Cervical Emphysema, Pneumomediastinum, and Pneumothorax Following Self-induced Oral Injury*

Report of Four Cases and Review of the Literature

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Spontaneous rupture of the pulmonary alveoli after a sudden increase in intra-alveolar pressure is a common cause of pneumomediastinum, which is usually seen in healthy young men. Other common causes are traumatic and iatrogenic rupture of the airway and esophagus; however, pneumomediastinum following cervicofacial emphysema is much rarer and is occasionally found after dental surgical procedures, head and neck surgery, or accidental trauma. We present four cases of subcutaneous emphysema and pneumomediastinum after self-induced punctures in the oral cavity. They constitute an uncommon clinical entity that, to our knowledge, has not been reported in the literature. Its radiologic appearance, clinical presentation, and diagnosis are described.

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Key words: cervical emphysema; chest CT; chest radiography; penetrating oral trauma; pneumomediastinum; subcutaneous pneumothorax

Spontaneous pneumomediastinum commonly occurs in healthy young men or parturient women in whom an increased intra-alveolar pressure (Valsalva maneuver, cough, emesis) leads to the rupture of the marginal pulmonary alveoli.1–3 The air ascends along the mediastinum toward the subcutaneous space of the neck, causing cervicofacial subcutaneous emphysema in 70 to 90% of cases.4–5 Inversely, pneumomediastinum following cervicofacial emphysema is very rare and has been reported in relation to dental surgical procedures, head and neck surgery, or orofacial trauma.6–8

We review four cases of subcutaneous cervical emphysema and subsequent pneumomediastinum secondary to self-induced injuries in the oral cavity in four young men from the same penitentiary center. Two of them also had pneumothorax demonstrated by chest radiography and CT. The radiologic appearance of this uncommon entity is described, and the English-language literature on the reported etiologies of pneumomediastinum is reviewed.

CASE REPORTS

Case 1

A 27-year-old man who smoked 10 cigarettes per day and had been treated for epilepsy since childhood was admitted to the hospital on November 3, 1997, complaining of sudden odynophagia, dyspnea, and cervicofacial emphysema. No history of trauma or surgery was reported. The initial physical examination showed important cervicofacial and thoracic subcutaneous emphysema. Results of the esophagogram, otolaryngologic examination, and bronchoscopy performed in the emergency department were normal. The initial chest radiograph showed subcutaneous emphysema in the cervicofacial, thoracic, and axillary regions with no evidence of rib fracture. The chest CT performed a few days later demonstrated air in the subcutaneous, visceral, and carotid spaces of the neck (Fig 1, top), extending along the anterior mediastinal space down to the aortic arch. A small right pneumothorax that collapsed the middle lobe slightly was also observed (Fig 1, bottom; arrows). Antibiotic therapy was administered to prevent mediastinitis, and the patient’s condition improved, making it possible for him to return to the prison.

Case 2

A 26-year-old man who was a former drug abuser, and was seropositive for HIV and hepatitis C, was admitted to hospital on November 4, 1997, reporting sudden periorbital and cervicofacial edema, chest pain, and dyspnea. He denied any autoprovocative maneuver. The initial physical examination, periorbital, cervicofacial, and thoracic emphysema were seen. The chest radiograph confirmed these findings, demonstrating a small left pneumothorax (Fig 2, arrows). The results of the esophagogram, otolaryng-

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gologic examination, and bronchoscopy ruled out any abnormality. Antibiotic therapy was administered to prevent mediastinitis, and his condition improved with progressive decrease of the emphysema and the pneumothorax.

Case 3

A 31-year-old former drug abuser who was seropositive for hepatitis C was admitted to hospital on January 5, 1998, with sudden cervicofacial swelling and chest pain. The patient reported fish ingestion the day before. In the initial physical examination, cervicofacial emphysema was found. The chest radiograph revealed important subcutaneous emphysema and pneumomediastinum (Fig 3, arrows). However, results of upper GI studies, otolaryngologic examination, and bronchoscopy performed in the emergency department were normal. During the following days, the patient’s condition improved notably, with almost total resolution of the cervical emphysema and pneumothorax.

Case 4

A 26-year-old former drug abuser arrived at the hospital on February 12, 1998, presenting with generalized pain and subcutaneous emphysema. He had reported having been punched in the prison. In the physical examination, the only positive finding was orbital and facial swelling, extending along the neck and anterior chest wall, down to the iliac fossa. However, there was no evidence of a traumatic lesion. The chest radiograph and CT confirmed the clinical findings and revealed the presence of pneumomediastinum.

On February 13, 1998, the patient was found unconscious with response only to intense painful stimulation. Naloxone and flumazenil were applied, and orotracheal intubation was performed. In the ICU, the patient continued to be unresponsive, hypotensive, and severely hypoxemic. As the results of cranial CT were normal and no toxins were found in the urine and stomach fluids, ischemic encephalopathy of unknown origin was suspected. Twenty-four hours later, the patient recovered consciousness with complete normalization of his neurologic functions. He progressed uneventfully until his total recovery.

Discussion

Spontaneous pneumomediastinum is usually seen in healthy young men or parturient women resulting from the rupture of peripheral pulmonary alveoli due to sudden increase of intra-alveolar pressure after exaggerated Valsalva maneuvers. Barotrauma during mechanical ventilation, ascent phase of a dive, or hyperbaric treatment are other possible causes. Spontaneous pneumomediastinum may also complicate obstructive airway processes such as asthma or foreign bodies. Subsequently, air descends along the connective tissue planes and vascular sheaths toward the mediastinum, ascending up to the communicating cervical spaces, producing subcutaneous cervical emphysema in 70 to 90% of cases. In 31% of cases, it has no known precipitating cause.
The mediastinum; likewise, air can ascend within the mediastinal space up to the root of the neck, producing subcutaneous emphysema.13

This diagnosis of pneumomediastinum is dependent on radiologic imaging: standard posteroanterior and lateral radiographs are usually sufficient for diagnosis, as posteroanterior chest radiographs typically demonstrate a radiolucent line between the left heart border and the mediastinal pleura.11,14 Other findings may include “highlighting” of the aortic knob and the “contiguous diaphragm” sign.1,11 However, posteroanterior chest radiographs by themselves may overlook 50% of cases, so that lateral chest radiographs should always be performed, which increases sensitivity to nearly 100%.3,13 With lateral views, air is visualized in the retrosternal space or as lucent streaks outlining the aorta and other mediastinal structures.11

In addition, radiographs may detect associated pneumothoraces.15 Lateral decubitus radiographs may sometimes be useful to differentiate a pneumothorax from pneumomediastinum: air will ascend to the highest point possible in the pneumothorax; however, mediastinal air shows little positional variations as it is relatively confined.1,11

Complementary diagnostic procedures (esophagogram, esophagoscopy, bronchoscopy, chest CT) are often performed following conventional radiographic imaging to rule out spontaneous (Boerhaave syndrome) or traumatic rupture of esophagus and tracheobronchial tree, among different causes of secondary pneumomediastinum.13 In our patients, results of the esophagogram, otolaryngologic examination, and bronchoscopy were normal. The chest CT was performed in two patients and confirmed pneumomediastinum in both patients and pneumothorax in one.

Initially, the four patients denied self-mutilation behavior. Two of them admitted their intentional insertion of sharp objects (needles and fish bones) into the oral cavity. Self-mutilation was subsequently confirmed in the other two patients. After the sublingual injuries were produced, all patients did the Valsalva maneuver. As no etiology for subcutaneous emphysema and pneumomediastinum was found, and the time frame of hospital admissions was so similar, the self-induced origin was initially suspected and subsequently confirmed. Such action could certainly suggest a possible collusion among the group of convicts from which the plan was elaborated, due to the implied intention of escaping from the penitentiary center by the simulation of an emergency.

The roots of the first, second, and third molars communicate directly with the sublingual and submandibular spaces.7,12 In addition, the visceral space of the neck communicates with the parapharyngeal, sublingual, and submandibular spaces anteriorly and superiorly11 and inferiorly directly with the mediastinum, between the trachea and great vessels (anteriorly) and down to the fourth thoracic vertebra (posteriorly). Thus, blood, pus, or air entering any of these spaces can migrate downwards into the mediastinum; likewise, air can ascend within the mediastinal space up to the root of the neck, producing subcutaneous emphysema.13

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Compressive Neuropathy of the Brachial Plexus and Long Thoracic Nerve*

A Rare Complication of Heparin Anticoagulation

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We present a case of a 69-year-old woman who developed brachial plexopathy and long thoracic nerve palsy secondary to compression from a hematoma while receiving heparin therapy for the treatment of a stroke. The patient was treated conservatively with discontinuation of heparin and had complete resolution of her compressive neuropathy. This is the first report of a patient with long thoracic nerve palsy with a brachial plexopathy complicating anticoagulation. We review the literature on hematoma-induced compressive neuropathies and treatment options. Our review concludes by emphasizing the importance of clinical judgment in determining the best therapeutic modality.

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Key words: anticoagulation; brachial plexus; compression; hematoma; long thoracic nerve

Abbreviation: APTT = activated partial thromboplastin time

Anticoagulant therapy is frequently used in both the inpatient and outpatient settings. As a consequence, clinicians frequently have to deal with the side effects of anticoagulation. In an attempt to increase general awareness, we report an unusual complication of heparin treatment and explore treatment options after a brief review of the literature.

CASE REPORT

A 69-year-old woman with a history of hemicolectomy for Dukes class D colon cancer was admitted to the hospital for evaluation and treatment of the ileus in association with findings of multiple mesenteric and omental soft tissue masses that were consistent with metastases on CT scans of the abdomen and pelvis. A chest radiograph was normal, and the results of a total body bone scan were negative.

Two days later, she developed acute binocular vision loss and was started on IV heparin therapy. A CT scan of the head revealed ischemic changes in both occipital lobes, an ultrasound of the neck showed normal flow in the vertebral arteries, and transesophageal echocardiography showed a small patent foramen ovale with trivial right-to-left shunting. No evidence of deep venous thrombosis was found on duplex ultrasonography of the lower extremities. The patient’s vision returned to normal over the next few days.

On day 5 of heparin therapy, the patient was grasping the bed-rail with her left hand in an attempt to roll over and developed a sudden onset of moderate left shoulder pain. Initially, there were no positive findings on physical examination, and she was treated with morphine. The results of laboratory tests at that time showed the following: activated partial thromboplastin time (APTT), 101 s (range, 21 to 33 s); hemoglobin level, 11.5 g/dL (range, 12 to 15.5 g/dL); platelet count, 233,000/μL (range, 150,000 to 450,000/μL). The rate of heparin infusion was decreased to achieve a target APTT of 60 to 90 s.

By the following day, the patient’s left shoulder pain had increased. Inspection revealed a large ecchymosis over the left axilla and flank, as well as mild left scapular winging, but no raised subcutaneous hematoma could be felt. The patient was not able to abduct her left arm beyond 50°, and the scapular winging was accentuated by having her push against the examiner’s hands. She was able to reasonably adduct, internally and externally rotate, and flex and extend the humerus, but a full assessment of left shoulder motor strength was limited by pain. Motor strength was normal in the remainder of the arm. Sensory examination, deep tendon reflexes, and upper extremity arterial pulses were normal. At that time, her APTT was 78 s, hemoglobin level was 10.4 g/dL, and platelet count was 224,000/μL. Heparin therapy was discontinued, and an MRI of the left brachial plexus was obtained, revealing a 7 × 4-cm mass subjacent to the left scapula in the region of the left axilla, which was consistent with a large hematoma (Fig 1). The hematoma involved the subscapularis muscle on the left and compressed the brachial plexus anteriorly (Fig 2). The hematoma also compressed the posterolateral left thorax in the region of the long thoracic nerve (Fig 3). An underlying metastasis was not seen.

Since the patient had only mild deficits, she was treated conservatively with analgesics and by withholding further administration of anticoagulant drugs. She had complete resolution of pain and a return of normal motor strength over the next 5 days, and winging of the scapula was no longer evident after 1 week.

DISCUSSION

The brachial plexus is formed by the union of the ventral rami of C5–T1. The plexus runs in the posterior triangle of...