Pneumomediastinum and Subcutaneous Emphysema in the Neck, Axilla, and Chest Regions in a Patient with Olfactory Neuroblastoma Treated with Chemoradiotherapy

Abstract
Spontaneous pneumomediastinum and subcutaneous emphysema in the neck, axilla, and chest do not commonly occur after neoadjuvant cisplatin/etoposide chemotherapy, followed by radiotherapy, and adjuvant cisplatin/etoposide chemotherapy in patients with olfactory neuroblastoma. There are few case reports of pneumomediastinum induced by and occurring during bleomycin/etoposide/cisplatin chemotherapy in testicular cancer. The present case differs from the previous cases in that our patient developed spontaneous pneumomediastinum and subcutaneous emphysema in the neck, axilla, and chest approximately 2 months after completion of chemoradiotherapy for olfactory neuroblastoma. These conditions may have been treatment induced or caused by breath-holding after forceful inspiration. The latter would have created a massive pressure gradient between the alveoli and surrounding structures, causing alveolar rupture, and subsequent passage of air into the mediastinum and subcutaneous tissue of the neck, axilla, and chest.

Keywords: cisplatin, etoposide, mediastinum, radiotherapy, subcutaneous emphysema

Introduction
Bleomycin-induced pneumomediastinum in testicular cancer has been reported,[1] as has pneumomediastinum, subcutaneous emphysema, and pneumothorax after pulmonary function testing (the Valsalva maneuver) in a patient with bleomycin-induced interstitial pneumonitis,[2] Herein, we describe and radiologically document pneumomediastinum and subcutaneous emphysema in the neck, axilla, and chest 2 months after cisplatin/etoposide chemotherapy and radiotherapy in a patient with olfactory neuroblastoma.

Case Report
A 45-year-old man presented with nasal bleeding for 2 months. Local examination and computed tomography (CT) scan revealed a mass in the right side of the nasopharynx extending to the posterior nasal cavity. The mass was diagnosed as an olfactory neuroblastoma through immunohistochemistry of a biopsy specimen of the primary mass. Complete blood counts, random blood sugar levels, and liver and renal function tests were within normal limits. A chest X-ray (posteroanterior view), abdomen/pelvis ultrasound, and 12-lead electrocardiography showed no other abnormalities.

The patient received neoadjuvant chemotherapy with etoposide (100 mg, days 1–3) and cisplatin (40 mg, days 1–3) every 21 days for 2 cycles followed by external beam radiotherapy (70 Gy in 35 fractions for 7 weeks). Twenty-eight days after completion of radiotherapy, the chemotherapy was repeated (every 21 days for 4 cycles). The patient visited the radiotherapy department 13 days after treatment completion (first follow-up). He was asymptomatic, with no evidence of residual local disease.

Two months after treatment completion (second follow-up), the patient presented with breathlessness. Chest X-ray (posteroanterior view) [Figure 1] and CT of the neck and chest showed extensive subcutaneous emphysema in the neck and axilla extending to the mediastinum [Figure 2]. He was referred to the chest unit where an intercostal drain (ICD) was inserted. The ICD relieved
the pneumomediastinum and subcutaneous emphysema, and
the patient was symptomatically better. After withdrawing
the ICD, he was discharged from hospital. One week later,
he died suddenly at home after an attack of coughing.

**Discussion**

The visceral space, which is continuous from the neck
and mediastinum to the retroperitoneum originates during
embryological development.[3] The visceral pleura overlies
both lungs in addition to the organs in the mediastinum
and encloses a space known as the visceral cavity.
The fascial planes connect cervical soft tissue to the
mediastinum and retroperitoneum, and the spaces between
the fasciae permit aberrant air arising in any of these
areas to spread. In patients with pneumomediastinum,
air can flow from the peribronchial space to the neck,
chest wall, pleural cavity, mediastinum, retroperitoneum,
pericardial cavity, pericardial space, and diaphragm.[3]
Although the body cavity is further divided into thoracic,
abdominal, and pericardial cavities by the pleuroperitoneal
and pleuropericardial membranes, the cavities remain
continuous throughout the visceral space.[4]

A chest X-ray usually detects pneumomediastinum and
subcutaneous emphysema although better visualization
is obtained through CT scan. In our case, CT scan
showed extensive pneumomediastinum and subcutaneous
emphysema. The patient underwent respiratory failure,
which resulted in his death 2 months after the completion
of treatment. The respiratory failure may have been
chemotherapy drug induced or caused by forced
expiration (e.g., the Valsalva maneuver with breath holding
for a prolonged period).[2]

Spontaneous pneumomediastinum is an uncommon event
characterized by retrosternal chest pain, subcutaneous
emphysema, dyspnea, and dysphonia. Classic signs
of pneumomediastinum include Hamman’s Crunch,
a crepitant sound that varies with the heartbeat on
auscultation of the precordium.[5,6] Known triggers,
which can be identified in some cases, include acute
asthma exacerbation and the Valsalva maneuver; such
complications are induced by structural changes associated
with interstitial and other lung diseases. Other known
triggers include those related to the Valsalva maneuver,
sterneous exercise, weightlifting, inhalation of illicit
drugs, coughing, forced evacuation, and labor as well as
vomiting, respiratory infections, foreign body aspiration,
and barotrauma. Pneumomediastinum usually follows a
benign, self-limiting course.[5,6]

Treatment of pneumomediastinum includes
mediastinotomy and supportive care consisting of
analgesic use, rest, and the avoidance of maneuvers
that increase transpulmonary pressure (e.g., the Valsalva
maneuver and spirometry). Radiotherapy is a treatment
option for patients with prolonged air leaks who are not
candidates for surgery.[7]

Our patient developed subcutaneous emphysema in the neck,
axillary, and chest regions as well as pneumomediastinum
and pneumothorax. He did not experience these
complications during chemotherapy/radiotherapy and had
no preexisting lung disease. An earlier study found that
most air leak syndromes resulted from alveolar rupture
rather than rupture of subpleural bullae.[8]

Intense respiratory efforts, adjacent atelectasis, and low
intravascular pressure may contribute to the development
of air leaks.[8] When a person inhales, the diameter of
the pulmonary vein increases owing to a decrease in
intrathoracic pressure and an increase in alveolar air volume
and the venous return. Because all compartments increase
in length and width in tandem, there is no pressure gradient
in the interstitial space. However, when a person “holds
his breath,” venous stasis occurs, which hinders pulmonary
venous filling, reduces vessel lumen size, and consequently
creates the pressure gradient necessary for alveolar rupture.
The Valsalva maneuver is classically related to air leak

**Figure 1:** A chest X-ray (posteroanterior view) showing subcutaneous
emphysema in the neck

**Figure 2:** Computed tomography of the neck and chest showed extensive
subcutaneous emphysema in the neck and axilla extending to the
mediastinum
syndromes because it increases intrapulmonary pressure.\textsuperscript{[8]} Therefore, labor and forced evacuation are associated with pneumomediastinum.\textsuperscript{[8]} Unreleased air pressure in the mediastinum can compress the heart and vessels and prevent lung inflation and deflation, resulting in death.

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**Conflicts of interest**

There are no conflicts of interest.

**References**