

CASE REPORT

Spontaneous pneumomediastinum, pneumoretroperitoneum, and cervicofacial subcutaneous emphysema after repeatedly and forcefully blowing into a bottle

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Spontaneous pneumomediastinum (SPM) is an uncommon, self-limiting condition associated with increased intra-thoracic pressure resulting in alveolar rupture. Search of the literature revealed no detailed case report about a 26-year-old psychiatric patient who repeatedly and forcefully blew air into a bottle for 5 days resulting in a combined condition of spontaneous pneumoretroperitoneum, pneumomediastinum, and cervicofacial subcutaneous emphysema. It is crucial to find a primary source and treat appropriately. Psychiatric patients may have psychotic behaviors mimicking Valsalva's maneuver that increases intra-thoracic pressure and causing SPM. Optimal medications should be given to control psychotic behaviors. Family members and caregivers should be explained about this unusual behavior so that they can prevent this rare condition.

Keywords: *pneumomediastinum; pneumoretroperitoneum; subcutaneous emphysema; blowing bottle; valsalva maneuver*

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Spontaneous pneumomediastinum (SPM) is characterized by the presence of air in the mediastinum without underlying lung disease, and it is a rare situation with incidence of 1 in 30,000 emergency department (ED) referrals (1). Alveolar rupture is caused by high intra-alveolar pressures, low peri-vascular pressures, or both. In this situation, air escaping from the alveoli tracks into the mediastinum during the respiration cycle because the pressure in the mediastinum decreases relative to the pulmonary parenchymal pressure. As a result, air may track into the cervical subcutaneous tissues and peritoneal cavity (2).

Spontaneous pneumoretroperitoneum is an infrequent condition without apparent underlying abdominal pathology such as perforation, iatrogenic instrumentation, or infections. Interestingly, the spaces between the retroperitoneum, mediastinum and subcutaneous tissue are anatomically continuous, and air emerging from a lesion in any of these areas can travel to another place along the fascial planes (3).

Case presentation

A 26-year-old male patient with past medical history of schizoaffective disorder, hypertension, and dyslipidemia

was brought to ED by his mother after she noticed swelling of his face and neck. According to his mother, patient was blowing into a water bottle with great force repeatedly for many times. Patient was under stress at work recently for about 5 days; he became irritable, agitated, and acting strangely like looking to ceiling, making some movement by his hands, and he was blowing into a bottle forcefully many times per day. Patient was not talking much but he denies cough, dyspnea, dysphagia, chest pain, palpitation, abdominal pain, nausea, excessive belching, vomiting, bowel problems, urinary problems, and suicidal or homicidal ideas. He has no previous medical history of lung diseases or any recent trauma. He denied any recreational drug use or smoking. Home medications include enalapril 2.5 mg per os daily and simvastatin 40 mg per os daily. According to his mother, he was not taking any psychiatric medications.

Initial vital signs included temperature 98°F, pulse rate 130 beats per minute, respiratory rate 23 breaths per minute, blood pressure 104/72 mm Hg, and oxygen saturation 98% on room air.

Head and neck examination was significant for facial swelling and diffuse crepitus on palpation around the neck without orbital swelling or lymph node enlargement.

Respiratory examination revealed moderate crepitus to palpation in the upper anterior chest bilaterally extending to neck and up to distal arms bilaterally. His chest wall was not hyper-resonant to percussion. There was no stridor or rhonchi. Peripheral pulses were equal and symmetric bilaterally. The remaining examinations including cardiovascular, abdominal, and central nervous system examinations were within normal limits.

Laboratory test showed normal arterial blood gas. Complete blood count and basal metabolic profile were unremarkable. Electrocardiogram was normal except sinus tachycardia. An initial chest X-ray (CXR) revealed extensive subcutaneous emphysema in chest and neck (Fig. 1). Soft tissue neck computed tomography (CT) scan showed cervicofacial soft tissue emphysema contiguous with the pneumomediastinum. CT scan of head revealed normal brain with extensive parapharyngeal, retropharyngeal, and scalp soft tissue emphysema. CT scan of chest with intravenous contrast showed extensive bilateral soft tissue chest wall emphysema with pneumomediastinum (Figs. 2, 3, and 4). Echocardiogram was within normal limits. CT scan of abdomen and pelvis revealed minimal punctate retroperitoneal air around left kidney without evidence of acute intra-abdominal pathology (Fig. 5). According to imaging studies, esophageal or tracheal rupture was ruled out.

Patient was managed conservatively in the intensive care unit with humidified oxygen 5 l/min *via* mask to facilitate clearance of SPM. Psychiatric medications were given to control his mood and psychotic behaviors. CT scan of chest 24 h later showed minimal improvement in SPM. CXRs were repeated daily and showed gradual improvement of subcutaneous emphysema and SPM. Patient's clinical condition continued to improve and he was discharged on day 7 of hospitalization. At a follow-up visit 2 weeks later, patient's symptoms

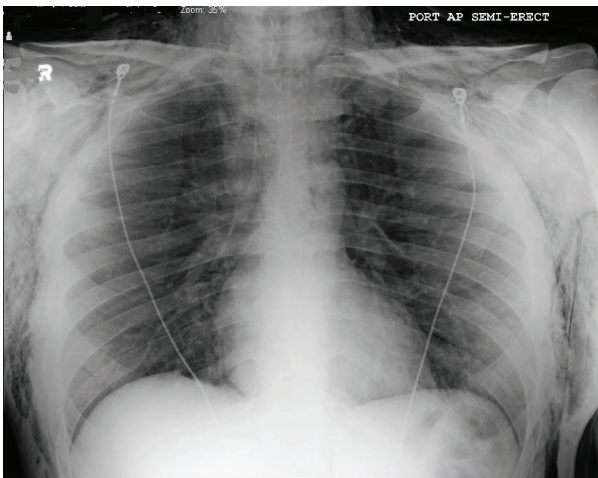


Fig. 1. Anteroposterior (AP) view of chest X-ray showing extensive subcutaneous emphysema in the chest and neck.

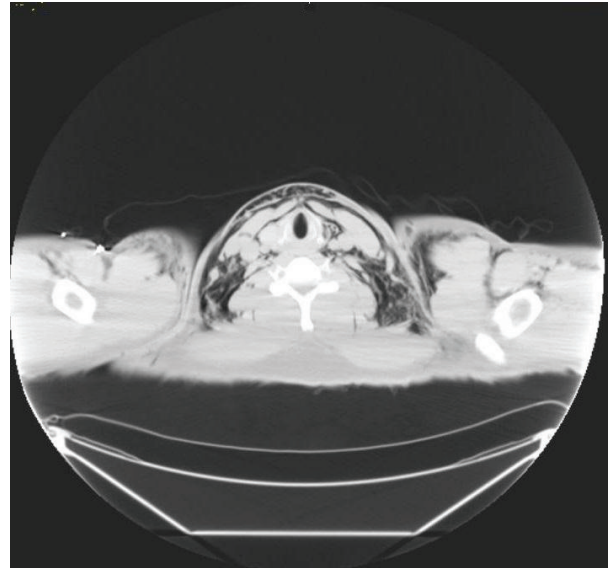


Fig. 2. Axial view of CT chest revealing cervical and facial subcutaneous emphysema.

improved and repeated CXR showed a normal chest with resolution of the previous subcutaneous emphysema and pneumomediastinum.

Discussion

SPM usually occurs from bronchial hyper-reactivity or barotraumas. Risk factors for bronchial hyper-reactivity are asthma and inhalation of illicit drugs (e.g., cocaine (4) and marijuana). Barotrauma may result from instrumentation, mechanical ventilation, or Valsalva's maneuver, which is the process of expiration through resistance that

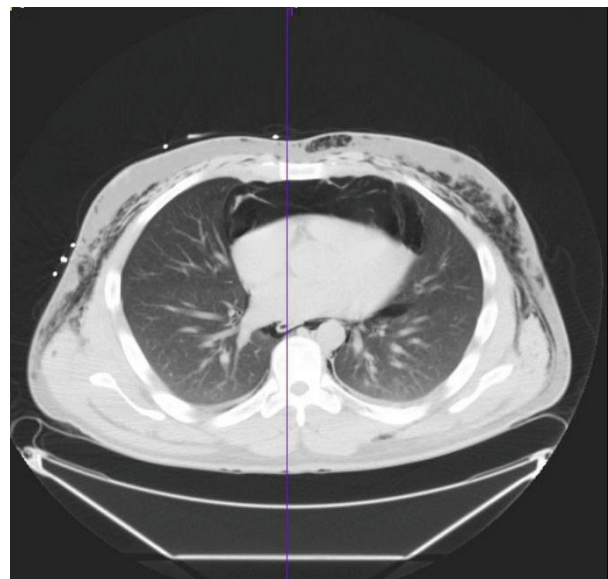


Fig. 3. Axial view of CT chest revealing extensive pneumomediastinum and subcutaneous emphysema.



Fig. 4. Coronal view of CT chest, abdomen, and pelvis revealing pneumomediastinum and subcutaneous emphysema from neck area to left flank of abdomen.

increases intra-thoracic pressure during coughing (5), straining during labor (6), or vomiting (2). Blowing into a closed space such as a bottle is also considered as a

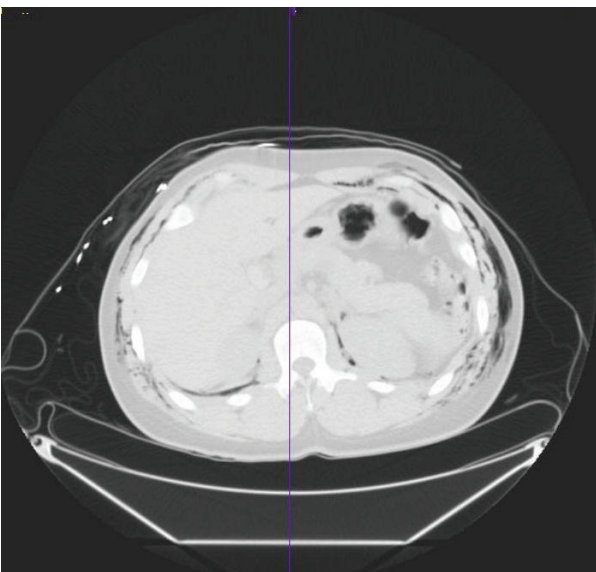


Fig. 5. Axial view of CT abdomen showing punctate retroperitoneal air around left kidney.

Valsalva's maneuver. Our patient had psychiatric disorder that led him to blow repeatedly and forcefully into the bottle for 5 days until his mother noticed swelling around his face, neck, and chest.

Secondary pneumomediastinum should be excluded first before diagnosing as SPM. The former has specific underlying pathologic event, such as trauma, esophageal rupture after vomiting, and infection by gas-forming organism (7). SPM should also be suspected in patients with dysphagia. Cho and Kim (7) reported two patients developed dysphagia after swallowing a peach seed and boned rib of pork, respectively.

Furthermore, Macia et al. published a retrospective study of 41 cases. The mean age of SPM is 21 years. Most common symptoms are chest pain (85%) and dyspnea (49%), and most common sign is subcutaneous emphysema in 71% of patients. Most of the patients respond well to medical treatment including analgesia, rest, and/or initial oxygen therapy. The average length of hospital stay was 5 days (range, 1–9 days) with only one case of early recurrence, which was improved satisfactorily. Imaging studies such as CXR can detect pneumomediastinum in all patients. CT scan, contrast-enhanced swallow, or bronchoscopy was performed in some cases (8). However, Kaneki et al. (9) reported a study of 33 cases; CXR alone was normal in 3 patients with mild SPM, and CT scan with contrast was needed to diagnose accurately. Antibiotics are not recommended in the absence of signs and symptoms of infection, and normal white blood cell count.

In order to diagnose spontaneous pneumoretroperitoneum, secondary causes such as prior history of endoscopy (10), gas-producing infections, trauma, and iatrogenic should be excluded. Witzig et al. reported a retrospective study of 15 cases of secondary pneumoretroperitoneum. Etiologies were infectious in six patients, traumatic in five, and iatrogenic in four. Surgical treatment was used in 12 patients. Conservative management was applied for the remaining three patients whose etiology was due to endoscopy (11). Gleeson and McMullin (12) reported a rare case of prednisolone-induced spontaneous pneumoretroperitoneum in a renal transplant patient.

Moreover, dental and oral procedures are known to be culprits for some cases of cervicofacial emphysema. Koukoutsis et al. (13) described a case of subcutaneous emphysema and pneumomediastinum after tonsillectomy.

It is important to note that there is anatomical connection of fascial planes because there are no barriers on the subcutaneous tissues in the body. Therefore, if the air pressure surpasses the strength of the tissues, or the time between the aggression and the imaging is too long, the air can travel to other areas hindering its primary location. The mediastinum is continuous directly with the visceral and retropharyngeal spaces (14). The reason behind using 100% oxygen is to replace nitrogen in the escaped air with oxygen; nitrogen is absorbed slowly by

capillaries, however, oxygen is absorbed rapidly (15). Although Sato et al. reported a case of secondary pneumoretroperitoneum, pneumomediastinum, and subcutaneous emphysema after colorectal endoscopy, we believe that there is no detailed case about spontaneously occurring pneumoretroperitoneum, pneumomediastinum, and cervicofacial emphysema in a psychiatric patient after repeatedly and forcefully blowing a bottle.

Spontaneously occurring air in the three different anatomical areas in the human body is a challenging medical vignette for a physician. It is crucial to find a primary source and treat properly. Psychiatric patients may have psychotic behaviors mimicking Valsalva's maneuver that increases intra-thoracic pressure and causing SPM. Optimal medications should be given to control psychotic behaviors. Family members and caregivers should be explained about this unusual behavior so that they can prevent this rare condition.

Conflict of interest and funding

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