

## CASE REPORT

# An Interesting Case of Spontaneous Pneumomediastinum with Subcutaneous Emphysema following Oral Provocative Manoeuvre

<sup>1</sup>CB Pratibha, <sup>2</sup>Deepthi Satish, <sup>3</sup>Suraj Gopal, <sup>4</sup>AM Balasubramanya

## ABSTRACT

**Aim:** To discuss a case of spontaneous pneumomediastinum with pneumothorax with subcutaneous emphysema presenting with stridor due to laryngeal edema with relevant review of literature.

**Background:** Spontaneous pneumomediastinum is a rare condition that has been described in healthy individuals following Valsalva manoeuvre, excessive and prolonged cough or emesis. Laryngeal involvement in these cases has not been reported so far.

**Case description:** We present an interesting case of spontaneous pneumomediastinum with pneumothorax with pneumopericardium and cervicofacial emphysema with suspected ingestion of foreign body. In view of stridor due to laryngeal edema tracheostomy was done. No obvious cause for the air leak was found on further investigations. The foreign body sensation could have led to oral provocative manoeuvres by the patient causing increased intra-alveolar pressures and air leak.

**Conclusion:** Spontaneous pneumomediastinum with cervicofacial emphysema with pneumopericardium with laryngeal involvement is very rare. Tracheostomy is essential in case of airway compromise. A thorough evaluation for the underlying condition is essential to prevent further air leak.

**Clinical significance:** In a case of spontaneous pneumomediastinum with airway compromise due to involvement of the larynx, tracheostomy is essential to secure the airway and could also help in resolution of emphysema.

**Keywords:** Emphysema, Laryngeal edema, Spontaneous pneumomediastinum.

**How to cite this article:** Pratibha CB, Satish D, Gopal S, Balasubramanya AM. An Interesting Case of Spontaneous Pneumomediastinum with Subcutaneous Emphysema following Oral Provocative Manoeuvre. *Int J Otorhinolaryngol Clin* 2015;7(2):93-96.

**Source of support:** Nil

**Conflict of interest:** None

## BACKGROUND

Pneumomediastinum, or mediastinal emphysema, was first described by Laennec in 1819 as the presence of air in the mediastinum due to traumatic injury. Spontaneous pneumomediastinum is defined as free air within the mediastinum, not associated with trauma, first described by Hamman in 1939. Secondary pneumomediastinum occurs following blunt thoracic trauma, endobronchial or esophageal procedures, head and neck surgery. Spontaneous pneumomediastinum is a rare condition, often self-limiting, can ascend along the tissue spaces to cause cervicofacial emphysema.<sup>1</sup> Evaluation of underlying causes is required to prevent further air leak.<sup>2</sup>

We present an interesting case of spontaneous pneumomediastinum with pneumothorax with subcutaneous emphysema presenting with stridor due to laryngeal edema with relevant review of literature.

## CASE DESCRIPTION

A 56-year-old male presented to the Emergency Department of St John's Medical College Hospital, Bengaluru a tertiary care centre in South India on a Saturday night with sudden onset throat pain, progressive swelling of the neck and face since morning. Subsequently over next few hours he had developed change of voice with progressive breathing difficulty. The patient reported having a chicken meal the previous night. Following this he had a foreign body sensation in the throat. He attempted to remove the foreign body by putting his fingers into his throat and subsequently had retching. However, he did not have cough or vomiting. Patient had diabetes and hypertension and was on oral medications. There was no other significant past history with no history of surgical intervention or external trauma to the neck.

On examination, patient had stridor and a breathy voice. Heart rate was 68 per minute and tachypnea was noted with saturation of 86% on room air. Neck examination revealed a diffuse swelling with crepitus extending from the eyelids down to the neck up to the nipples suggestive of extensive subcutaneous emphysema. Auscultation revealed normal breath sounds and heart sounds. Arterial blood gases showed hypoxia with PO<sub>2</sub> 55.9 mm Hg and PCO<sub>2</sub> 35.9 mm Hg

<sup>1</sup>Assistant Professor, <sup>2</sup>Senior Resident, <sup>3</sup>Intern, <sup>4</sup>Professor

<sup>1-4</sup>Department of ENT and Head and Neck Surgery, St John's Medical College Hospital, Bengaluru, Karnataka, India

**Corresponding Author:** CB Pratibha, Assistant Professor Department of ENT and Head and Neck Surgery, St John's Medical College Hospital, Bengaluru, Karnataka, India, Phone: 08022065735, e-mail: cb.pratibha@gmail.com

and respiratory acidosis with pH of 7.37 and bicarbonate of 20.4 mmol/L. Lateral neck radiograph showed subcutaneous emphysema and no radiopaque shadow seen. Chest X-ray showed double lining over the cardiac silhouette (Fig. 1). Electrocardiography (ECG) showed a normal sinus rhythm. Contrast enhanced computed tomography (CT) scan was done which showed extensive air in bilateral subcutaneous and muscular planes extending from face to chest. Pneumomediastinum and pneumopericardium with narrowing of the laryngeal inlet was also noted (Figs 2 to 4). There was a small pneumothorax noted in the anterior lung fields on the left side. No foreign body was seen.

In view of stridor and worsening respiratory distress, emergency tracheostomy was done and airway secured. Intraoperatively on laryngoscopy, laryngeal edema was noted. Postoperatively he was maintaining saturation on room air. Cardiothoracic surgeons opined conservative management for pneumopericardium and pneumomediastinum. As foreign body of the upper aerodigestive tract was suspected, barium swallow with thin

barium was done postoperatively, which did not reveal any intraluminal pathology.

On postoperative day 3, resolution of emphysema was noted (Fig. 5). Office laryngoscopy showed edema of the posterior larynx with normal mobility of the vocal cords. In view of aspiration on oral feeds, nasogastric tube

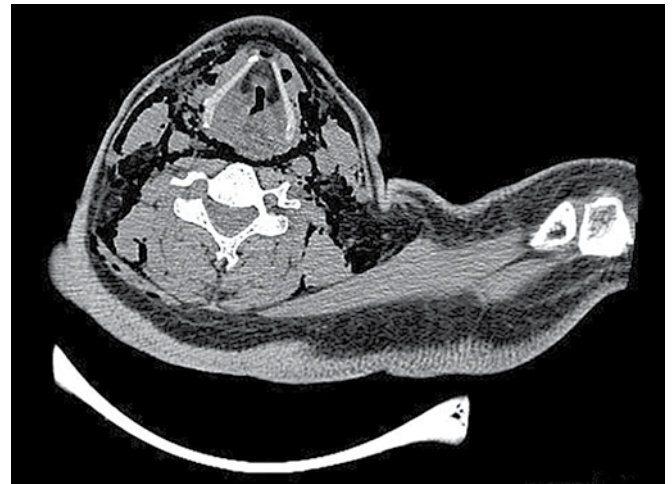


Fig. 3: Contrast CT scan axial section showing laryngeal edema



Fig. 1: Chest X-ray showing air shadow surrounding the cardiac silhouette

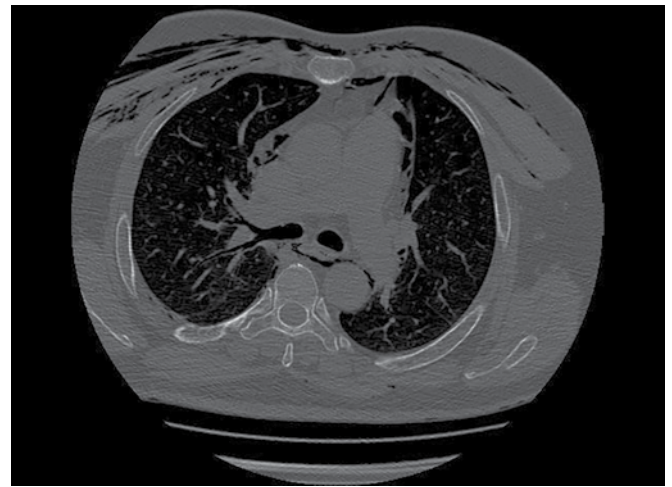


Fig. 4: Contrast CT scan axial section showing pneumomediastinum with air around the arteries



Fig. 2: Contrast CT scan coronal section showing extensive cervicofacial emphysema



Fig. 5: Postoperative image of the patient

was inserted and swallowing manoeuvres was initiated. Gradually patient improved and was able to tolerate oral feeds well. On postoperative day 18, tracheostomy tube was downsized and later decannulated.

Patient was discharged after 3 weeks of admission and on a regular follow-up thereafter. Examination revealed complete resolution of emphysema and videolaryngoscopy showed normal vocal cords.

## DISCUSSION

Pneumomediastinum may be due to intrathoracic and extrathoracic causes. Blunt trauma to the chest could lead to rupture of the alveoli or a tear in the bronchus or trachea leading to escape of air into the mediastinum. It could also occur due to vomiting, weight lifting or straining against a closed glottis. Extrathoracic causes though rare, can be due to subcutaneous emphysema involving head and neck or dissection of air from the retroperitoneum.<sup>3</sup>

Spontaneous pneumomediastinum occurs mostly in healthy young men or parturient women. In healthy individuals, Valsalva manoeuvre, excessive and prolonged cough, emesis could lead to increased intra-alveolar pressure. Preexisting weakness of either the alveolar or bronchial wall could cause rupture of the marginal pulmonary alveoli at a weakened point allowing escape of air into the tissue. Air escapes via peribronchial or perivascular channels to the mediastinum into loose alveolar tissue, which could ascend into the neck and subcutaneous plane, causing cervicofacial subcutaneous emphysema.<sup>2,4</sup> Reports of pneumomediastinum following cervicofacial emphysema is very rare mostly following dental surgical procedures, head and neck surgery, or orofacial trauma.<sup>4</sup> In the patient described here, the foreign body sensation leading to oral provocative manoeuvres would have caused retching which in turn led to increased intra-alveolar pressure causing pneumomediastinum and pneumothorax with subcutaneous emphysema.

The presence of pneumomediastinum is in itself of little significance, but the underlying condition causing the air leak (particularly bronchial, esophageal or pharyngeal perforation) may be of great significance.<sup>3</sup> In our case, the patient presented with stridor with extensive subcutaneous emphysema of the face, neck and upper chest wall and imaging showed pneumomediastinum with laryngeal edema. In view of the airway compromise tracheostomy was done to secure the airway.

The escape of air into subcutaneous tissue is known as subcutaneous emphysema. It presents as a diffuse swelling with crepitus commonly involving the neck and chest wall though rarely scalp, palm of the hands,

soles of the feet may be involved. Secondary spontaneous subcutaneous emphysema is described when leakage of air occurs as a result of a recognizable coexisting structural abnormality in the lungs. It is termed traumatic if it occurs following either blunt or penetrating external injury. Spontaneous emphysema is usually self-limiting, however, the underlying cause should be evaluated and treated to prevent any further air leak.<sup>2</sup>

Pneumopericardium is defined as a collection of air or gas in the pericardial cavity. Symptoms of pneumopericardium include chest pain, dyspnea, cyanosis, hypotension, bradycardia or tachycardia, and pulsus paradoxus. On auscultation, muffled heart sounds have been described and a mill wheel murmur (bruit de moulin) is characteristic. The pathogenesis of pneumopericardium is similar to pneumomediastinum with spread of air to the pericardial reflection.<sup>5</sup>

Gas surrounding the heart may be confused with pneumomediastinum. Therefore, studies other than posteroanterior roentgenograph are helpful to distinguish pneumopericardium from pneumomediastinum: A left-side-down decubitus radiograph will show a rapid shift of air in the pericardial sac, while air in the mediastinum will not move in the short interval between films.<sup>5</sup>

Patients with pneumomediastinum present with thoracic pain (usually retrosternal and pleuritic in nature), subcutaneous emphysema and dyspnea. Other symptoms include cough, fever, dysphonia, odynophagia, and dysphagia. The Hamman sign is pathognomonic and characterized by systolic crackles in the left sternal border, best heard in left lateral decubitus position.<sup>1</sup>

Radiologic imaging is essential in the diagnosis of pneumomediastinum. Chest radiography would show a double line outlining the mediastinum. If this is not apparent on radiography, chest CT can be done to confirm diagnosis.<sup>1</sup>

Once diagnosis is confirmed, patient should be admitted for monitoring and treatment, avoidance of the trigger factor, oxygen and bed rest. Complications are rare, though mediastinitis can be a serious complication with high morbidity and mortality. Role of antibiotics is mainly in cases of suspected mediastinitis. Intercostal drainage is recommended in case of pneumothorax and/or hypertensive pneumomediastinum.<sup>1</sup>

## CONCLUSION

Spontaneous pneumomediastinum with cervicofacial emphysema with pneumopericardium is a rare entity. Laryngeal involvement in these cases has not been reported so far. In case of airway compromise due to laryngeal involvement tracheostomy is essential to secure the airway and could also help in resolution of

emphysema. A thorough evaluation for the underlying condition is essential to prevent further air leak.

### CLINICAL SIGNIFICANCE

In a case of spontaneous pneumomediastinum with airway compromise due to involvement of the larynx, tracheostomy is essential to secure the airway and could also help in resolution of emphysema.

### REFERENCES

1. Meireles J, Neves S, Castro A, França M. Spontaneous pneumomediastinum revisited. *Respir Med CME* 2011;4(4) 181-183.
2. Pandey D, Jaret P, Sharma R, Sharma A, Thakur S. Subcutaneous emphysema secondary to pulmonary cavity in absence of pneumothorax or pneumomediastinum. *Respir Med* 2007 Feb;101(2):363-365.
3. Parkar N, Javidan-Nejad C, Bhalla S, et al. The Mediastinum including the pericardium. In: Adam A, Dixon AK, Gillard JH, et al, editors. *Grainger's and Allison's diagnostic radiology*. 6th ed. Churchill Livingstone, Elsevier; 2015. p. 209-246.
4. Lopez-Pelaez MF, Roldan J, Mateo S. Cervical emphysema, pneumomediastinum, and pneumothorax following self-induced oral injury. *Chest* 2001;120(1):306-309.
5. Westermann GW, Suwelack B. Spontaneous pneumopericardium due to exertion. *South Med J* 2003;96(1):1-3.

