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Case report - Thoracic non-oncologic

# Negative-pressure pulmonary edema presented with concomitant spontaneous pneumomediastinum: Moore meets Macklin

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**Abstract**

Negative-pressure pulmonary edema is an unusual complication mainly associated with general anesthesia. It is caused by excessive negative intrathoracic pressure following a deep inspiration against an acute airway obstruction. The resultant decreased intrathoracic pressure amplifies venous return to the right heart and increases pulmonary capillary wedge pressure that can be further amplified by massive sympathetic discharge due to hypoxia. The combination of increased venous return and pulmonary capillary wedge pressure favours the shift of fluid into the pulmonary interstitium with resultant pulmonary edema. Conversely, spontaneous pneumomediastinum (SP) results from alveolar rupture following an excessive positive intrathoracic pressure. The air leaks out of the alveoli and along the perivascular space toward the mediastinum. We experienced a case of negative pulmonary edema which presented in association with SP. Pneumomediastinum is probably caused by an excessive positive intrathoracic pressure for a subsequent expiration against a closed airway. In the present case, both complications resolved with conservative management.


*Keywords:* Negative-pressure pulmonary edema; Pneumomediastinum; General anesthesia

## 1. Introduction

Negative-pressure pulmonary edema (NPPE) is an unusual complication mainly associated with general anesthesia. It is caused by excessive negative intrathoracic pressure following a deep inspiration against an acute airway obstruction [1]. Conversely, spontaneous pneumomediastinum (SP) results from alveolar rupture subsequent to an excessive positive intrathoracic pressure [2]. We report an exceptional case where NPPE with concomitant SP developed after general anesthesia.

## 2. Clinical summary

A 19-year-old male was admitted for elective breast reduction for gynecomastasia. His physical examination and laboratory studies were unremarkable. Surgery was done under general anesthesia and trachea intubated with cuffed endotracheal tube size 7.5 mm without any difficulty. Bilateral breast reduction was attempted with excision of the gland combined with subcutaneous liposuction. No complications were encountered and the procedure lasted for approximately one and half hours. Just following extubation we observed the common signs of laryngospasm, i.e. audible respiration accentuated by an impaired ability to inhale. The patient then began coughing forcefully with a large amount of frothy pink sputum and oxygen desaturation. He was placed on a non-rebreather mask and oxygen saturation climbed rapidly to 100%; he was then transferred to the intensive care unit (ICU) for monitoring.

Bilateral pulmonary infiltrates were seen on the chest X-ray (CXR) (Fig. 1), while the chest computed tomography (CT)-scan surprisingly showed the presence of coexisting pneumomediastinum that had been missed on CXR (Fig. 2). Thus, an endotracheal perforation was suspected. Bronchoscopy was attempted, but no endotracheal lesions were found. Gastrografin swallow excluded any esophageal perforation. At this stage our working diagnosis included fluid overload, acute myocardial infarction, and fat embolism. Fluid overload was excluded as he was given only 500 ml of Ringers lactate solution, and acute myocardial infarction was rejected on the basis of echocardiogram (ECG). Laboratory investigations revealed no abnormalities, fat embolism is characterized by laboratory abnormalities including a rise in the serum lipase level, anemia, thrombocytopenia, and hypocalcemia [3]. Yet, D-dimer was not elevated.

Target questioning of the patient revealed a previous episode of laryngospasm which resolved spontaneously. On the basis of such clinical considerations, the clinical suspicion of NPPE was high. Thus, an intravenous injection of diuretics was given. A CXR, repeated after 24 hours, confirmed radiological improvement with clearencing of pul-

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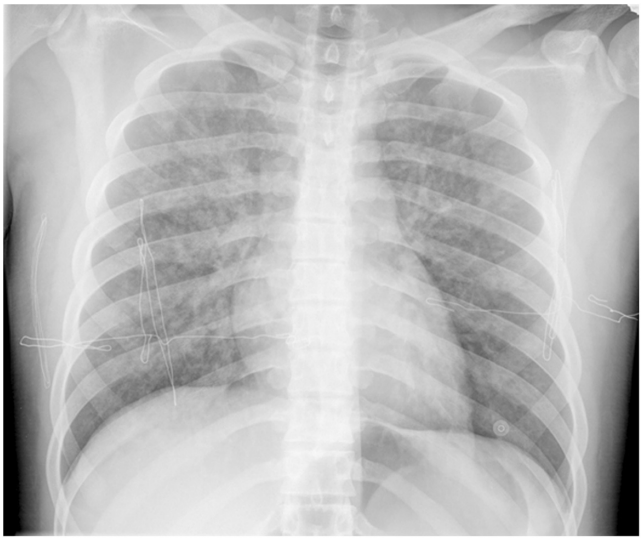

Fig. 1. Chest radiography shows bilateral diffuse pulmonary infiltrates consistent with pulmonary edema. The cardiac size is within normal.

monary edema. Chest CT-scan showed no progression of pneumomediastinum. Thus, conservative management led to gradual improvement of his condition. The patient was discharged on postoperative day 7. The pneumomediastinum completely resolved and follow-up showed no residual effects.

## 3. Discussion

NPPE, first hypotized by Moore in 1927, is an uncommon complication following general anesthesia that can occur in otherwise healthy patients. The pathogenesis is multifactorial. The main mechanism is a large inspiratory force generated against an obstructed upper airway. The resultant decreased intrathoracic pressure amplifies venous return to the right heart and increases pulmonary capillary wedge pressure that can be further amplified by massive sympathetic discharge due to hypoxia. The combination of increased venous return and pulmonary capillary wedge pressure favors the shift of fluid into pulmonary interstitium with resultant pulmonary edema [1]. The typical associated signs of NPPE are: agitation, tachypnea, frothy pink secretions, and progressively decreasing oxygen saturation [1, 4], all of which we observed in our patient. If recognized early, NPPE is a self-limiting condition; the mainstay of treatment is usually supportive with administration of diuretics or re-intubation, if necessary. Conversely, the mortality and morbidity has been reported as high as 40% in the case of delayed diagnosis [4]. Despite this during the last 15 years increasing numbers of cases of NPPE have been reported, to our knowledge the present case is the first in which NPPE presented in association with SP.

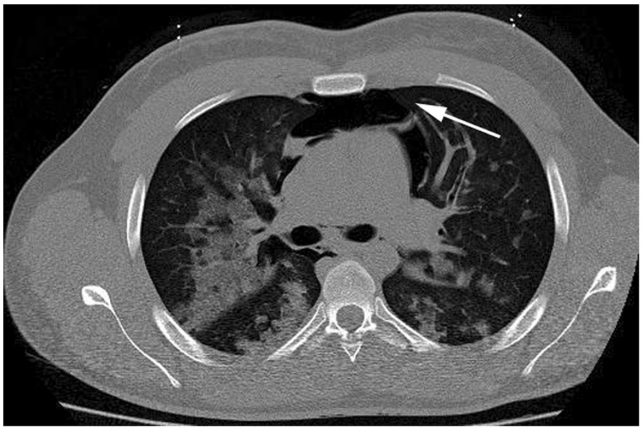

Fig. 2. Chest CT-scan shows bilateral diffuse pulmonary infiltrates and the presence of coexisting pneumomediastinum (white arrow). CT, computed tomography.

Macklin first postulated the mechanism for SP in 1944 [5]. Alveolar rupture occurs if a large enough pressure gradient is generated against a closed glottis [6]. The gas takes the line of least resistance along the fascia surrounding the bronchovascular tree to reach the mediastinum [2].

Pneumomediastinum may present in the early postoperative period from a number of possible causes reviewed in our case.

Trauma of the esophagus or trachea following difficult intubation is rejected because intubation is atraumatical. Yet, bronchoscopy and gastrografin swallow show any perforation of trachea and oesophagus, respectively.

The overinflation of the right lung due to accidental positioning of the endotracheal tube in the right main bronchus is reported by Bembridge and Bembridge [7] as a cause of pneumomediastinum. However, it was excluded because following intubation the breath sounds are equal on auscultation.

Other authors report that air may also reach the mediastinum by tracking down the fascial planes of the neck [8] or through the diaphragmatic hiatuses [9], but in our case surgery was far from these areas. Yet, the negative pressure of the anterior chest wall during liposuction may have a partial effect on the occurrence of pneumomediastinum because the mediastinum and subcutaneous layer are connected to each other. According to this hypothesis, we would expect the simultaneous presence of subcutaneous emphysema and pneumomediastinum, this was not observed in our patient.

Other causes of SP including intoxication with cocaine, marijuana, paraquat, and chloridine gas [6], were not present in our case.

Thus, we hypotize that pneumomediastinum may be the result of the same pathophysiological mechanism of NPPE (acute closed airway) but occurred in a different time of respiratory cycle such as:

First, a vigorous inspiration against an acute upper airway obstruction probably caused by laryngospasm leads to a high-level of negative intrathoracic pressure with resultant pulmonary edema according to above-mentioned mechanism. As confirmation of that, we observed a normal level of albumin (4.5 g/dl).

Second, deep inspiration may be followed by strong expiration against a closed airway. The Valsalva maneuver and coughing generates an excessive positive-pressure ventilation which increases intralveolar pressure and results in alveolar rupture. Theoretically, the loss of integrity of the alveolar capillary membrane and the coughing due to pulmonary edema may also contribute to a weakening of the alveolar wall. Following alveolar rupture, air can track

along the perivascular tissue planes to the mediastinum (the Macklin phenomenom).

Finally, thoracic surgeons and anesthesiologists need to consider NPPE and SP as possible complications after extubation. A high index of suspicion for the diagnosis and prompt treatment is required to avoid morbidity and mortality.

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