

## CASE REPORT

# Pneumomediastinum secondary to non-traumatic anterior tracheal wall defect in myasthenia gravis

A. Goyal<sup>1</sup>, M. Krishnakumar<sup>1</sup>, V. Murumkar<sup>2</sup>, G. Krishna<sup>1</sup>

Department of <sup>1</sup>Neuroanaesthesia and Neurocritical Care, and <sup>2</sup>Neuroimaging and Interventional Radiology, National Institute of Mental Health and Neuro Sciences, Bengaluru, Karnataka/India

## Correspondence

A. Goyal - amitgoyal26@gmail.com

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## Abstract

Spontaneous pneumomediastinum is a rare occurrence in patients on mechanical ventilation. It usually occurs as a complication of endotracheal intubation, causing a defect in the membranous posterior wall of the trachea. Rarely, a defect in the anterolateral tracheal wall results in pneumomediastinum. The most likely pathophysiology resulting in weakening of the anterior fibrocartilaginous trachea is reduction in the vascular supply causing ischaemia. We report here a case of pneumomediastinum in a patient with myasthenia gravis on mechanical ventilation due to an anterior tracheal wall defect, which possibly resulted from ischaemia of the tracheal wall secondary to steroid use or previous prolonged endotracheal intubation.

## Background

Pneumomediastinum is a clinical condition involving the presence of air in the mediastinum. It usually develops secondary to trauma, surgery, malignancy or infection which cause an anatomical defect in the lung, airway or oesophagus. Rarely, it occurs as a complication of endotracheal intubation, most commonly due to a defect in the membranous posterior wall of the trachea.<sup>[1]</sup> We report a case of spontaneous pneumomediastinum due to an anterior tracheal wall defect in a patient with myasthenia gravis on mechanical ventilation.

## Case report

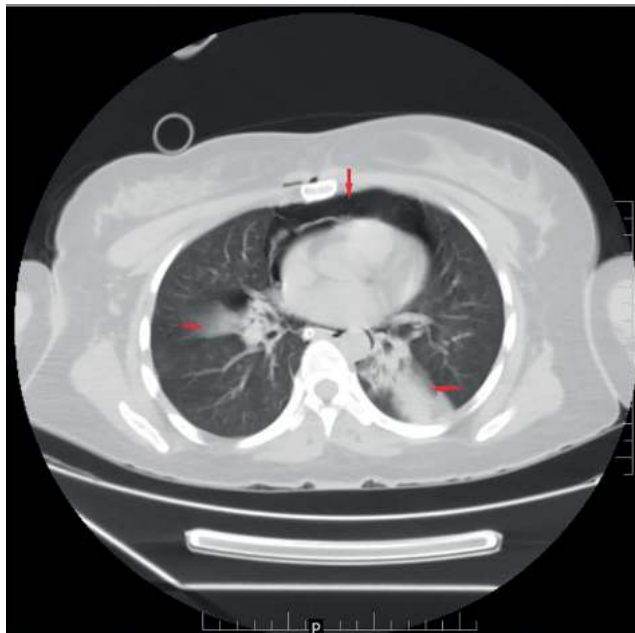
A 26-year-old non-asthmatic, non-smoking female, who was known to have myasthenia gravis, came to our hospital with a chief complaint of difficulty in breathing in the last two days. There was no history of limb weakness, extra-ocular symptoms, bladder, bowel or sensory involvement or any pre-existing lung disease. One year previously, she was evaluated for complaints of nasal intonation of her voice and generalised weakness with diurnal variation, which progressed to difficulty in chewing

food and swallowing. She was diagnosed with myasthenia gravis. Later, she developed respiratory distress for which she was intubated and was on mechanical ventilation for a period of one month. She was treated with oral prednisolone for three months and pyridostigmine with which she was clinically stable; a thymectomy was scheduled at a later date.

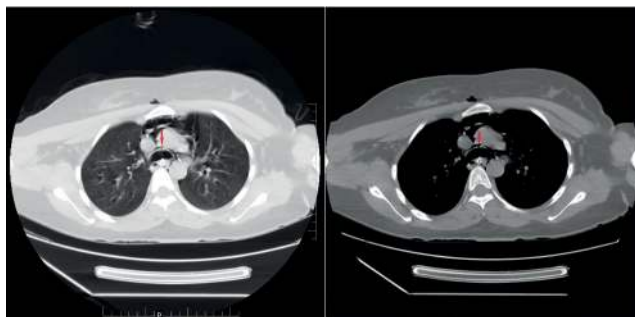
On examination, her vital signs were within normal limits. Routine blood investigations, thyroid profile, chest X-ray and electrocardiogram were unremarkable and CT thorax showed thymic hyperplasia with maintained morphology and a normal trachea.

The next day, she developed tachypnoea with a single breath count of 8, suggestive of myasthenia crisis. She was transferred to the ICU, the airway was secured with a 7.5 mm cuffed endotracheal tube by direct laryngoscopy, in a single attempt without stylet or bougie and she was put on pressure controlled mechanical ventilation with PEEP of 5 cmH<sub>2</sub>O, pressure support of 12 cmH<sub>2</sub>O, respiratory rate of 12 and FiO<sub>2</sub> of 40%. She was started on neostigmine, methylprednisolone, antibiotics and large volume plasmapheresis. On day 6 of mechanical ventilation, she developed a cough along with a gradual fall in SpO<sub>2</sub> (up to 86%) and tachycardia, in the absence of hypotension. On auscultation, air entry was equal on both sides. Her chest X-ray showed haziness in the left lower lung field and arterial blood gases (ABG) showed pH 7.41, pO<sub>2</sub> 35.3 mmHg, pCO<sub>2</sub> 49.6 mmHg with a calculated shunt of 110%. Her lung ultrasonography showed bilateral multiple B-lines and two-dimensional echo could not be performed because of a poor window. Later, her SpO<sub>2</sub> improved to 92% following an increase in FiO<sub>2</sub> 0.7 and PEEP 10 cmH<sub>2</sub>O after intravenous sedation with opioids. Repeat ABG showed pH 7.48, pO<sub>2</sub> 67.6 mmHg, and pCO<sub>2</sub> 43.2 mmHg. On day 7, she again had a fall in SpO<sub>2</sub> (up to 82%) and tachycardia without hypotension. She also complained of palpitations and became restless. The electrocardiogram showed no significant changes. Her blood

reports and cultures did not show any evidence of infection. On examination, air entry was equal on both sides and she had subcutaneous emphysema in the region of neck and face. She was immediately transferred for computed tomography (CT) of the thorax, which showed consolidation in both lungs, pneumomediastinum (*figure 1*), a defect in the anterior tracheal wall just above the level of carina (*figure 2*), and the position of the tip of the tube was well above the defect. Her saturation and symptoms improved with reduction in PEEP 0 cmH<sub>2</sub>O and pressure support.



**Figure 1.** CT thorax showing the pneumomediastinum along with consolidation in the bilateral lung fields (arrows)



**Figure 2.** CT thorax (lung window and soft tissue window) showing the pneumomediastinum along with a defect (arrow) in the anterior tracheal wall just above carina

As pneumomediastinum has the potential to rapidly increase in size and cause lethal haemodynamic and respiratory effects, and due to lack of multispecialty care in our institution, we decided to transfer the patient to a multispecialty health centre after discussion with the patient's family. On follow-up over

the phone, we were informed that she had been managed conservatively and was extubated after one week.

## Discussion

Non-traumatic pneumomediastinum is a rare presentation in critically ill patients on mechanical ventilation. Risk factors for tracheal rupture following intubation include female sex (owing to a weak pars membranacea), presence of anatomical abnormality, difficult intubation with multiple attempts, use of a bougie or stylet protruding beyond the tip of tube, high cuff pressure, incorrect positioning of the tube tip, severe cough and prolonged intubation causing tracheal necrosis. The time interval from intubation to diagnosis may vary from 1 hour to 5 days.<sup>[2,3]</sup> Patients usually complain of difficulty in breathing, palpitations and chest pain, along with the presence of swelling (subcutaneous emphysema) in the region of chest, neck or face. Diagnosis of pneumomediastinum is based on high clinical suspicion and radiological features. As chest X-ray may be inconclusive in many cases, diagnosis is confirmed by CT scan.<sup>[4,5]</sup> Bronchoscopy or oesophagoscopy may be used to ascertain the underlying pathology and to exclude oesophageal perforation. Although air in the mediastinum is absorbed by the tissues, with most benign cases requiring a conservative approach along with symptomatic treatment, there is no consensus regarding the choice between conservative and surgical treatment.

Most of the literature reports defects in the membranous posterior tracheal wall as causative factor for non-traumatic pneumomediastinum, whereas defects in the anterolateral tracheal wall are rare. Various factors known to cause non-traumatic rupture of the anterolateral tracheal wall include radiotherapy, ulcerative tracheobronchitis due to aspergillosis and thoracic surgery.<sup>[6]</sup> The most likely pathophysiology resulting in weakening of the anterior fibrocartilaginous trachea is reduction in the vascular supply causing ischaemia.

Our patient initially showed a fall in oxygen saturation which was thought to be secondary to developing pneumonia in the left lung, as seen on the chest X-ray, and the PEEP was increased on the ventilator. This high PEEP led to an increase in the air leakage into the mediastinum causing worsening of the symptoms. We also failed to suspect pneumomediastinum when we could not find the window for 2D echo. It was only the development of subcutaneous emphysema which raised the suspicion towards the presence of an air leak from the airway. CT of the chest is the gold standard for diagnosis of pneumomediastinum; in addition a defect in the anterior tracheal wall was seen in this case.

Since we had to transfer the patient to a multispecialty centre, we could not confirm the pathology in this case. Steroids are known to reduce vascularity and angiogenesis in the tracheobronchial tree<sup>[7-9]</sup> and can induce gross cartilage damage within eight to twelve weeks of therapy.<sup>[10]</sup> In the background of the disease-induced cartilage loss, steroid use or previous

mechanical ventilation and intubation could have a plausible vascular or mechanical effect causing weakening of anterior tracheal wall in our patient, which gave way during positive pressure ventilation.

### Conclusion

Respiratory distress and hypoxaemia is common in patients with myasthenia gravis secondary to respiratory failure, atelectasis and aspiration, but the possibility of other causes should be ruled out if the patient does not show any improvement with initial therapeutic measures. A triggering event such as coughing or vomiting causing a sudden increase in intrathoracic pressure is frequently seen in spontaneous pneumomediastinum. Patients with spontaneous pneumomediastinum should be completely evaluated with (1) radiological imaging preferably CT thorax, (2) detailed history of traumatic endotracheal intubation, previous thoracic surgery, radiotherapy, prolonged steroid usage and mechanical ventilation, (3) bronchoscopy findings, and (4) laboratory tests for bacterial, mycobacterium, fungal infection and any malignant pathology.

### Disclosures

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Written consent for publication of this case has been obtained from the patient.

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