Pneumothorax in dysferlin myopathy associated with mechanical ventilation

Martín Hunter, Patricia Karina Aruj

ABSTRACT

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CASE REPORT

A 38-year-old female with dysferlin myopathy (and domiciliary use of nocturnal non-invasive positive pressure ventilation and mechanical insufflation-exsufflation respiratory aid) presented with progressive right upper quadrant abdominal discomfort. The vital signs at presentation were: temperature 36.6°C, heart rate 100 beats per minute, blood pressure 140/80 mmHg, respiratory rate 30 breaths per minute, oxygen saturation 96% while breathing ambient air. On examination, breath sounds were absent on the right side. An anteroposterior chest radiograph showed a large collection of gas in the right hemithorax with inversion of the right hemidiaphragm (Figure 1A). Computed tomography (CT) scan of the chest confirmed the diagnosis of large right-sided pneumothorax, with cardiomeastinal shift to the left (Figure 1B). Despite the size of the pneumothorax, the patient was hemodynamically stable and, surprisingly, she did not develop hypoxemia. Pneumothorax was managed with tube thoracostomy. However, since lung expansion was not successfully achieved, a Heimlich valve was placed. After discharge, a new CT scan showed resolution of the pneumothorax with adequate right lung expansion. The patient is still using mechanical ventilation through a tracheostomy at home without further complications.

DISCUSSION

Mechanical insufflation-exsufflation (MI-E) is a respiratory aid used by patients with weak respiratory muscles to improve cough effectiveness. Machine pressures can range from +60 cm H₂O of insufflation to -60 cm H₂O of exsufflation [1]. A few limb-girdle muscular dystrophies are associated with weakness of respiratory or oropharyngeal muscles and an increased risk of respiratory failure with disease progression [2]. This patient had a severe restrictive pattern in pulmonary function tests and diminished cough peak flows from inspiratory and expiratory muscle weakness. The cause of pneumothorax was probably due to barotrauma related to high insufflation pressures.

Figure 1A: Anteroposterior chest radiograph showing a large collection of gas in the right hemithorax with inversion of the right hemidiaphragm.

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Figure 1B: Computed tomography scan of the chest confirms the diagnosis of large right-sided pneumothorax, with cardiomeastinal shift to the left.

CONCLUSION

Although the use of mechanical insufflation-exsufflation (MI-E) alone may or may not increase the risk of barotrauma, clinicians should be aware that the use of MI-E may add to other risk factors to increase the risk of pneumothorax. Although rare, pneumothorax associated with mechanical ventilation has been reported previously in this patient population.

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Author Contributions

Martín Hunter – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Patricia Karina Aruj – Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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