An infrequent but important complication of primary spontaneous pneumothorax

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Introduction

In primary spontaneous pneumothorax, before revision of the American College of Chest Physicians (ACCP) management guideline in 2001, chest drains with or without connection to suction under water-seal were commonly performed as the initial management. We here report a case of an infrequent but important complication secondary to chest drain insertion for primary spontaneous pneumothorax.

Case presentation

TC, 17 years old gentleman, was admitted to United Christian Hospital because of acute onset of pleuritic chest pain for 4 days and shortness of breath for 3 days. All along, he enjoyed good past health except allergic rhinitis. There was no preceding chest wall injury, fever or symptoms of upper respiratory tract infection. Physical examination showed no Marfanoid features. Upon chest examination there was no chest deformity. Air entry was reduced over left chest on auscultation with hyper-resonance on percussion. Chest radiograph (Figure 1a) showed complete left pneumothorax with deviation of trachea to the right. Chest drain was inserted without connection to suction under water seal. \( \text{SpO}_2 \) immediate after chest drain insertion was 92% with 2 litres per minute nasal oxygen. Respiratory rate was 18/min. Chest X-ray taken immediately after chest drain insertion (Figure 1b) showed near complete re-expansion of left lung with just a small rim of residual pneumothorax. Mild hazziness over left middle and lower zones were noted. At 4 hour after chest drain insertion, patient complained of increasing chest pain and increasing cough with sputum. Oxygen requirement was increased from 2 litres per minute to 4 litres per minute and then to 50% via rebreathing mask, with \( \text{SpO}_2 \) of 89-90%. An increase in respiratory rate was noted. Blood pressure at that time remained normal with tachycardia of 129/min.

Another chest X-ray was taken in view of his symptoms (Figure 1c) which showed an increase in lung field infiltrates over left middle zone. Diagnosis of re-expansion pulmonary oedema (REPE) was made. Patient was then managed with 60% oxygen via rebreathing mask. His condition gradually improved with resolution of clinical symptoms and repeated chest X-ray showed decrease in haziness. No fluid resuscitation was required. However, repeated chest X-ray showed persistent air leak at the lung apex. Cardiothoracic surgeon was consulted and video-assisted thoracoscopic surgery (VATS) was performed 3 days after admission. Presence of apical lung bleb was noted intra-operatively. Resection of lung bleb and chemical pleurodesis were performed. He was finally discharged after 17 days after hospitalisation. He was offered follow-up by surgeons. He did not have recurrence of pneumothorax requiring further hospital admission so far.

Discussion

Re-expansion pulmonary oedema (REPE) occurring after drainage of pneumothorax is a clinical entity first
described in 1958 by Carlson et al. Since then, there had been increasing numbers of case reports in both adult and paediatric population. Apart from pneumothorax, other conditions necessitating expansion of previously collapsed lung by insertion of chest drain, e.g. pleural effusion, haemothorax, are recognised conditions that might cause REPE. Incidence of REPE after drainage of pneumothorax varies from 1% to 14%, with fewer reports in paediatric population. However, it carries a high mortality up to 20% if unrecognised or untreated. In most cases, supportive treatment is adequate as gradual resolution is the usual course. Clinically, it is important to differentiate this condition from acute cardiogenic pulmonary oedema as they share the same presentation but the respective management is in completely different directions.

There had been different speculations concerning the pathogenesis of REPE. In the editorial by Thiagarajan 2007, REPE was classified under the group of negative pressure pulmonary oedema (NPPE) which accurately described and proposed the mechanism of this condition. As the lung collapses for a long period of time, there would be decreased amount of surfactant inside the alveolar units. Both the lymphatic flow and the microcirculation in the involved alveolar unit are reduced. Influx of neutrophils to the hypoxic lung segments with subsequent production of free radicals and inflammatory mediators such as IL-8 and MCP-1 (monocyte chemotactic protein) would increase the capillary permeability by damaging the endothelial lining. Hydrostatic pressure in the capillary wall would increase once normal perfusion in the collapsed alveoli ensues, with flooding in the vascular network due to negative intrapleural pressure. This effect can be localised in one or both side of lungs in rabbit model. Mechanical stretch on vessel wall after lung re-inflation by insertion of chest drain, especially after application of suction, provided another precipitating factor for lung injury.

In the study by Tan, the pulmonary wedge pressure remains normal but there would be increase in cardiac output in REPE subjects, which might shed some light concerning the cardiorespiratory interaction in the development of REPE. At risk population include, in summary, young age (20-39 years old) with late presentation (after few days of symptoms), presence of comorbidities in adults such as hypertension, large pneumothorax occupying 30% lung field, or tension pneumothorax at presentation.
with rapid re-expansion of previously collapsed lung by application of high negative suction of more than 20 cm water after chest drain insertion.\textsuperscript{1-3,11}

In spontaneous pneumothorax, suspicion of REPE should be raised when patient complains of shortness of breath, persistent pleuritic chest pain and severe cough within 1 to 24 hr after insertion of chest drain.\textsuperscript{7} Evidence of hypoxemia would be reflected as decreased SpO\textsubscript{2}, increased respiratory rate and heart rate accompanied by an increased oxygen requirement. Some patients may develop symptoms and signs of poor tissue perfusion or, if severe enough, with hypotension and shock. Some patients may be asymptomatic.\textsuperscript{4} CXR would show an increase in lung infiltrates either ipsilateral, contralateral or bilateral.\textsuperscript{11} The extent of infiltrates, however, does not predict disease severity. The size of the cardiac shadow remains normal, which contrasts with cardiomegaly in acute cardiogenic pulmonary oedema. REPE can remain either static, or progress for further 1-2 days and lasts for 4-5 days.\textsuperscript{3} Complete resolution is the rule and patient would not suffer from chronic sequelae from REPE itself.

Other possible differential diagnosis could include pneumonia or cardiogenic pulmonary oedema involving one side. To differentiate from REPE, the time frame in symptoms progression, normal cardiac size on CXR, normal white cell count with absence of fever all can help to pinpoint the correct diagnosis.\textsuperscript{12}

For REPE, once developed, the strategy of management lies in fluid resuscitation by crystalloid while closely monitor the fluid status of the patient; and use of positive pressure ventilation, either invasive ventilation by intubation and mechanical ventilation with higher positive end-inspiratory pressure (PEEP), or non-invasive ventilation via use of nasal continuous positive airway pressure (CPAP).\textsuperscript{1-3,5} In very severe cases, use of asynchronous differential lung ventilation had been tried with success.\textsuperscript{1,13} There had not been any studies comparing the efficacy between uses of invasive or non-invasive ventilation at the moment. Other supportive treatment include intravenous fluid replacement and intravenous morphine.\textsuperscript{1,3} Use of diuretics is detrimental as it would further reduce the systemic perfusion with potential development of organ failure due to hypovolaemia.

As prevention measures, as recommended in BTS guideline, use of high flow oxygen in order to provide good oxygenation would be beneficial to the patient.\textsuperscript{2} Chest X-rays should be performed after chest drain insertion within four hours. Application of suction to promote lung re-expansion should be avoided in the first 48 hours after chest tube insertion.\textsuperscript{7} Whenever use of negative pressure suction, it should be judiciously practiced and only be used when deemed necessary with high vigilance towards the potential development of REPE.

**Conclusion**

We here report a case of REPE post chest drain insertion in primary spontaneous pneumothorax stressing the importance of high clinical suspicion, judicious use of negative suction and clinical management of REPE once occurs.

**References**