

Re-Expansion Pulmonary Edema in Children - A Rare Complication After Pneumothorax Drainage: A Case Report

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Background: Re-expansion Pulmonary Edema (RPE) is a non-cardiogenic form of pulmonary edema which occurs following rapid lung expansion after drainage of significant pneumothorax or pleural effusion, and rarely following resection of obstructive mediastinal mass. RPE is a rare but potentially fatal phenomenon with only few case reports in the pediatric literature.

Methods: We are reporting a case of RPE in a 5-year-old girl following drainage of pneumothorax who succumbed to worsening hypoxemia despite therapy with mechanical ventilation and other supportive care.

Conclusion: RPE should be anticipated, and early preventive, diagnostic and therapeutic measures should be instituted in high-risk patients who require significant pleural fluid or air drainage.

Keywords: pulmonary edema, re-expansion, pneumothorax, pleural effusion, chest tube, Ethiopia

Introduction

Re-expansion Pulmonary Edema (RPE) was first reported in the medical literature in the 1860s.¹ Pediatric RPE cases are less common in the reports than adults.¹⁻³ RPE has also been reported following COVID-19 infection.⁴ The diagnosis of RPE depends on high index of suspicion aided by chest imaging after a clinical deterioration following lung expansion. RPE has no specific treatment, and management is mainly supportive with positive pressure ventilation based on severity of illness. Additionally, proactive measures are recommended to modify the occurrence and severity of RPE in high-risk patients. Because of the failure of consideration of RPE in the differential diagnosis, especially in low-income settings such as ours, where TB and other pleuropulmonary infections are very prevalent, it is likely that RPE is easily missed in clinical practice.^{1,2}

Here we report a pediatric case from a low-income setting who succumbed to RPE after chest tube drainage of pneumothorax. We also present a brief review of the literature of this rare but potentially serious condition to highlight the need to anticipate, prevent, and consider RPE in the differential diagnosis of clinical deteriorations accompanying lung expansions.

Case Presentation

A 5-year-old female child was transferred from a local hospital for further management after 5 weeks of inpatient treatment with oxygen, parenteral broad-spectrum antibiotics, and anti-tuberculous (TB) drugs for non-resolving pneumonia and suspected pulmonary tuberculosis. Her symptoms started acutely with high fever, cough, and difficulty of breathing 8 weeks before admission. After 4 weeks of treatment, the fever subsided but the respiratory distress and hypoxemia persisted for which treatment for pulmonary tuberculosis was initiated empirically without strong clinical or laboratory/imaging evidence. Finally, the child was referred for better workup and management after she failed to respond to the anti-TB.

Physical examination at presentation demonstrated an acutely sick and emaciated child who had significantly increased work of breathing (WOB) and was hypoxemic ($\text{SPO}_2 = <70\%$ at room air and 90–94% on 6–7 L/min

facemask). She had crackles and decreased aeration over the left lower posterior chest with significantly decreased air entry over the right hemithorax and the remaining part of the systemic exam was unremarkable except for presenting as a grossly emaciated child.

PA chest X-ray (CXR) revealed a significant right pneumothorax (Figure 1); other investigations including CBC, COVID-19 and HIV test were unremarkable except for a mildly elevated ESR (30 mm/hr) but negative C-reactive protein.

Her antibiotics and anti-TB treatment were continued, and a chest tube was inserted for pneumothorax. Within the first hour of drainage, she started to have worsening cough and increased WOB with worsening hypoxemia. A repeat CXR 4 hours after her clinical deterioration showed an expanding but opacified right lung and worsening opacity of the left lung suggestive of bilateral pulmonary edema (Figure 2).



Figure 1 Initial CXR showing right pneumothorax. Right lung is collapsed and left paracardiac opacities are also present from the underlying lung disease.



Figure 2 Follow-up CXR after a few hours of chest tube insertion and drainage. Note the expanding but still opacified right lung despite decreasing pneumothorax, and worsening opacities noted in the left lung compared with the previous imaging demonstrating bilateral involvement of the pulmonary edema.

The patient was then started on treatment for severe pulmonary edema with diuretics, and noninvasive ventilation (NIV) without success for >12 hours, subsequently requiring admission to the PICU. She was put on mechanical ventilation with PRVC mode and prior management was continued but she failed to respond and died after 16 hours of PICU stay from therapy-resistant and worsening pulmonary edema.

To the best of our knowledge, this is the first case of RPE reported from Ethiopia. This case report aims to alert clinicians to this rare but potentially fatal complication following drainage of significant pleural effusions and pneumothorax, both of which are fairly common for the practicing pediatrician, especially in low- and middle-income countries (LMICs).

Discussion and Literature Review

Although data are scarce for most settings including ours, pleural effusion was reported in up to 15% of children admitted with pneumonia in LMICs.⁵ RPE was first recognized more than 160 years ago and it usually follows drainage of effusions and pneumothorax. Pediatric reports of RPE are generally few, and incidence estimates are variable. This is likely because of publication biases, and inclusion criteria as most cases are likely asymptomatic and may be detected only with imaging.^{1,2} It is also possible that most of these cases are under-recognized, especially in low-income settings, where post-infectious pleural diseases are more prevalent.⁵

Pathogenesis and Risk Factors

Different mechanisms may cause RPE including increased hydrostatic pressure and inflammatory injury following collapsed lung endothelial reperfusion injury as main mechanisms. Others reported decreased lymphatic flow as a possible contributing factor.^{2,3,6–11} As re-expansion can cause inflammatory changes to the expanded lung, which can then cause a temporary contracted state, this may lead to persistent pneumothorax after drainage (Figure 2). The pathogenesis is even less clear for contralateral pulmonary edema. Overperfusion of the contralateral lung and possibly contralateral lung underlying disease (evidenced by clinical exam and CXR; Figure 1 in our patient) are thought to be the main mechanisms. Although studies have shown the fundamental role of inflammation (both leucocytes and platelets) for contralateral RPE, inflammation is expected to be less severe contralaterally.¹²

The most important risk factor for RPE is the chronicity of lung collapse, RPE risk increasing especially after more than 3 days, and rate of lung expansion with faster re-expansion increasing risk of edema.^{2,6–8} Other factors are the amount of pleural fluid and pneumothorax drained. Adult studies have reported increasing risk with drainage of more than 1500 mL at a time, although pediatric data are limited.^{2,6,9,10}

Clinical Presentation

RPE has been documented in all pediatric ages from early infancy to adulthood and usually develop within a few hours' post-procedure although it sometimes can occur after a few days. The typical presentation is with new-onset or worsening cough, dyspnea, and increasing hypoxemia. Other rare symptoms may include diarrhea and vomiting.^{2–4,6–15} CXR and CT will show features of worsening opacities mainly on the ipsilateral lung but possibly bilaterally as in our patient.^{13–15}

Management and Prevention

RPE has no specific treatment. Asymptomatic and mildly symptomatic patients may not be diagnosed and will not require treatment other than close monitoring if diagnosed. Symptomatic children with RPE are mainly treated with severity-based respiratory support with oxygen, noninvasive positive pressure, and mechanical ventilation in most severe cases.² Diuretics can be considered in those with normal circulatory status while others may require vasopressors and inotropes. Corticosteroids may ameliorate the pro-inflammatory state and were given to some children but their benefit is not proven.^{1,2,4,9–16}

Prevention strategies include timely and limited (<1500 mL fluid at a time and avoiding excessive negative pressure for pneumothorax) drainage of fluid, slow rate of drainage, and possibly the use of ultrasound to guide management of fluid drainage.^{9,10,17} RPE is a serious complication with a fatality rate reaching up to 20%, thus early consideration and prompt diagnosis and treatment are warranted.² However, mortality is likely higher in the resource-scarce settings such as ours, meaning that prevention should be emphasized in high-risk patients. It is also important to note the lack of specific measures that could

improve outcomes specifically dealing with the constraints of resource-limited environments. As a result, practitioners in LMICs should emphasize risk prediction and prevention of RPE as advanced management and timely PICU admission may not be possible even though RPE is diagnosed. However, the current trend towards wider availability of ultrasound in emergency departments and ICUs in LMICs coupled with anticipation of this rare complication could help predict, prevent and early recognition when it happens, facilitating optimal clinical care in such settings.¹⁷

Although our case report highlights the importance of RPE in resource-scarce settings, it has several important limitations. The majority of these limitations are related to the availability of diagnostic and therapeutic measures. The lack of readily available chest CT and ICU beds are simple examples in our case. The quality of CXR images and difficulty to exclude other differentials and the underlying chronic respiratory disease are also to be considered as our challenges. Delayed admission to PICU because of bed unavailability with delay of institution of optimal care including MV will further compromise patient outcome as in our child, who was admitted to the PICU after 12 hours of waiting.

Conclusion

We reported a fatal case of RPE in a 5-year-old child following drainage of a prolonged pneumothorax. RPE should be anticipated in children with massive effusion and pneumothorax, especially after a prolonged period. Preventive measures, as well as preparation for treatment of RPE, should be in place in case preventive measures fail. A fragile health system with diagnostic limitations and delayed institution of advanced treatment is likely to impact the outcome of such severe cases in low resource settings as seen in our patient.

Ethics Statement

Informed consent was obtained from parents of the child to publish this case report. According to the Institutional Review Board of St Paul's Hospital Millennium Medical College, publishing a case report is exempted from the board review.

Author Contributions

Both authors made a significant contribution to the work reported, that is in the conception, execution, acquisition of data, analysis and interpretation, took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

Disclosure

The authors declare that they have no conflicts of interest to disclose for this work.

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