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Case report

Reexpansion pulmonary edema after surgery for spontaneous pneumothorax in a patient with anorexia nervosa

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HIGHLIGHTS

- Ligation of bullae for spontaneous pneumothorax in a patient with AN has never been reported.
- Anorexia nervosa results in critical complications such as RPE.
- Malnutrition due to AN changes the architectural changes in the lung.

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ABSTRACT

Introduction: Several adverse effects on the pulmonary system in patients with anorexia nervosa (AN) have been reported. We present a case of AN who presented with a complicated reexpansion pulmonary edema (RPE) after video-assisted thoracic surgery (VATS) for spontaneous pneumothorax.

Presentation of case: A 23-year-old woman with severe anorexia nervosa (weight: 25 kg, body mass index: 8.96 kg/m^2) underwent VATS for spontaneous pneumothorax. Five hours after the surgery, she immediately presented acute cardiorespiratory insufficiency. Chest radiography showed an infiltrating shadow in the entire right lung. She was diagnosed with reexpansion pulmonary edema that was treated with methylprednisolone pulse therapy and mechanical ventilation. She recovered and was extubated on postoperative day 4. The chest drain tube was removed on postoperative day 5.

Discussion: Bullectomy or ligation of bullae for spontaneous pneumothorax in a patient with AN has never been reported. In our case, bullae were identified in preoperative CT and we chose ligation of the bullae instead of the bullectomy using automatic suture device because of poor wound healing concerned.

Conclusion: We present a case of RPE after VATS for spontaneous pneumothorax in a patient with AN. Malnutrition owing to AN results in critical complications such as RPE.

1. Introduction

Malnutrition resulting from anorexia nervosa (AN) causes various medical complications. Several studies have reported adverse effects of the pulmonary system such as spontaneous pneumothorax due to AN; however, there are no reports in the English literature on reexpansion pulmonary edema (RPE) in such patients. RPE is a rare and lethal complication that occurs after the treatment of pneumothorax. We present a case of RPE after video-assisted thoracoscopic surgery (VATS) for spontaneous pneumothorax in a patient with AN.

2. Presentation of case

A 23-year-old woman, diagnosed with AN three years previously, was admitted to a psychiatric ward of a hospital for psychotherapy and nutritional rehabilitation. Although she was asymptomatic, chest radiography on admission showed pneumothorax in the right lung. A 20-French tube thoracostomy catheter attached to a water-seal was inserted. After two weeks, the patient experienced persistent air leakage that appeared as inadequate expansion of the lung on radiography (Fig. 1A). Subsequently, she was transferred to our hospital for the surgical treatment of the

Abbreviations: AN, anorexia nervosa; RPE, reexpansion pulmonary edema; VATS, video-assisted thoracoscopic surgery.

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pneumothorax. On admission, she weighed 25 kg, and her body mass index was 8.96 kg/m^2. Her neck muscles were too weak to maintain an erect posture while simultaneously opening her mouth. Chest radiography showed a moderate degree of right pneumothorax. A chest computed tomography scan demonstrated several bullae in the upper lobe of the right lung (Fig. 2A). Therefore, video-assisted ligation of the bullae (Fig. 2B) was performed, with which the persistent air leakage resolved. A chest drain tube (−8 cm H_2O suction) was placed in the right pleural cavity, and the right lung was fully expanded perioperatively. Although chest radiography before extubation of the tracheal tube showed an infiltrating shadow at the hilum of the right lung (Fig. 1B) and bronchoscopy revealed frothy secretions, her respiratory condition was stable. Later, extubation of the tracheal tube was performed immediately after the operation. Five hours after the surgery, she developed hypotension, hypoxemia, and anuria. Chest radiography showed that the infiltrating shadow had spread over the entire right lung (Fig. 1C). Arterial blood gas revealed a pH of 7.33, PCO_2 of 35 mm Hg, and PO_2 of 70 mm Hg on FiO_2 of 0.6. She was diagnosed with acute heart failure on the basis of these findings. Re-endotracheal intubation and mechanical ventilation were initiated. The patient was administered albumin, and frozen fresh

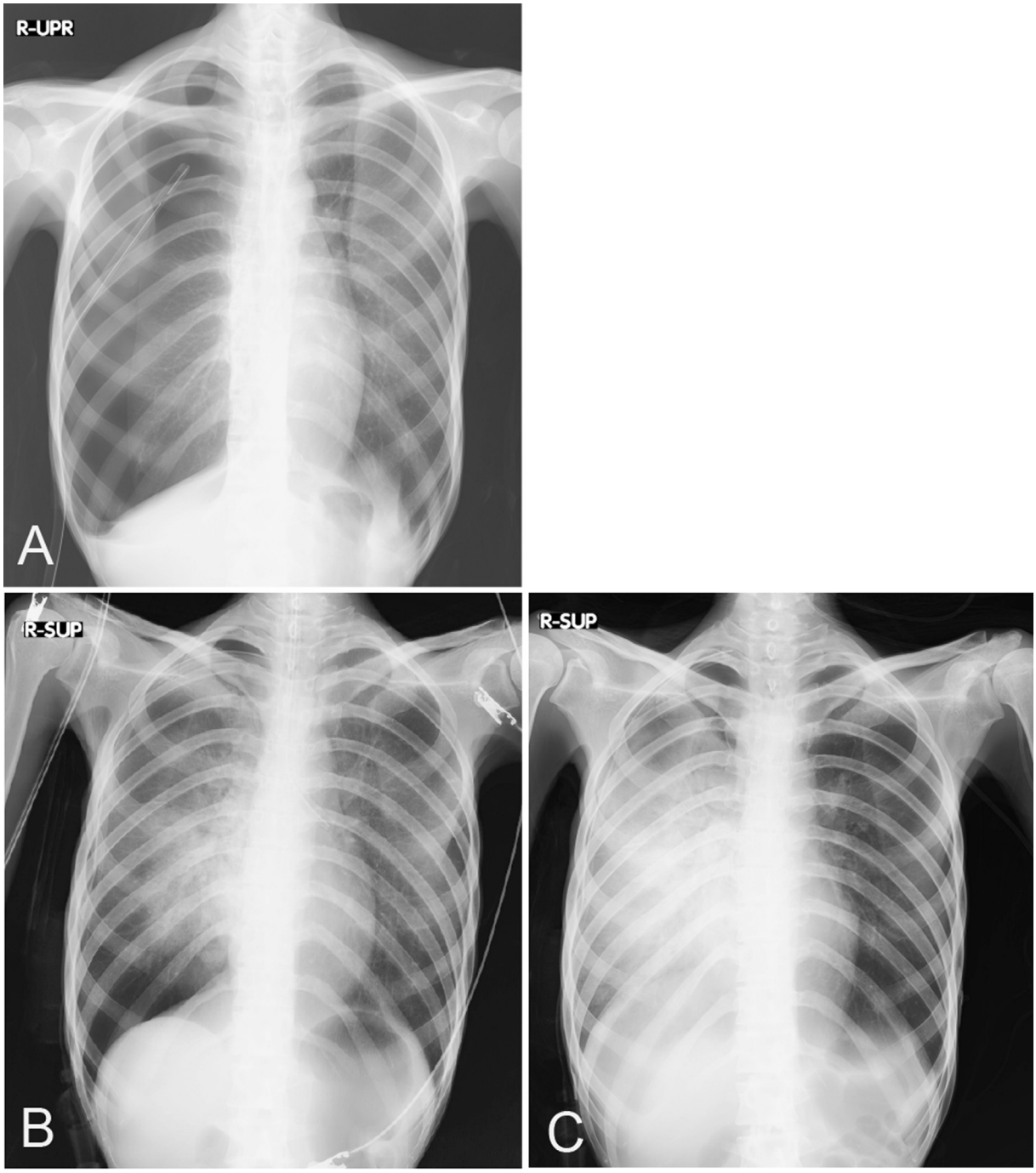

Fig. 1. (A) Chest radiograph obtained on admission showing pneumothorax in the right lung, and placement of the chest drain in the right pleural cavity. (B) Chest radiograph before extubation showing early infiltration in the mid zone of the right lung. (C) Chest radiograph 5 hours after extubation showing full-blown right-sided reexpansion pulmonary edema.

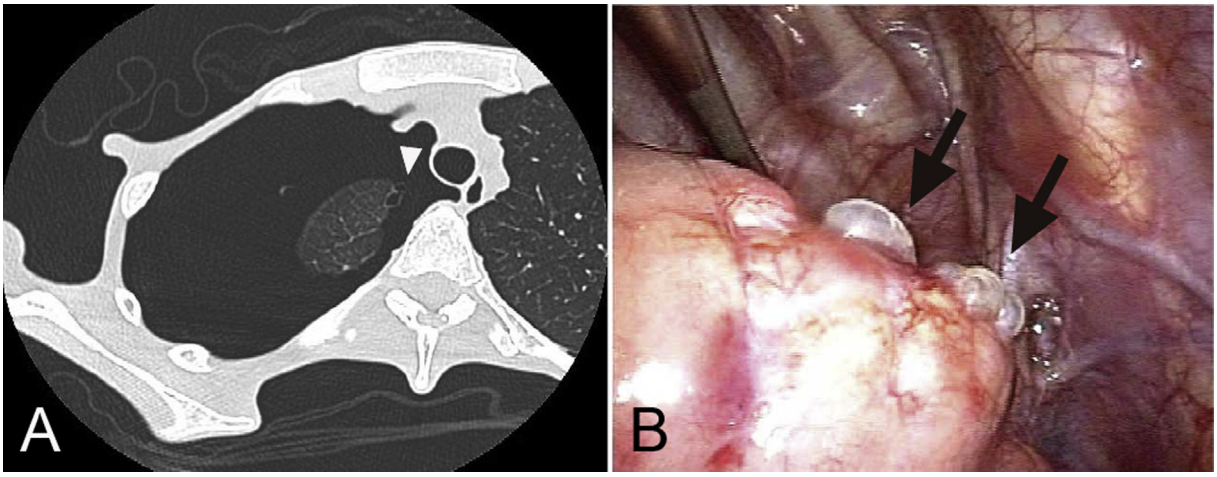

Fig. 2. (A) Chest computed tomography before VATS showing two bullae. The white arrowhead indicates the bullae. (B) Intraoperative image showing several bullae at the apex of the right lung. The arrow indicates the bullae.

plasma preparations and packed red blood cells for circulatory failure, and diuretics and pulse intravenous steroids with 500 mg of methylprednisolone daily for three days for lung edema. Her respiratory and circulatory statuses improved gradually, and the tracheal tube was extubated on postoperative day 4. The chest drain tube was removed on postoperative 5, and there was no recurrence of pneumothorax. The patient was transferred back to another hospital for psychotherapy and nutritional rehabilitation 11 days after the surgery.

3. Discussion

There are five cases including the present case of spontaneous pneumothorax without spontaneous pneumomediastinum in patients with anorexia nervosa in the literature [1–4]. Two cases were improved by chest drainage [1,2], One had spontaneous remission with resting in bed for 3 weeks [3]. One had bilateral VATS pleurectomy in addition to chest drainage, which could not find any abnormality in lung parenchyma [4]. In our case, bullae were identified in preoperative CT and we chose ligation of the bullae instead of the bullectomy using automatic suture device. Coxson and colleagues [5] described that patients with AN may develop architectural changes and decreased surfactant production. This could results in a lung that heals poorly and will not seal itself. Thus, we did not resect the bullae using surgical staplers and instead performed ligation of the bullae to prevent postoperative air leakage. We suggest that an advantage of this management is remediable for a vulnerable lung protectively. However, we consider that ligation of bullae is not effective, when there are numberless bullae or could not find any ruptured bullae.

Extent and duration of lung collapse are reported as a factors susceptible to RPE [6]. In this case, difficulty of the lung expansion retrieval in the preoperative clinical course resulted in protraction of the moderate lung collapse, which might be related in the onset of the RPE. Sahebjami and colleagues [7] reported that prolonged starvation leads to decreases in total lung protein content, connective tissue, hydroxyproline and elastin in rat lung. Based on their findings, we speculate lung in patients with AN decrease elasticity and structural retainability, which increases the resistance to lung re-expansion. Additionally, Sakuma T et al. [8] reported malnutrition causes reduction in alveolar fluid clearance in rat lungs indicating functional deterioration. These mechanical and functional modifications in AN patient's lungs could participate in susceptibility to RPE.

As regard to treatment of RPE, although the standard therapy has not been established, steroid could be an option when the cardiopulmonary insufficiency would be sustained in spite of the appropriate intensive treatment.

4. Conclusion

We present a case of RPE after VATS for spontaneous pneumothorax in a patient with AN. We should recognize the possibility of RPE in patients undergoing VATS. Postoperative RPE is rare but extremely critical complication for poor general condition such as AN. It should be noted that malnutrition of AN develops critical complication such as RPE.

5. Consent

Written informed consent was obtained from the patient and her mother for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Ethical approval

N/A.

Sources of funding

No.

Author contribution

Yuichiro Ozawa and Hideo Ichimura participated in the operation or management of the patient in this case. The manuscript was prepared by Yuichiro Ozawa under the supervision of Hideo Ichimura and Mitsuaki Sakai.

Conflicts of interest

The authors declare that have no competing interests.

Guarantor

Dr. Yuichiro Ozawa.

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