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Case Report

Spontaneous Bilateral Pneumothorax in a Patient with Anorexia Nervosa: The Management of Prolonged Postoperative Air Leakage

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A 24-year-old Japanese female with anorexia nervosa presented to our hospital for bilateral pneumothorax, and 12-Fr thoracostomy catheters were inserted into the bilateral pleural cavities. On hospital day 9, a thoracoscopic bullectomy was performed. However, air leakage relapsed on both sides on postoperative day 1. The air leakage on the right side was particularly persistent, and we switched the drainage to a Heimlich valve. Both lungs expanded gradually and the chest tube was removed on postoperative day 19. Passive pleural drainage might be an option for prolonged air leakage after a bullectomy in patients with anorexia nervosa.

Key words: spontaneous pneumothorax, anorexia nervosa, Heimlich valve

Spontaneous pneumothorax is commonly observed in tall and thin adolescent males, and chest-tube drainage is the standard treatment for the first episode. Prolonged air leakage, whether relapsed or bilateral, is an indication for surgical treatment. The most well known and minimally invasive surgical treatment for spontaneous pneumothorax is stapling of the bullae by video-assisted thoracic surgery [1, 2].

Anorexia nervosa (AN) is a serious eating disorder characterized by severely low weight and fear of becoming obese; AN can cause medical complications in the gastrointestinal, cardiovascular, hematologic, and endocrine systems, and especially the lungs [3]. The occurrence of refractory spontaneous pneumothorax in patients with AN has been encountered [4-8]. We report the case details of a young woman with AN and the successful treatment of prolonged air leakage that occurred after she underwent surgery for bilateral spontaneous pneumothorax.

Case

A 24-year-old Japanese female with AN, a nonsmoker, presented to our hospital complaining of chest discomfort and dyspnea. Chest X-ray imaging revealed bilateral pneumothorax. Based on the British Thoracic Society guidelines [9], the patien's right lung was completely collapsed and the left was moderately collapsed (Fig. 1A). One month prior to this presentation, the patient had been diagnosed with AN and was advised to be hospitalized for nutrition therapy, but she refused. She was 150 cm (~4' 11") tall and weighed only 26 kg (57.3 pounds), making her body mass index 11.5 kg/m^2. Her vital signs were as follows: heart rate, 50 beats/min; blood pressure, 97/58 mmHg; and oxygen saturation, 91% (room air). Laboratory analyses revealed slightly elevated aspartate aminotransferase (AST) and alanine aminotransferase (ALT) levels (53 and 117 U/L, respectively). Platelet and electrolyte counts were normal.

The patient was hospitalized urgently, and a 12-Fr thoracostomy catheter was inserted into the bilateral

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pleural cavities. Both chest drains were on water seal for 3 days due to the high risk of re-expansion pulmonary edema [4]. On hospital day 4, we started using suction and gradually increased the pressure. Computed tomography (CT) images after the lungs re-expanded showed bullae at the apex of both lungs (Fig. 1B, C). Nutritional rehabilitation was indicated simultaneously.

On hospital day 9, a thoracoscopic bullectomy was performed and covered with oxidized cellulose. Ruptured bullae at the apex of both lungs and an air leakage were detected. The bullae were then resected with the use of endoscopic staplers, and the surgery was completed after no air leakage was detected (Fig. 2). However, the air leakage relapsed on both sides on postoperative day 1. The collapse on the right side was persistent, whereas that on the left side was resolved conservatively and became a smaller collapse on postoperative day 3. The left-side chest drain was thus removed. Reoperation for the right lung air leakage was not performed because the alveolopleural fistula was considered to be on the staple row from weakened lung architecture. After confirmation that the patient's right lung collapse was moderate on the water seal, the drain was switched to an 8-Fr trocar catheter and a Heimlich valve was connected. She was discharged on postoperative day 6 (hospital day 15).

The patient was then followed up in an outpatient department, and it was observed that both lungs gradually expanded. On postdischarge day 13 (postoperative day 19), the chest tube was removed after it was confirmed that the patient's lungs were fully expanded after a 3-h clamping trial. Chest X-ray imaging at 3 months postdischarge detected no evidence of collapse (Fig. 3).

Discussion

This patient's case involved a gradual re-expansion of collapsed lungs and passive drainage for postoperative air leakage due to pneumothorax with AN. Chronic malnutrition causes organic and functional disorders, and pulmonary complications also occur in patients suffering from AN. It was reported that the lungs of patients with AN (even those without a smoking history) show emphysema-like changes [10]. Corless *et al.* described bilateral spontaneous pneumothorax in a patient with AN, treated by chest drainage and pleurectomies [7]. In a model using young rats, prolonged starvation resulted in reduced total lung protein content, connective tissue, hydroxyproline, and elastin, indicating that malnutrition can weaken the lung architecture [11]. The fragility of the visceral pleura and lung parenchyma would be a factor in refractory pneumothorax and consequent complications. In addition, Ozawa *et al.* presented a pneumothorax case of postoperative re-expansion pulmonary edema in a patient with AN [4]. Reduced alveolar fluid clearance in starved rats was described [12]. In patients with AN, gentle drain-

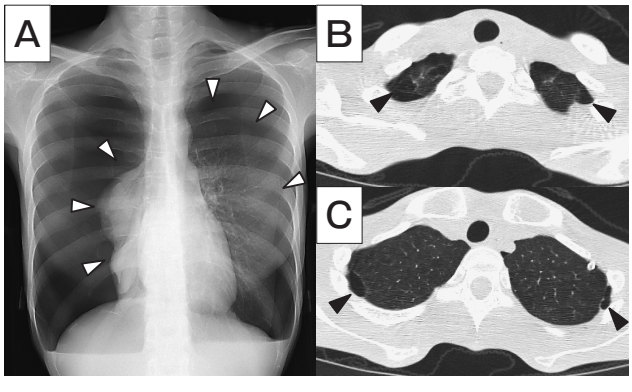

Fig. 1 **A**, A chest X-ray image obtained on admission revealed bilateral pneumothorax. The right lung was completely collapsed, and the left lung was moderately collapsed (*white arrowheads*); **B**, **C**, Computed tomography images showed bullae at the apex of both lungs (*black arrowheads*).

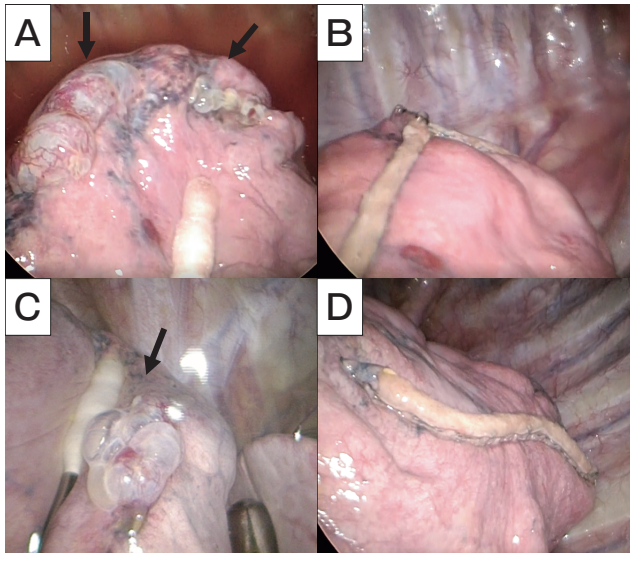

Fig. 2 **A**, **B**, The thoracic intraoperative images of the right and (**C**, **D**) left sides. On thoracoscopic exploration, two bullae were detected at the right lung apex and one bulla was detected at the left lung apex (*black arrows*). Air leakages were identified, and the bullae were resected with the use of endoscopic staplers.

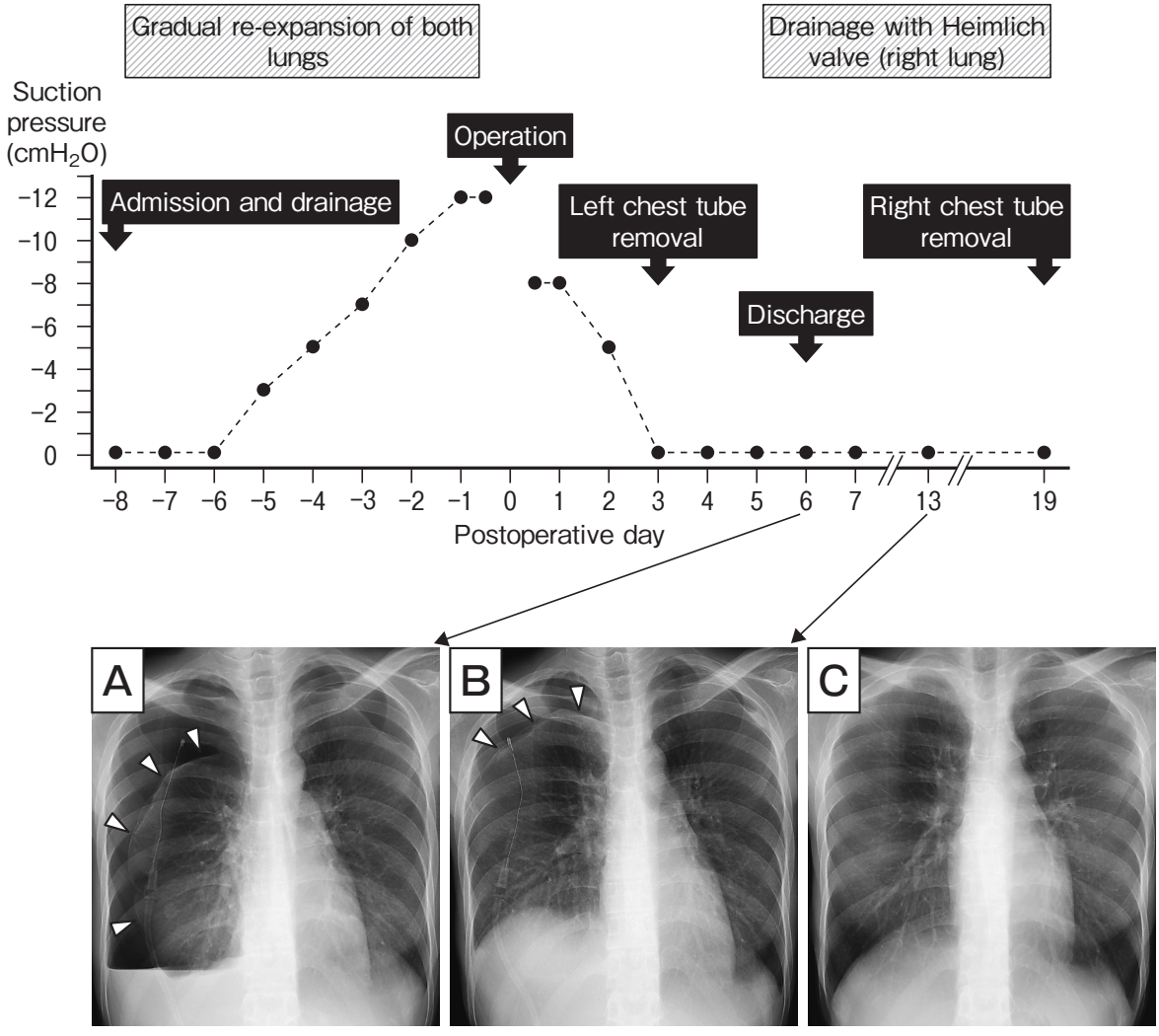

Fig. 3 The patient's clinical course. **A**, The chest X-ray image obtained just before discharge on postoperative day 6; **B**, Gradual re-expansion of the right lung was observed on postoperative day 13 (*white arrowheads*); **C**, Full expansion of both lungs was observed on postoperative day 27 (8 days after the catheter removal).

age of the thoracic cavity would be crucial to prevent re-expansion pulmonary edema.

From a surgical perspective, there are concerns that weakened lung architecture causes an intraoperative lesion, resulting in persistent postoperative air leakage. This can be prevented by the ligation of bullae instead of a bullectomy with surgical staplers, which may be useful in reducing the tension of the visceral pleura [4]. In the present patient's case, ligation was not performed because the bullae were sessile and another bulla was adjacent. However, if possible, ligation of the bullae should be considered for the prevention of postoperative air leakage.

Prolonged air leakage is a common complication in spontaneous pneumothorax surgery [13]. In patients with AN, the effects and risks of follow-up surgery and medical pleurodesis, including minocycline or talc, are not well known. In addition, given that our patient's right lung collapsed, we felt that it would not be effective to perform pleurodesis. The water seal drainage was switched to a Heimlich valve, and the postdischarge prognosis was good. Water seal drainage has been reported to shorten the duration of chest drain placement after spontaneous pneumothorax surgery compared to suction drainage [14]. Biffl *et al.* also described a patient with pneumothorax and AN who was treated

using a Heimlich valve for persistent air leakage [6]. Excessive negative pressure in the thoracic cavity may worsen the air leakage and weaken the visceral pleura.

Our patient's case suggests that conservative treatment with passive drainage is an option for prolonged air leakage after a bullectomy. Further studies are needed to establish a standard therapy for spontaneous pneumothorax in patients with AN.

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