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Surgical Treatment of Postpneumonectomy Syndrome with Tissue Expanders in Children

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Postpneumonectomy syndrome (PPS) is a rare late complication of pneumonectomy. It occurs more often in children than in adults, and is characterized by respiratory failure resulting from bronchial compression caused by severe mediastinal shift. Various methods have been used to treat PPS, including aortopexy and the insertion of plastic balls, silastic implants, and saline-filled breast prostheses. We describe two cases of PPS corrected with tissue expanders after right pneumonectomy in patients with esophageal atresia.

Key words: 1. Pneumonectomy
2. Tissue expansion devices
3. Esophageal atresia

CASE REPORTS

1) Case 1

A three-month-old male presented with cough, sputum, and intermittent cyanosis that had lasted for one week. He underwent an operation for esophageal atresia (type C). A postoperative esophagogram showed a bronchoesophageal fistula, and a secondary operation involving fistulectomy and right lower and middle bilobectomy was performed. Nevertheless, he required right pneumonectomy due to recurrent pneumonia in the remaining right upper lobe. On readmission five months after the pneumonectomy, a chest computed tomography (CT) scan revealed post-pneumonectomy changes, with mediastinal shift to the right and collapse of the posterior wall of the trachea compressed by the aortic arch (Fig. 1). An operation was performed via right thoracotomy. Aortopexy was performed, followed by insertion of a tissue expander into the right pleural cavity. After the insertion of the tissue expander, 45 mL of normal saline was instilled, and the thoracotomy wound was closed. A postoperative CT scan demonstrated that the mediastinum had returned to its normal position. The patient was discharged 29 days after the operation. At a one-year follow-up, he had no symptoms.

2) Case 2

A 22-month-old female with esophageal atresia (type C) underwent fistula resection and an esophageal anastomosis. A postoperative esophagogram revealed a bronchoesophageal fistula. A fistulectomy between the right intermediate bronchus and the distal esophagus was performed, as well as right pneumonectomy for persistent collapse of the right lung. Twenty-two months after the pneumonectomy, she experienced unspecific respiratory symptoms of cough and sputum for five days. A chest CT scan demonstrated mediastinal shift

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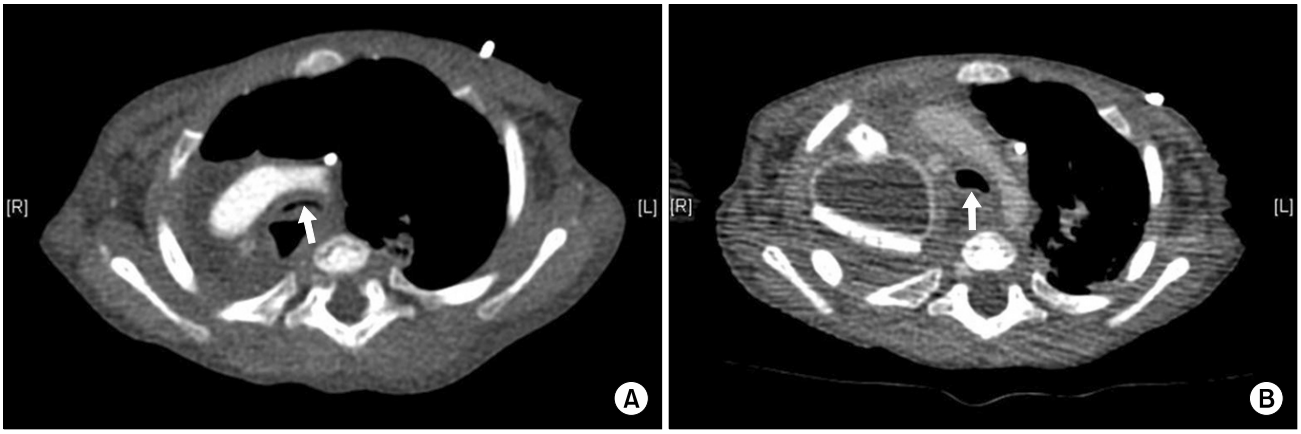

Fig. 1. (A) A preoperative computed tomography (CT) scan shows narrowing of the trachea (arrow), which is compressed by the aortic arch. (B) A postoperative CT scan shows the tissue expander with the mediastinum in normal position.

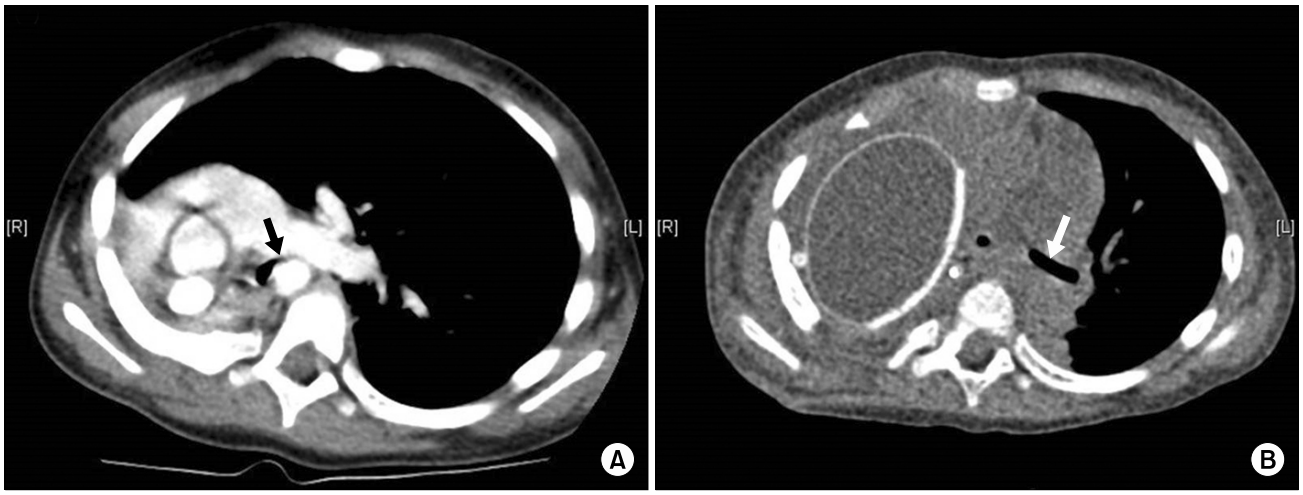

Fig. 2. (A) A preoperative computed tomography (CT) scan shows narrowing of the left main bronchus (arrow), which is compressed between the left main pulmonary artery and descending aorta. (B) A postoperative CT scan shows the tissue expander and the resolved left bronchus (arrow).

to the right and a narrowed left main bronchus, which was compressed between the left main pulmonary artery and the descending thoracic aorta (Fig. 2). We performed mediastinal repositioning by inserting a tissue expander with 120 mL of saline. During the first two postoperative days she required ventilator therapy, and venovenous extracorporeal membrane oxygenation was used to provide support due to acute respiratory failure resulting from a bronchospasm. The patient was discharged on the 77th postoperative day, following successful weaning from extracorporeal membrane oxygenation. At a one-year follow-up, the patient showed no symptoms and a chest X-ray showed no specific findings.

DISCUSSION

Postpneumectomy syndrome (PPS) is a rare disorder involving the extrinsic compression of the trachea or bronchus due to shifting of the mediastinum and over-inflation of the remaining lung parenchyma. It results in a counterclockwise rotation of the heart and great vessels, along with herniation of the remaining lung into the contralateral hemithorax. PPS is more common in children, occurring in up to 15% of pa-

tients who undergo right pneumonectomy (or, rarely, left pneumonectomy in patients with a right aortic arch). The frequency with which this syndrome occurs in infants and young children is thought to be related to the increased elasticity and pliability of their lungs and mediastinum, compared with those of adults. They have more pliable and mobile mediastinal tissue than older patients, with a greater tendency to shift that can easily lead to anatomic distortion. PPS is characterized by progressive dyspnea, cough, and recurrent pneumonia at least six months after pneumonectomy. Because of the rarity and unspecific symptoms of the disease, most patients already have bronchomalacia by the time of diagnosis.

PPS can be life-threatening if not treated. In the past, various procedures have been proposed, including phrenectomy or crushing of the phrenic nerve, tracheal or bronchial transection with anastomosis, suspension of the aorta to the sternum, and endoscopic bronchial stent insertion [1-4]. More recently, surgical repositioning of the mediastinum with a nonabsorbable material has been widely accepted as a standard treatment. Many studies have reported that the insertion of an intrathoracic prosthesis can improve clinical symptoms and relieve functional obstructions [5,6]. Shen et al. [7] reported that placement of a saline-filled tissue expander led to successful mediastinal repositioning. At a median follow-up of 32 months, 10 of 13 patients (77%) reported a significant improvement in their symptoms, with a relatively low operative mortality rate of 27.8%.

The early insertion of a tissue expander when pneumonectomy is performed may prevent the possible detrimental consequences that result from the delayed diagnosis and treatment of PPS. Furthermore, some reports have described an inability to place a tissue expander due to postoperative adhesion when the PPS is developed after pneumonectomy [8]. Choi et al. [8] have suggested that prophylactic implantation of a tissue expander may prevent the potentially very serious complication of PPS.

In our two cases, tissue expanders improved the ventilation of the remaining lung by alleviating the obstruction of the trachea or bronchus and shifting the mediastinum to the midline. Although our follow-up duration was short, we observed decreased respiratory compromise and no complications involving leakage or rupture related to the tissue expander. In our opinion, PPS can be completely and successfully treated using a tissue expander, which can restore the position of the mediastinum.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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