Thoracic Cancer ISSN 1759-7706

ORIGINAL ARTICLE

Management of post-pneumonectomy syndrome using tissue expanders

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Keywords
Pneumonectomy; post-pneumonectomy syndrome; tissue expander.

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Received: 24 March 2015;
Accepted: 3 May 2015.

doi: 10.1111/1759-7714.12282

Thoracic Cancer **7** (2016) 88–93

Abstract

Background: Post-pneumonectomy syndrome (PPS) is a rare syndrome characterized by trachea-bronchial stenosis and severe dyspnea. In this study, we retrospectively evaluated the clinical outcomes in patients who underwent placement of tissue expanders for PPS.

Methods: Data from patients who underwent placement of tissue expanders for PPS were analyzed for preoperative characteristics, surgical techniques, and postoperative outcomes. Between 1997 and 2014, a total of 10 patients were treated for PPS by tissue expanders.

Results: The median age of the 10 patients was 45 years (range, 16–70). Four patients had undergone right pneumonectomy, three patients had undergone left pneumonectomy, and three patients had post-pneumonectomy-like syndrome. Preoperatively, seven patients initially received high oxygen therapy for hypoxemia but progressed to respiratory failure, and three patients required mechanical ventilator support. Among these three patients, one required intraoperative extracorporeal membrane oxygenation support because of sudden cardiac arrest during preparation for surgery. The median follow-up was 59.5 months (range, 2–204). The median interval between pneumonectomy and repositioning was 13 months (range, 8–581). Two patients underwent placement of a single tissue expander, and two tissue expanders were placed in eight of the 10 patients. The median volume of tissue expanders inflated with saline was 450 cc (range, 60–850 cc) per tissue expander. There was no perioperative mortality in our study. Complications occurred in four patients (40%).

Conclusions: Repositioning of the mediastinum with placement of a saline filled tissue expander for PPS is very effective for the relief of symptoms, with low mortality.

Introduction

Post-pneumonectomy syndrome (PPS) is very rare complication that occurs after pneumonectomy. However, this syndrome is not exclusive to patients that receive a pneumonectomy, it can also occur in patients that experience destructive change, agenesis, and other conditions. In these situations, it is called post-pneumonectomy-like syndrome (PP-like syndrome); however, symptoms, diagnosis, and treatment are similar to PPS. Progressive dyspnea is a typical symptom of PPS and other symptoms include recurring pneumonia, stridor, laryngeal nerve palsy, and dysphagia.[1] Respiratory symptoms may be caused by an external airway obstruction as a result of a mediastinal shift and rotation to an empty hemithorax. Therefore, it is important to check airway condition and mediastinal status because, without treatment, this syndrome may cause respiratory failure and death. Previous reports have shown that surgical correction using a saline-filled tissue expander restored the normal mediastinal position and allowed the compressed airway to return to its normal position.[2–4]

We present our experience with 10 patients that were treated with tissue expanders for PPS or PP-like syndrome in our hospital.

Material and methods

From March 1997 to March 2014, 10 patients with PPS underwent surgical correction in the Department of Thoracic and Cardiovascular Surgery at Samsung Medical Center, Seoul, Republic of Korea. The Samsung Medical Center institutional review board approved our study.

Patient characteristics

Patient records were retrospectively reviewed in detail. We collected and analyzed patient characteristics including: gender; age; original disease; interval between original and corrective surgery; surgical procedure performed during first surgery and corrective surgery; preoperative and postoperative radiographic and pulmonary function tests; postoperative morbidity and mortality; length of hospital stay; and last follow-up visit or date of death.

Post-pneumonectomy syndrome was diagnosed on the basis of physical examination, chest radiography, pulmonary function test, chest computed tomography (CT) scanning, and bronchoscopy. Preoperative bronchoscopy was performed in patients when possible, and intraoperative bronchoscopy was performed in all patients to examine the status of the airway during surgery.

The selection criteria were as follows:

1 Any patients who were diagnosed with PPS.
2 No significant metastatic disease.
3 No severe co-morbidity, such as irreversible cardiac or pulmonary disease in the remaining lung.
4 Eligible for general anesthesia.
5 No evidence of empyema in the post-pneumonectomy space.

Surgical technique

After general anesthesia, patients were positioned in the lateral decubitus position. A postero-lateral thoracotomy incision was performed and the intercostal space above the original incision was reopened. Adhesions were dissected in patients who had a previous thoracotomy wound. Close attention must be paid while accessing the pleural cavity for adhesiolysis; severe anatomic distortion and dense fibrosis may be present and may place mediastinal structures at risk during this portion of the procedure. Once the pleural cavity was accessed, the mediastinum was completely released from the chest wall using blunt and sharp dissection to return the mediastinum to the midline. The junction between the pneumonectomy stump and the main bronchus needs to be extensively dissected and released, which can be challenging. That junction is sometimes scarred to either the vertebral body or the posterior mediastinum, and complete release and lysis of the scar tissue are very important. The surgical groove was filled with saline and was temporarily inserted to determine the amount of fluid volume required to fix the mediastinum for appropriate repositioning of tissue expanders (Fig 1) before the size and number of prostheses were chosen. Natrelle (Allergan Inc., Irvine, CA, USA) and Mentor Siltex (Mentor Corporation, Santa Barbara, CA, USA) tissue expanders were used. The pneumonectomy space was filled before returning the mediastinum and heart to a midline position to avoid compressing the heart and lung. Central venous, blood, and pulse pressures were carefully monitored during this part of the procedure. If the central venous pressure increased by more than 5 mmHg or was greater than 10 mmHg, we adjusted the fluid volume of the tissue expander to correct the blood

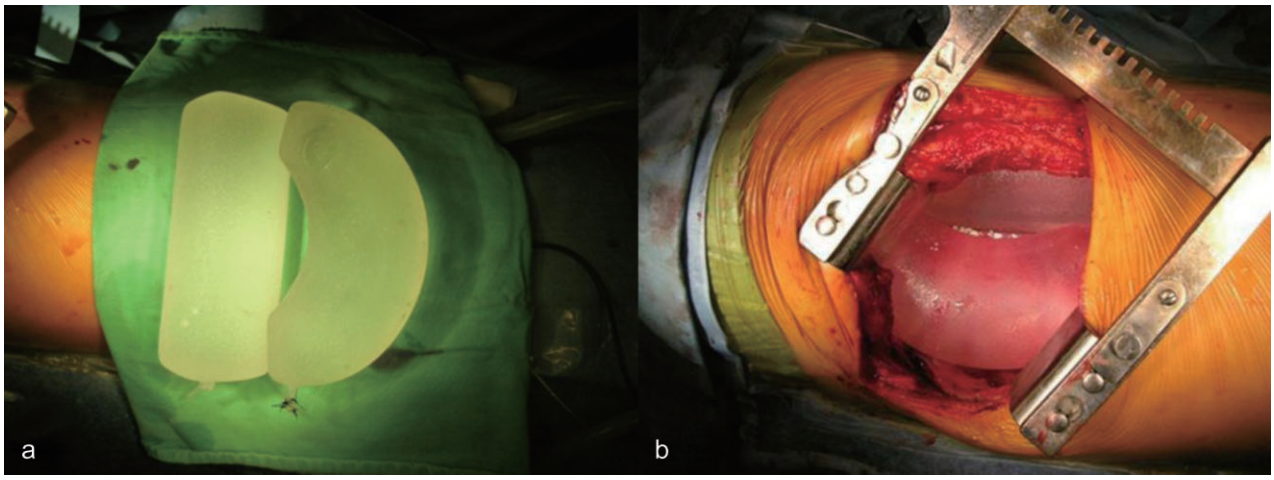

Figure 1 (**a**) Two types of tissue expanders were used. One expander is rectangular-shaped and the other is a crescent type. (**b**) After thoracotomy, two types of tissue expanders were inserted in the thoracic cavity.

pressure. Finally, the thoracotomy wound was closed with a single drainage tube.

Follow-up

Follow-up evaluation consisted of patient history, physical examination, laboratory testing, pulmonary function test, chest radiography, bronchoscopy, and CT.

Results

Patient characteristics

The study included 10 patients (9 women and 1 man) with a median age of 45 years (range, 16–70) who underwent mediastinum repositioning using tissue expanders. The median follow-up was 59.5 months (range, 2–204). The median time interval between intervention and diagnosis of PPS was 25.5 days (range, 2–99). The major clinical characteristics of the patients are shown in Table 1. Four patients underwent right pneumonectomy (1 patient had an adenoid cystic carcinoma, 1 a pulmonary tuberculosis, and 2 received pneumonectomies for pulmonary tuberculosis 45.4 and 52.3 months after lobectomy), three patients underwent left pneumonectomy (all 3 patients had pulmonary tuberculosis), and three patients had PP-like syndrome (2 patients had severely destroyed lungs as a result of pulmonary tuberculosis, and 1 patient had congenital pulmonary agenesis). All patients had symptoms of progressive dyspnea that occurred at rest. Other presenting symptoms included recurrent infection (30%), chest pain (20%), and dysphagia (10%).

Table 1 Patient characteristics (n = 10)

Variable	Number	Percentage (%)
Age, mean (range)	45 (16–70)	
Gender		
Male	1	10
Female	9	90
Original disease		
PTB	7	70
NTM	1	10
ACC	1	10
Congenital pulmonary agenesis	1	10
Asthma	2	20
COPD	1	10
Pneumonectomy type		
Right	4	40
Left	3	30
Pneumonectomy-like	3	30
Symptom		
Dyspnea	10	100
Chest pain	2	20
Recurring pneumonia	3	30
Dysphagia	1	10
Preoperative management		
High oxygen therapy	7	70
Mechanical ventilator	3	30
ECMO	1	10

ACC, adenoid cystic carcinoma; COPD, chronic obstructive pulmonary disease; ECMO, extracorporeal membrane oxygenation; NTM, nontuberculosis mycobacteria; PTB, pulmonary tuberculosis.

Preoperatively, seven patients initially received high oxygen therapy for hypoxemia, which progressed to respiratory failure, and three patients required mechanical ventilator support. Among these three patients, one patient needed intraoperative extracorporeal membrane oxygenation support because of sudden cardiac arrest while preparing for surgery.

Tissue expander placement and surgical outcomes

Seven patients received mediastinum repositioning and tissue expander implantation. Three patients underwent mediastinum repositioning and tissue expander implantation after pneumonectomy. Two patients underwent a single tissue expander placement, and two tissue expanders were placed in eight of the 10 patients. The median fluid volume, inflated with saline, per tissue expander was 450 cc (range, 60–850 cc) (Table 2).

Preoperative and postoperative imaging scan

Preoperative chest CT scans of the patients after pneumonectomy showed deviation of the trachea and heart to the empty hemithorax. Hyperinflation of the remaining lung with a shift of the great vessels was observed (Fig 2a). A chest CT scan after replacement of the tissue expander showed a well-positioned tissue expander with a midline mediastinum (Fig 2b). Mediastinal repositioning relieved the mechanical obstruction of the bronchus.

Surgical outcomes

There was no perioperative mortality in our study. Complications occurred in four patients: pneumonia in one patient, venous thrombosis of the arm in one patient, and tissue expander leakage in two patients. One patient developed pneumonia, which improved with antibiotic treatment. One patient developed a venous thrombosis of the arm, but this improved after anticoagulation therapy. Two patients required reoperation to address tissue expander leakage. Surgical results are summarized in Table 2. Nine patients (90%) showed symptom improvement after surgery and they were discharged without oxygen. The median hospital stay was 11 days (range, 4–83).

Table 2 Patient details: clinical features and surgical management

Patient No.	Original disease	Interval between original and corrective surgery (year)	No. of implants	Volume of implants (mL)	Length of hospital stay (d)	Follow-up interval (months)	Early result (within 30 days)	Late result (after 6 months)	Cause of death
1	Tuberculosis	0.7	2	850	12	204	Improved	Good	Survival
2	Tuberculosis	0.8	2	400	14	61.7	Improved	Death	Pneumonia
3	NTM	1.1	2	420	6	69.7	Improved	Dyspnea aggravation	Survival
4	ACC	0.9	2	780	5	97.7	Improved	Good	Survival
5	Tuberculosis	(-)	2	380	4	65.7	Improved	Good	Survival
6	Tuberculosis	49	1	300	60	58.9	Improved	Good	Survival
7	Tuberculosis	(-)	2	580	10	29.7	Improved	Good	Survival
8	Tuberculosis	2.0	2	220	4	26.8	Improved	Dyspnea aggravation	Survival
9	Tuberculosis	(-)	2	620	23	6.5	Improved	Good	Survival
10	Tuberculosis	12.2	1	60	83	2.7	No improvement	No improvement	Survival

ACC, adenoid cystic carcinoma; NTM, nontuberculosis mycobacteria.

Six patients underwent preoperative and postoperative pulmonary function testing. The remaining four patients could not undergo a pulmonary function test. The forced expiratory volume in one second (FEV1) decreased in three patients and improved in three patients. The vital capacity decreased in four patients and improved in two patients.

Discussion

Post-pneumonectomy syndrome is a rare complication that can occur after pneumonectomy and is caused by excessive mediastinal shifting and rotation that results in extrinsic airway compression.[1] In some cases, a PP-like syndrome can occur, even after a lobectomy.[5] This syndrome was observed in a patient with complete lung destruction resulting from tuberculosis.[6] In 1972, the first report of severe airway obstruction after pneumonectomy was observed in a child.[7] The term PPS was introduced in 1979 by Wasserman *et al*.[8] Mediastinal shifting and rotation toward the empty hemithorax can cause pulmonary and esophageal compression. The mediastinum shifts posteriorly and to the right, with the heart and great vessels rotating in a counter-clockwise direction after right pneumonectomy. This results in dynamic airway extrinsic compression of the left main bronchus between the left pulmonary artery and aorta. Additionally, the mediastinum rotates clockwise after left pneumonectomy. The hyperinflation and herniation of the remaining lung caused by the mediastinal shift lead to progressive clinical worsening. This results in dynamic airway extrinsic compression of the right main bronchus between the right pulmonary artery and the thoracic spine after left pneumonectomy.[1,9] PPS appears to be much more common after right pneumonectomy.[10]

The overall incidence of this condition is unknown; however, in children, the incidence is thought to be one in 640 cases.[11–13] PPS is more common in children than in adults and it is hypothesized that this is because of the increased elasticity and compliance of the remaining lung and mediastinal tissue. This results in a narrowing and twisting of the main

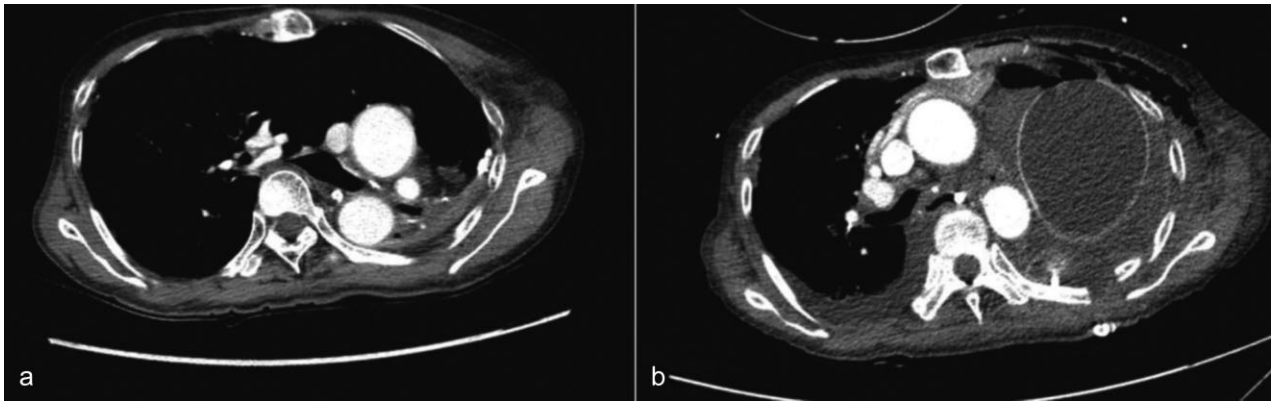

Figure 2 Preoperative and postoperative computed tomography scans of a patient. (**a**) The preoperative scan shows complete displacement of the mediastinum to the left chest. (**b**) The postoperative scan shows a well-positioned tissue expander with a midline mediastinum.

bronchus.[2,3,13] Furthermore, several studies have reported that this syndrome is more frequent in women than in men.

Patients typically present with dyspnea, stridor, atelectasis, recurring pneumonia, and laryngeal nerve palsy, which may appear weeks to years after pneumonectomy. They may also present with feeding difficulty, although it is masked by dramatic airway symptoms.[1] Diagnosis is based on chest radiography, CT, pulmonary function test results, and bronchoscopy. Other pathologic and physiologic conditions, such as recurrent cancer, progressive chronic obstructive pulmonary disease, pulmonary thromboembolism, pulmonary hypertension, asthma, aspiration, and heart failure, must be ruled out.[1,14]

Without treatment, PPS can be life threatening and the treatment options are very limited. Most of the previously reported cases were single case reports; a small series described up to 20 patients.[2–22]

Treatment options include conservative management and surgery. Division of the ligamentum arteriosum, fixation of the aorta or pulmonary artery to the sternum, and placement of expandable stents have been described with varying outcomes.[7,17–20] However, endoscopic bronchial stent insertion does not always enable adequate placement of a stent, and is usually applied in the presence of gross distortion. Moreover, with this treatment, the cause of compression of the main bronchus does not affect mediastinal displacement. Therefore, endobronchial stents should only be considered for treating patients with PPS who are not candidates for surgical treatment strategies.[19]

Various methods for repositioning the mediastinum have also been described. These include suture fixation of the pericardium to the back of the sternum, surgical and chemical phrenectomy, and placement of prostheses to fill the empty hemithorax.[10,16] Lucite plastic balls, silastic implants, injection of sulfur hexafluoride into the empty hemithorax space, and saline-filled breast prostheses have been used for prevention and therapy.[2,3,7,8,12,16,21,22]

Besides these treatments, saline-filled tissue expanders in the empty hemithorax are widely used for mediastinal repositioning to the midline and to decompress the bronchial obstruction. This method has been used to achieve successful surgical outcomes.[2–4] One report indicated that there was a 77–100% significant improvement in syndromes; operative morbidity was 27.8–32% and mortality was 0–5.6%.[2,3] In our study, 90% of patients achieved significant improvement of respiratory symptoms and there was no perioperative mortality.

To determine the efficacy of the surgery, patient symptoms, chest radiography, bronchoscopy, and pulmonary function are evaluated. Improved symptoms and a mediastinum that returns to the midline on chest radiography are considered successful outcomes. Bronchoscopy shows the successful relief of bronchial compression, as well as the absence of bronchomalacia after successful repositioning. Several studies have reported a change in pulmonary function tests after corrective surgery.[2,16,23] Two key changes are the increase of the flow rate and decrease of hyperinflation. The flow rate improvement is reflected in both the increased FEV1/forced vital capacity (FVC) percent ratio and the peak expiratory flow rate. FVC is decreased after surgery when hyperinflation of the remaining lung is corrected. FEV1 may increase or decrease depending on the situation. FEV1/FVC increases to reduce FVC and is absolutely greater than the reduction in FEV1. These changes were more obvious in cases with right-side PPS than in left-side PPS.[2,14,16] In our study, the mean FEV1 increased from 1.08–0.98 L, the mean FVC decreased from 1.50–1.32, the mean FEV1/FVC ratio increased from 0.68–0.75, and the mean peak expiratory flow rate increased from 1.94–2.26. Although pulmonary function tests are objective indicators that can predict the outcome after surgery, there are many variables, such as the degree of airway obstruction, compliance, and underlying lung disease. For this reason, clinical symptom improvement is the most important factor in assessment of surgical outcomes.

In patients with recurrent symptoms, the onset of a complication of implantation, such as inadequate filling of or leaking from the tissue expander, malposition, and luxation, must be ruled out. CT is the best tool for confirming this.[3] In our study, leakage occurred in two patients. One patient showed improved symptoms after revision surgery, and the remaining patient had no improvement of symptoms.

Post-pneumonectomy syndrome is a rare complication after pneumonectomy surgery. Recently, the tissue expander has been used as a treatment approach for this disease. Ten patients were operated on in our hospital, and achieved good results with lower morbidity and no mortality. We believe that the results of this study can assist physicians to determine appropriate surgical treatment and therapeutic strategies in patients with PPS.

Conclusion

Repositioning of the mediastinum with placement of saline-filled tissue expanders for PPS is very effective for relieving symptoms and is associated with a low mortality rate.

Disclosure

No authors report any conflict of interest.

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