Postpneumonectomy Syndrome: Results of mediastinal repositioning vs. stent placement

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Citation

Abstract
Background: Postpneumonectomy syndrome (PPS) is a late complication of pneumonectomy characterized by mediastinal shift and bronchial compression. It is most common following right pneumonectomy but is also seen left pneumonectomy. It can be treated with mediastinal repositioning and tissue expander placement in the postpneumonectomy space, but there is some interest in less invasive modalities. Endobronchial stent placement may be an option. We looked at our experience treating PPS with these two modalities. Methods: All patients with PPS treated with mediastinal repositioning/tissue expander placement or bronchial stenting at our institution from 1991 to 2005 were reviewed. Results: Mean age at the time of pneumonectomy was 45 years. Mean follow-up was 33 months. Six patients underwent tissue expander placement. They had relief of symptoms with the following complications: wound infection, atrial fibrillation, expander leak and esophageal dysmotility. Two patients underwent silastic stent placement with immediate resolution of symptoms, however they suffered from frequent mucous plugging, stent migration, and granulation tissue formation requiring repeat bronchoscopic treatment and stent replacement. Conclusions: Mediastinal repositioning with tissue expander placement provides durable relief of symptoms. Endobronchial stenting is a less invasive treatment option for PPS, however patients require close follow-up due to a high complication rate. Need for frequent bronchoscopy often emergently present serious limitations in this small group.

INTRODUCTION
Postpneumonectomy syndrome (PPS) is a rare complication occurring months to years after pneumonectomy. Lung and mediastinal contents herniate into the empty postpneumonectomy space causing displacement and rotation of mediastinal contents. The displaced distal trachea, primary bronchus or lower lobe bronchus is stretched and compressed between the pulmonary artery anteriorly and the aorta or spine posteriorly. This results in severe and progressive dyspnea and may lead to secondary malacia of the airway cartilages. Postpneumonectomy syndrome was initially described exclusively after right sided pneumonectomy, but subsequently cases following left pneumonectomy have been reported with and without a right aortic arch. The syndrome occurs in all age groups but is most common in children and young adults.

Numerous treatment modalities for PPS have been described over the years. Mediastinal repositioning with hopes that scar tissue formation would prevent relapse was associated with an unacceptably high recurrence rate. Aortic division and bypass graft is has been used with varied results. The lowest recurrence rates were achieved when mediastinal repositioning was augmented by placement of a prosthetic device in the post pneumonectomy space to prevent relapse. Although this is generally successful, mediastinal repositioning is a major operation that may not be tolerated by some patients. Further, obstructive symptoms may persist after mediastinal repositioning in cases of severe malacia of the airways. Alternative treatment has consisted of bronchoscopically placed endobronchial stents, however the long term outcome and complication rates following stent placement remains unknown. In an effort to examine the long term outcomes and complications associated with each technique, we present our experience treating postpneumonectomy syndrome with standard mediastinal repositioning-implant placement and endobronchial stent placement.

MATERIAL AND METHODS
Approval for this study was obtained from the hospital’s internal review board. All patients with the diagnosis of post-pneumonectomy syndrome were identified from the hospital’s medical record database. A retrospective chart
Review was performed based on information from the patient’s clinical chart including records that were obtained from outside institutions at the time of patient evaluations. Outcomes data is based on review of the medical record.

Standard workup for post-pneumonectomy syndrome included history and physical examination, laboratory studies, formal pulmonary function testing (PFT), chest radiograph, chest CT and flexible bronchoscopy. Patients met criteria for post-pneumonectomy syndrome based on the appropriate history along with objective evidence of disease. Patients were seen and evaluated by specialists in pulmonary medicine, thoracic surgery and plastic surgery. Final treatment was based on consensus recommendations. Surgery was performed using a multidisciplinary approach. Stents were placed in the operating room by the pulmonologists. Patients had follow up care at our institution and through correspondence in some cases with their local specialists.

Surgical correction was performed in collaboration with the thoracic surgeon, plastic surgeon and pulmonologist. Pre and post-operative bronchoscopy was obtained to verify the existence of airway narrowing and ensure post-operative resolution. (Figure 4a and 4b) The surgical procedure was performed as follow: the patient was placed in a lateral decubitus position, and the thoracotomy incision was re-opened. The prior pneumonectomy space was then entered and adhesions to the mediastinum were released. This allowed the lung and mediastinal structures to fall back into their normal location. After fixing the mediastinum in position with permanent suture, expandable breast prosthetics were placed in the post-pneumonectomy space. Fluid was added or removed from the prosthetics to allow complete filling without compromise of venous return to the heart. Prosthetic filler ports were then secured in a subcutaneous location for subsequent fluid adjustment as needed. These ports either remained in place indefinitely (3) or were removed in 6-9 months (3). The ribs were closed with interrupted pericostal Vicryl sutures. The muscles and skin were then closed in layers. A CT scan after the repair is shown in figure 3. (Figure 3)

One patient underwent a modified version of the above operation. After performing the thoracotomy, the intercostals muscles 1-9 on the side of the pneumonectomy were mobilized on their pedicles en bloc and reduced into the thoracic cavity to help fill the void and reduce expansion of the right hemithorax. The denuded rib cage was then stabilized in a lattice of stainless steal mediastinal wire. A single implant was then placed to fill the additional void. The remainder of the operation was performed in a similar manner.

Bronchoscopic stent placement was performed by our staff pulmonologist in a surgical suite under general anesthesia. Flexible bronchoscopy was used to examine the location of airway narrowing followed by placement of a silastic stent.
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using the rigid bronchoscope.

RESULTS
Thirteen cases of postpneumonectomy syndrome were identified and treated at our institution from 1991 to 2005. Two patients with mild symptoms elected conservative management. Two patients treated in the early 1990’s underwent mediastinal repositioning alone with good results. Mediastinal repositioning and implant placement was planned in 7 patients. The surgery was abandoned in 1 patient secondary dense adhesions making it difficult to free the right ventricle from the chest wall. Six patients underwent successful mediastinal repositioning and tissue expander placement at our institution. Two patients underwent stent placement.

Patients had right pneumonectomy (6) and left pneumonectomy (2) for the following conditions: squamous cell carcinoma (3), adenocystic carcinoma (1), pulmonary carcinoid (2) and pulmonary histoplasmosis (2). Male to Female ratio was 1:7. Average age at pneumonectomy was 44 ±12 years. Baseline characteristics and comorbid conditions are listed in table 1. Patients presented on average 19 ±23 months after pneumonectomy with symptoms of worsening shortness of breath, stridor, cough, dyspnea on exertion and frequent bronchitis. Most patients were seen numerous times in local emergency departments and primary care clinics before the diagnosis of post-pneumonectomy syndrome was made.

Figure 3
Table: 1. Patient demographics: (Abbreviations: DOE: dyspnea on exertion, Ca: cancer, COPD: chronic obstructive pulmonary disease, GERD: gastroesophageal reflux disease, L: left, R: right, SCC: squamous cell cancer, SOB: shortness of breath)

<table>
<thead>
<tr>
<th>Patient</th>
<th>Gender</th>
<th>Lesion</th>
<th>Treatment</th>
<th>side of pneumonectomy</th>
<th>side of symptoms</th>
<th>Preneumonectomy side right/left</th>
<th>symptoms</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>M</td>
<td>SCC</td>
<td>stent</td>
<td>R</td>
<td>L</td>
<td>SOB, DOE, O2, H1, H2</td>
<td>none</td>
</tr>
<tr>
<td>2</td>
<td>M</td>
<td>histoplasmosis</td>
<td>stent</td>
<td>L</td>
<td>L</td>
<td>dyspnea, O2, H1, H2</td>
<td>none</td>
</tr>
<tr>
<td>3</td>
<td>F</td>
<td>histoplasmosis</td>
<td>stent, implant failure</td>
<td>50, 12</td>
<td>R</td>
<td>O2, O4, H1, H2</td>
<td>none</td>
</tr>
<tr>
<td>4</td>
<td>F</td>
<td>carcinoid</td>
<td>implant</td>
<td>L</td>
<td>L</td>
<td>SOB, O2, H1, H2</td>
<td>none</td>
</tr>
<tr>
<td>5</td>
<td>F</td>
<td>carcinoid</td>
<td>implant</td>
<td>L</td>
<td>R</td>
<td>SOB, O2, H1, H2</td>
<td>none</td>
</tr>
<tr>
<td>6</td>
<td>F</td>
<td>squamous cell</td>
<td>implant</td>
<td>L</td>
<td>L</td>
<td>SOB, O2, H1, H2</td>
<td>none</td>
</tr>
<tr>
<td>7</td>
<td>F</td>
<td>squamous cell</td>
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<td>L</td>
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<td>SOB, O2, H1, H2</td>
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<tr>
<td>Average</td>
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<td></td>
<td></td>
<td>R</td>
<td>L</td>
<td>SOB, O2, H1, H2</td>
<td>none</td>
</tr>
</tbody>
</table>

Standard workup for post pneumonectomy airway obstruction was performed on all patients. After a thorough history and physical exam, chest radiographs were obtained demonstrating tracheal deviation and mid-line shift with hyperinflation of the remaining lung. CT scan of the chest revealed marked hyperinflation of the lung with herniation of mid-line structures into the post-pneumonectomy space. There was a posterolateral rotation of the mediastinum in the direction of the empty hemithorax. In cases after right pneumonectomy the left mainstem bronchus was narrowed as it passed posterior to the left pulmonary artery and anterior to the spine or aorta. (Figure 1) In the cases following right pneumonectomy the right mainstem bronchus and right bronchus intermedius was compressed between the right pulmonary artery and vertebral body. (Figure 2) Pulmonary function testing revealed a dynamic obstructive picture with decreased forced expiratory volume in 1 second (FEV1). The ratio of FEV1 to forced vital capacity was markedly reduced. Average pre operative FEV1 was 41.3 with an average FEV1/FVC of 67.3. Pre-operative bronchoscopy revealed severe extrinsic compression as noted by flattening and fishmouth narrowing of the bronchus. Post-operative bronchoscopy revealed resolution of the extrinsic compression with a circular shape to the bronchus. (Figure 4a and 4b) Average operative times were 4 hours 36 minutes (range 2:28-6:55)

Figure 4
Figure 1: CT scan of post-pneumonectomy syndrome following right pneumonectomy. Note, left mainstem bronchus compressed between the pulmonary artery (anterior) and the aorta and spine (posterior).
Results from surgical mediastinal repositioning and implant placement into the post-pneumonectomy space were excellent at our institution. All 6 patients had short and long term improvement in dyspnea, shortness of breath and stridor. Minor complications occurred in 2 of 6 patients at 30 days and 3 of 6 patients after 30 days. Clinically insignificant partial implant failure occurred in 2 patients. There were no major complications. One patient died of metastatic disease during the follow up period at 17 months post-operatively. 30 day complications included superficial cellulitis and atrial fibrillation. Long term complications included partial implant deflation in 2 patients 1 of which had a sub-clinical partial herniation of an implant. One patient experienced difficulty swallowing 2 years following surgery. After a thorough work-up including CT chest, esophagoscopy, barium swallow and esophageal manometry the patient was given a diagnosis of primary esophageal dysmotility which was treated conservatively (Table 2).

The 2 patients who underwent stent placement are a heterogeneous group. One patient (patient #1) was treated with primary silastic stent placement. Surgical correction was discussed with the patient but not performed secondary to the patient’s poor prognosis and functional status. She underwent flexible bronchoscopy which demonstrated an obvious obstruction from the posterior wall of the right bronchus intermedius that nearly completely obliterated the airway. Distally there was compression of the middle and lower lobe bronchi from the anterior wall of the spine. The rigid bronchoscope was then used to dilate the lower lobe area. A 10 x 20 mm silastic stent was placed in the bronchus intermedius. Peak and mean airway pressures measured before and after stent placement: 28, 13 and 20, 10 mm Hg respectively. Operative time was 1 hour 12 minutes.

The patient had instant resolution of airway obstruction. Long term results were not as impressive. Over the next 3
months the patient experienced recurrent wheezing, retained secretions, cough and shortness of breath. This prompted repeat bronchoscopy 4 times for stent replacement (1), stent displacement (1), retained secretions and mucous plugging (2). She was ultimately placed in hospice and succumbed to respiratory distress secondary to severe COPD 3 months following stent placement.

The second patient (patient #2) presented to our institution after treatment elsewhere for post-pneumonectomy syndrome following left pneumonectomy. She had undergone thoracotomy, mediastinal repositioning and saline implant placement. At 3 months post-operatively her symptoms of dyspnea, inability to clear secretions and stridor returned. Repeat workup revealed normal airway patency with relaxed breathing, however near complete airway collapse with mild forced exhalation or cough. A diagnosis of bronchomalacia was made and a 10 x 2 mm Wallstent (Schneider Inc, Minneapolis, MN) was placed into the right mainstem bronchus. The patient returned 2 years later with recurrent shortness of breath. Granulation tissue growth was found around the metallic stent. This was cryo-ablated several times unsuccessfully over the next 7 months. At this point, she was referred to our institution.

The stent was painstakingly removed wire by wire with the use of rigid bronchoscopy until the entire stent was removed. Extensive granulation tissue throughout the bronchial tree was removed with forceps. The airway was diluted and a tapered 30 x 12 mm silicone stent was placed across the right mainstem bronchus with fenestration open to the right upper lobe. (Figure 5a, Figure 5b) Operative time was three hours.

Her symptoms resolved immediately following the procedure. However over the next 28 months, the patient returned 16 times with airway symptoms prompting repeat bronchoscopic procedures. Near complete occlusion of the distal stent or fenestration to the right upper lobe from granulation tissue was noted on multiple occasions. Treatment modalities included: removal of granulation tissue with forceps, Fogarty balloon dilation, application of...
There is little experience in using primary stent placement for post-pneumonectomy syndrome. Several case reports are limited by lack of follow-up. This may be due to a selection bias toward those with a short life expectancy or those not fit for surgery. A clear benefit to stent placement is the immediate relief of symptoms without the need for extensive recovery and risk of mortality. Silicone stents have the benefits of ease of customization, repositioning and removal. Self-expanding metallic stents have the advantage of ease of insertion, conformation to the airways, stent stability and a low profile in the airways. These advantages may be offset by the development of problematic granulation tissue and difficulty in removal once the stent is seated as was observed in our patient. Our bias is toward surgical correction with implant placement for post-pneumonectomy syndrome except in select cases where there is a short life-expectancy or poor functional status. In these situations stent placement can offer palliation of symptoms. We prefer the use of silastic stents because they can be easily removed and manipulated. More effective stents are needed before widespread use of stent should be considered for treatment of post-pneumonectomy syndrome.

**SUMMARY**

Post-pneumonectomy syndrome is a rare complication following pneumonectomy. It is most reliably treated by redo thoracotomy reduction of mediastinal structures and implant placement in the post pneumonectomy space. Primary stent placement may be considered in cases where there is need for short term palliation. The current role of stent placement in the treatment of patients with severely malacic airways is not well understood. Frequent complications may limit their role until improved stents can be developed.

**References**

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