Right Native Lung Pneumonectomy due to over Inflation Three Years after Left Single Lung Transplantation for Pulmonary Lymphangioleiomyomatosis

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Native lung hyperinflation (NLH) is one of the known complications after single lung transplantation (SLT). Generally, satisfactory results are achieved in patients undergoing SLT when simultaneous (or second stage) volume reduction of the contralateral native lung is performed. Contralateral native lung pneumonectomy after SLT is rarely reported. In this article, we report a case of a successful, right pneumonectomy of the native lung, 3 years after a left single lung transplant for pulmonary lymphangioleiomyomatosis (PLAM). The patient’s pulmonary function and quality of life improved significantly after a right pneumonectomy of the native lung.

Keywords: lung transplantation, native lung, pneumonectomy, pulmonary lymphangioleiomyomatosis

Introduction

Lung transplantation is an established treatment for patients with advanced emphysema. NLH is one of the known complications after SLT. After a patient with pulmonary lymphangioleiomyomatosis receives SLT, primary disease of the native lung continues to progress and deteriorate. Damaged alveoli gradually combine and lead to the development of cystic lesions and honeycomb changes. The hyperinflated native lung suffers with respect to the function of gas exchange. In addition, the hyperinflated native lung pushes the mediastinum into the contralateral chest cavity and compresses the transplanted lung, which leads to further deterioration and loss of transplanted lung function.

Any surgical intervention in lung transplant patients was previously thought to carry an excessively high risk due to concerns regarding impaired wound healing and immunosuppressive regimes. However, many centers have published their experience of post-lung transplant pulmonary resections and demonstrated the feasibility of surgery within this group. Indeed, lung volume reduction within this patient group has surgical risk, and when comes to pneumonectomy, it is associated with higher morbidity and mortality. In view of this, patient selection and preoperative work-up are vital in improving postoperative outcomes. Hence, our objective was to report our institutional experience and to analyze the indications, surgical procedures as well as outcomes of patient who underwent native lung pneumonectomy after SLT.

Case Report

A 56-year-old woman underwent a left SLT on
September 27, 2005 for severely impaired pulmonary function due to PLAM. Postoperatively, she received “triple antirejection therapy” including: tacrolimus, mycophenolate, and prednisone. The complications of tracheomalacia and anastomotic stoma stenosis occurred in the patient, 3 months after the lung transplantation. These complications were treated with a tracheal stent. Subsequently, lung function improved significantly: FEV$_1$ of 1300 ml and a 6-minute “walking test” up to 500 meters. The patient had a favorable postoperative course for 3 years, no signs of transplant rejection, no severe infection, and the ability to participate in social activities. After 3 years from the operation, the patient began complaining of dyspnea aggravated by exertion. Pulmonary function worsened with a significant decline in FEV$_1$ to 600 ml and the 6-minute “walking test” distance dropped to 50 meters. Chest high resolution computerized tomography (HRCT) scan showed diffuse, thin-walled, cystic lesions in the right lung. The lesions varied in size, some of which coalesced into bullous changes accompanied by significant lung herniation. The mediastinum was shifted to the left side, which resulted in severe compression of the transplanted lung (Fig. 1). Bronchoscopy has ruled out alternative causes for deterioration in pulmonary function test. A ventilation/perfusion scan demonstrated that the right native lung ventilation was difficult to discern and radioactive aggregation could only be seen in the main bronchus. The contour of the left transplanted lung was completely visualized, and the radioactive material was evenly distributed. The assessment convinced us that the transplanted lung function can tolerate the right lung operation including pneumonectomy.

In accordance with the conventional regimen, our initial surgical plan was to perform an anatomical bilobectomy (right middle lung and lower lung lobectomy) in June 2009. The patient received general anesthesia. Double-lumen endotracheal intubation was performed and the position was confirmed by direct visualization via bronchoscopy. The bilateral lungs were alternatively ventilated. A CO$_2$ probe determined that the right trachea end-tidal CO$_2$ (ETCO$_2$) was 0, while the left ETCO$_2$ was 28 (which proved that there was no gas exchange occurring in the right, native lung). Blood pressure, heart rate, and oxygen saturation were normal after the occlusion of the right lumen of endotracheal intubation. Therefore, the transplanted lung function was believed to be able to tolerate the right lung operation. Various sized vesicles and bullae with diffuse cystic lesions could be seen on the right lung upon entering the right chest cavity (Fig. 2). The tension of the vesicles and bullae in the right lung could be easily palpated. It was observed that the right lung would collapse (with fissure dysplasia) during ventilation of the left lung. The operation was approached by first ligating the lower pulmonary vein. Next, we dealt with the pulmonary fissure by applying a linear cutter (TLC75, Ethicon Endo-Surgery, Inc., Somerville, New Jersey, USA) and absorbable polyglycolic acid mesh (Neoveil, Gunze Limited, Osaka, Japan). The arterial branches to the middle and lower lobes of the right lung

**Fig. 1** Preoperative chest computed tomography (CT) scan: diffuse cystic lesions in the right, native lung, lung herniation, the left transplanted lung with obvious compression.

**Fig. 2** Diffuse cystic lesions in right native lung.
were ligated and dissected in sequence. Finally, the intermediate bronchi were clamped with the aid of a linear stapler (TLH-30, Ethicon Endo-Surgery, Inc., USA). The thoracic cavity was irrigated with distilled water after the middle and lower lobes were removed. The tracheal stump showed no signs of an air leak during a pressurized ventilation test. However, a large volume of air escaped from the fissure and the lung surface through the pores made by the needles and nails. Attempts were made to seal the air leak with bio-glue, chemical gel, and absorbable, hemostatic gauze. However, these measures were ineffective. Therefore, the pneumonectomy of the native lung was performed at last.

The patient was discharged 3 weeks after the surgery. Postoperative recovery of the patient was uneventful and lung function improved significantly. FEV1 increased to 900 ml and the 6-minute “walking test” increased to 300 meters. Chest HRCT showed that the mediastinum had nearly returned to its original position and the transplanted lung showed good expansion (Fig. 3). A lung ventilation/perfusion scan demonstrated that the contours of the left transplanted lung had expanded and radioactivity showed an even distribution. The lung ventilation/perfusion ratio showed coordination. There was no radioactive aggregation in the area of right thoracic cavity. The patient has been closely followed for nearly 4 years (almost 8 years since the single left lung transplantation). The patient remained in stable, was able to manage her activities of daily living, and received regularly scheduled pulmonary function tests and chest CTs.

Discussion

NLH is one of the common complications after SLT. Lung volume reduction for NLH is an effective strategy with acceptable mortality and chance for success. Contralateral, native lung pneumonectomy after SLT is rarely reported. In fact, native lung pneumonectomy is an option and has a median survival time similar to patients who receive SLT without any complications in the native lung.\(^3,4\)

Lung volume reduction or native lung pneumonectomy within this patient group is not without risk. The most common cause of morbidity and mortality in this patient group is bronchiolitis obliterans (BO) and infective complications. In view of this, the preoperative evaluation such as chest HRCT and bronchoscopy is vital to exclude those with deterioration in lung function caused by some other pathology. Chest HRCT must show atelectasis of the transplanted lung. If there is NLH alone, then the patient cannot gain significant improvement following Lung volume reduction. In addition, ventilation/perfusion scans should be performed in all cases to assess the contribution of each lung. The native lung must have a minimal contribution to the overall function, so it is possible to remove parts of the native lung with little function in surgery.

The decision was made after careful consideration by the transplant team. Considering native lung pneumonectomy has a higher surgical risk, generally, if the patient had relatively normal native lung parenchyma, lung volume reduction would have been undertaken. When the severity of the underlying bullous disease of native lung prevented the use of standard lung volume reduction procedure, a native lung pneumonectomy should be considered to avoid the problems with pulmonary healing or prolonged airleak.

As another option, lung retransplantation can be performed after native lung pneumonectomy.\(^5\) If the patient is with severe graft dysfunction while there is no other therapy that can prolong life, lung retransplantation is the only treatment option. However, retransplantation has the potential to add increased, surgical risks. Taking into account the serious risk of surgery and limited donor organ availability, we advocate that if the patients are

Fig. 3 Postoperative chest computed tomography (CT) scan: a well expanded, transplanted lung and a high density shadow indicating the tracheal stent.
without severe graft dysfunction, the retransplantation should be performed with caution.

In this case, the success of the operation depended on proper assessment of the function of the transplanted and native lung and correct recognition of the underlying disease process. A preoperative lung ventilation/perfusion scan and an intra-operative end-tidal CO₂ test are helpful in the assessment of the function and status of both the transplanted and native lung. The successful reoperation of this patient indicates that native lung pneumonectomy after SLT can be performed successfully when approached with sound clinical reasoning.

Disclosure Statement

The authors declared that they had no conflict of interest.

References


