Chylopericardium following orthotopic lung transplantation

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Chylopericardium is an uncommon condition, reported to occur following routine cardiac surgery, orthotopic heart transplantation, cardiac trauma, intrathoracic tumors, or infection. It has not, to date, been reported following uncomplicated orthotopic lung transplantation. This article describes chylopericardium following bilateral orthotopic lung transplantation.

Chylopericardium was first described by Hasebrock in 1888 (1). In 1954, the term “primary chylopericardium” was offered by Groves and Effler for idiopathic cases without an apparent cause (2). Chylopericardium has been reported also as a consequence of various diseases and as a complication of surgical procedures (3). It has been reported rarely following cardiac transplantation (4), but has never been reported as a complication of lung transplantation. This report is the first to describe chylopericardium following lung transplantation.

CASE REPORT

A 54-year-old woman, who had asthma as a child and smoked cigarettes as an adult, suffered from chronic obstructive pulmonary disease complicated by pneumothorax requiring tube thoracostomy in the right hemithorax at age 39 (1997). She did not have alpha-1-antitrypsin deficiency or lymphangioleiomyomatosis, and her chronic obstructive pulmonary disease was treated with inhaled beta-agonist bronchodilators, steroids, and acetylcholine receptor antagonists. She had blood type A. She weighed 142 pounds and was 64 inches tall. She had impaired bronchspirometry with a forced expiratory volume in 1 second of 20% predicted, diffusion of carbon monoxide of 14% predicted, and BODE score (body mass index, airflow obstruction, dyspnea, and exercise capacity) of 9. Her right lung perfusion was 54% and left lung perfusion, 46%. Right heart catheterization demonstrated normal pulmonary arterial pressures.

In May 2012, she underwent an uncomplicated bilateral sequential lung transplantation through a thoracosternotomy based off of the fourth intercostal space. Cardiopulmonary bypass was not required. Her immediate postoperative intensive care unit course was uneventful. Immunosuppression induction therapy and postoperative immunosuppression maintenance

were accomplished with corticosteroids, azathioprine, and tacrolimus. She was discharged 2 weeks later.

Her early posttransplant course was complicated by atrial fibrillation treated with flecainide and with hyponatremia thought to be secondary to a syndrome of inappropriate antidiuretic hormone, which responded to free water restriction. She also experienced anemia (hemoglobin 6.5 g/100 mL), with a high serum ferritin level indicative of an iron reutilization defect. Her lung function following transplantation was excellent, with a peak postoperative forced vital capacity of 3.26 L.

Beginning 4 months following her transplant, routine imaging studies demonstrated an asymptomatic increase in the size of her pericardial silhouette (*Figure 1*). Transthoracic echocardiogram demonstrated a large effusion with mild right atrial compression. In October 2012, she underwent a subxyphoid pericardial exploration and drainage, with evacuation of 650 mL of opaque, lactescent sanguinous fluid (*Figure 2*) with a triglyceride level of 2139 mg/dL and leukocyte count of 619/ μ L, with a differential count of 80% lymphocytes. Viral, mycobacterial, and fungal cultures, as well as Gram stain and acid-fast stains, were negative.

Postoperatively she was maintained on a nonfat diet and then transitioned to a low-fat diet. Three days after operation, her chest tube output remained low and serous in nature, despite escalation of her dietary fat intake. She was discharged without recurrence of her effusion and is convalescing well on an unrestricted diet with no evidence of recurrent chylopericardium on evaluation with a chest radiograph and echocardiogram 1 month later.

DISCUSSION

Chylous effusions in the pericardial space can arise spontaneously, following penetrating or blunt chest trauma, or following surgical procedures (2, 3). Known specific causes include

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Figure 1. The patient's chest x-ray (a) immediately after transplant and (b) 4 months after transplant.

congenital mediastinal lymphangiectasia (cystic hygroma) (5), gastric carcinoma (6, 7), lymphangiomatosis of the chest/Gorham syndrome (8, 9), primary mediastinal neoplasms such as lymphoma and germ cell tumor (6, 10), mediastinal hamartoma (8), infection with tuberculosis (11), deep vein thrombosis with superior vena cava syndrome (12), pancreatitis (13), allergic alveolitis (14), and Behçet's disease (15). A review in 1935 identified only three cases of chylopericardium (12), and a more comprehensive review in 2006 identified 33 reported cases, the most common etiology being idiopathic (3). Postoperative causes of chylopericardium reported in the literature include mitral valve replacement (4, 16, 17), coronary artery bypass grafting with left internal thoracic artery harvesting (18), congenital heart surgery (19), and orthotopic heart transplant (4, 20).

Figure 2. Subxyphoid drainage fluid.

Although chylothorax is a commonly reported complication of orthotopic lung transplantation (21) and a rare complication of combined heart/lung transplantation (22), a review utilizing Ovid, MEDLINE, PubMed, and the Cochrane Library databases failed to isolate a single report of chylopericardium following isolated orthotopic lung transplantation. Accordingly, the current case report represents the first

time chylopericardium has been reported following orthotopic lung transplantation.

The exact mechanism of chylopericardium in this case is unknown, but the postulated mechanism is that lymphatic leak from the subcarinal, peribronchial lymph nodes and lymphatic channels (which are dissected to allow for bronchial anastomosis) creates efflux of lymph into the posterior mediastinum. The posterior parietal pericardium is routinely widely opened during orthotopic lung transplantation in an effort to afford intrapericardial mobilization of the pulmonary veins, such that a direct left atrial cuff-to-left atrial cuff anastomosis between donor and recipient can be performed, instead

of a direct individual pulmonary vein anastomosis. This area of posterior parietal pericardial dissection around the paired pulmonary veins could allow a path of ingress of posterior mediastinal, peribronchial lymph to the pericardial space; absorbed oral fat in the form of chylomicrons could then enter into the pericardial space in this fashion.

The treatment of chylopericardium should be disease specific, with the goals of decreasing chyle production, relieving or treating tamponade (16, 23, 24), or preventing secondary constrictive pericarditis (10, 25); treatment is also tailored to mitigate against the deleterious consequences of lymph depletion in regards to nutritional status, fluid and electrolyte depletion, and immunologic side effects of chronic lymphocyte reduction (25). For idiopathic chylopericardium, successful treatment has most often been reported with simple drainage, either by pericardiocentesis or surgical transmediastinal drainage (19). Dietary modifications to a nonfat or medium-chain triglyceride diet have been reported as useful; refractory cases have required total parenteral nutrition and occasionally the addition of octreotide to suppress chylomicron production. Idiopathic chylopericardium refractory to drainage and dietary modifications and postsurgical or posttraumatic chylopericardium most often require either pericardial drainage or creation of a pleuropericardial window, with or without thoracic duct ligation. Malignancy-related chylopericardium, reported after signet-cell gastric cancer (7), may require the addition of chemotherapy and radiation. Surgical pericardial drainage can be transmediastinal (through the subxyphoid space of Larrey, as occurred in this case) or transthoracic via thoracostomy or video-assisted thoracoscopic surgery, especially if creation of a pleuropericardial window is necessary (3, 4, 16, 18, 19, 24). Creation of a pleuropericardial window following lung transplantation, in the absence of thoracic duct ligation, may not represent an optimal approach, as there remains the possibility that the chylopericardium will only be replaced with a refractory chylothorax.

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