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Successful Treatment With Lymphovenous Anastomosis for Lower Extremity Edema Secondary to Lipiodol Lymphangiography

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Abstract

Background: Chylothorax is one of the complications after thoracic surgery and treated by conservative or surgical means. Lipiodol lymphangiography is one of the options and it causes obliteration of chylous leak by inflammatory manner. In this article, we describe a case of lymphedema of the bilateral lower extremities occurs after lipiodol lymphangiography and it is treated successfully by lymphovenous anastomosis.

Case presentation: A 67-year-old man presented with refractory chylothorax after subtotal esophagectomy and thoracic lymph node dissection. His chylothorax developed 4-month later of subtotal esophagectomy and was refractory to the conservative treatment (i.e. tube thoracostomy). He was referred to our department to treat chylothorax. We chose lipiodol lymphangiography as the treatment. Lymphatic duct of left foot was detected with indocyanine green and exposed to inject lipiodol into lymphatic duct directly. Chylothorax improved immediately after lipiodol lymphangiography and his edema of right lower extremity emerged 22-month later of lipiodol lymphangiography. We considered that his lower extremity edema was caused by lipiodol lymphangiography and performed lymphovenous anastomosis. Lymphovenous anastomosis was performed at the proximal of right thigh and the dorsum of the foot. At six-month later of lymphovenous anastomosis, we revealed that his right lower extremity had become thinner significantly, nevertheless laterality remained.

Conclusion: To our best knowledge, this is the first report of lymphedema of the bilateral lower extremities after lipiodol lymphangiography for chylothorax. Lymphovenous anastomosis is a treatment option for such condition.

Keywords: Chylothorax; Lipiodol; Lymphangiography; Lymphovenous anastomosis

Introduction

Chylothorax is a rare complication of thoracic surgery. Esophagectomy has the highest incidence of postoperative chylothorax among thoracic surgical procedure (1% to 9%) [1,2]. Typical symptoms of chylothorax includes dyspnea, chest pain, cough and fatigue. Long-term complication is lymphopenia which put the patient into immunosuppressive status [1].

Management of chylothorax is conservative and surgical [1]. Conservative management is to reduce chylous leakage. To reduce chyle production, long-chain triglycerides free diet and nil per os regimen are effective. Tube thoracostomy and repeated thoracentesis can keep the lung expanded. Octreotide and somatostatin can be effective. When chylothorax is refractory to conservative treatment, surgical means (e.g., thoracic duct ligation and pleurodesis) are considered.

Lymphangiography is typically required to identify the location of leakage for surgical treatment [1,3,4]. When lipiodol is chosen as the agent for lymphangiography, that can act as curative method as well. Lipiodol causes obliteration of chylous leakage by inflammatory manner. According to O'Brien [5], lipiodol lymphangiography exaggerates edema in one-third of patients of obstructive lymphedema. Lymphovenous anastomosis is surgical gold standard to treat secondary lymphedema. In this article, we describe a case of successful treatment with lymphangiography for right lower extremity which occurred after lipiodol lymphangiography.

Case Report

A 67-year-old man was suffered from refractory bilateral chylothorax after subtotal esophagectomy and thoracic lymph node dissection. His chylothorax developed 4-month later of subtotal esophagectomy. Despite of tube thoracostomy, chylous leakage continued with the average of 620 mL/day. Ten weeks after his chylothorax emerged, he was referred to our department to treat chylothorax. CT showed massive pleural effusion (Figure 1).

We chose lipiodol lymphangiography as the treatment. Indocyanine green was injected subcutaneously at the first web space of the left foot to detect lymphatic duct. Lymphatic duct was exposed and 4 mL of lipiodol was injected directly into the lymphatic duct with 30 G needle. Lymphatic duct in calf, kenn, groin, L4 and L2 was enhanced 14, 24, 63, 240 and 420 minutes later of lipiodol injection, respectively (Figure 2). Lymphatic duct above L2 was seldom enhanced and lymphatic duct injury at the site of lymph node dissection was suspected. No apparent finding indicating lymphatic duct injury (i.e. lipiodol leakage and pooling) was detected.

Chylothorax improved right after lipiodol lymphangiography. Chylous leakage decreased to 20 mL/day until postoperative day 16, and at the postoperative day 20, he was discharged without thoracic drain tube. CT showed dcreased pleural effusion (Figure 1). He remained free of chylothorax and his right lower extremity edema emerged 22-month later of lipiodol lymphangiography. His symptoms developed gradually and he was re-referred to our department (Figure 3). Physical examination showed right lower extremity edema and limited range of motion of knee. Echography did not reveal any deep vein thrombosis.

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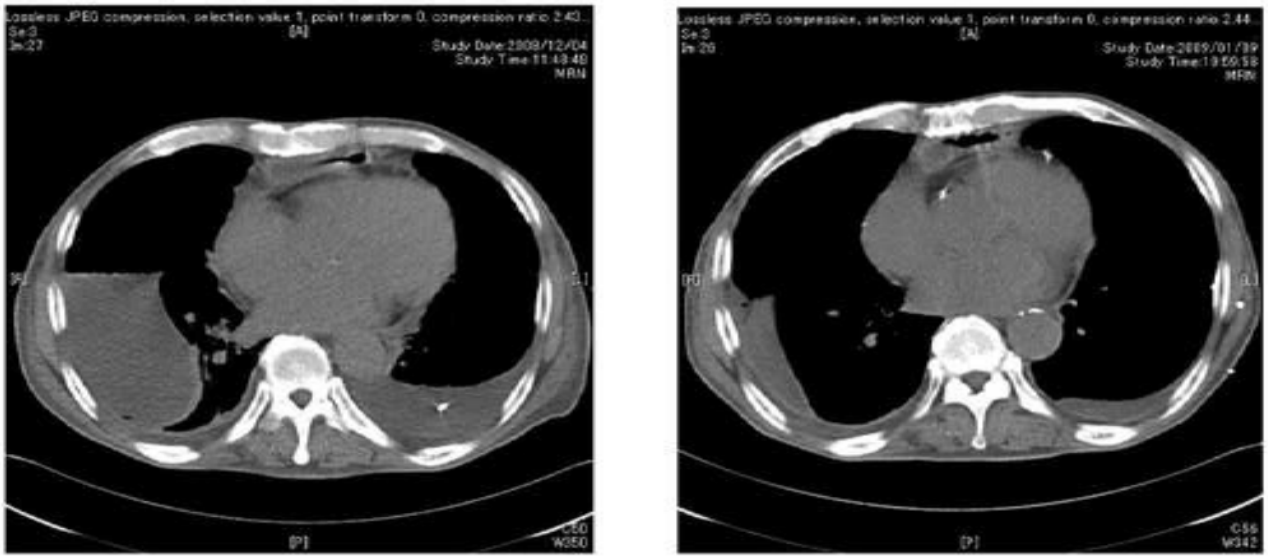

Figure 1: Computed tomography of thorax; (Left) before lipiodol lymphangiography, bilateral chylothorax are revealed; (Right) after lipiodol lymphangiography, chylothorax improved.

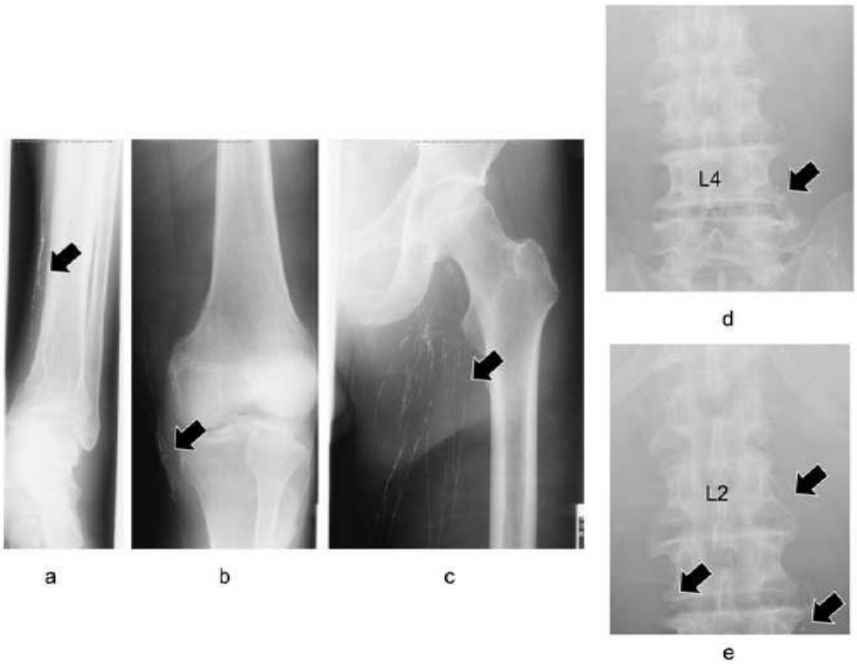

Figure 2: Radiography of lipiodol lymphangiography, black arrows, enhanced lymphatic ducts; (a) calf; (b) knee: (c) groin; (d) L4; (e) L2.

According to the history of lipiodol lymphangiography and physical examination, we considered that his lower extremity edema was caused by lipiodol lymphangiography and decided to perform lymphovenous anastomosis. End-to-end lymphovenous anastomosis was performed at the proximal of right thigh and the dorsum of the foot and the diameter of lymphatic duct was 0.8 mm and 0.6 mm, respectively (Figure 4). At six-month later of lymphovenous anastomosis, we revealed that his right lower extremity edema improved significantly, nevertheless laterality remained (Figure 3).

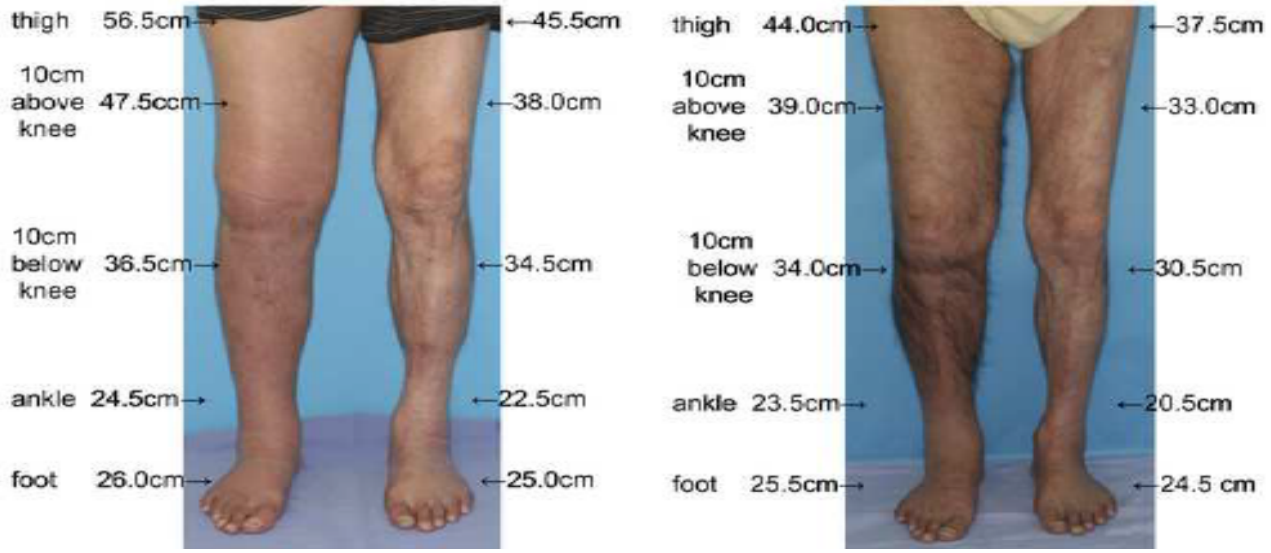

Figure 3: Circumference at each site; (Left) pre-operative; (Right) Post-operative month of 6.

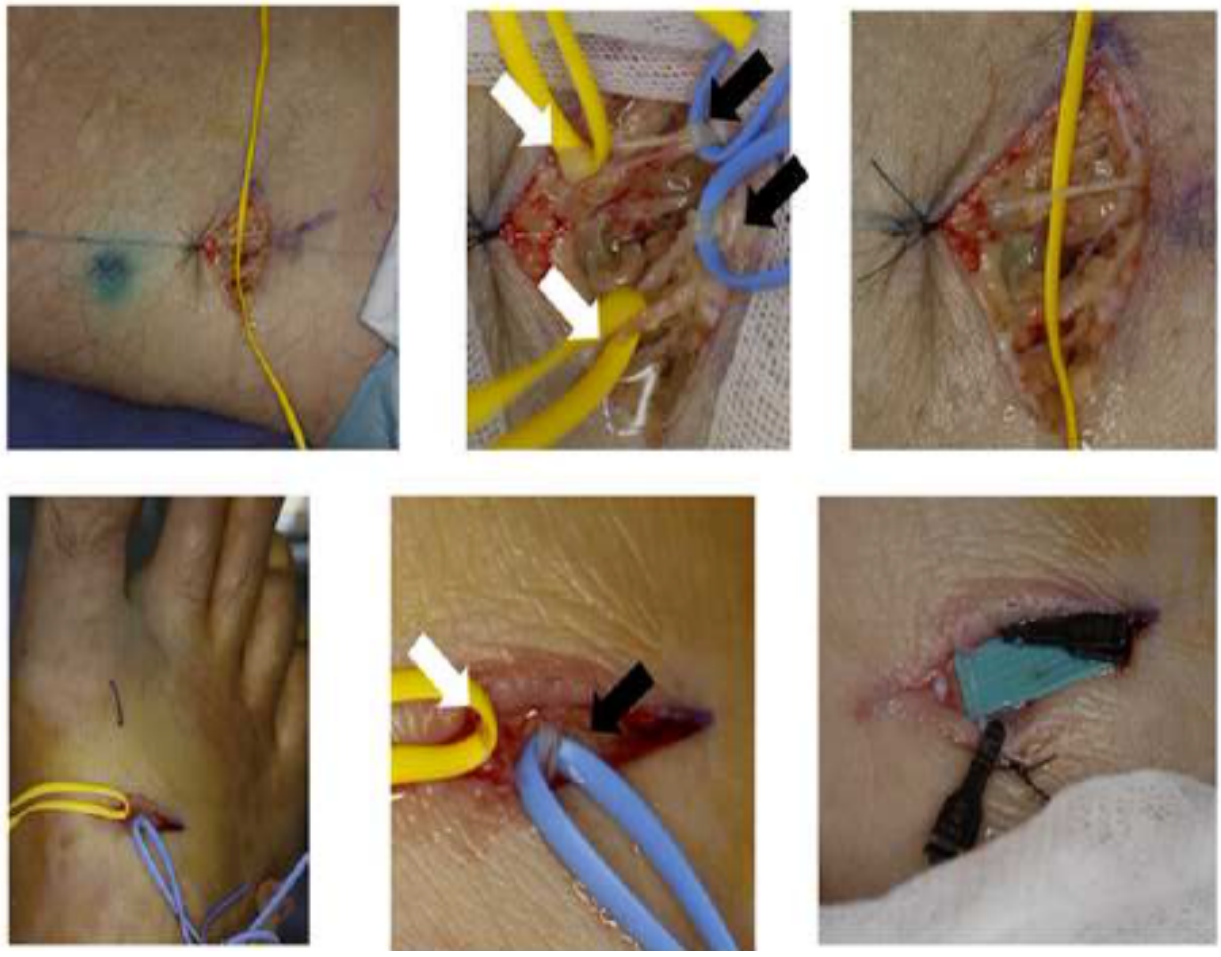

Figure 4: Lymphovenous anastomosis, white arrows, lymphatic ducts, black arrows, veins; (above left) surgical site at proximal thigh; (above middle) before anastomosis; (above right) after anastomosis; (below left) surgical site at dorsum of foot; (below middle) before anastomosis; (below right) after anastomosis.

Discussion

Chylothorax is a complications of esophagectomy and causes decompression of the lungs and heart. Conservative treatment (i.e. total parenteral nutrition and long-chain triglycerides free diet) is the first line treatment [1]. Thoracentesis and tube thoracostomy are effective to relief symptoms [1]. These types of treatment were instituted to our patient and his chylothorax was refractory to conservative treatment. It is reported that lipiodol lymphangiography is diagnostic and curative method for chylothorax [3,4]. To reduce patient's physical burden, we chose lipiodol lymphangiography as a treatment. His chylothorax improved right after this treatment nevertheless his right lower extremity edema developed 22-month later. Lipiodol induces the inflammatory process and occlude chylous leakage site by adhesion [3]. In one-third of patients with obstructive edema, their edema can be worsened by lipiodol lymphangiography [5]. We hypothesized that his right lower extremity edema was caused by lymphatic duct obstruction because of lipiodol. Lymphovenous anastomosis is a gold standard surgical treatment for secondary lymphedema [6,7]. This surgery is to create a bypass between lymphatic and venous system by anastomose lymphatic duct and vein and mainly performed on upper and lower extremities of patients with lymphedema after lymph node dissection for breast and gynecologic cancer, respectively. In our patient, lymphedema caused by lipiodol lymphangiography improved by lymphovenous anastomosis. This indicates that lipiodol lymphangiography may cause obstructive lymphedema and lymphovenous anastomosis can be effective in patients with this complication.

Conclusion

To our best knowledge, this is the first case report which shows successful lymphovenous anastomosis for lymphedema secondary to lipiodol lymphangiography instituted to treat chylothorax. Our case report indicates that lymphovenous anastomosis is one of treatment options for lymphedema secondary to lipiodol lymphangiography.

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