







Direct Percutaneous Needle Puncture and Intrapulmonary Lymphatic Embolization for Treatment of Chylothorax in a Patient with Lymphoma

림프종 환자의 비외상성 유미흉에서 폐림프종의 피부경유 직접천자를 통한 색전술

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Lymphoma is a common cause of nontraumatic chylothorax. Clinical success rates of thoracic duct embolization are lower in patients with nontraumatic chylothorax compared to patients with traumatic chylothorax. Herein, we report a case of nontraumatic chylothorax and lymphoma in a 77-year-old man managed with thoracic duct embolization. The chest tube drainage decreased but not was sufficient to enable removal of the chest tube. Therefore, a second embolization was performed through a direct puncture of the lymphatic mass in the lung, following which the chyle leakage ceased, and the chest tube was removed. The treatment strategy discussed in this report may be an effective therapeutic option for select patients with nontraumatic chylothorax.

Index terms Chylothorax; Embolization, Therapeutic; Lymphatic System; Lymphoma

INTRODUCTION

Thoracic duct embolization (TDE) is a prominent alternative and a first line therapy for patients with chylothorax. However, nontraumatic chylothorax is a relatively rare con-

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dition with a lower clinical success rate than that of traumatic chylothorax. A case of non-traumatic chylothorax in a patient with lymphoma is presented, in which intrapulmonary mass embolization was successfully performed.

CASE REPORT

A 77-year-old man patient undergoing chemotherapy for mantle cell lymphoma complained of dyspnea. A computed tomography (CT) scan of the chest showed pleural effusion in the both hemithoraces and a mass in the left lower lobe of the lung that appeared to be lymphomatous involvement (Fig. 1A). Following chest tube insertion, 1000–1200 cc of milky effusion was drained per day. Pleural fluid cytology showed an enrichment of lymphocytes. A biochemical analysis of the fluid revealed a high triglyceride content (206 mg/dL), which was diagnostic of chylothorax. The patient underwent a medium-chain triglyceride diet af-

Fig. 1. Embolization of intrapulmonary lymphoma for the treatment of continuous chylothorax after TDE.

A. Axial contrast-enhanced computed tomography demonstrates pleural effusion in both the hemithoraces and a mass in the left lower lung lobe consistent with lymphomatous involvement.

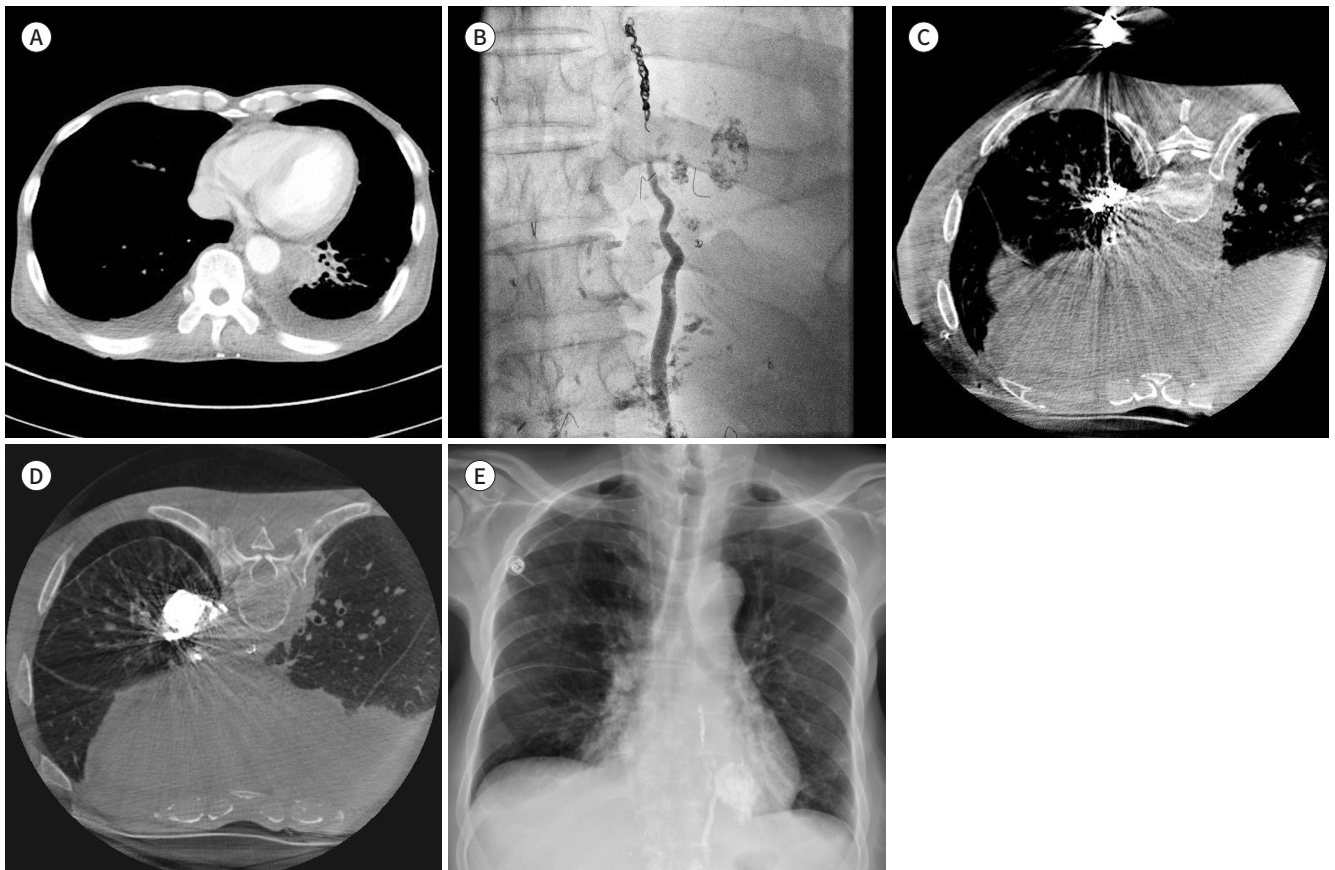
B. Fluoroscopy after TDE demonstrates multiple microcoils and an NBCA-lipiodol cast in the thoracic duct.

C. Axial cone-beam computed tomography demonstrates a spinal needle placed in the intrapulmonary mass.

D. Cone-beam computed tomography after TDE demonstrates proper injection of the NBCA-lipiodol cast in the mass and intrapulmonary lymphatics. A small pneumothorax developed during the procedure, but the patient was asymptomatic.

E. Follow-up chest radiography demonstrates complete resolution of pleural effusion and successful removal of the chest tube.

NBCA = N-butyl cyanoacrylate, TDE = thoracic duct embolization



ter 2 weeks of nil per os diet as conservative management. However the chest tube drainage volume persisted above 800 cc/day.

Consequently, the patient underwent intranodal lipiodol lymphangiography (Lipiodol Ultrafluid, Laboratoire Guerbet, Paris, France) for TDE. TDE was performed by a direct puncture of the cisterna chyli with a 21-gauge Chiba needle (Cook, Bloomington, IN, USA) through a transabdominal route. Digital subtraction angiography was performed to find chyle leakage sites in the microcatheter inserted into the thoracic duct. Lymphatic leakage was suspected in the lymphatic duct around the left lower hemithorax. The leakage site on the fluoroscopy correlated with the location of the lymphatic mass in the left lower lobe. Coil embolization was performed using Concerto microcoils (Medtronic, Jacksonville, FL, USA) in the proximal portion of the thoracic duct above the leakage site. The leakage site of the thoracic duct was embolized using a mixture of N-butyl cyanoacrylate (NBCA; Histoacryl, B Brown, Melsungen, Germany) plus lipiodol in a ratio of 1:1 (Fig. 1B). Although the chest tube drainage volume decreased to 200 cc/day on the 5th day after TDE, this was not adequate to indicate removal of the chest tube. The cause for residual chest tube drainage was speculated to be a leakage of the pulmonary lymphatics owing to the mass on the left lower lobe. Therefore, second session of embolization was performed via direct puncture of the mass in the left lower lobe.

In the prone position, the mass was directly punctured through the intercostal space using 26-gauge spinal needle. The position of the needle was confirmed by a conebeam CT (Fig. 1C). A small amount of lipiodol was injected to confirm proper communication of the needle with the intrapulmonary lymphatics. The intrapulmonary lymphatic mass was embolized using a mixture of NBCA plus lipiodol in a ratio of 1:3. Post embolization cone beam CT revealed proper injection of the NBCA plus lipiodol mixture into the mass and the intrapulmonary lymphatics (Fig. 1D). A small amount of pneumothorax developed during the procedure, but the patient had no symptoms.

Two days following the procedure, the chest tube drainage was completely arrested, and the chest tube was removed successfully (Fig. 1E).

DISCUSSION

Nontraumatic chylothorax is a condition caused by leakage of the intestinal lymph into the chest. The leak can originate from the chest or abdominal cavity, the cause of which could be idiopathic, malignant, lymphatic vessel disease, systemic disease or congenital. The most common cause of chylothorax is idiopathic and due to lymphoma (1, 2). Traditionally, chylothorax is managed conservatively by thoracentesis and dietary modification (3). If conservative management fails, surgical intervention with pleurodesis and thoracic duct ligation is attempted (4).

TDE has become an effective alternative treatment for chylothorax patients. However its clinical success rates have been reported to be low in patients with nontraumatic chylothorax compared to those in patients with traumatic chylothorax. This is usually attributed to a less predictable location of the site of chylous leak (5). In the largest series of 109 patients with traumatic chylothorax, Itkin et al. (6) reported a success rate of 88% after TDE and needle in-

terruption. For cases of nontraumatic chylothorax, while Maldonado et al. (7) reported an overall clinical success rate of 27% using a combined approach of conservative and surgical management, Nadolski and Itkin (1) reported an overall clinical success rate of 52% using pedal lymphangiography and TDE. In addition, it is difficult to find an interventional method for when TDE fails in chylothorax patients.

In the presented case, the chest tube drainage decreased after a successful TDE. However, there was persistent pleural effusion, that was treated successfully by a direct puncture of the intrapulmonary lymphoma. To the best of our knowledge, this is the first reported case of embolization via direct puncture of an intrapulmonary lymphoma.

In cases of malignant nontraumatic chylothorax, a tumorous invasion of the posterior mediastinal or retroperitoneal lymph nodes can obstruct lymphatic flow, resulting in high pressure in the small lymphatic channels and thereby causing lymphatic leakage (5). In the present case, it was speculated that an erosion of the pulmonary lymphatics due to an intrapulmonary mass was attributed to a prolonged lymphatic leakage after TDE. In addition, the flow direction of pulmonary lymphatics was opposite to that of the chyle refluxed from the thoracic duct. Thus, the pulmonary leakage could not be resolved with TDE alone.

The potential complications of direct puncture of intrapulmonary mass include bleeding, tumor seeding and pneumothorax. However, serious complications of conventional TDE using 22-gauge Chiba needles, particularly related to bleeding have not been reported in the literature. The risk of complications related bleeding or tumor seeding is very low in the 26-gauge spinal needle in this report. Pneumothorax is most concerned complication in this treatment method. Nonetheless, patients with chylous effusions are likely to have chest tubes at the time of the procedure and small amount of pneumothorax due to fine needles can be easily resolved by observation or conservative management.

The treatment strategy adopted in this case report may be an effective therapeutic option, when applied selectively for patients with nontraumatic chylothorax.

Author Contributions

Conceptualization, K.H., K.C.W.; investigation, K.H.; methodology, K.H.; resources, K.H., J.C.H., K.C.W.; supervision, K.H., K.C.W.; visualization, H.L., K.H.; writing—original draft, H.L., K.H.; and writing—review & editing, H.L., K.H., J.C.H.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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림프종 환자의 비외상성 유미흉에서 폐림프종의 피부경유 직접천자를 통한 색전술

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림프종은 비외상성 유미흉의 원인 중 높은 빈도를 차지한다. 문헌에 따르면 비외상성 유미흉에 대해서 흉관 색전술을 시행할 경우 외상성 유미흉의 경우에 비해 치료성적이 낮은 것으로 알려져 있다. 이 논문에서는 비외상성 유미흉이 있던 림프종 환자에서 흉관 색전술을 시행하고, 추가적으로 피부경유 종양 직접 천자 색전술을 시행하여 좋은 결과를 얻었기에 이에 대한 증례를 보고하고자 한다. 본 증례는 77세 남자 환자로 림프종에 동반된 유미흉을 치료하기 위해 흉관 색전술을 시행 받았고 그 후 유미흉의 양이 줄어들었으나 지속되는 흉막삼출을 호소하였다. 이에 피부경유로 폐를 침범한 림프종을 직접 천자하여 이차 색전술을 시행하였고, 이후 흉막삼출이 호전되어 성공적으로 흉관을 제거하였다. 이 기술은 림프종과 동반된 유미흉이 있는 경우에 시행할 수 있는 새로운 치료 선택지라 할 수 있다.

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