

Chylothorax After Esophagectomy Cured by Intranodal Lymphangiography: A Case Report

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Abstract. *This is a case report of chylothorax after esophagectomy for esophageal cancer cured by intranodal lymphangiography in the groin. A 62-year-old man was admitted to our Hospital with dysphagia. The diagnosis was made of advanced esophageal cancer (T3N1M0, stage IIIA). After neoadjuvant chemotherapy, radical surgery was performed. On postoperative day (POD) 9, left pleural effusion drainage increased sharply and changed to a milky white color, leading to a diagnosis of chylothorax. Although chylothorax was managed with conservative treatment, there was an inadequate response to it. On POD 15, lymphangiography was performed. Inguinal nodes were directly accessed under ultrasound guidance and lipiodol was injected into nodes. After the lymphangiography, the left pleural effusion drainage immediately decreased. On POD 40, the patient was discharged. To the best of our knowledge, this is the first report of a chylothorax after esophagectomy for esophageal cancer to be cured by ultrasound-guided intranodal lymphangiography in the groin.*

Chylothorax is a leakage of intestinal lymph into the pleural cavity and is a result of traumatic or nontraumatic (e.g. iatrogenic, malignancy) injury to the thoracic duct. Chylothorax is a relatively rare complication among various complications after esophagectomy, reportedly occurring in 1.1% to 3.9% (1), but it is a serious complication that can lead to pulmonary dysfunction and pneumonia (2-4). Although conservative treatment including chest drainage, total parenteral treatment and discontinuation of enteral feeding is frequently applied as initial treatment for

chylothorax, surgical treatment is often required due to failure of such treatment (5). However, surgical treatment may be highly invasive for patients with chylothorax who are frequently in poor nutritional condition and at an immunosuppressive condition (6). Lymphangiography with lipiodol not only helps determine the site of chyle leakage but can also be an effective therapeutic modality for treating chylothorax (7). Indeed, several previous reports have shown that chylothorax can be cured after performing lymphangiography (7, 8). The traditional procedure of lymphangiography is a bilateral pedal approach. Because the pedal approach is both time-consuming and technically challenging, it remains a significant barrier to performing a lymphangiography. Recent studies have suggested that ultrasound-guided intranodal lymphangiography in the groin is technically simple and has potential for reduction in procedure time compared with the traditional pedal approach (9, 10). To the best of our knowledge, this is the first report of a chylothorax after esophagectomy for thoracic esophageal cancer cured by ultrasound-guided intranodal lymphangiography in the groin.

Case Report

A 62-year-old man noted difficulty with swallowing, and consulted a local physician. He was referred to our Hospital after a diagnosis of thoracic esophageal cancer was made from an upper gastrointestinal endoscopy.

Physical findings. Body height was 173 cm, body weight was 52.2 kg and body mass index (BMI) was 17.4 kg/m². The patient was undernourished, had neither anemia nor icterus, and no remarkable findings were noted in the neck, chest or abdomen.

Laboratory tests on admission. The hematological and biochemical tests returned normal results and the tumor markers (carcinoembryonic antigen and squamous cell carcinoma related antigen) were within normal range, but p53 antibody was high (71.6 U/ml).

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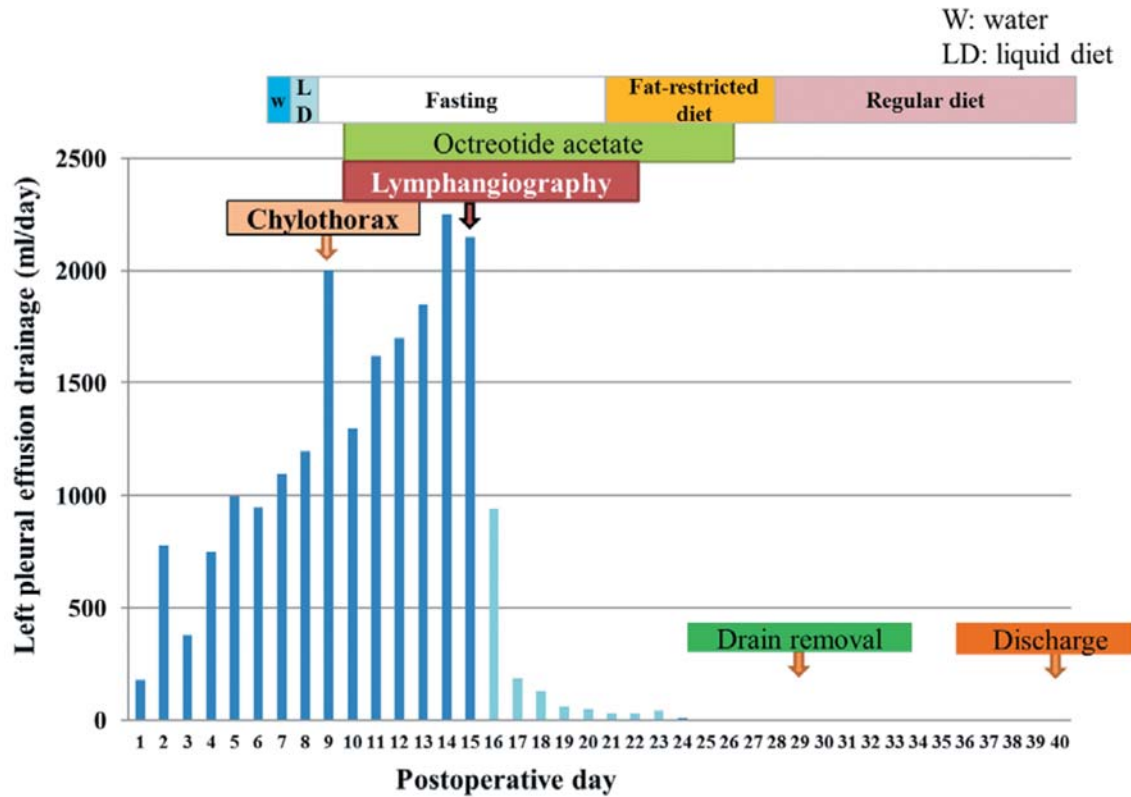


Figure 1. Postoperative progress and left pleural effusion drainage.

Upper gastrointestinal endoscopy. A 3-cm long irregular ulcerated lesion with marginal elevation was noted in the middle and lower thoracic esophagus, circumferentially involving the esophageal wall, extending 30 to 33 cm from the incisal dentition. The extent of tumor invasion was estimated to be T3. Microscopic examination of a biopsy specimen revealed the lesion to be squamous cell carcinoma.

Computed tomography scans of the neck, chest and abdomen. Computed tomography scans revealed that the tumor, which was located in the lower thoracic esophagus, did not invade locally other organs, although there was a metastasis in the paraesophageal lymph node of the neck.

From the findings described above, the diagnosis was made of locally advanced esophageal cancer, T3N1M0 stage IIIA by the Union for International Cancer Control (UICC) TNM staging system (11). Neoadjuvant chemotherapy consisting of docetaxel, cisplatin, and 5-fluorouracil (DCF) was performed before surgery. The patient received two cycles of the regimen, intravenous infusions of docetaxel (70 mg/m²) and cisplatin (70 mg/m²) on day 1 and a continuous intravenous infusion of 5-fluorouracil (700 mg/m²) from day 1 to day 5, every three weeks. The tumor and the lymph node lesion were reduced and the neoadjuvant chemotherapy evaluation was a partial

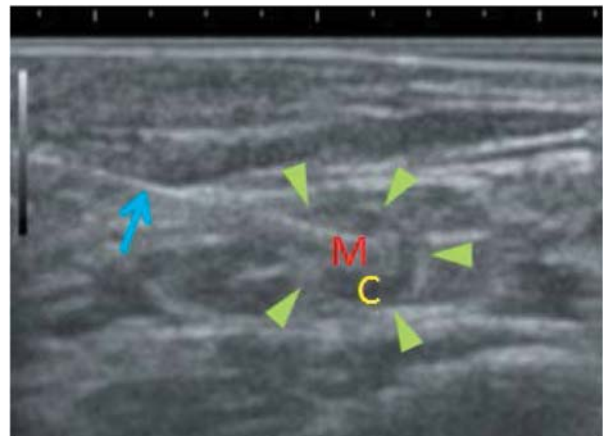


Figure 2. Ultrasonography showing access to the groin lymph node (arrowhead) with a needle (arrow). The cortex (C) is a low-echoic lesion, the medulla (M) is a high-echoic lesion.

response by Response Evaluation Criteria in Solid Tumor (12). **Surgical procedure.** The patient underwent subtotal esophagectomy and 3-field lymphadenectomy via right thoracotomy and laparotomy. Esophagogastrostomy was

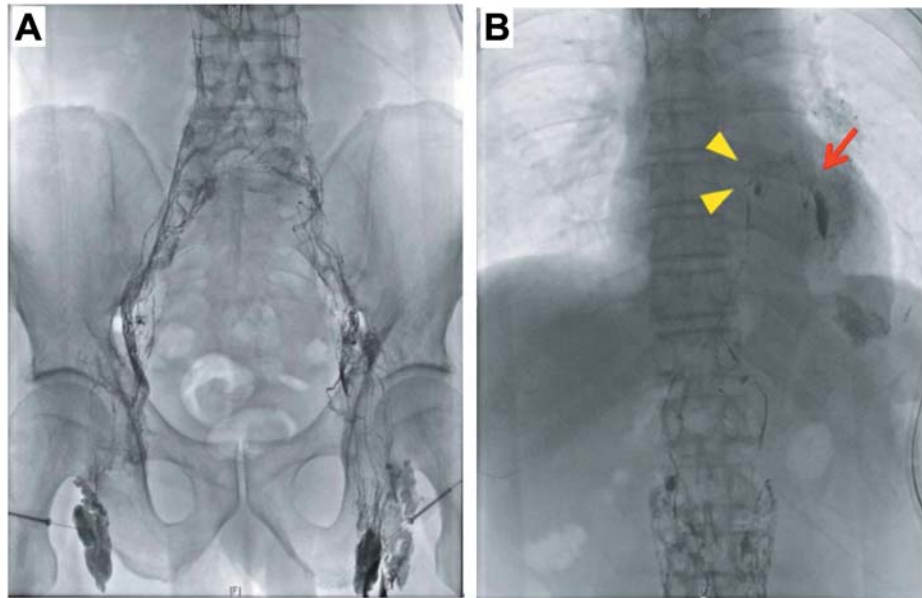


Figure 3. Spot radiograph of the bilateral groin nodes shows the initial phase of intranodal injection of lipiodol with opacification of the bilateral inguinal lymph nodes (A). Spot radiograph obtained 30 min after starting the injection shows clips (arrowhead) and a leakage (arrow) of lipiodol to the left thoracic cavity at the level of the lower mediastinum (B).

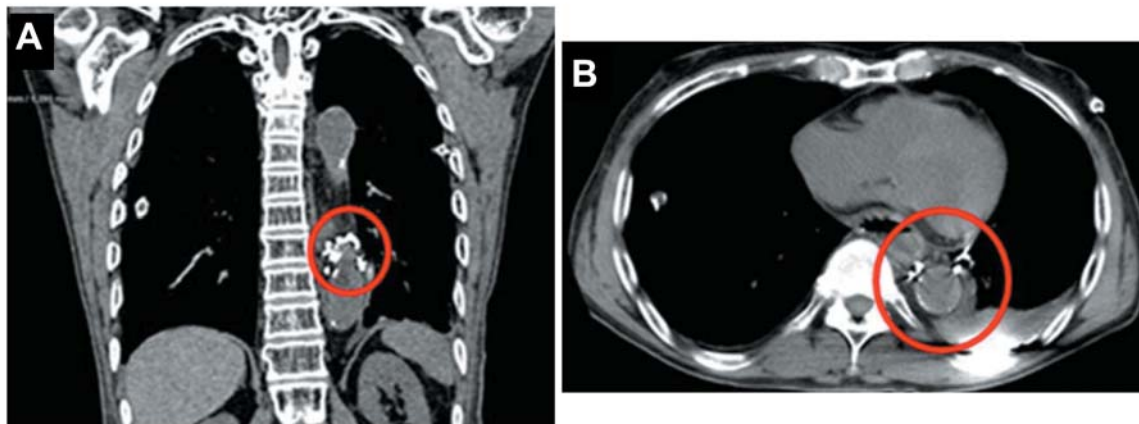


Figure 4. Coronal (A) and axial (B) sections by computed tomography obtained 50 min after starting the injection shows leakage (circle) of the lipiodol in the left hilar area.

performed for reconstruction at the neck *via* the posterior mediastinal route. The thoracic duct was ligated and resected at the lower mediastinum. There was no remarkable damage the thoracic duct during the operation.

Histological findings. The histological diagnosis was moderately differentiated squamous cell carcinoma of the esophagus (pT3, N3, M0, pStage IIIC) and the response evaluation of the neoadjuvant chemotherapy was mild effect, grade 1b (the percentage of viable residual tumour cells

within the total cancerous tissue was 1/3 to 2/3 residual tumor cells) by the Japanese Classification of Esophageal Cancer tenth edition (13).

Postoperative course (Figure 1). On POD 1, the endotracheal tube was extracted and the patient left the intensive-care unit. Although the left pleural effusion drainage remained at around 500-1000 ml/day, a wait-and-see approach was taken because the effusion was of a clear yellow fluid. On POD 7, the patient began to drink water. On POD 8, the patient began to take a

liquid diet. On POD 9, the volume of left pleural effusion drainage increased sharply and changed from clear yellow to milky white, leading to a diagnosis of chylothorax. Although the patient was given nothing to eat and administered total parenteral nutrition (TPN) through a central venous catheter and octreotide acetate (300 µg/day) as subcutaneous injections, there was an inadequate response to these treatments. The drain output gradually increased, reaching about 2,000 ml per day.

On POD 15, lymphangiography was performed with the aim of identifying the site of leakage. The transitional zones between the cortex and hilum of the bilateral inguinal nodes were directly accessed under ultrasound guidance with a 25-gauge needle (Figure 2). Lipiodol was injected by hand at a rate of about 1 ml per 5 min. Initial injection was observed under fluoroscopic guidance to identify the efferent lymphatic and lymph nodes to confirm proper positioning of the needle (Figure 3A). After a total volume of approximately 6 ml of lipiodol was injected into each lymph node, the lipiodol had leaked to the left thoracic space on the lower mediastinum and the site of leakage was located 1.5 cm cephalad to the clips used to resect the thoracic duct during surgery (Figure 3B). After a total volume of approximately 10 ml (6 ml on the right side and 4 ml on the left side) of lipiodol was injected, CT scans showed the site of leakage from the left hilar area (Figure 4). Neither damage to the thoracic duct nor the leakage from the resection stump of the thoracic duct was detected.

After lymphangiography, the left pleural effusion drainage decreased sharply. On POD 22 (seven days after the lymphangiography), the patient began to receive a fat-restricted diet. On POD 25 (10 days after the lymphangiography), the left pleural effusion drainage disappeared. On POD 29 (14 days after the lymphangiography), the left chest tube was removed. Pleural effusion was not seen at all after the removal of the tube. Decreasing pleural effusion was not associated with such complications as fever and leg edema. On POD 40 (25 days after the lymphangiography), the patient was discharged.

Discussion

Chylothorax is a comparatively rare complication of esophagectomy for carcinoma. However, occasionally patients with chylothorax do not respond to treatment. Depletion of the lymphatic fluid leads to dehydration, electrolyte abnormality, undernutrition and lymphopenia; it follows that such patients have low immunity and their condition becomes severe and life-threatening. Therefore early treatment is important (6).

The therapeutic strategy for chylothorax is divided into conservative and surgical therapy. The first approach attempts to reduce leaked lymphatic flow by conservative treatment centering on fasting, TPN, and thoracic drainage. But if the patient does not show signs of improvement, lymphangiography is considered before surgical therapy.

Lymphangiography has been widely used as an investigative tool for chylothorax since the first report by Kinmonth *et al.* in 1954 (14). The traditional procedure of lymphangiography involves the subcutaneous injection of an oily contrast medium, such as lipiodol, into each foot. The traditional procedure has some problems. First by, it is an invasive procedure requiring dorsal incisions, so that it has the potential to lead infection associated with the incisions. Secondly, it is technically challenging because it requires the isolation and cannulation of the fine pedal lymphatic vessels. Even after successful placement of lymphangiogram needles, minimal movement of the patient can dislodge the needles (10).

Intranodal lymphangiography was first reported by Rajebi *et al.* in 2011 as an improved method for the traditional procedure (9). Intranodal lymphangiography using ultrasound is a less-invasive approach in terms of requiring no incision compared to the traditional procedure. Intranodal lymphangiography is simpler and easier because an inguinal node is directly accessed under ultrasound guidance with a 23-gauge to 26-gauge spinal needle with use of a local anesthesia. The needle tip is positioned in the transitional zone between the cortex and hilum of the lymph node. Contrast agent such as lipiodol is injected by hand at a rate of about 1 ml per 5 min under fluoroscopic guidance, up to a total volume of approximately 4-8 ml in each lymph node. Nadolski *et al.* reported that the mean time from start of the procedure until initiation of the lymphangiogram was 20.5 ± 9 min in the intranodal lymphangiography group compared with 46.5 ± 23 min in the traditional group (10). Thus, intranodal lymphangiography can reduce not only the amount of time, but also the radiation dose and volume of an oily contrast medium.

Lymphangiography with lipiodol not only helps determine the site of chyle leakage but can also be effective for curing chylothorax (7). The reported success rate of lymphangiography for curing chylothorax ranges from 64.3% to 80% (15, 16). Lymphangiography is also considered to be effective for curing not only minor leakage but also major leakage (16). It is estimated that the functional mechanisms are embolization of the site of leakage by lipiodol and granulation by inflammatory stimulus at the site of the injury (6). Even if lymphangiography cannot completely embolize the site of chyle leakage, it can contribute to reducing the amount of chyle leakage, leading to a cure of chylothorax.

When chylothorax cannot be cured by conservative treatment including lymphangiography, ligation of the thoracic duct as surgical treatment is considered. The indication and timing of operation have been variously proposed but are still a subject of debate. Some authors proposed that surgery should be selected if the medical management is not effective because reoperation is too invasive for patients who have undergone esophagectomy (17-20), whereas others demonstrated that early operation is recommended (1, 21, 22). Selle *et al.* demonstrated that surgery should be considered if leakage of

the chyle continues for more than two weeks, if more than 1,500 ml/day chyle continues to leak for five days, and if malnutrition becomes severe during conservative treatment (17). Patterson *et al.* reported that surgical ligation of thoracic duct is recommended if chyle of more than 1000 ml/day continues to leak for seven days (18) and Robinson *et al.* also reported that surgery should be considered if chyle of more than 500 ml/day continues to leak for two weeks (19). On the other hand, other authors demonstrated that early reoperation has some advantages in that it is possible to reoperate in a pleural space without adhesions and it makes easier to identify the site of chyle leakage, and that patients are not severely impaired by loss of fluid, electrolytes, proteins, and lymphocytes (1). In our case, successful treatment of intranodal lymphangiography allowed surgery to be avoided although the chyle leakage of more than 1,500 ml/day continued for five days. If intranodal lymphangiography had not been successful for curing chylothorax, we would have had to consider surgery.

In conclusion, we describe a case of chylothorax after surgery for esophageal cancer which was cured by ultrasound-guided intranodal lymphangiography in the groin. Because this procedure is technically simple and can reduce the procedural time for lymphangiography, we anticipate the growing availability of ultrasound-guided intranodal lymphangiography for chylothorax after esophagectomy.

Conflicts of Interest

There are no conflicts of interest regarding this article.

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