

Lymphangiography Was Useful in Postoperative Intractable Chylothorax after Surgery for Esophageal Cancer: A Case Report

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Postoperative chylothorax after surgery for esophageal cancer is a rare but serious complication. Treatment initially consists of conservative therapy and, if it fails to provide improvement, it is important to perform surgical treatment without delay. We report on a recent case of intractable chylothorax. This report describes a 72-year-old man with Stage III esophageal squamous cell carcinoma. Subtotal esophagectomy, through a right thoracoabdominal approach with two-field lymphadenectomy, and cervical esophagogastric anastomosis via the retrosternal route, were performed. On the 12th postoperative day, a diagnosis of chylothorax was made. Conservative treatment was initiated, but it proved to be ineffective. Therefore, ligation of the thoracic duct via a thoracotomy was performed, but this was not effective, either. Lymphangiography undertaken to identify the site of the leak in the thoracic duct enabled a diagnosis of an extremely rare double thoracic duct and identification of the site of the leak in the thoracic duct, thereby allowing curative direct ligation of the site. This case underscores the remarkable usefulness of lymphangiography in dealing with intractable postoperative chylothorax.

(J Nippon Med Sch 2017; 84: 268–273)

Key words: esophageal cancer, chylothorax, lymphangiography

Introduction

Postoperative chylothorax after surgery for esophageal cancer is a rare but serious complication that gives rise to significant impairment of the cardiovascular and respiratory systems, nutritional status, and immune system unless appropriately treated^{1,2}. The thoracic duct is a trunk of the lymphatic vessel coursing in the proximity of the esophagus. Consequently, an injury incurred by an accident during an esophagectomy may lead to the development of a chylothorax. The incidence of chylothorax as a postoperative complication after surgery for esophageal cancer is reported to be 1–4%^{3,4}. Treatment initially consists of conservative therapy and, if it fails to provide improvement, it is important to perform surgical treatment without delay^{1,5}.

We have recently treated a case of intractable chylothorax which developed after surgery for cancer of the esophagus and failed to respond to conservative therapy,

and in which ligation of the thoracic duct also failed to be effective. Lymphangiography, then undertaken to identify the site of the leak in the thoracic duct, enabled a diagnosis of an extremely rare double thoracic duct and identification of the site of the leak in the thoracic duct, thereby allowing curative direct ligation of the site. This paper documents the case.

Case Presentation

A 72-year-old man diagnosed with thoracic esophageal cancer was referred and admitted to our hospital for medical workup and treatment. His status on admission was unremarkable, and preoperative hematologic and blood biochemical tests showed no abnormalities, including any elevation of tumor markers. Upper gastrointestinal endoscopic examination disclosed a circumferential type 2 esophageal cancer in the lower intrathoracic esophagus, and biopsies revealed the lesion to be

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Journal Website (<http://www2.nms.ac.jp/jnms/>)

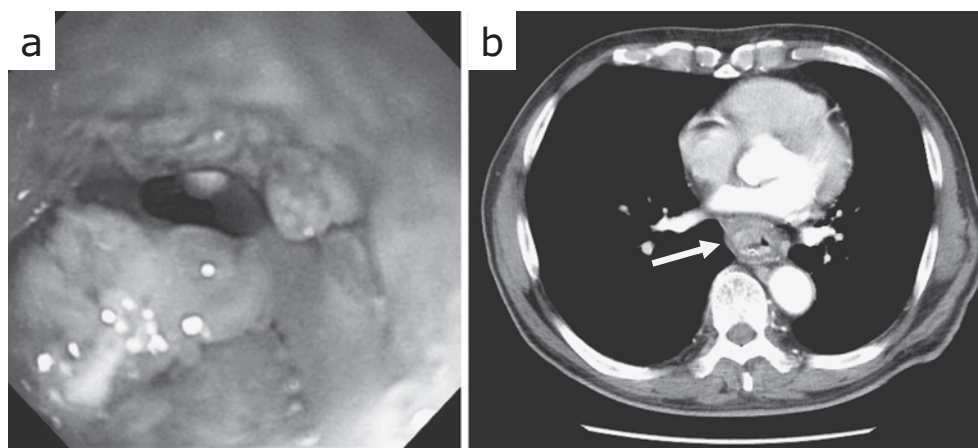


Fig. 1 Upper gastrointestinal endoscopic examination showed a circular type 2 esophageal cancer in the lower intrathoracic esophagus (a). CT scan demonstrated that the depth of invasion was T3 with no evidence of lymph node metastasis (N0) or distant metastasis (M0) (b).

squamous cell carcinoma (**Fig. 1a**). A preoperative computed tomography (CT) scan demonstrated that the depth of invasion was T3 with no evidence of lymph node metastasis (N0), or distant metastasis (M0), leading to a diagnosis of Stage II (**Fig. 1b**), (The Japan Esophageal Society: Japanese Classification of Esophageal Cancer, 11th Edition⁶). With the above diagnosis, subtotal esophagectomy through a right thoracoabdominal approach with two-field lymphadenectomy, cervical esophagogastric anastomosis via a retrosternal route, and gastrostomy were performed. Histopathologic examination of resected tissue specimens showed that the esophageal tumor was a well-differentiated squamous cell carcinoma, with no tumor remnants, a T3 depth of invasion, and no lymph node metastasis.

The total amount of bilateral pleural effusion drainage was 0.43 L on the day of operation, and diminished thereafter. On the 3rd postoperative day, administration of nutrients via a gastrostomy tube was started. As there was no increase in pleural effusion even after the start of enteral nutrition, the left thoracic drain was removed on the 6th postoperative day and the right thoracic drain on the 9th postoperative day. On the 12th postoperative day, however, he suddenly became more dyspneic and a chest X-ray demonstrated a large left-sided pleural effusion. Immediate insertion of a thoracic drain yielded more than 3.0 L of milky fluid, and a diagnosis of chylothorax after surgery for esophageal cancer was made. As for nutritional management, the enteral nutrition was discontinued and feeding was limited solely to total parenteral nutrition. Because the pleural effusion drainage continued to exceed 1 L per day thereafter, he was begun on a somatostatin analogue at 300 µg/day on the 14th postop-

erative day.

Nevertheless, the pleural effusion did not decrease and, judging that it seemed difficult to further continue the conservative treatment, ligation of the thoracic duct via a thoracotomy was performed on the 19th postoperative day. Although he had been administered high-fat enteral nutrients by means of an enteral nutrition tube before the operation, our intraoperative attempt to identify the site of a chyle leak failed. Therefore, we performed convergent ligation of the thoracic duct in the right vicinity of the descending aorta on the diaphragm, taking account of the anatomical coursing of the thoracic duct.

As chylous effusions exceeding 1.0 L per day were noted on the ensuing consecutive days, lymphangiography was carried out for the purpose of identifying the thoracic duct leak site on the 24th postoperative day (**Table 1**). Following an interdigital injection of 1% lidocaine hydrochloride (Xylocaine[®]) and Indigotindisulfonate Sodium (Indigo carmine) into the foot, a minor incision was made, a blue-stained lymph vessel was then identified, and fluoroscopic examination was performed while injecting Lipiodol Ultra-Fluid[®], a contrast medium-embolizing agent, at a rate of 3 mL/hr through the same site. As a result, interruption of the contrast medium just above the diaphragm was noted along with the presence of a separate thoracic duct showing a leak of the contrast medium. Thus, the patient was found to have an extremely rare duplicated thoracic duct with a leakage from the thoracic duct distinct from the previously ligated one (**Fig. 2**). As there was still a large pleural effusion on the day following the lymphangiography (25th postoperative day), a surgical repair was carried out again. Thoracotomy was performed through the sixth in-

Table 1 Postoperative pleural effusion (1)

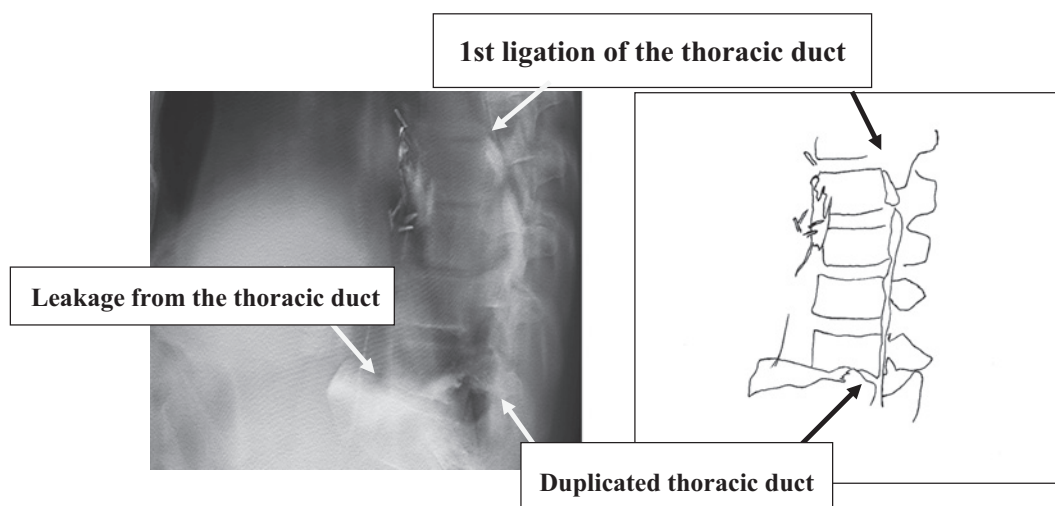
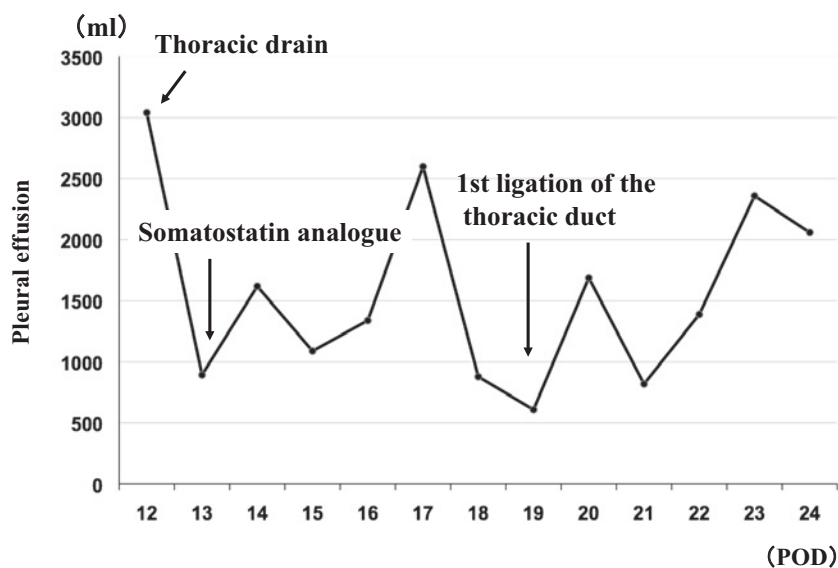


Fig. 2 Lymphangiography demonstrated an interruption of the contrast medium just above the diaphragm along with the presence of a separate thoracic duct showing a leak of the contrast medium. Thus, the present case was found to be of an extremely rare duplicated thoracic duct with a leakage from the thoracic duct, distinct from the previously ligated one.

tercostal space close to the site of the leakage, verified on lymphangiography. A leak of chyle was identified, left-posteriad to the descending aorta. A direct ligation was performed and chylous leakage was confirmed to have stopped. There was a remarkable reduction in the drained volume of pleural fluid postoperatively; the drained fluid volume decreased to 0.3 L in 24 hours post-operation.

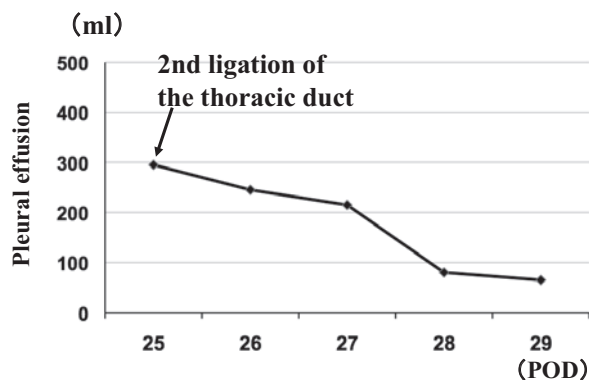
On the 28th postoperative day, the patient was started on a liquid diet and the left pleural tube drain was removed on the 30th day after the initial operation (Table 2). A pleural effusion was noted on a chest X-ray exami-

nation performed on the 39th postoperative day, and complete resolution of the effusion was evident on the 54th postoperative day. He was discharged home on the 57th postoperative day, and there was no recurrence of tumor or chylothorax present 3 years after the operation.

Discussion

Chylothorax is a rare complication seen in 1–4%^{3,4} and 0.5%⁷ of cases of chest surgery for esophageal cancer or lung cancer, respectively. The cause of the disorder is a thoracic duct injury, associated with surgical intervention, in virtually all cases. Conservative treatment provides

Table 2 Postoperative pleural effusion (2)



improvement in 70–80% of cases caused by surgery for lung cancer, whereas in cases occurring after surgery for esophageal cancer, the percentage of responders to conservative therapy is not as high due to a large chylous effusion⁸. Another report stated that surgical intervention was required in 62% of cases of chylothorax, that had developed after esophageal surgery, and that thoracotomy was required in 71% of those surgically treated cases⁹.

Chyle is a milky bodily fluid consisting of long-chain fatty acids mixed with emulsified lymph and, because it does not contain fibrinogen, there is no mechanism for the occlusion of stoma in the thoracic duct. Once it has developed, therefore, chylous leakage persists. Furthermore, chyle also contains an abundance of proteins, lymphocytes (especially T-cells), immunoglobulins, vitamins, and electrolytes. Since the thoracic duct has a flow rate of 4 L/day in healthy adults, persistent loss of chyle results in the loss of a large amount of lymphocytes and calories. Eventually, if a chylothorax remains untreated, the disorder gives rise to drastic impairment of the cardiovascular and respiratory systems and nutrition, in addition to an increased incidence of viral and bacterial infections¹⁰.

Whilst high triglyceride levels, and the demonstration of Sudan III-stained fat droplets in the pleural effusion on thoracocentesis, are diagnostic for chylothorax, this disorder is readily diagnosed by the detection of milky white fluid drained from the chest after the initiation of enteral nutrition¹¹. To prevent the development of chylothorax, measures undertaken include exercising great caution to prevent injury to the thoracic duct during thoracotomy by keeping the anatomical course of the thoracic duct in mind and conducting a prophylactic ligation of the thoracic duct in the case of a possible accidental injury incurred to the thoracic duct during a course of regional lymphadenectomy. Matteo et al. reported that, in

their retrospective study, prophylactic ligation of the thoracic duct enabled complete prevention of the development of postoperative chylothorax without the involvement of any complication associable with the surgical procedure¹².

Treatment may be conservative or surgical. As with our case, continued drainage of chyliform pleural fluid is performed at first, followed by nutritional management with parenteral fluid infusion, NPO and total parenteral nutrition against massive loss of chyle. Moreover, the efficacy of pleural adhesions by intrathoracic administration of OK-432 (Picibanil[®]) or minocycline hydrochloride (Minomycin[®]) was reported¹⁰. However, there has also been a report that the attempt failed to provide sufficient effectiveness despite the administration of two or more doses.

In recent years, a report stated that the administration of octonucleotide, a long-acting somatostatin analogue, was effective in the treatment of chylothorax¹³. As for underlying mechanisms of action, the inhibition of fat absorption from the gut, and diminished lymph flow due to decreased visceral blood flow, may account for the event, and there are receptors for somatostatin distributed in the blood vessels, lymph vessel endothelium and smooth muscles, whereby the lymph flow is decreased, giving rise to adhesions in the injured thoracic duct with the surrounding tissues. Other recent studies reported good treatment outcomes after thoracic duct embolization following interventional radiology (thoracic ductography)^{14,15}. Itkin et al.¹⁴ performed thoracic duct lymphangiography and embolization using a vascular coil, or a lipid material, in 109 patients with chylothorax associated with thoracic duct injuries and reported a low invasiveness and usefulness of the technically challenging procedure based on a 71-percent cure rate. Atie et al.¹⁶ also reported a case of chylothorax cured with thoracic duct lymphangiography and embolization. The authors explained the treatment success rate was from 61% to 71%, and immediate surgical treatment would be required if the procedure failed. While thoracic duct embolization is a very useful treatment option for patients who would not tolerate surgery, the surgeon needs to be thoroughly skilled at the procedure. Itkin et al.¹⁴ and Yamamoto et al.¹⁷ reported the simplicity and usefulness of intranodal lymphangiography, a new technique involving inguinal lymph node puncture.

According to Selle et al.¹⁸, surgery should be indicated when pleural effusions of 1.5 L per day persist for ≥ 5 days, if no indication of resolution is seen despite conser-

vative therapy for 14 days or longer, or in the case of worsened nutritional status. Cerfolio et al. described that surgical treatment should be instituted without aimless continuation of conservative therapy in the case where the mean chest drain fluid volume during 7 days post-operation exceeds 1 L per day^{19,20}. Rottoli et al.²¹ and Barbetakis et al.²² reported cases in which acute aggravation of the respiratory condition with destabilization of the hemodynamic status occurred. Regarding the pathogenetic mechanism of the event, Rottoli et al.²¹ explained that extrinsic compression of the left pulmonary vein and atrium by retained fluid brought about a cardiac tamponade-like pathophysiologic state. Surgical treatment should be undertaken without delay in such instances as well.

Surgical treatments commonly include thoracic duct ligation, whereby the thoracic duct is ligated on the diaphragm and direct repair of the thoracic duct leak site is performed. The usefulness of endoscopic ligation of the thoracic duct has also been reported in recent years. One report deals with the Denver pleuroperitoneal shunt²³.

Unsuccessful ligation of the thoracic duct may be attributed to an anomaly of the thoracic duct, such as a duplicated thoracic duct, the incidence of which has been reported to be about 10%²⁴. Anatomic anomalies of the thoracic duct include a bisymmetric double thoracic duct and a complete right thoracic duct, and their incidence has been documented to be approximately 5% within the Japanese population²⁵. In the case reported herein, chest drain insertion was performed immediately after the diagnosis of chylothorax post-operation for esophageal cancer had been made, and conservative treatment was instituted, but proved to be ineffective. Therefore, surgical treatment was then undertaken. As the intraoperative attempt failed to identify the point of chyle leakage, we conducted convergent ligation of the thoracic duct on the diaphragm. However, there was no appreciable decrease in pleural effusions after the operation, and lymphangiography was carried out with the aim of identification and embolization of the site of chyle leakage. Thoracic duct embolization could not be achieved and, consequently, a diagnosis of a duplicate thoracic duct and identification of the site of chyle leakage were made with gratifying results to enable direct surgical repair. Thoracic duct embolization might have been performed following lymphangiography if we had been skilled at the procedure; however, there was no time to develop the skill because immediate action was required due to the previous failure of thoracic duct ligation in this patient. Subse-

quently, the pleural fluid drain ceased and the circulatory-respiratory condition stabilized.

The diagnosis of a duplicate thoracic duct and identification of a chyle leak site were achieved with the aid of lymphangiography, enabling treatment of the disorder in this case. The experience in this case stresses the remarkable usefulness of lymphangiography in dealing with intractable postoperative chylothorax.

Conflict of Interest: None of the authors have any conflict of interests to declare.

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(Received, July 26, 2016)

(Accepted, December 5, 2016)