American Journal of Case Reports

e-ISSN 1941-5923
© Am J Case Rep, 2022; 23: e936590
DOI: 10.12659/AJCR.936590

Received: 2022.03.07
Accepted: 2022.05.24
Available online: 2022.06.16
Published: 2022.07.22

Thoracic Duct Embolization for Delayed Chyle Leak After Lewis-Tanner Esophagectomy

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

ABCDEFG 1 Marzia Franceschilli
CDF 2 Renato Argirò
CDEF 1 Leandro Siragusa
BCDE 1 Valeria Usai
AEFG 3 Simone Sibio
ABCDEFG 1 Sara Di Carlo

1 Department of Surgery, University of Rome Tor Vergata, Rome, Italy
2 Department of Diagnostic Imaging and Interventional Radiology, Tor Vergata University of Rome, Rome, Italy
3 Department of Surgery "Pietro Valdoni", University "Sapienza" of Rome, Viale del Policlinico, Rome, Italy

Corresponding Author: Marzia Franceschilli, e-mail: marzia.franceschilli@gmail.com
Financial support: None declared
Conflict of interest: None declared

Patient: Male, 54-year-old
Final Diagnosis: Chyle leak
Symptoms: Abdominal pain
Medication: —
Clinical Procedure: —
Specialty: Radiology • Surgery

Objective: Unusual or unexpected effect of treatment

Background: Radical esophagectomy for cancer is a potentially curative treatment that requires two/three-field lymphadenectomy. Serious complications can occur, including chyle leak (CL). CL has an incidence rate of 1-9% and is associated with a higher rate of postsurgical morbidity and mortality. It usually occurs in the early postoperative period; delayed CL is less common and is thought to be due to an occult leak or late diagnosis.

Case Report: A 54-year-old man with adenocarcinoma of the esophagus underwent Lewis-Tanner esophagectomy after neoadjuvant chemotherapy with FLOT. During en bloc lymphadenectomy, the main thoracic duct was identified, clipped, and divided. The postoperative course was uneventful. One month after hospital discharge, he was readmitted with severe abdominal, scrotal, and lower-limb edema. A chest-abdomen CT scan revealed massive pleural effusion with left shift and compression of the mediastinum. The patient was initially treated with fasting and fat-free total parenteral nutrition, and the drain output was 2800-3000 mL/dL. Lymphoscintigraphy with ethiodized oil eventually revealed a thoracic duct leak, and lymphatic embolization was successfully performed with a 4-mm metallic spiral and glue. Drain output dramatically reduced, and after 11 days the thoracic drain was removed and the patient was safely discharged.

Conclusions: Thoracic duct embolization seems be an effective therapy in treating high-output (>1000 mL/dL) CL that has occurred more than 2 weeks after esophagectomy. It can be considered as a first-line treatment due to its simplicity and effectiveness.

Keywords: Chyle • Esophagoscopy • Lymphoscintigraphy • Squamous Cell Carcinoma of Head and Neck

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/936590

Indexed in: [PMC] [PubMed] [Emerging Sources Citation Index (ESCI)] [Web of Science by Clarivate]

Background

Esophageal cancer is the seventh most common neoplasia worldwide and is responsible for approximately 450 000 deaths per year [1]. Radical esophagectomy is a potentially curative treatment, requiring two/three-field lymphadenectomy and carrying a high risk of postoperative complications [2]. Chyle leak (CL) has a reported incidence of 1-9% and it has been associated with a higher rate of postsurgical morbidity and mortality [3]. Chylothorax usually occurs early in the postoperative period and treatment should be timely and effective to prevent patient deterioration. Delayed CL is less common and is thought to be due to an occult leak or late diagnosis [4]. Treatment options range from fully conservative approaches to surgical ones. Interventional radiology has been described as an effective treatment in the management of main thoracic duct (TD) fistulas.

Case Report

Our patient was a 54-year-old man referred to our institution with an ulcerated, bleeding mass localized 1 cm from the gastroesophageal junction, occupying 2/3 of the esophageal lumen and extending cranially for 8 cm. Biopsies confirmed a poorly-differentiated, high-grade, esophageal adenocarcinoma. PET-CT scan revealed cT3 N0 M0. Multidisciplinary discussion indicated neoadjuvant chemotherapy with FLOT: 4 cycles Docetaxel 50 mg/m^2+ Oxaliplatin 1.85 mg/m^2+ Leucovorin 200 mg/m^2+ 5-Florouracil 600 mg/m^2 i.v. The patient had a good response to the neoadjuvant treatment and underwent radical subtotal Lewis-Tanner esophagectomy and gastric pull-up reconstruction. During en bloc lymphadenectomy, the TD was identified at the base of the right chest, dissected, clipped, and divided. The chest was drained with 2 closed water-seal drains; 1 abdominal drain was placed at the level of the hiatus.

Postoperatively, total parenteral nutrition with Olimel N9E and vitamins was administered and then discontinued 6 days after surgery. The abdominal drain was removed on postoperative day 3, when drainage was less than 80 mL. The apical chest tube was removed after a negative chest X-ray (**Figure 1**), also on postoperative day 3. Finally, the basal chest drain was removed on day 6, with draining less than 100 mL in the last 24 h. No quality test of drain fluids was performed. The postoperative course was uneventful, and the patient was discharged in good clinical condition on postoperative day 8.

The patient was clinically assessed in the outpatient department 1 week after discharge. He had no concerns and the clinical examination was negative. Skin staples were removed and his diet increased from soft to frequent light meals. One month after hospital discharge, his general practitioner referred him

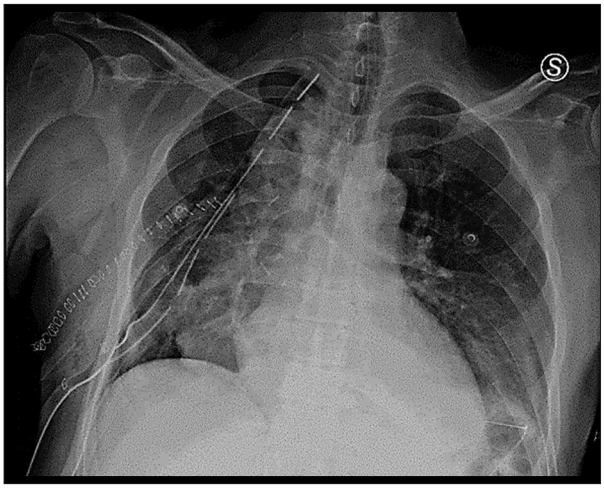

Figure 1. Day 3 postoperative chest X-ray after Ivor-Lewi esophagectomy, showing no sign of consolidation or effusion before apical drain removal.

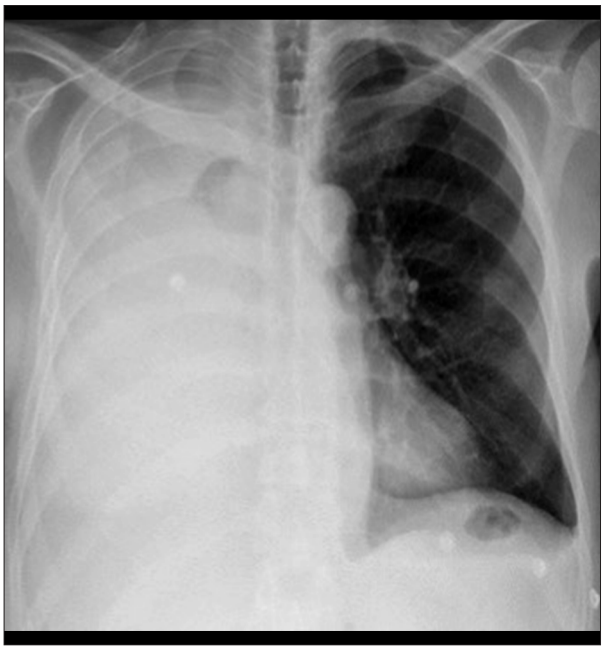

Figure 2. Re-admission chest X-ray 1 month after surgery, showing right unilateral pleural effusion with consensual pulmonary atelectasis.

to the emergency department because of severe abdominal, scrotal, and lower-limb edema. The patient had noticed scrotal edema 2 weeks earlier, with subsequent insurgence to the lower limbs and abdominal edema. A blood test revealed a mild increase of WBC (11.94), hypoalbuminemia (2.99 g/dL), hyponatremia (133 mEq/L), and BUN elevation (83 mg/dL). The chest X-ray (**Figure 2**) showed unilateral right pleural effusion with atelectasis. A chest-abdomen CT scan confirmed the X-ray findings, revealing compression and left shift of the mediastinal structures with ground-glass appearance in the

Indexed in: [PMC] [PubMed] [Emerging Sources Citation Index (ESCI)] [Web of Science by Clarivate]

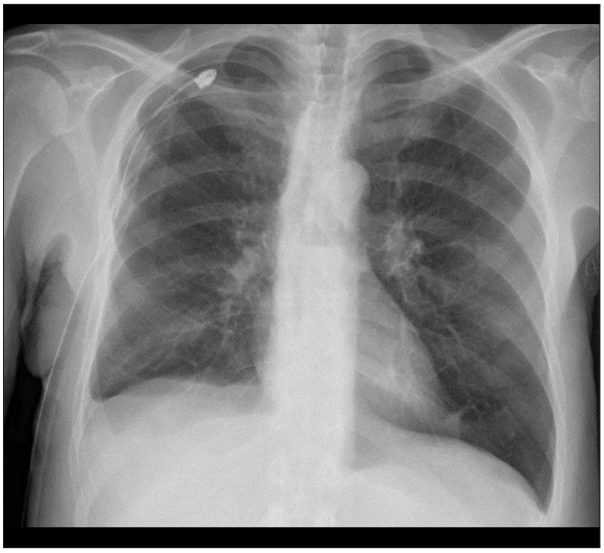

Figure 3. Chest X-ray on day 5 post re-admission after 24-French chest drain inserted showing right lung re-expansion with atelectasis resolution and residual pleural effusion.

left hilum and inferior left lung lobe. Abdominal sequences showed presence of fluid in the upper quadrants in the subphrenic spaces. Oral contrast with soluble solution was also given, showing good passage through the neo-esophagus and pylorus, and no fistulas.

The patient was admitted to the surgical ward and a 24 French chest drain was inserted: 1400 mL of milky fluid was drained, thus confirming CL. Triglyceride dosage on the pleural fluid sample was 399 mg/dL and a microbiological culture was negative. Treatment was initially conservative, with fasting and fat-free parenteral nutrition with Clinimix N12G20E (1400 kcal/day) and Octreotide 0.1 mcg 3 times a day. Hyponatremia and hypoalbuminemia were managed with albumin and NaCl infusions. X-rays on day 5 showed right lung re-expansion and residual pleural effusion (**Figure 3**).

For the following 10 days, drain output remained stable at 2800-3000 mL per day.

On day 11, TDE was carried out by ultrasound-assisted percutaneous injection of iodinated contrast agent into the right inguinal lymph node [5]. A slow diffusion of contrast was observed in the abdominal and pelvic lymphatic network up to the cisterna chyli (CC) and TD. After identification of the target ducts, cannulation was performed with a right transabdominal approach using a 21 G sterile needle and fluoroscopic guidance. A 3-French microcatheter was advanced into the TD over a 0.014" guidewire. Two sources for leaking were identified just at the level of the clips on the TD. A 4-mm metallic coil was placed at the superior aspect of the thoracic duct to provide a

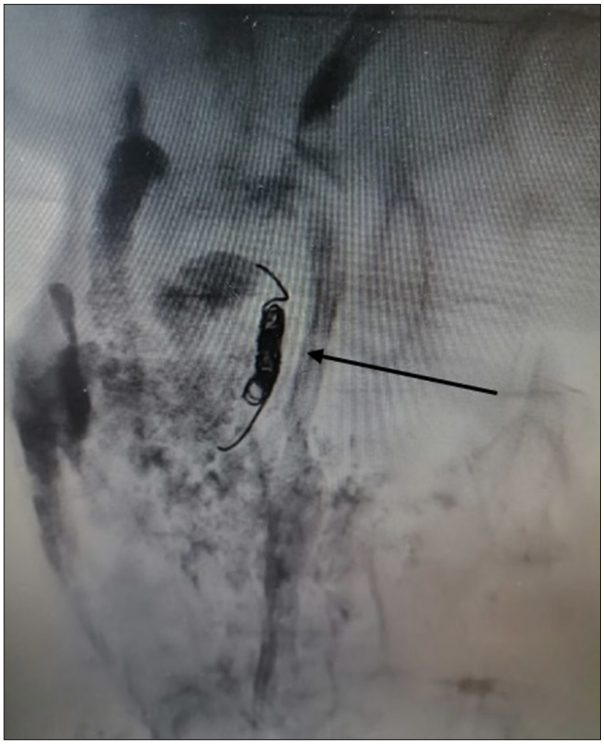

Figure 4. Lymphoscintigraphy images after thoracic duct embolization with 4-mm metallic spiral and glue (arrow) to obtain complete thoracic duct closure.

Figure 5. Radiological drain output during hospital stay before and after thoracic duct embolization (green line) (mL on y axis, days on x axis).

matrix for glue polymerization. TD was embolized with a mixture of glue proximally injected in the duct. The microcatheter was removed immediately after glue injection (**Figure 4**).

No immediate complication was observed, and the following day an X-ray showed further reduction of pleural effusion and complete lung expansion. A progressive reduction of the drainage

Indexed in: [PMC] [PubMed] [Emerging Sources Citation Index (ESCI)] [Web of Science by Clarivate]

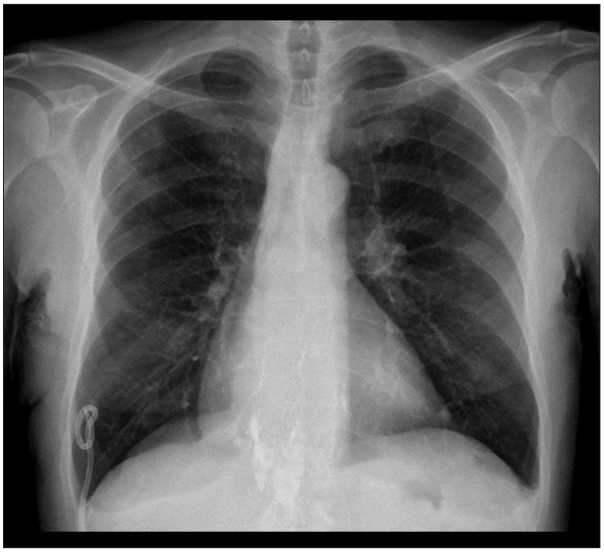

Figure 6. Chest X-ray after discharge showing complete resolution of pleural effusion and complete lung re-expansion.

was observed during subsequent days (**Figure 5**). Parenteral nutrition was gradually replaced by a Medium-Chain-Fat dietary scheme and the patient was discharged for home-care aid with 90 mL of output per day. Fat food was progressively reintroduced in the diet, and the chest drain was finally removed 11 days after thoracic ducts embolization (**Figure 6**).

Discussion

We report what appears to be a true case of delayed CL. Chylothorax after esophagectomy is not common but is indeed a serious complication, with a reported incidence of 1% to 9% [6,7]. CL is associated with considerable morbidity, mainly related to pneumonia with respiratory failure. Clinical management requires a prompt assessment of patients' general condition, since they often become severely malnourished and immunocompromised due to massive loss of immunoglobulins, lymphocytes, proteins, and fat [8]. Recent studies reported mortality rates as high as 20% [9]. CL is generally considered a milky chest drain output that commonly occurs within the first week after surgery, although cases have been identified up to 13 days postoperatively [7]. Diagnosis is confirmed by triglyceride levels higher than 110 mg/dL after starting enteral feeding. Postoperative CL is mainly due to TD or damage to its tributaries or ligate failure during esophagectomy in the abdominal or thoracic phase. In addition, CL can result from damage to the cisterna chyli during nodal dissection around the celiac trunk. To prevent chylothorax, several approaches for thoracic duct identification and ligation during esophagectomy have been identified. Although some esophageal surgeons advocate for routine supradiaphragmatic thoracic duct ligation [10], others recommend excluding thoracic duct dissection as a standard procedure during esophagectomy [11]. The open transthoracic Lewis-Tanner operation seems to be associated with higher risk of thoracic duct injury in comparison to the trans-hiatal operation or to minimally invasive procedures [12]. In the present patient, identification, closure with clips, and division of the TD was carried just above the right diaphragm. Clipping the duct seems to be a perfectly reasonable thing to do and CL could be attributed to damage to tributaries of the main duct rather than injury to the thoracic duct per se or slippage of the clips [13]. An increased output (>13.5 mL/kg/day) seems to be linked to an increased incidence of CL from main thoracic duct [14], but in our patient there was no high postoperative chest drain output, thus suggesting a true delayed CL.

The Esophagectomy Complications Consensus Group categorized CL into 3 types, depending on treatments (I, enteric dietary modifications; II, total parenteral nutrition; and III, interventional or surgical therapy) and 2 types based on severity (A, <1000 mL output per day; and B, >1000 mL output per day) [15].

CL management is highly variable and there is no consensus on a predefined algorithm. Traditionally, conservative management based on dietary modification (fat-free diet) and drainage is the first-line treatment for low output (<1000 mL/day) of chylothorax. Some studies suggest the adjuvant use of somatostatin analogs, such as octreotide and etilefrine, with or without pleurodesis [16,17]. Wemyss et al [4] based the therapeutic approach on daily drain output. They reported a success rate of 65.6% with a 7-day low-fat diet in CL <500 mL/day. Drain output of >1000 mL is an indication for surgical intervention at any time point [18-20].

When conservative management fails, radical options are necessary. Most studies did not specify objective criteria for treatment failure or indications for treatment escalation, while others advocated a weight-based quantification of chyle output (eg, mL/kg), or applied the same threshold to all patients (eg, >1000 mL).

Pleurodesis is a relatively common procedure. Among the most commonly used agents that have shown effectiveness in pleurodesis are tetracycline, minocycline, bleomycin, OK-432, povidone iodine, and talc [21]. One study trialed platelet-rich fibrin glue pleurodesis in 26 patients following conservative management, with a success rate of 100% after 2 procedures [22].

Overall, surgical therapy is successful in 67-100% of cases [23]. Thoracoscopic or thoracotomy direct thoracic duct ligation are popular and successful means to achieve postoperative CL resolution. Lai et al [24] described thoracic duct ligation below

Indexed in: [PMC] [PubMed] [Emerging Sources Citation Index (ESCI)] [Web of Science by Clarivate]

the inferior pulmonary vein at the diaphragmatic hiatus encircling all the lympho-fatty tissue located along the pleura, aorta, and spine, including the azygos vein. However, multiple leaks can occur concomitantly at different sites, primarily from one of the tributaries. Therefore, ligation proximal to the aortic hiatus is preferred when a leak site is not well visualized. Pre- or intraoperatively butter, cream, or methylene blue administration can facilitate direct visualization of the leak site. Clip ligation is the most common procedure performed even if fibrin glue application to the leak site appears effective when the exact leak site cannot be identified [25]. If surgery fails and chylothorax becomes intractable, a pleuroperitoneal or pleuro-venous shunt could be considered as a last resort. Two different types of shunts are available: the active Denver pleuroperitoneal shunt and the passive Le Veen shunt, which is most commonly used [26]. The shunt is implanted in the chest and the catheter is used to transport chyle into the peritoneal cavity, where its components can be absorbed. Pleuroperitoneal shunts should not be overused, as complications are significant, including shunt occlusion, infections, bacterial translocation skin erosions, shunt relocation, and pneumoperitoneum [27,28].

Thoracic duct embolization (TDE) has become a treatment alternative for CL with high-output chylothorax, given its high success rate and minimal complications. In 1998, Cope et al [29] described TDE for first time. Lymphangiography before TDE detects the lymphatic leakage and is helpful to visualize the anatomy of lymphatic vessels before embolization. After identification of the cause of CL (extravasation or obstruction), embolization of the TD is performed proximally. N-Butyl cyanoacrylate (n-BCA) (Truefill1 Cordis, Johnson & Johnson, Warren, NJ) diluted 1: 1 in Ethiodol is used for embolization. The use of fibrin glue and cyanoacrylate has been successfully employed in the treatment of various sinuses and fistulas [30]; the TD is filled with the glue mixture proximal to the leak or occlusion. Clinical success rates of this technique range from 70% to 90%, and complications are rare, including the transgression of the needle through adjacent organs, chronic lower-limb swelling, chronic diarrhea, and ascites [31,32].

Conclusions

TDE is already described in the literature as a good alternative to surgery and the first-line treatment of high-output (>1000 mL/dL) CL. More research is needed on delayed CL, which is rare and less described. TDE can be an effective first-line approach to delayed CL, obtaining good results with a minimally invasive technique.

Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

References:

1. Institute of Health Metrics. Global health data exchange. Available at: http://ghdx.healthdata.org/gbd-results-tool (cited January 2021)
2. Sica GS, Djapardy V, Westaby S, Maynard ND. Diagnosis and management of aortoesophageal fistula caused by a foreign body. Ann Thorac Surg. 2004;77(6):2217-18
3. Milito P, Chmelo J, Dunn L, et al. Chyle leak following radical en bloc esophagectomy with two-field nodal dissection: Predisposing factors, management, and outcomes. Ann Surg Oncol. 2021;28(7):3963-72
4. Wemyss-Holden SA, Launois B, Maddern GJ. Management of thoracic duct injuries after oesophagectomy. Br J Surg. 2001;88:1442-48
5. Rossi P, Sileri P, Gentileschi P, et al. Percutaneous liver biopsy using an ultrasound-guided subcostal route. Dig Dis Sci. 2001;46(1):128-32
6. Sica GS, Iaculli E, Benavoli D, et al. Laparoscopic versus open ileo-colonic resection in Crohn's disease: Short- and long-term results from a prospective longitudinal study. J Gastrointest Surg. 2008;12(6):1094-102
7. Mishra PK, Saluja SS, Ramaswarmy D, et al. Thoracic duct injury following esophagectomy in carcinoma of the esophagus: Ligation by the abdominal approach. World J Surg. 2013;37:141-46
8. Fina D, Franze E, Rovedatti L, et al. Interleukin-25 production is differently regulated by TNF-alpha and TGF-beta 1 in the human gut. Mucosal Immunology. 2011;4(2):239-44
9. Brinkmann S, Schroeder W, Junggeburth K, et al. Incidence and management of chylothorax after Ivor Lewis esophagectomy for cancer of the esophagus. J Thorac Cardiovasc Surg. 2016;151(5):1398-404
10. Hoeppner J, Hopt UT. Transabdominal mass ligation of the thoracic duct for the prevention of chylothorax following en bloc oesophagectomy. Eur J Cardio- thoracic Surg. 2013;44:1134-36
11. Batool S, Akbar SA, Khan M, et al. Risk factors for chyle leak after esophagectomy. J Ayub Med Coll Abbottabad. 2019;31(4):506-11
12. Gentileschi P, Camperchioli I, Benavoli D, et al. Laparoscopic single-port sleeve gastrectomy for morbid obesity: Preliminary series. Surg Obes Relat Dis. 2010;6(6):665-69
13. Nair SK, Petko M, Hayward MP. Aetiology and management of chylothorax in adults. Eur J Cardiothorac Surg. 2007;32(2):362-69
14. Marts BC, Naunheim KS, Fiore AC, Pennington DC. Conservative versus surgical management of chylothorax. Am J Surg. 1992;164(5):532-35
15. Low DE, Alderson D, Cecconello I, et al. International consensus on standardization of data collection for complications associated with esophagectomy: Esophagectomy complications consensus group (ECCG). Ann Surg. 2015;262:286-94
16. Fujita T, Daiko H. Efficacy and predictor of octreotide treatment for postoperative chylothorax after thoracic esophagectomy. World J Surg. 2014;38:2039-45
17. Lambertz R, Chang DH, Hickethier T, et al. Ultrasound-guided lymphangiography and interventional embolization of chylous leaks following esophagectomy. Innov Surg Sci. 2019;4:85-90
18. Kim D, Cho J, Kim K, Shim YM. Chyle leakage patterns and management after oncologic esophagectomy: A retrospective cohort study. Thorac Cancer. 2014;5:391-97
19. Rao DVLN, Chava SP, Sahni P, Chattopadhyay TK. Thoracic duct injury during esophagectomy: 20 years experience at a tertiary care center in a developing country. Dis Esophagus. 2004;17:141-45
20. Milito P, Chmelo J, Dunn L, et al. Chyle leak following radical En bloc esophagectomy with Two-Field nodal dissection: Predisposing factors, management, and outcomes. Ann Surg Oncol. 2021;28(7):3963-72
21. Huang PM, Lee YC. A new technique of continuous pleural irrigation with minocycline administration for refractory chylothorax. Thorac Cardioc Surg. 2011;59:436-38

Indexed in: [PMC] [PubMed] [Emerging Sources Citation Index (ESCI)] [Web of Science by Clarivate]

22. Alamdari DH, Asadi M, Rahim AN, et al. Efficacy and safety of pleurodesis using platelet-rich plasma and fibrin glue in management of postoperative chylothorax after esophagectomy. World J Surg. 2018;42:1046-55
23. Bender B, Murthy V, Chamberlain RS. The changing management of chylothorax in the modern era. Eur J Cardiothorac Surg. 2016;49(1):18-24
24. Lai FC, Chen L, Tu YR, Lin M, Li X. Prevention of chylothorax complicating extensive esophageal resection by mass ligation of thoracic duct: A random control study. Ann Thorac Surg. 2011;91:1770-74
25. Natta V, Nguyen AT, Benharash P, French SW. Thoracoscopic thoracic duct ligation for persistent cervical chyle leak: utility of immediate pathologic confirmation. JSLS. 2009;13:430-32
26. Gupta D, Ross K, Piacentino V 3rd, et al. Use of LeVeen pleuroperitoneal shunt for refractory high-volume chylothorax. Ann Thorac Surg. 2004;78:e9-12
27. Foroulis CN, Desimonas NA. Massive pneumoperitoneum: A late complication of the Fenver pleuroperitoneal shunt. Ann Thorac Surg. 2005;80:13
28. EuroSurg Collaborative. Body mass index and complications following major gastrointestinal surgery: A prospective, international cohort study and meta-analysis. Colorectal Dis. 2018;20(8):O215-25
29. Cope C. Diagnosis and treatment of postoperative chyle leakage via percutaneous transabdominal catheterization of the cisterna chyli: Aa preliminary study. J Vasc Interv Radiol. 1998;9:727-34
30. Sileri P, Mele A, Stolfi VM, et al. Medical and surgical treatment of chronic anal fissure: A prospective study. J Gastrointest Surg. 2007;11(11):1541-48
31. Chen E, Itkin M. Thoracic duct embolization for chylous leaks. Semin Intervent Radiol. 2011;28:63-74
32. Itkin M, Kucharczuk JC, Kwak A, et al. Nonoperative thoracic duct embolization for traumatic thoracic duct leak: Experience in 109 patients. J Thorac Cardiovasc Surg. 2010;139:584-89

Indexed in: [PMC] [PubMed] [Emerging Sources Citation Index (ESCI)] [Web of Science by Clarivate]