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Case Report

Treatment of High-Output Thoracic Chylous Fistula with Transabdominal Embolization of Cisterna Chyli: A Case Report and Review of the Literature

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ABSTRACT

Postoperative thoracic chylous fistula is an infrequent complication after esophageal surgery. It represents a difficult management problem due to the serious mechanical, nutritional, and immunological consequences of the constant loss of protein and lymphocytes. A 55-year-old woman sequentially developed a high-output (2500 mL/day) thoracic chylous fistula and right-sided chylothorax, after a transhiatal total esophagectomy for adenocarcinoma of the distal esophagus. In this study, we discuss the case and treatment modalities in view of the literature. Multimodal procedures including low-triglyceride diet, sclerosing agents, repeated thoracentesis, and closed thoracostomy tube drainage were applied for treatment within two months after surgery. Finally, embolization of the cisterna chyli with liquid embolic agents produced rapid clinical and radiographic improvement. The procedure of opacification, catheterization, and embolization of the cisterna chyli was successful. Percutaneous transabdominal duct embolization is a safe, effective, and minimally invasive option to treat chylous fistula.

Keywords: Chyle fistula, embolization, esophagus, lymphangiography, surgery

INTRODUCTION

Chylous fistula is an uncommon form of ascites. It is defined as the leakage of the lipid-rich lymph into the peritoneal cavity. Damage or obstruction to the lymphatic system or one of its tributaries produces ascites with an opaque or milky appearance from the high-triglyceride content (1). Although the incidence of chyle fistula post surgery is low (1%-4%), this complication can present significant challenges including fluid and electrolyte abnormalities, malnutrition, and overwhelming infections, including peritonitis and empyema (2, 3).

Here in, we described and discussed a successful case of image-guided percutaneous embolization of cisterna chyli as a treatment modality for chylous fistula after failed conservative treatment modalities in an adult.

CASE PRESENTATION

A 55-year-old woman sequentially developed a high-output thoracic chylous fistula after a transhiatal total esophagectomy for adenocarcinoma of the distal esophagus. The mean daily drainage was 800 mL/day (range: 250-2500 mL/day). The mean concentration of triglycerides of 135 mg/dL in drainage fluid was accepted as chylous fistula. First right-sided and then left-sided chylothorax (bilateral) developed that lasted two and one month, respectively (Figures 1, 2). Multimodal procedures including low-triglyceride diet, sclerosing agents, repeated thoracentesis, and repeated closed thoracostomy tube drainage (bilateral) were applied for treatment within two months after surgery.

The cisterna chyli and lymph nodes were successfully embolized in the first intervention, but there was continuation of the chylothoraxes through development of collateral lymphatic ducts seen leaking into both hemithorax. Then, our radiology team tried second image-guided percutaneous cannulation and embolization of cisterna chyli (Figures 3, 4). Finally, embolization of the cisterna chyli with liquid embolic agents (Lipiodol Ultra-Fluide; Guerbet, France) produced rapid clinical and radiographic improvement (Figure 5). The patients remained recurrence-free for 24 months. As a formal procedure, the patient signed the informed consent form before treatment, interventive procedures, and publication.

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Figure 1. Chest X-ray; first left-sided chylothorax developed

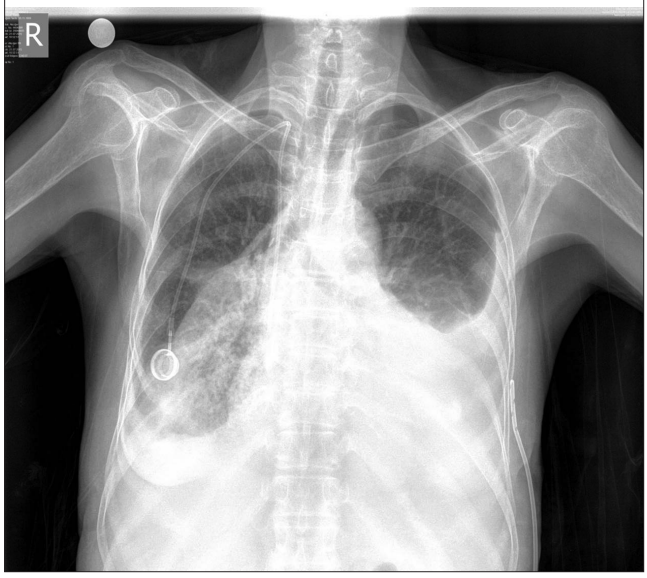
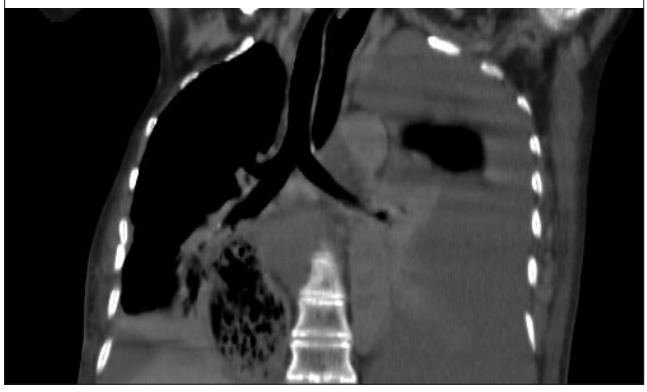


Figure 2. CT thorax demonstrating bilateral chylothorax
CT: computed tomography



DISCUSSION

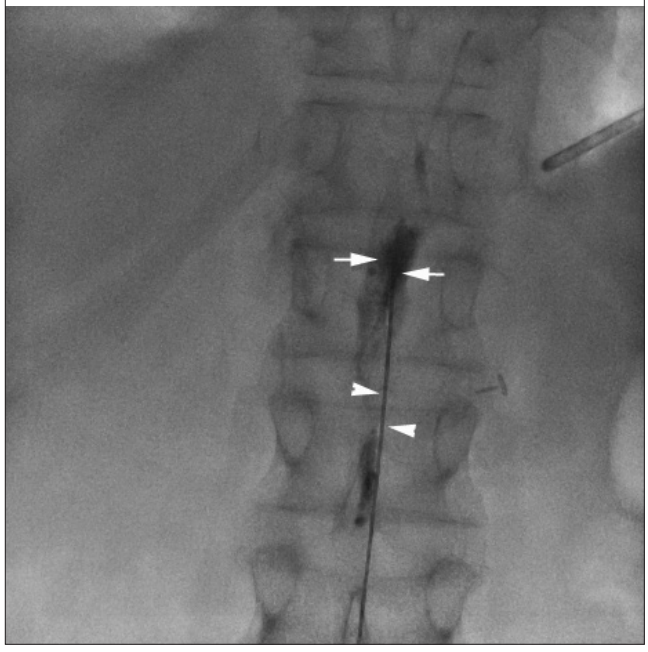
Damaged lymphatics most often heal spontaneously or direct lymph centrally via rich interconnected lymphatic collaterals, without any significant morbidity (1). High-output thoracic chyle fistula is a rare but potentially devastating and morbid condition.

Conservative approaches including fasting, dietary modification with low fat medium, thoracentesis, tube drainage, and octreotide administration are generally used as first treatment. It is well known that fasting and the administration of low fat medium chain triglycerides by mouth resolves approximately 50% of traumatic chylous fistulas (1, 2). The healing rate of nonoperative treatment modalities enormously varies; the maximum success rate in series is 70% (3).

In cases refractory to treatment, malnutrition, infection, immunologic complications, and high mortality rate of up to 50% have been reported. Several authors recommend interventional or surgical approaches (thoracic duct ligation, pleurodesis, and pleuroperitoneal shunt) in such cases (3, 4).

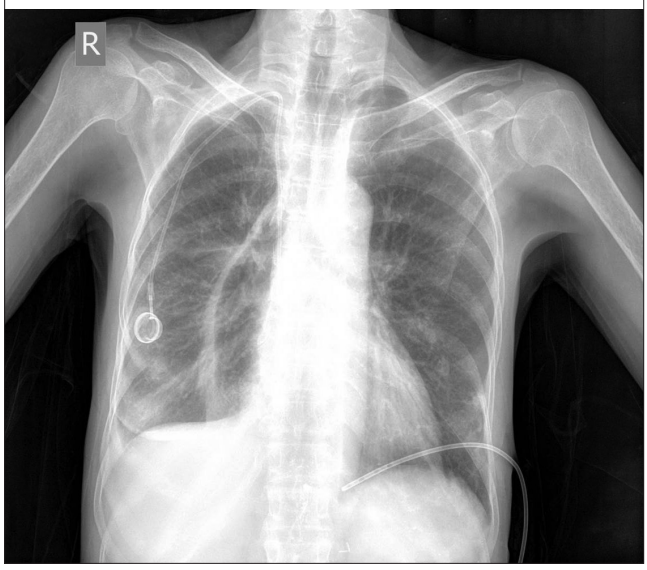
Figure 3. Fluoroscopic image shows 25 G needle in the inguinal lymph node (arrowheads) and opacification of the lymphatic vessels (arrows)

Figure 4. Fluoroscopic image shows 22 G needle (arrowheads) in the opacified cisterna chyli (arrows)



Pleurodesis, defined as promotion of adhesions between pleura and lung, may be a good option for the treatment of chylous fistula. It is reported that pleurodesis may be successful in 73%-

Figure 5. Chest X-ray; last condition



83% of cases with chylous fistula (3, 4). Also, there are many reports on intrapleural instillation of octreotide, concentrated glucose solution, tetracycline, streptokinase, or fibrin glue therapy (5-7). Huang (7) recommended pleurodesis using via continuous intrapleural irrigation with minocycline instead of conventional intermittent pleurodesis. There are new reports related with using orlistat and etilefrine for medical treatment (6). Picibanil (OK-432) is a lyophilized mixture of *Streptococcus pyogenes* with antineoplastic activity. It has capacity to produce a selective fibrosis of lymphoid tissue (7). Shimizu et al. (4) reported that OK-432 injection of into the thoracic cavity is effective and safe in the management of postoperative chylous fistula, and they revealed a success rate of 87%.

Sziklavari et al. (5) reported that the combination of radiotherapy and dietary restriction in the treatment of postoperative thoracic chylous fistula is more rapid response, safe, and successful. They recommend that radiotherapy should be indicated if the daily chylous drainage exceeds 450 mL after cessation of oral food intake. The major restriction to using radiation therapy for thoracic chylous fistula is the probability of acute and long-term side effects.

Cope reported that two-thirds of the patients with life-threatening thoracic chylous fistula can be safely treated with percutaneous transabdominal embolization of the thoracic duct (8). Matsumoto et al. (9) reported that lymphangiography is effective not only for differential diagnosis but also for treatment of chylous leakages. Schoellnast et al. (10) reported that the needle interruption of the cisterna chyli with subsequent resolution of the chylous fistula is successful in 50% of cases. In our case, our radiology team performed twice image-guided percutaneous cannulation and embolization of cisterna chyli. On the second attempt, embolization of the cisterna chyli with liquid embolic agents (Lipiodol Ultra-Fluide, France) was successfully performed, and it then produced rapid clinical and radiographic improvement.

Radical dissections increase the incidence of chylous fistulas more than in functional dissections and leads to prolonged hospital stay. It is reported that chylous fistula during the initial operation can be prevented by performing protective dissection and ligating lymphatic vessels, particularly in the posterior mediastinum. Surgical treatment of chylous fistula usually involves ligation of the thoracic duct. Repeated surgical procedures because of complications with postoperative chylous fistula have to be underwent up to 11% of the patients, and 9% experienced recurrence of their chylous fistula (2). Furthermore, surgical treatment also includes pleurodesis and decortication, resection and overlapping of leaking lymphatics, microsurgical or surgical repair of chylous fistula, video-assisted thoracoscopic surgery, closing with locoregional flaps, thoracotomy, pleura-venous or pleura-peritoneal shunts, and pericardial "window" (2). Shimizu et al. (4) found that surgical procedures give better results than conservative treatment modalities when the daily chyle flow exceeds 500 mL/day in adults. Zabeck et al. (2) reported that repeat surgery should be performed as soon as possible in postoperative chylous fistula with a high flow of >900 mL/day, because conservative treatment does not lead to an improved chyle flow. The success rate of the ligation of thoracic duct was reported to be 70%-90% in the largest clinical series (1, 10).

CONCLUSION

Currently, the morbidity and mortality associated with chylous fistula have been better with more energetic management strategies. In cases refractory to conservative treatment, transabdominal embolization may be a good treatment option. It is not only a possible first-line treatment in selected cases but also a primary diagnostic and therapeutic procedure in patients with massive chylous fistula.

Informed Consent: Written consent was obtained from the patient to reproduce information and photographs appearing in this case.

Peer-review: Externally peer-reviewed.

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