CASE REPORTS

TREATMENT OF POST-OPERATIVE CHYLOTHORAX: THE ROLE OF INTRANODAL LYMPHANGIOGRAPHY AND THORACIC DUCT DISRUPTION

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Abstract

Thoracic duct embolization has been increasingly adopted as a first-line therapy of chylothorax and this procedure includes lipiodol lymphangiography, thoracic duct access and embolization. Lymphangiography itself has a therapeutic role, with volume-dependent success rates of 37%-97% and even a reported 100% success rate in outputs of < 500 mL/day. We present a clinical case of a 48-years-old man diagnosed with esophageal squamous cell carcinoma, who underwent esophagectomy and presented with post-operative high-output (> 1L/day) chylothorax; thoracic duct embolization was proposed. Even though thoracic duct access and embolization were not achieved due to technical and anatomical factors, lipiodol lymphangiography and possibly thoracic duct maceration (after several punctures/attempts) contributed to the clinical success of the procedure, and this chylothorax with output values superior to those reported in the literature resolved within three days. As such, the therapeutic role of intranodal lymphangiography and thoracic duct disruption should be taken into account.

Keywords: *Chylothorax; Lymphangiography; Lipiodol; Thoracic duct; Interventional Radiology.*

INTRODUCTION

Chylothorax (the presence of chyle in the pleural space) is an uncommon event with high risk of morbidity and mortality.[1] Traumatic/iatrogenic etiology accounts for 54% of cases,2 with rates of up to 4% after esophagectomy.[3]

Clinically, chylothorax most commonly presents with dyspnea.[2] Thoracentesis is performed for palliative and diagnostic purposes. Chyle is usually an odorless fluid, milky in its macroscopic appearance.[2] Sample triglyceride levels of > 110 mg/dL and chylomicrons are diagnostic of chylothorax.[3]

Management can be conservative, surgical or a percutaneous image-guided treatment. Thoracic duct embolization (TDE) has been gaining traction as a first-line therapy. However, the therapeutic roles of lymphangiography and thoracic duct disruption (TDD) should also be taken into account.

In this paper, we present a case of post-esophagectomy high-output chylothorax treated with lipiodol lymphangiography and a brief literature review of chylothorax percutaneous radiological management.

CASE REPORT

A 48-years-old male active smoker with history of p16-positive oropharyngeal squamous cell carcinoma cT2cN2cM0 three years prior, with poor adherence to CT-RT treatment and follow-up, presented with de novo dysphagia and weight loss. A stenosing esophageal squamous cell carcinoma located 24 cm from incisors was diagnosed. The patient underwent McKeown esophagectomy and Stamm feeding jejunostomy procedures.

In the following days, bilateral pleural effusions developed and an output of 900-1700mL/day of milky fluid was detected in the cervical and thoracic drains.

Figure 1 *Right inguinal intranodal lymphangiography: fluoroscopic view of lipiodol injection after ultrasound guided access to the lymphatic system with a 26G needle. The same approach was used in the contralateral side.*

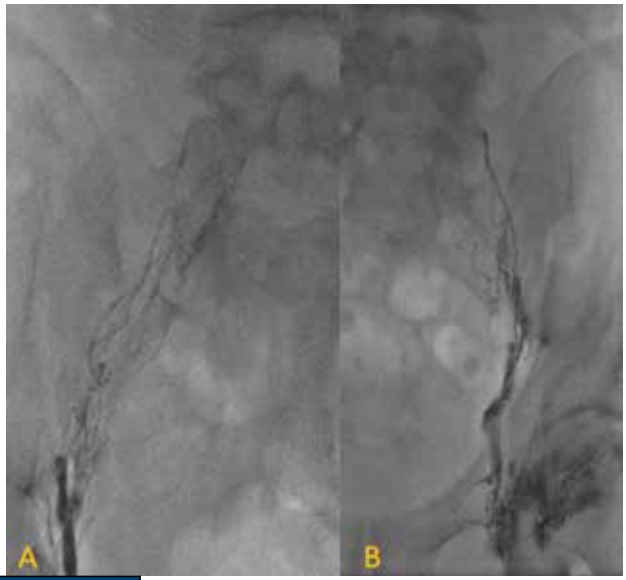

Figure 2 *Pelvic spot radiographs showing bilateral (A-right side; B-left side) progression of lipiodol into the lumbar lymphatic channels.*

Pleural fluid was analysed, with sample triglyceride levels of 294 mg/L and presence of chylomicrons – the diagnosis of chylothorax was made, and TDE was proposed.

Bilateral inguinal intranodal lymphangiography was performed with injection of 10mL lipiodol on each side (Fig. 1).

Slow cranial progression of lipiodol into the lumbar lymphatic channels was documented in spot radiographs (Fig. 2).

Although cisterna chyli was not identified, an attempt was made to puncture the main dilated retroperitoneal lymphatic channels (Fig. 3). Several punctures were made, without successful TDA.

Even though TDE could not be performed, the post-lymphangiography cervical and thoracic drain output was reduced to only 50mL the following day, eventually becoming vestigial three days after the procedure. The patient did not present lower limb lymphedema nor other procedure-related symptoms, afterwards.

This case of high-output (> 1L/day) chylothorax was treated with therapeutic lymphangiography, with TDD possibly contributing to the clinical success, as discussed below.

DISCUSSION

The optimal clinical management of postoperative chylothorax is yet to be established.[1] In low-volume leaks (<1 L/day), an initial conservative approach is preferred.[1,3,2]

Criteria for non-conservative treatment are not well defined,[2] but invasive procedures are often required in high-output leak (>1 L/ day), if the chylothorax has not resolved after 2 weeks and in underlying neoplastic etiologies.[1,4,2] This includes surgical treatment (talc pleurodesis, pleurectomy, thoracic duct ligation) with success rates of 67-93%[1,5], but rates of morbidity and mortality of 38.8% and 25%, respectively.[1]

Thoracic duct embolization (TDE) has gradually gained acceptance as first-line therapy for post-traumatic chylothorax.[1] The major steps of this procedure are ethiodized oil (lipiodol) lymphangiography, thoracic duct access (TDA) and TDE.[1,4,3,2]

Lymphagiography was commonly obtained with transpedal access,[1,5] an approach largely replaced by intranodal lymphangiography[6] in which bilateral inguinal lymph nodes are punctured (under ultrasound-guidance) by a 25-26G needle with its tip positioned between the nodal cortex and hilum.[1,2,5] Then, lipiodol is injected (rate of about 1 mL/ 5 minutes)[1,2] and spot radiographs are acquired to assess the cranial progression of lipiodol[2] until upper lumbar lymphatics and the cisterna chyli are opacified.[1]

To obtain TDA, a 21G Chiba needle is advanced to the target structure (cisterna chyli or main retroperitoneal

lymphatic channels) under fluoroscopy.[1,2,5] When successful puncture is achieved, a microcatheter is inserted over a relatively stiff guidewire into the thoracic duct – at this point, digital subtraction lymphangiography with iodinated contrast injecton is performed to better depict the leakage site.[1]

After the lymphatic leak is identified, the thoracic duct is embolized with a combination of microcoils and Onyx glue or n-butyl-2-cyanoacrylate (NBCA) liquid glue mixed with lipiodol (usually at a 1:2 ratio),3 with success rates of 90%.7

Of these three steps, TDA is the most challenging and heavily operator-dependent, with technical success rates of 70%,8 thus dictating the technical outcome of TDE.[1]

However, intranodal lymphangiography itself can be curative, with success rates depending on the volume of the leak.[3] Lymphangiography is a necessary step to TDE procedure and has been shown to stop leakage in 37% to 70% of cases without additional procedures.[3,2] In fact, Gruber-Rouh et al. reported a 97% success rate of lymphangiography for iatrogenic lymph leak volumes of < 200 mL/day,[9] and Yannes et al. reported a 100% success rate in outputs of < 500 mL/day and without extravasation.10 Authors suggest the leak is reduced due to the viscosity and embolic properties of lipiodol,[4] and the inflammatory reaction that occurs during lipiodol extravasation,[4] producing scar tissue and promoting the resolution of the leak within days or weeks.[1]

Additionally, if TDA is not possible, thoracic duct disruption (TDD) may be attempted, with multiple needle punctures. The goal is to macerate the cisterna chyli or main retroperitoneal lymphatic channels that can be visualized, thus reducing lymph flow and promoting spontaneous healing of the leak site.[1] Also, the effect of lipiodol, the inflow of venous blood through perforated lymph channels and the pressure exerted by hematomas at puncture sites can contribute to the mechanism of action.[1] Itkin et al reported a 72% clinical resolution of chyle leak with TDD7, with many others reporting its therapeutic effects. [8]

In our clinical case, although TDA and TDE could not be achieved, the high-output leak could be successfully managed by therapeutic lipiodol lymphangiography; maceration of lumbar lymphatic channels may also have contributed to the therapeutic success, as described in the literature[7,8] thus resolving the chylothorax in the following days.

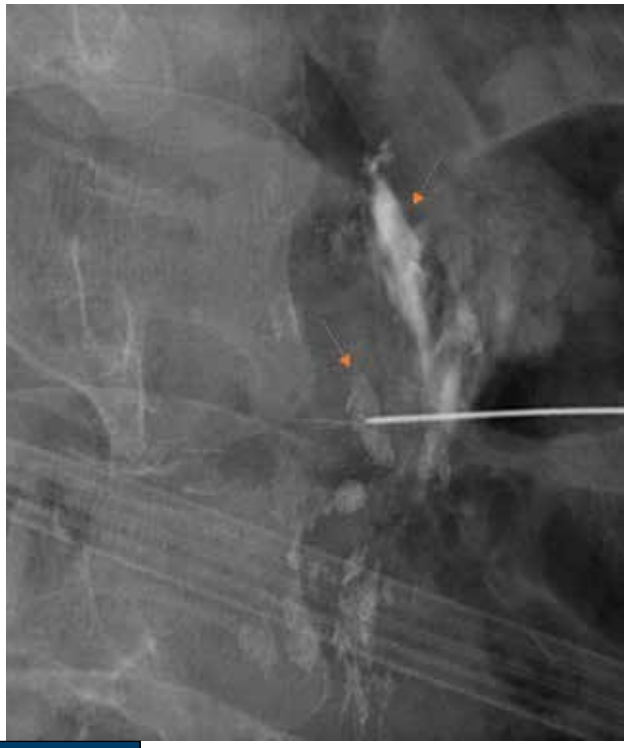

Figure 3

Attempt of puncturing the main dilated retroperitoneal lymphatic channels at L2 vertebra level (arrow), with 21G Chiba needle. Cisterna chyli was not unequivocally identified. Several punctures were made, without successful access to the thoracic duct. A small, contained lipiodol leak or hematoma can be seen (dotted arrow), possibly due to multiple punctures at this site.

CONCLUSION

Thoracic duct embolization (TDE) is becoming the preferred method of treatment for chylothorax. Even if TDA and/or TDE are not feasible due to anatomical or technical issues, the implied lipiodol lymphangiography can be therapeutic, as presented in our high-output leak clinical case. Additionally, thoracic duct disruption may be attempted with multiple needle punctures, also contributing to the clinical resolution of the chylothorax.

Protection of humans and animals

The authors declare that the procedures were followed according to the regulations established by the Clinical Research and Ethics Committee and to the 2013 Helsinki Declaration of the World Medical Association.

Data confidentiality

The authors declare having followed the protocols in use at their working center regarding patients' data publication.

Patient consent

Obtained.

Conflicts of interest

All authors report no conflict of interest.

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