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Chlyous leak after radical oesophagectomy: Thoracic duct lymphangiography and embolisation (TDE)—A case report

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ABSTRACT

INTRODUCTION: Chyle leak after oesophagectomy is highly morbid and may carry significant mortality if treatment is delayed. Identification of the site of leakage and surgery may be plagued by failure.
PRESENTATION OF CASE: We describe a case of chyle leak after oesophagectomy. Lymphangiography revealed the site of chyle leak to be an aberrant duct that would have been difficult to identify surgically. Radiological coiling and embolization successfully treated the leak.
DISCUSSION: The gold standard for treatment of chyle leak or chylothorax after oesophagectomy was a re-operation, either open or thoracoscopic, to ligate the thoracic duct. The interventional radiological technique employed in our case was not only efficacious in stopping the leak, but had the added advantage of identifying the site and highlighting the anatomy hence avoiding a morbid reoperation. The literature is reviewed.
CONCLUSION: The report and review confirm that lymphangiography followed by coiling and embolization for chylothorax post oesophagectomy is safe and effective in a majority of cases.

1. Introduction

Thoracic duct anatomy must be understood in the context of its embryology. The first lymph sacs to develop in the human body are the paired jugular lymph sacs at the junction of the internal jugular and subclavian veins. The jugular lymph sacs communicate inferiorly with the cisterna chyli. Channels that join the jugular lymph sacs to the cisterna chyli become the thoracic duct (left lymphatic duct) and the right lymphatic duct. Disturbances in processes that govern the formation of these lymphatic channels can result in anatomical variations of the thoracic duct [1–3].

The thoracic duct is a tubular structure that is 2–3 mm in diameter and varies in length from 38 to 45 cm. It begins in the abdomen at the level of the second lumbar vertebra. It enters the thorax through the aortic opening of the diaphragm between the aorta and the azygous vein. The thoracic duct then passes cephalad on the right side of the aorta and crosses to the left side at the level of the fifth cervical vertebra where it joins the venous system. The previously described course of the thoracic duct has an incidence of 60–65%. The thoracic duct may also be partially duplicated in 15–20% of the time or fully duplicated in 15%. These anatomical variations provide a reason why the thoracic duct gets damaged during surgery despite the surgeon's vigilance [3–5].

The complication rate of oesophageal surgery is relatively high, in the region of 30–40%. The thoracic duct can often be damaged during mobilisation of advanced oesophageal cancers, whether via a right thoracotomy or through the *trans*-hiatal route. A comprehensive review reports chylothorax occurring in up to 10% of patients after blunt *trans*-hiatal oesophagectomy [6]. An incidence of 2–3%, during open trans-thoracic resection, is commonly reported [7].

In the event of thoracic duct injury, chylothorax usually presents in the first 7 days after surgery, when the patient has commenced oral intake, or jejunostomy feeds. A massive increase in chest drainage occurs that if left untreated, results in malnutrition and significant immune suppression, with a markedly reduced CD4 count from the subsequent white cell loss [8].

Leaks of less than 500 mL/day may resolve with enteral feeding using medium-chain triglyceride. Octreotide/somatostatin and etilefrine therapy may also be highly efficacious in the conservative management of low volume chylothorax. High volume leaks, however, warrant immediate re-exploration as the damaged thoracic duct is usually easily identified, following a bolus of "cream" at the time of re-exploration [7]. Open or thoracoscopic exploration had been established as the gold standard of treatment.

We describe a case of chyle leak post radical 2- stage oesophagectomy (Ivor-Lewis) with two-field lymph node dissection. The gold standard of treatment that we grew accustomed

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to was not employed in the management of our case but rather lymphangiography was used to identify the anatomy and the site of the leak. The leak was then successfully treated by means of radiological coiling and embolization.

2. Case report

A 69-year-old male presented with dysphagia to solids and vomiting. Weight loss was denied, and appetite was good. Past medical history was significant for hypercholestrolaemia, diverticular disease, renal calculi, parathyroidectomy for hyperparathyroidism and benign prostatic hypertrophy.

Endoscopy revealed a Seiwert I type junctional/distal oesophageal cancer. Biopsies confirmed adenocarcinoma. Radiology including CT, EUS and PET staged this tumour at T3N0M0. The cancer multidisciplinary meeting consensus was that the criteria for "Magic Protocol" were met.

Treatment with 3 cycles of ECF was followed by radical subtotal oesophagectomy (Ivor-Lewis) with two-field lymph node dissection. A feeding jejunostomy was fashioned during the abdominal stage as is routine in our practice. The thoracic duct was excised during the chest stage of the operation. An apical, basal and mediastinal chest tubes were inserted for drainage. Thoracic and abdominal "pain buster" catheters were positioned intra-operatively for analgesia.

The patient was managed according to our enhanced recovery program. He was mobilized out of bed on the evening of surgery. Vivonex (Nestle Health Science, U.S.A), low fat jejunostomy feeding, was commenced at a rate of 25 mls/hour on day 1 post surgery and was increased by 25 mls every 24 h to a maximum of 75 mls/hour on day 3 post surgery. The output in the chest drains combined averaged less than 200 mls/day on day 3. There was minimal output in the apical drain and therefore it was removed. The basal and mediastinal drains were left in place until a time when radiological assessment of the anastomosis was feasible. Contrast swallow on day 5 confirmed anastomotic integrity and demonstrated good gastric emptying. Apical and basal chest drains were removed by day five, as there was minimal drainage. Chest X-rays showed clear lung fields bilaterally.

Acute respiratory failure developed on day 8 post operatively. The patient was re-admitted to the intensive care unit. Investigations including CT pulmonary angiogram demonstrated the presence of a large effusion on the left side, almost obliterating the entire pleural space (Fig. 1a and b). The right pleural space was unremarkable. All other pathology was excluded. Ultrasound guided drainage of the left pleural effusion revealed that it was chylous in nature. A pigtail drain was inserted in the left chest cavity and a trial of conservative therapy was commenced. Jejunostomy feeds were ceased. Total paraenteral nutrition was commenced. Pantoprazole 40 mgs IV twice daily and Octreotide 200 units SC three times daily were given to slow down gut function.

It is our experience that when a major chyle leak ensues as a result of thoracic duct damage, the volume produced is usually high from the onset. What was intriguing in this case report was the delayed onset of the chyle leak despite having commenced jejunostomy feeding from day 1 post operatively.

Unfortunately, a 5-day trial of conservative therapy was fraught with failure. At almost 2 weeks since surgery, it was felt that the condition of the tissues in the thorax at that point of time would not favour surgical intervention. A radiological approach was therefore sought.

Percutaneous thoracic duct embolization was arranged. Technique: a 25-gauge needle was introduced into the largest right superficial inguinal lymph node under ultrasound guidance. Lipiodol was injected to opacify the lymphatic system up to the cysterna chyli, which was at the level of T12/L1 disc (Fig. 2a). The cysterna chyli was then punctured with a 22-gauge needle and a V18 guidewire (Boston Scientific, U.S.A) was introduced. The latter was exchanged for a Renegade microcatheter (Boston Scientific,U.S.A), which was used to perform a lymphangiogram with the water-soluble contrast medium ioversol 370 flushed with 5% dextrose (anticipating the use of cyanoacrylate which sets in the presence of ionic solutions). The lymphangiograms showed the presence of an aberrant thoracic duct running to the left of the thoracic vertebral column. There was extravasation of contrast medium at the level of T9 (Fig. 2b). A small channel draining towards the right main duct was also seen. The main duct did not opacify consistent with surgical obliterated. Three Tornado coils 50 mm in length and 0.018 in. in diameter (Cook Medical, U.S.A) were deployed in the aberrant duct near the region of extravasation. This was followed by injection of 1 ml of cyanoacrylate (Histoacryl by Braun, Germany) mixed with Lipiodol ultra fluid (Guerbet, France). A repeat lymphangiogram confirmed the absence of further extravasation (Fig. 2c)

Flow from the left pleural drain slowed and drainage had almost ceased by day 2 after the procedure. No pleural fluid was visible on a radiograph of the chest nine days after the lymphangiogram and embolization. The patient's diet was upgraded and he was discharged day 5 post procedure (13 days post-operation).

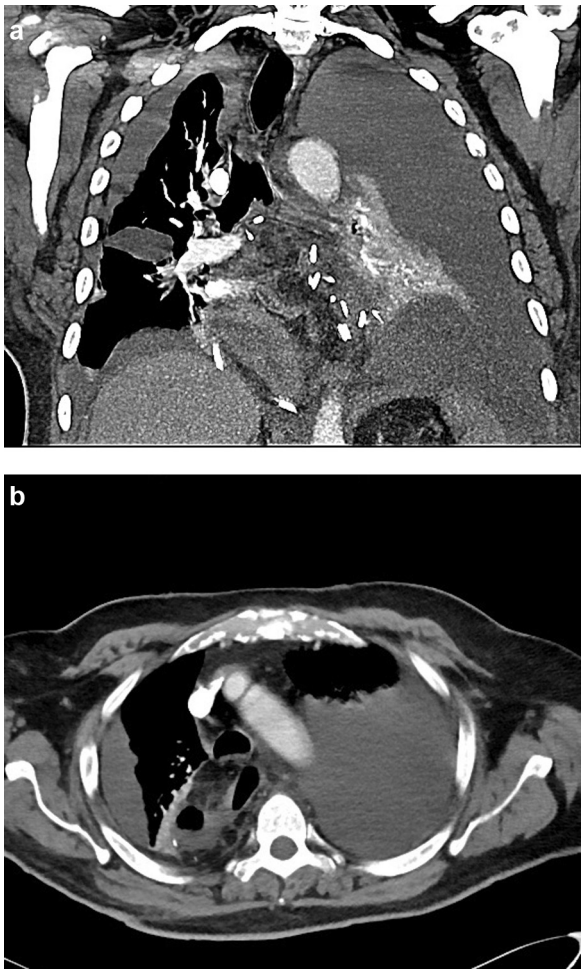

Fig. 1. (a) and (b) coronal and transverse views: "white out" resulting from a large left pleural effusion.

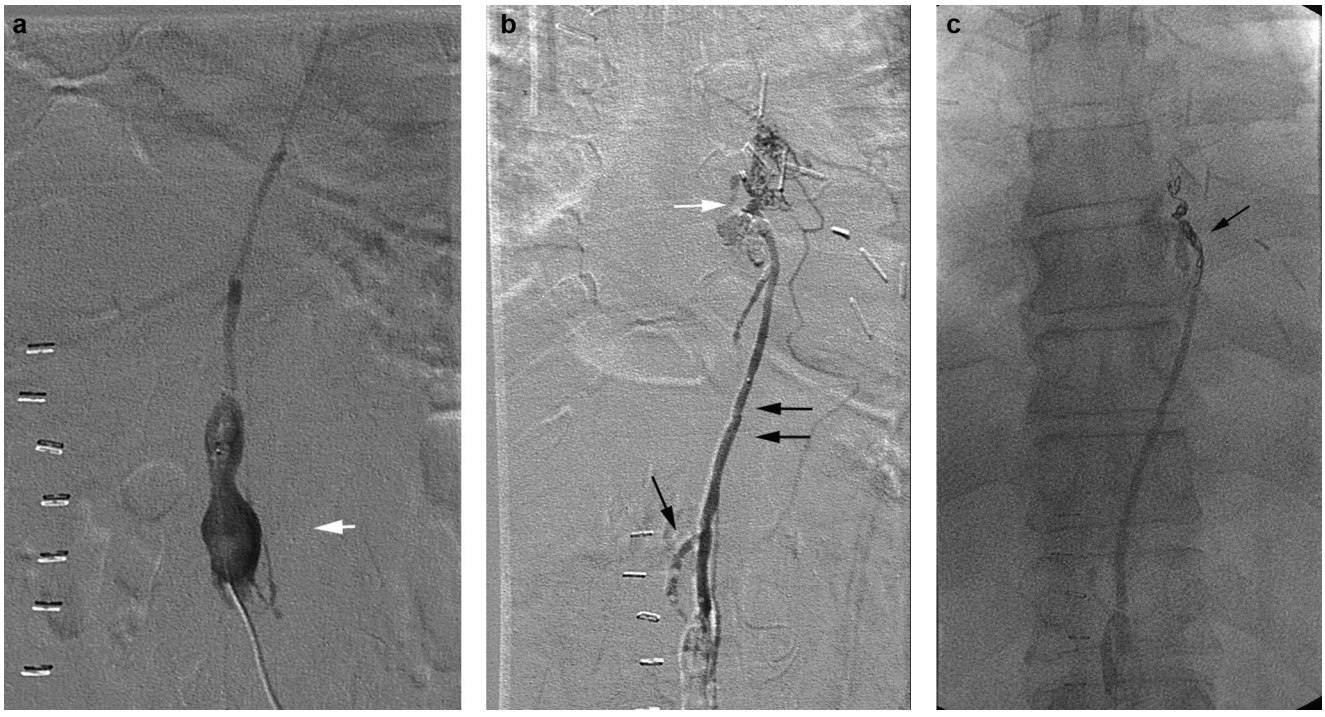

Fig. 2. (a) Lymphangiogram: Lipoiodol injected into superficial inguinal node fills cysterna chyli (white arrow). (b) Lymphangiogram: Aberrant left thoracic duct (2 black arrows) extravasation at T9 (white arrow). A single black arrow points towards a side branch of the thoracic duct. (c) Lymphangiogram: Occlusion of leak following coiling and embolization (black arrow). The side branch has also been occluded.

3. Discussion

Chylothorax after oesophagectomy should be suspected when there is an unexplained high-volume chest tube output which could appear milky after enteral tube feeding is commenced. The exact diagnosis of chylothorax is based on the presence of chylomicrons in the pleural fluid. Chylomicrons are molecular complexes of proteins and lipids that are synthesised in the jejunum and transported via the thoracic duct to the circulation. They are only found in the circulation post-prandially with a peak 3 h after eating [9]. Cytological analysis of fluid stained with Sudan III will demonstrate chylomicrons, which although sensitive, is not specific and therefore should be combined with complementary fluid analysis. In centres with available facilities, lipoprotein analysis demonstrating chylomicrons is the gold standard. Where this facility is not available, institutions rely on the measurement of fluid cholesterol and triglyceride levels. A pleural fluid triglyceride levels >1.24 mmol/l (110 mg/dl) with a cholesterol <5.18 mmol/l (200 mg/dl) is diagnostic of chylothorax [5]

Alexiou et al. [10] carried out a retrospective analysis of 523 patients with cancer of the oesophagus or the gastro-oesophageal junction who underwent oesophageal resection between January 1987 and November 1997. Chylothorax occurred in 21 patients (4.0%). Radicality of dissection was the only apparent predisposing factor.

The management of early post-operative chylothorax requires rigorous scrutiny. The aetiology of the chylothorax, the flow rate and patient condition dictate the preferred management. Interventions are only required if unresponsive to medical management.

Surgical therapy is recommended in cases where despite conservative management, the patient drains more than 1.5/day in an adult or >100 ml/kg body weight per day in a child [11], leaks chyle at a rate of >1 l/day × 5 days [8] or has persistent chyle flow for more than 2 weeks [12]. Surgery is also recommended if there has been a rapid decline in nutritional or immunological status despite conservative management [13–15]. Chylothorax following oesophageal surgery managed with re-exploration is associated with a mortality of 10% compared to a mortality of 50% if managed conservatively [16,17].

Patients with iatrogenic chylothorax after oesophagectomy who are good surgical candidates and in whom the site of the leak is identified, do well following surgical re-exploration. When re-operation is not delayed and simple duct closure of any type is performed, patients have little added morbidity and the reported success rates are around 90% [18]. Ligation of the thoracic duct via a thoracotomy has been considered to be the gold standard approach. Video-assisted thoracic surgery and ligation of the duct has also proven to be a safe and effective approach [19].

Ligation of the thoracic duct is successful in 90% of patients when performed just above the right hemi-diaphragm [20]. Ligating at that site has the advantage of halting flow from any unidentified accessory ducts [21,22]. Collateral circulation re-directs the chyle around the ligation point ensuring that the chyle still completes its journey to the circulation. If the leak is in the region of the neck or upper thorax, the thoracic duct is ligated in the area known as Poirier's triangle between the arch of the aorta, internal carotid and vertebral column [23].

In thoracoscopic ligation, up to 3 ports are inserted strategically between different ribs and the thoracic duct is sought. A short segment of the duct is excised before clipping the remaining ends [13].

If the leak is not identifiable on either thoracoscopy or thoracotomy, then mass ligation of all the tissue between aorta, spine, oesophagus and pericardium is performed [13]. Extensive dissection to find the duct is discouraged reducing the risk of further trauma and leak. Pleurectomy or pleurodesis with talc or glue have been described as alternative options [24]. In cases of loculated or complicated chylothorax, pleural decortication with pleurodesis may be performed [13]. In patients that are unfit for major surgery,

a pleuroperitoneal shunt may be useful. It minimises the nutritional or immunological deficits seen in chylothorax [20].

The accumulation of experience in treating chylous effusions has significantly broadened the adoption of thoracic duct embolization to treat chylothorax. A cannulation and embolisation technique used by Cope et al. [25] to treat chylothorax was curative in patients with demonstratable duct leakage. However, reproducibility and success rates have varied in different centres. More recently Boffa et al. [26] have used the technique of thoracic duct embolisation or disruption with very good effect in patients with chyle leak post thoracic surgery and Litherland et al. [27] described a case report where CT guided disruption of the lymphatics had good effect in the management of high output chylothorax.

Matsumoto et al., performed lymphangiography on 9 patients that were unlikely to respond to conservative measures. They found that lymphangiography not only identified the site of the leak but also led to the leak resolving in all cases. They recommend early lymphangiography in cases unlikely to be cured by conservative methods only [28].

The feasibility and effectiveness of percutaneous thoracic duct embolization or interruption have been reported in four papers.

Marcon et al. [29] reviewed the existing literature on percutaneous management of chyle leaks. The authors evaluated five case series and three case reports inclusive of 90 patients in whom percutaneous treatment for chylothorax was attempted between 1998 and 2004. Percutaneous treatment resulting in successful resolution of the chylothorax was achieved in 69% of the patients. The authors concluded that such percutaneous management of chyle leaks is feasible, with low morbidity and mortality rates and a high rate of effectiveness.

A retrospective review of 34 patients was similarly conducted by Nadolski et al. [30] to assess the technical and clinical success of thoracic duct embolization for iatrogenic chylous effusions. Thoracic duct embolization was technically successful in 24 of 34 patients (70.6%).

A retrospective review of 109 patients was conducted by Itkin et al. [31] to assess the efficacy of thoracic duct embolization or interruption for the treatment of high-output chyle leak caused by injury to the thoracic duct. The authors concluded that catheter embolization or needle interruption of the thoracic duct is safe, feasible, and successful in eliminating a high-output chyle leak in the majority (71%) of patients. Further more, the authors stated that this minimally invasive procedure, although technically challenging, should be the initial approach for the treatment of a traumatic chylothorax.

Pamarthi et al. [32] retrospectively report the indications, technical approach, and clinical outcomes of thoracic duct embolization and thoracic duct disruption in 105 patients with symptomatic chylous effusions. The technical success rate was 79% in this series. The authors concluded that thoracic duct embolization and thoracic duct disruption are safe and effective minimally invasive treatments for traumatic thoracic duct injuries.

4. Conclusion

TDE in this case was technically satisfactory. Chyle leak may occur from aberrant sites as in this case and lymphangiograohy is helpful. High volume chyle leak requires prompt management before immune compromise occurs. TDE is successful in a majority of cases (61–71%) and so should be utilised in reserving surgery for failed percutaneous management.

Conflict of interest

There is no conflict of interest.

Funding

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Ethical approval

This was purely an observational case study. The patient's management and outcome were unaltered. There was no research that involved the patient.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

All of the authors have participated in caring for the patient, preparing and designing the intervention under review, acquiring the data, analysing and interpreting the data, drafting the article, revising it critically for important intellectual content and approving the final version to be submitted.

Guarantor

Professor Greg L. Falk.

Disclosure

All of the authors have participated in caring for the patient, preparing and designing the intervention under review, acquiring the data, analysing and interpreting the data, drafting the article, revising it critically for important intellectual content and approving the final version to be submitted.

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