Postoperative chylothorax with a duplicated left-sided thoracic duct: a case report and review of the literature

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Abstract

Postoperative chylothorax is a potentially lethal complication after esophagectomy. A 53-year-old woman underwent subtotal esophagectomy. The thoracic duct was resected due to swollen lymph nodes. Postoperative chylothorax was diagnosed but conservative treatment was ineffective. Lipiodol lymphangiography revealed leakage from a duplicated left-sided thoracic duct. Left-sided video-assisted thoracoscopic ligation of the left-sided thoracic duct was performed. Because anatomical variations in the thoracic duct contribute to refractory chylothorax, lymphangiography is useful in detecting the position of thoracic duct injury as well as any duct anomalies. Based on lymphangiography, left-sided video-assisted thoracoscopic surgery could be considered in case of left-sided thoracic duct injury.

Introduction

Postoperative chylothorax is a serious complication. The occurrence of chylothorax is associated with an increase in the incidence of major complications and malnutrition, leading to a risk of hospital mortality. Early diagnosis and effective management determine the outcome. This is a case of postoperative chylothorax caused by injury to the left-sided thoracic duct (TD), ligated by left-sided video-assisted thoracoscopic surgery in right decubitus position.

Case presentation

A 53-year-old woman presented with obstruction to food passage. Computed tomography (CT) revealed tumor invasion to the descending aorta (Fig. 1a) and swollen lymph nodes at the left paracardial area (Fig. 1b,1c). Clinical findings, determined based on the Japanese Classification of Esophageal Cancer, 11th edition [1], showed squamous cell carcinoma (cT4b, cN1, cM0, cStageIVa). The patient received three courses of chemotherapy of docetaxel, cisplatin and 5-fluorouracil.

Subtotal esophagectomy by thoracotomy with three-field lymphadenectomy in left decubitus position and reconstruction at the neck via the retrosternal route was performed. The left mediastinal pleura was partially resected around the tumor. TD was resected at the lower intrathoracic esophagus because the lymph nodes along the TD were swollen. A 24-Fr thoracic drain was inserted into the right thorax and negative pressure up to 10 cmH_2O was applied. The histological diagnosis was no residual cancer cell (pT0, N0, M0, pStage0).

Peripheral parenteral nutrition was initiated, but a large amount of pleural effusion flowed on postoperative day (POD) 1. After refilling phase, pleural effusion slightly decreased and an elemental diet started (Elental; EA pharma, Japan) via a gastric fistula catheter on POD 2. However, the amount of discharge again increased and postoperative

chylothorax was diagnosed. Octreotide acetate (300 μg/day) was administered from POD 4, but ineffective. On POD 6, lipiodol lymphangiography was performed. The bilateral inguinal lymph nodes were accessed under ultrasound guidance and lipiodol was injected. A fluoroscopic apparatus showed accumulation at cistern of chyle. A 3-Fr sheath was inserted there under CT guidance. Interruption of the right-sided TD and injury to a duplicated left-sided TD were shown (Fig. 1d). Temporal embolization of the left-sided TD by cyanoacrylate was performed. This, however, achieved only a small decrease of effusion. Etilefrine hydrochloride (120 mg/day) was administered from POD 7, but effusion continued and serum albumin level decreased. Because further conservative treatment had a high risk of nutritional disorder, surgical intervention was deemed necessary. Ligation of the left-sided TD was performed by left-sided video-assisted thoracoscopic surgery (VATS) in right decubitus position on POD 8. The cut-end of the left-sided TD was easily detected by wiping with gauze and magnifying the leakage point detected by lymphangiography under VATS and was ligated precisely (Fig. 2a, b). The thoracic drain was relocated into the left thorax. The amount of pleural effusion decreased and the thoracic drain was removed on POD 10 (2 days after reoperation). Finally, the patient was discharged on POD 18 (10 days after reoperation).

Discussion

Postoperative chylothorax is a rare but lethal complication after esophagectomy because chylous leaks result in the loss of both humoral and cell-mediated immunity. We previously reported the incidence of this complication as 2.4% in open esophagectomy and 2.2% in minimally invasive esophagectomy from a Nationwide Database in Japan [2].

Embryologically, the TDs are formed bilaterally. The cranial side of the left-sided TD and the caudal side of the right-sided TD are usually regressed. Adachi reported a classification, specifying the incidence of a duplicated left-sided TD at 4% [3]. Anomalies of the TD, including a duplicated left-sided TD, may hinder surgeons to accurately identify the TD, which increases the risk of TD injury during esophagectomy. In this case, adhesion around the tumor was strong and partial resection of the left mediastinal pleura was required. However, the existence of the left-sided TD was not suspected because there was no significant lymphorrhea.

Lipiodol lymphangiography is one of the modalities for the identification of chylous leakage as well as for the minimally invasive embolization [4]. However, the success rate of this procedure is uncertain.

The treatment strategy for postoperative chylothorax is not well-established. Marts et al.

reported that conservative managements alone allow closure of the fistula within 1-62 days (median 13.3 days) in up to 79% [5]. In contrast, Orringer et al. suggested rapid reoperation to close chylous leak before postoperative adhesions developed [6]. There is no consensus on how early and what volume of chyle output should indicate a need for surgical intervention. Persistent chest tube drainage exceeding 10–12 ml/kg after the initiation of conservative measures would be an indication for early ligation [7]. An important factor to consider in all cases is whether the patient could withstand surgery. Only when surgery is expected to improve the patient's postoperative general condition, it would be an appropriate treatment option. Recently, VATS has been suggested as an alternative to thoracotomy in the treatment of chylothorax, due to the magnified view for detection of an injury to the TD. When the cases of postoperative chylothorax caused by injury to the left-sided TD in PubMed were researched, two reported cases were found [8, 9], both from Japan. In all cases, the right-sided TD was resected. In contrast to the reported cases, which were performed in prone position, ours was performed by left-sided thoracoscopy in right decubitus position. The advantage of right decubitus position was that it allowed us to perform a transition from thoracoscopy to thoracotomy in case of any difficulty of ligation or strong adhesion. Retraction of the left lung was considered as a problem in right decubitus position. However, by detecting the leak point from lipiodol

lymphangiography, partial retraction was easy.

The present case has clinical significance for the detection of the cause of refractory postoperative chylothorax as a duplicated left-sided TD discovered by selective lymphangiography.

Conclusion

Anatomical anomalies of the TD increase the risk of TD injury during operation. Injuries to the TD due to anomalies often result in persistent postoperative chylothorax. Lymphangiography is useful to detect the location of TD injury including anomaly. Based on lymphangiography images, surgeons may be able to consider less invasive approaches for the treatment of TD injuries. Left-sided VATS could present a useful tool to the left-sided TD.

Ethics declarations

The authors declare we obtained permission from ethics committee in our institution.

Informed consent

Written informed consent for publication was obtained from patient.

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Figure legends

Fig 1 (a) Preoperative CT revealed tumor invasion to the descending aorta. (b, c) Swollen lymph nodes at the left paracardial area (yellow arrow). (d) A lymphangiography revealed interruption of lymph flow of the right-sided TD (red arrow) and leakage from a duplicated left-sided TD (yellow arrow).

Fig 2 (a) Thoracoscopic findings revealed a duplicated left-sided TD (dashed line) and a point of TD injury (arrowhead). (b) Ligation of the TD was completed.

D; diaphragm, *A*; aorta, *L*; lung

Figure 1

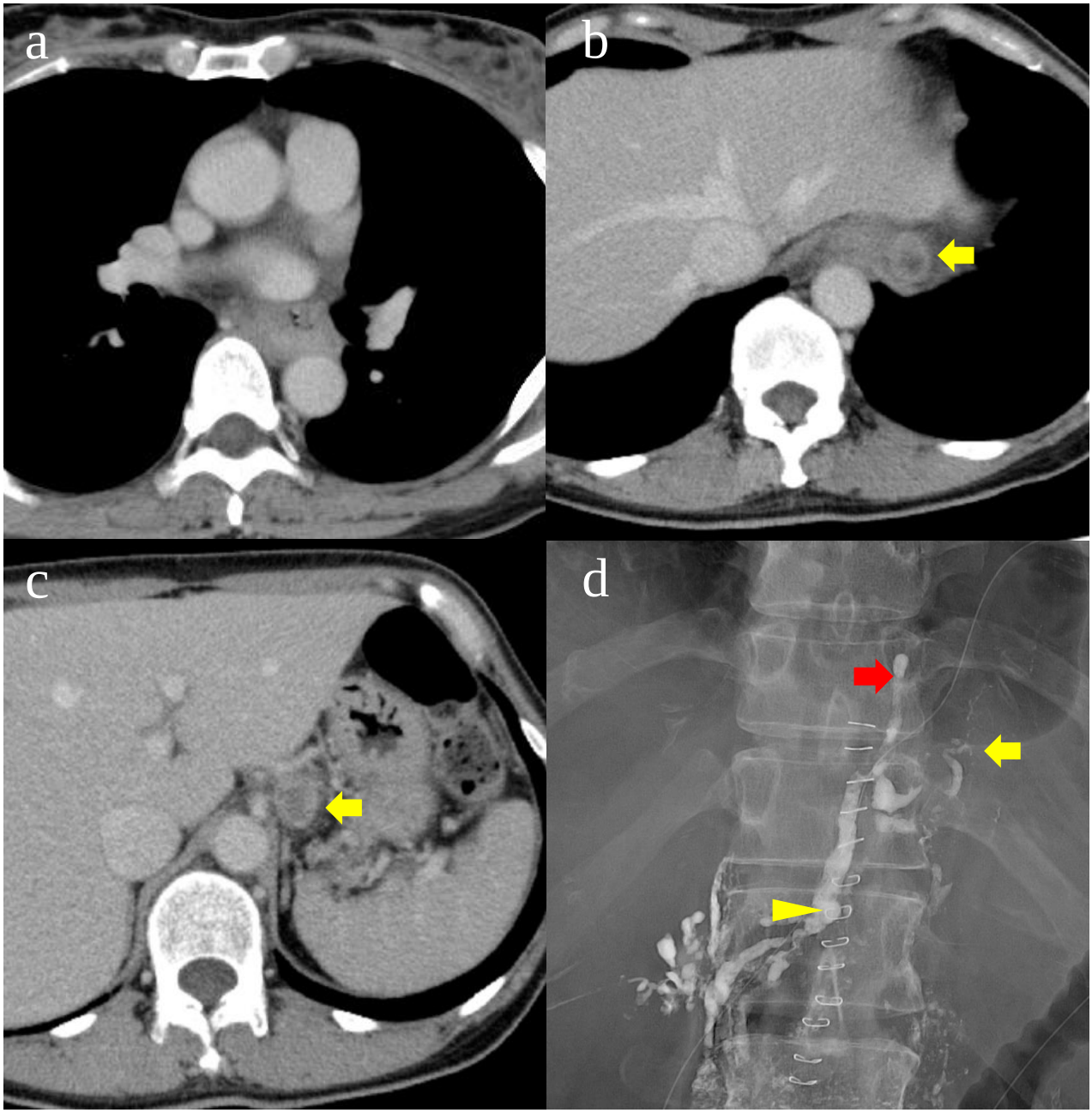
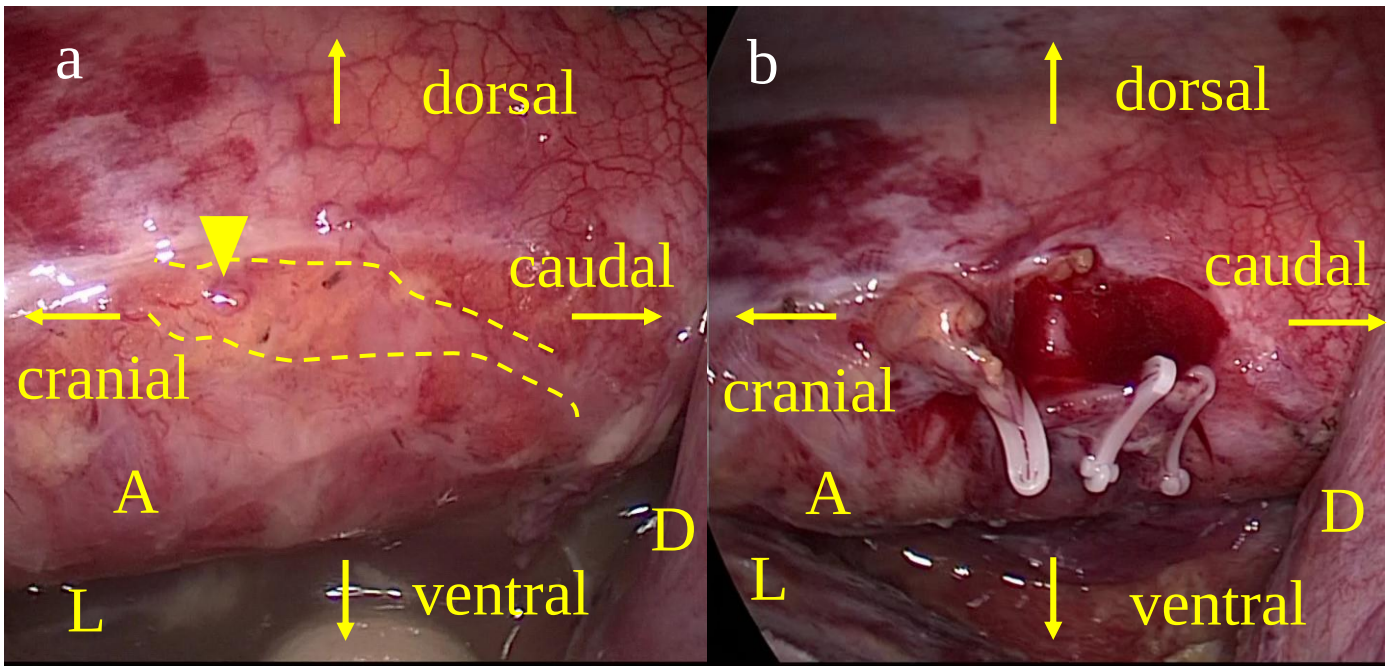


Figure 2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