

Bilateral Chylothorax Following Modified Radical Neck Dissection in Thyroid Cancer: A Case Report

Youngpeck Song, Jeoghun Lee, Ji Woo Choi, Euy Young Soh

Department of Surgery, Ajou University School of Medicine, Suwon, Korea

Bilateral chylothorax is an extremely rare complication of modified radical neck dissection. It is a potentially life-threatening condition that can lead to severe respiratory, nutritional, metabolic, and immunologic disorders. Use of a multi-disciplinary approach including drainage of chyle, reduction of chyle formation, adequate nutritional support, use of somatostatin or its analogue, surgical ligation of the thoracic duct, and thoracic duct embolization is the best method for treatment of bilateral chylothorax. We report on a case of bilateral chylothorax following total thyroidectomy with modified radical neck dissection and discuss its management.

Key Words: Chylothorax, Neck dissection, Thyroidectomy

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Correspondence: **Euy Young Soh**
Division of Surgery, Thyroid Center, Ajou
University School of Medicine, 164,
Worldcup-ro, Yeoungtong-gu, Suwon
443-380, Korea
Tel: +82-31-219-5201
Fax: +82-31-219-5755
E-mail: sohey@ajou.ac.kr

INTRODUCTION

Chylous fistula occurs in 1~2% of patients who undergo neck dissection, with the majority being localized to the left side of the neck.⁽¹⁾ It can be recognized by drainage of milky fluid from the neck. In almost all of the cases, it can be diagnosed promptly. However, postoperative bilateral chylothorax is rare, and hence, it is difficult to detect and treat this condition.⁽²⁾ Stuart⁽³⁾ reported the first case of bilateral chylothorax following neck dissection in 1907. Persistent high-output chylothorax can result in a life-threatening condition because of malnutrition, susceptibility to infection, metabolic derangements, and cardiopulmonary problem.^(1,4) Recently we experienced a case of bilateral chylothorax without evidence of chyle leakage from the neck after bilateral neck dissection.

The aim of this article is to present a case of chylothorax following modified radical neck dissection and to discuss the appropriate treatment through literature review.

CASE REPORT

A 38-year-old woman presented with a right thyroid

mass detected 2 years ago. Ultrasonography revealed that the mass was 0.82 cm in size and had spiculated margins and microcalcification. The mass was cytologically suspicious for malignancy according to the Bethesda system. Metastatic lymphadenopathy was detected by computed tomography in the level VI and the left level IV area. We had planned thyroidectomy, but the patient wanted to put off the operation because she became pregnant. Two years later, we reexamined the tumor by performing ultrasonography and the tumor had grown up to 1.0 cm in size. And then we performed fine needle aspiration cytology about suspicious lymph node in left level IV area. The result of cytology was positive for malignant cells. She had no underlying diseases and family history of thyroid cancer. Preoperative thyroid function test was normal. She underwent total thyroidectomy with bilateral central compartment neck dissection and bilateral modified radical neck dissection. Intraoperative findings were as follows: 0.7 cm sized mass in the right thyroid without capsular invasion, a 3.0 cm sized lymph node in the pretracheal area, and multiple enlarged lymph nodes in the left level IV area including a 3.0 cm sized lymph node and other small lymph nodes in the bilateral levels III and IV areas. There were

multiple engorged lymphatic ducts with lymphatic spillage in the left level IV area after node dissection. Therefore, we performed multiple suture ligations in the level IV area including the thoracic duct and no more spillage of chyle was observed. Postoperative amount of drainage was decreased from 270 mL to 41 mL in 3 days. No milky fluid was drained postoperatively. The drain was removed on the third postoperative day (POD). After that, the patient experienced dyspnea without any abnormal breath sounds. On POD 4, dyspnea was aggravated with decreased breath sounds. Oxygen saturation measured by pulse oximetry was 95% after 3 L/min oxygen supplement via nasal prongs. Chest X-ray revealed large, bilateral pleural effusion (Fig. 1). We inserted percutaneous drains (PCD) in the bilateral pleural space, and 1,240 mL of milky fluid was drained from the right pleural space and 1,520 mL of milky fluid was drained from the left pleural space on POD 4. Triglyceride (TG) level in the fluid drained from the right pleural space was 597 mg/dL and that in the fluid drained from the left pleural space was 700 mg/dL. Dyspnea was improved after PCD insertion. 'Nil per os' (NPO) order was applied immediately. On POD 6, the serum TG level in the fluid drained from the right pleural space was 20 mg/dL and that in the fluid drained from the left pleural space was 18 mg/dL. Amount of drainage was decreased to 16 mL from the right PCD and to 12 mL from the left PCD, and the color of drainage was serous on POD 7. No more

milky drainage was detected. Normal diet was started on POD 8. The color of drainage continued to be serous and chest X-ray returned to normal; therefore, we removed both the PCD (Fig. 2). She was discharged uneventfully on POD 11. Final pathology was papillary thyroid carcinoma and lymph node metastasis existed in central and bilateral lateral neck area.

DISCUSSION

Chylous fistula is a rare complication of neck dissection and three-quarters of them occur on the left side.⁽¹⁾ It is usually recognized as a milky fluid in the neck drains postoperatively. Bilateral chylothorax can occur following neck dissection, but it rarely occurs after thyroid operation. Only 39 cases of bilateral chylothorax associated with neck dissection have been reported in the literature. Among them, 18 cases were associated with thyroidectomy (Table 1). The exact pathophysiology of chylothorax is not fully understood. However, two hypotheses have been postulated. One hypothesis is that the chyle escaping from the base of the neck flows directly into the mediastinum.⁽¹⁴⁾ This may be possible because of rupture of the mediastinal pleura caused by tissue maceration and the inflammatory reaction.⁽²⁾ Another hypothesis is that chylothorax leads to high intraluminal hydrostatic pres-

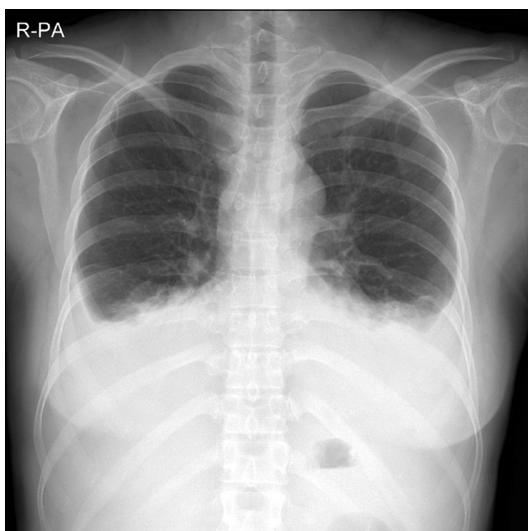


Fig. 1. Chest X-ray shows bilateral pleural effusion (Fourth postoperative day).

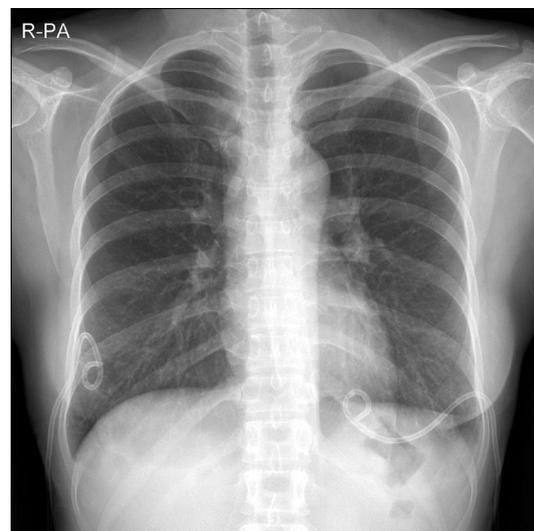


Fig. 2. Chest X-ray shows complete resolution of bilateral pleural effusion (Eighth postoperative day).

Table 1. Review of the literature for bilateral chylothorax following neck dissection in thyroid cancer

Study	Year	Age (yr)	Sex	Primary lesion	Side of neck dissection	Thoracic duct injury/ligation	Primary symptom	Conservative management	Secondary surgical ligation
Runge, et al.(5)	2014	40	F	Thyroid	Left	No	Chest discomfort and dyspnea, POD 2	TPN, octreotide	Yes
Li, et al.(6)	2013	48	F	Thyroid	Bilateral	Yes	Chest discomfort, POD 2	TPN, drainage	No
		65	F	Thyroid	Bilateral	Yes	Chest discomfort and dyspnea, POD 8	TPN, drainage	No
		40	F	Thyroid	Bilateral	Yes	Chest discomfort, POD 2	TPN	Yes
		38	F	Thyroid	Left	Yes	Chest discomfort and dyspnea, POD 4	TPN	No
		31	F	Thyroid	Bilateral	Yes	Chest discomfort and dyspnea, POD 3	TPN, drainage	No
Tian et al.(7)	2012	40	F	Thyroid	Bilateral	Yes	Chest discomfort, POD 2	TPN, drainage	No
		38	F	Thyroid	Bilateral	Yes	Chest discomfort, POD 4	TPN	No
		31	F	Thyroid	Bilateral	Yes	Chest discomfort and dyspnea, POD 3	TPN, drainage	No
Han et al.(2)	2009	42	F	Thyroid	Bilateral	No	Chest discomfort and dyspnea, POD 8	TPN, drainage	No
Tsukahara, et al.(8)	2007	72	F	Thyroid	Bilateral	Yes	Dyspnea, POD 2	TPN	No
Bae, et al.(9)	2007	46	F	Thyroid	Left	No	Chest discomfort and dyspnea, POD 5	TPN, drainage	No
		47	F	Thyroid	Left	No	Dyspnea, POD 3	LF, drainage	No
		42	F	Thyroid	Left	Yes	Dyspnea, POD 2	TPN, drainage	No
Shin, et al.(10)	2006	31	M	Thyroid	Left	Yes	Dyspnea, POD 2	TPN, drainage, somatostatin	No
		70	F	Thyroid	Left	Yes	Dyspnea, POD 2	TPN	No
Kim, et al.(11)	1999	70	F	Thyroid	Left	Yes	Dyspnea, POD 2	TPN	No
Jabbar and Al-Abulkareem (12)	1995	35	F	Thyroid	Bilateral	Yes	Chest discomfort and dyspnea, POD 4	Unknown	No
Har-El, et al.(13)	1985	34	F	Thyroid	Left	No	Chest discomfort and dyspnea, POD 3	LF	No

POD = postoperative day; TPN = total parenteral nutrition; Drainage = continuous closed chest drainage; LF = low fat diet.

sure after thoracic duct ligation, which can cause chyle extravasation or rupture of the thoracic duct in the mediastinum.(15) Intraluminal pressure exceeds the capacity of the lymphatic collateral channels, and this is coupled with the negative intrathoracic pressure caused by inspiration.(2) There was no evidence of chyle leakage in the lateral neck portion in our case, but chylothorax developed. This can be explained by the second hypothesis. We performed multiple suture ligations in the level IV area including the thoracic duct because there were multiple engorged lymphatic ducts with lymphatic spillage. The area was rich in lymphatics, and hence, the intraluminal hydrostatic pressure might be high after thoracic duct ligation. There was a high probability of chyle extravasation, but not of thoracic duct rupture because chyle leakage was dramatically improved after PCD insertion combined with NPO.

Chylothorax can be lethal because of severe compli-

cations such as cardiopulmonary problems.(16,17) Hence, prompt and accurate diagnosis is important. Our patient experienced dyspnea, whitish sputum, and chest discomfort. Chest X-ray is a convenient way to confirm pleural effusion. If the chest X-ray confirms pleural effusion, a diagnostic thoracentesis can be proposed. Milky appearance of the pleural fluid is helpful for making the diagnosis of chylothorax. But, the classical milky appearance is seen in only 50% of the cases. It can also manifest as a serous or serosanguineous effusion.(18) Measurement of the fluid cholesterol and TG levels or Sudan III staining are also useful for making the diagnosis of chylothorax. A TG level above 110 mg/dL is diagnostic for chylothorax, while a TG level below 50 mg/dL can exclude chylothorax in patients on a normal diet.(18) Our patient was diagnosed as having chylothorax because the TG levels in the drainage were more than 110 mg/dL on POD 4.

Chylothorax should be treated at an early stage because

it can be a harmful condition.(1,4,12) Conservative treatment is the initial line of treatment. It includes drainage of chyle, reduction of chyle formation, and adequate nutritional supplementation. Nutritional program for reducing chyle formation consists of medium-chain TG (MCT)-based oral diet or NPO with total parenteral nutrition.(19,20) Chest PCD for chyle drainage, NPO for reduction of chyle formation, and total parenteral nutrition for nutritional support were used in our patient. Somatostatin and octreotide may inhibit chyle production and reduce the thoracic duct flow; hence, they could be incorporated in the conservative management.(21) However, we did not administer somatostatin in this case.

The use of surgical intervention is controversial; but, it can be employed in cases where patients fail to respond to the conservative management.(22) The indications for surgical management are as follows: persistent leak for more than two weeks despite optimal conservative treatment; severe metabolic or nutritional complications; chyle leakage of more than 1 L per day for more than five days.(23)

Thoracic duct embolization can also be used to treat chylothorax. While the effect of thoracic duct embolization is doubtful, the literature suggests that it can replace more invasive surgical procedures; thus, reducing patient morbidity and mortality.(24)

A multi-disciplinary approach with optimized conservative management was successful in treating chylothorax and preventing complications in our patient. This condition can be easily overlooked because of its rarity, but surgeons have to consider the possibility of chylothorax in patients with chest discomfort or dyspnea after neck dissection. Early diagnosis and treatment seem to be important for reducing life-threatening complications and for achieving good results.

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