Bilateral Chylothorax Following Total Thyroidectomy with Neck Dissection for Papillary Thyroid Cancer

Case report

*Musallam Kashoob,1 Saleh Bawain,2 Yahya Al Badaai3

1Department of Ear, Nose and Throat, Oman Medical Specialty Board, Muscat, Oman; 2Department of Radiology & Molecular Imaging, Sultan Qaboos University Hospital, Muscat, Oman; 3Department of Surgery, Sultan Qaboos University, Muscat, Oman

*Corresponding Author’s e-mail: doctorkashoob@hotmail.com

Abstract

Bilateral chylothorax is a rare entity that occurs after a thyroidectomy and neck dissection. This rare condition can lead to severe morbidity, and potentially death, if not managed properly. We report a rare complication of neck surgery and subsequent bilateral pleural effusion, regarding a 35-year-old female who presented at Sultan Qaboos University Hospital in 2018 with shortness of breath and respiratory distress. Particularly, the bilateral pleural effusion and related symptoms occurred one week after a total thyroidectomy with central and left lateral neck dissection. Intraoperative findings, management and literature review will be discussed.

Keywords: chylothorax, thyroid cancer, papillary, thyroidectomy, neck dissection, Case report; Oman.

Introduction

Chylothorax can be classified based on its etiology, such as traumatic and non-traumatic/idiopathic forms. The most common traumatic cause of Chylothorax is chest surgery.1 A rarer yet serious complication is iatrogenic chyle leak (CL) from a thoracic duct injury during head and neck surgery, which occurs in 0.5–1.4% 2–5 of thyroidectomies and 2–8% 6–9 of neck
dissections. Bilateral chylothorax is an extremely rare complication which occurs after neck dissection and is potentially life-threatening. In a systematic review by Merki et al, the reported incidence of chylothorax after total thyroidectomy and neck dissection was 1.85%. We report a case of bilateral chylothorax following total thyroidectomy with central and left lateral neck dissection, which was managed conservatively.

Case report
A 35-year-old female with no known comorbidities was evaluated for a left thyroid nodule during a routine neck ultrasound. Which was later proven to be consistent with papillary thyroid cancer (PTC) on Fine Needle Aspiration Cytology (FNA). Her preoperative mapping neck ultrasound and contrast-enhanced head and neck Computed Tomography (CT) scan did not reveal any suspicious lymphadenopathy [Figure 1 (A,B)]. Then, she underwent total thyroidectomy. During the surgery, thyroid tissue was adhered to the tracheal wall with multiple central compartment lymph nodes positive for malignant cells on frozen section. Hence, she underwent total thyroidectomy with bilateral central compartments level (VI – VII) and left lateral level (II - V) neck dissection.

CL was encountered during neck dissection and was repaired using surgical clips, prolene 5–0 suture and tissue glue. There was no more leak noted, even with positive intrathoracic pressure ventilation post repair. The wound was closed after insertion of two large negative pressure neck drains. The patient was doing well during the immediate post-operative period, the neck drains were removed after ensuring minimal output and the patient on full normal diet. She was then discharged on the second day after surgery.

However, on the seventh post-operative day, she presented to the emergency department with shortness of breath and chest tightness. Her primary examination showed oxygen saturation of 93%, mild tachypnea and tachycardia (respiratory rate 32/minute, heart rate 112/minute). Physical examinations revealed flat neck with a healing incision site and decreased breath sounds bilaterally from the lung bases to the mid-lung fields. Imaging demonstrated bilateral moderate pleural effusions with bilateral compressive atelectasis of adjacent lung parenchyma [Figure 2].
Intrathoracic pleural catheters were placed bilaterally, which drained approximately 2.5 litres of chylous fluid from both sides. Pleural fluid analysis showed a triglyceride level of 5.7 mmol/L (normal range: 0.0–2.3 mmol/L) which confirmed chylous fluid. The patient was started on a fat-free diet and Somatostatin analogue injection (Octreotide 100 mcg every 8 hours subcutaneously) along with chest physiotherapy, intravenous antibiotics and daily monitoring of drain output and electrolytes. Chest drains were draining <50 ml on the consecutive follow-up days, and a chest X-ray confirmed marked improvement of the effusions bilaterally after use of the above conservative measures alone. [Figure 3 (A) & (B)]. The chest drains were kept for seven days and the patient did not show any radiological signs of re-accumulation or persistent leak.

The histopathological diagnosis showed a tall cell variant of papillary thyroid carcinoma (tumour size 2.5 cm) with 11 metastatic lymph nodes out of a total of 88 lymph nodes (largest 1.4 cm). The patient recovered very well and received a treatment dose of radio-iodine therapy and consent for the publication of this case report was obtained from the patient.

**Discussion**

The pathophysiology of bilateral chylothorax is still not fully understood, but there have been two proposed mechanisms of injury.\(^\text{10}\) The first mechanism includes direct injury or incomplete ligation of the duct during surgery, due to variable duct anatomies described earlier. This first mechanism results in chyle leakage into the mediastinum. The second possible mechanism of injury is due to increased retrograde hydrostatic pressure in the thoracic duct, resulting in an increased intraluminal pressure. This, combined with negative intrathoracic pressure, causes a secondary rupture within the chest. Based on the patient’s Intraoperative and post operative clinical profile and radiology, we presume the second mechanism is responsible for bilateral chylothorax in our case.

Early diagnosis and appropriate management are essential in order to avoid chylothorax-related morbidity. This can be in the form of either cardiopulmonary compromise or metabolic derangement, including loss of proteins, electrolytes, fluid, fat, fat-soluble vitamins and T cell lymphocytes.\(^\text{15}\)
Although a comprehensive clinical assessment with imaging may point to the diagnosis of chylothorax, chemical analysis of the pleural fluid is essential for diagnosis. Chylothorax is present in 99% of patients with an aspirate triglyceride content of >110 mg/dL (1.24 mmol/L) and a cholesterol content >200 mg/Dl (5.1 mmol/L). Moreover, the presence of chylomicrons in the pleural fluid is considered diagnostic.\textsuperscript{16–18} The pleural aspirate triglyceride content in our patient was 5.7 mmol/L.

Operating Surgeons should have a clear concept of the tortuous anatomy of the thoracic duct to avoid complications. At the level of the upper lumbar spine, the thoracic duct starts as cisterna chyli. It has different anatomical relations when it passes within the abdomen, thoracic cavity, root of the neck and before it terminates. Initially, it ascends upward along the abdominal aorta and azygos vein through the aortic orifice overlying the vertebral column to enter the thoracic cavity. Then, it turns to the left side and continues to pass upwards in the posterior mediastinum to enter the root of the neck. At this level, it passes lateral to the esophagus, medial to the omohyoid muscle, posterior to the left carotid sheath, and anterior to the vertebrae. Then, the thoracic duct courses superiorly and laterally before it descends inferiorly to arch over the anterior scalene muscle, phrenic nerve, subclavian artery, and ends within 1 cm from the confluence of internal jugular vein and subclavian vein.\textsuperscript{11–13} However, there are anatomical variations of the terminal course of the thoracic duct, in 46% it enters the internal jugular vein, in 32% it enters the junction of the internal jugular and subclavian veins. Less commonly, it may enter at the subclavian vein in 18%. Although two or three endpoints of thoracic duct terminations have been described, usually it ends as a single duct in about 76% of cases.\textsuperscript{8,14}

Management of chylothorax should be tailored to each individual case. The management varies between conservative approaches, interventional radiological approaches and surgical options. Conservative measures include the drainage of chylothorax in addition to diet modifications in the form of medium-chain triglycerides (MCT), fat-free diet or total parenteral nutrition to reduce the chyle production. In addition, certain medications are used such as somatostatin or its analogue, or octreotide to reduce chyle production.\textsuperscript{19–20} however, there is no consensus as to how long these medications should be used. If the drained chyle is < 500 ml/day, spontaneous
recovery with conservative measures is more favourable.\textsuperscript{19} In our case, the conservative treatment was effective in reducing the leak to less than 50 ml/day. Depending on the available expertise of each centre, percutaneous lymphangiography guided cannulation with embolization of the leakage is another minimally invasive option.\textsuperscript{21}

Surgical intervention, including thoracic duct ligation via video-assisted thoracoscopic surgery (VATS) or open surgery, should be considered if conservative measures fail, drain flow is more than 1 L per/day or if severe metabolic complications are present. However, there are currently no prospective studies available to compare efficacy of different approaches.

**Conclusion**

Bilateral chylothorax is a rare and potentially life-threatening complication, which should be considered in all patients with post-operative dyspnea occurring after central or left lateral neck dissection. Early diagnosis and intervention are crucial for best outcomes. Management is tailored according to the clinical scenario, level of expertise available and patient response to initial treatment.

**References**


**Figure 1 (A,B)** Contrast enhancing axial cut CT in a 35-year-old female who presented at Sultan Qaboos University Hospital in 2018 with shortness of breath and respiratory distress showing: **A:** 10 mm hypoattenuating nodule on the left thyroid lobe (yellow arrow). **B:** 7 mm nodule with rim of calcification on the right thyroid lobe (red arrow).
Figure 2: PA chest radiograph for a 35-year-old female who presented at Sultan Qaboos University Hospital in 2018 with shortness of breath and respiratory distress showing bilateral moderate chylothorax with associated atelectasis and post-thyroidectomy surgical clips at the base of the neck.

Figure 3: Chest radiograph PA view for a 35-year-old female who presented at Sultan Qaboos University Hospital in 2018 with shortness of breath and respiratory distress. A: Near total resolution of the Chylothorax post placement of the bilateral intrathoracic pleural catheters. B: Clear lungs and costophrenic angles on follow-up chest X-ray after 7 days.