CASE REPORT

Bilateral chylothorax following reoperative central neck dissection for metastatic papillary thyroid cancer

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SUMMARY

Locoregional recurrence of papillary thyroid cancer (PTC) can occur in up to 30% of patients within 10 years after initial treatment. Reoperative surgery can be challenging due to scarring, fibrosis and distorted anatomy and may be associated with increased complications compared with initial operations. Chylothorax is an extremely rare and potentially life-threatening complication that can be associated with neck dissections and thyroid surgery. In this report, we describe an unusual case of bilateral chylothorax following a left central neck dissection for recurrent PTC.

BACKGROUND

Papillary thyroid cancer (PTC), the most common type of thyroid cancer, has excellent prognosis after surgical treatment, with 10-year survival rates reported to be greater than 90%.¹ However, approximately 5%–30% of patients will develop a locoregional recurrence within 10 years from the initial diagnosis.² The most common site of recurrence involves the central compartment of the neck, but may also include the lateral neck to a lesser extent. Reoperations for thyroid cancer can be challenging due to scarring, fibrosis and distorted anatomy and may be associated with increased complications compared with initial operations.³ Complications include temporary or permanent recurrent laryngeal nerve injury and hypoparathyroidism. Chyle leak after lateral neck dissection is a known complication that occurs in 1%–2% of patients, usually after a left-sided dissection.⁴ However, a chylothorax is an extremely rare and potentially life-threatening complication that can be associated with neck dissections and thyroid surgery. In this report, we describe an unusual case of bilateral chylothorax following a left central neck dissection for recurrent PTC.

CASE PRESENTATION

A 41-year-old Hispanic woman, known to have a 10-year history of Hashimoto's thyroiditis on levothyroxine, presented in 2012 to an outside hospital in Honduras with a 1 cm left thyroid lobe biopsy-proven PTC. At the time, she underwent a total thyroidectomy with sampling of lymph nodes in the left central neck compartment. Final pathology demonstrated classical PTC without evidence of angiolymphatic invasion. Excised lymph nodes did not show evidence of tumour. Postoperatively, she received 100 mCi radioactive iodine therapy.

In 2015, an enlarged, 0.8 cm suspicious-appearing left level VI nodule was identified on a follow-up ultrasound and biopsied. The patient was referred to our institution for surgical consultation. Review of her fine-needle aspiration biopsy at our institution demonstrated thyrocytes suspicious for PTC. Imaging of the neck demonstrated no other evidence of recurrent disease or lymphadenopathy. Lab work at the time showed a thyroid stimulating hormone level of 0.19 IU/mL (normal range: 0.50–4.50 IU/mL) and free T4 (FT4) of 1.4 (normal range 0.8–1.8 ng/dL). Her thyroglobulin level was undetectable but was deemed to be unreliable given that she tested positive for thyroglobulin antibodies. The patient denied any compressive symptoms of hoarseness, dysphagia, dysphonia, dyspnoea, neck pain or tenderness. On physical examination, she did not have any palpable lymphadenopathy, and flexible laryngoscopy demonstrated bilateral vocal cord motion. An extensive multidisciplinary discussion of the options of active surveillance versus reoperation took place with the patient and her family. Given the knowledge of a cancerous lymph node in her neck, the patient strongly wished for reoperation.

The patient underwent left central neck dissection with nerve monitoring. Intraoperatively, the thoracic duct was not identified and clips were used for haemostasis. A Valsalva manoeuvre was performed at the end of the case. Her final pathology revealed metastatic PTC in 1/5 lymph nodes. She tolerated the procedure well without complications and was discharged on postoperative day 1 with no complaints.

On postoperative day 3, the patient presented to the emergency department complaining of posterior chest pain along her costal margin, shortness of breath, orthopnoea and a dry cough. Physical examination revealed a flat neck with a healing incision site and decreased breath sounds bilaterally from the lung bases to the midlung fields. Fiberoptic laryngeal examination demonstrated normal bilateral vocal cord motion. Imaging demonstrated bilateral pleural effusions with compressive atelectasis, greater on the left (figure 1).

Pigtail catheters were placed bilaterally with drainage of the fluid. Pleural fluid analysis showed a triglyceride level of 1996 mg/dL (normal range: <50 mg/dL), suggesting chylous fluid. The patient was started on a fat-free diet. A follow-up chest X-ray confirmed marked improvement of the effusions bilaterally. A lymphangiogram demonstrated obstruction of the thoracic duct in the neck,

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Figure 1 Chest X-ray from the emergency department demonstrating bilateral pleural effusions.

approximately 4 cm below surgical clips. Enlarged collateral thoracic duct branches extending to the left and right from the thoracic duct throughout the chest were noted and suspected to have led to the bilateral pleural effusions. Lipiodol injection revealed no clear area of leakage for further intervention (figure 2). Similarly, follow-up CT imaging was non-revealing for definitive leak, but re-demonstrated enlarged collateral thoracic duct branches (figure 3). Her chest tubes were eventually removed and follow-up chest X-ray did not demonstrate any further reaccumulation of fluid.

OUTCOME AND FOLLOW-UP

At a follow-up visit 2 weeks after discharge, the patient endorsed significant improvement in her symptoms and reported that

Figure 3 Representative CT images obtained immediately after lymphangiogram demonstrating enlarged thoracic duct collaterals (yellow asterisk).

her breathing had returned to normal. Follow-up chest X-ray confirmed no residual chylothorax (figure 4).

DISCUSSION

Chylothorax is a rare, but major complication seen anecdotally after thyroid surgery combined usually with a lateral neck dissection. Herein, we report a case of bilateral chylothorax following a central neck dissection that was managed conservatively.

To the author's best knowledge, this is the first case report to describe a bilateral chylothorax following a central neck dissection for thyroid cancer recurrence. This uncommon complication has been described only in case reports in patients after a thyroidectomy and a left lateral neck dissection or a sternotomy due to the large thyroid size. A systematic review by Merki *et al*⁵ report the incidence of chylothorax after total thyroidectomy and neck dissection was 1.85% and 7.3% when using a thoracic approach.

This uncommon complication has been described only in case reports. Two possible mechanisms of injury have been proposed.⁵ One possible mechanism includes direct injury or incomplete ligation of the duct during surgery leading to chyle leakage into the mediastinum. Incomplete ligation may occur due to the variable anatomy of the cervical portion of the thoracic duct. After originating in the abdomen at the cisterna chyli, the thoracic duct crosses to the left side of the body near the fifth and sixth thoracic vertebrae, and then ascends until it drains into a vein in the region of the junction of the left subclavian vein and the internal jugular veins. However, the entry of the thoracic duct into the venous system is highly variable with branches entering either at the internal jugular vein, subclavian vein or the jugulo-venous angle. In less than 5% of cases, the duct can terminate in the external jugular vein, vertebral vein, transverse cervical vein, brachiocephalic vein and suprascapular vein.⁶ The duct can

Figure 2 Lymphangiogram demonstrating an enlarged thoracic duct (red arrow) that obstructs at the level of clips.

Figure 4 Chest X-ray at postoperative follow-up visit demonstrating complete resolution of effusions and re-expansion of the lungs.

also terminate at multiple sites with many people having up to three terminal vessels.⁷ The cervical portion of the thoracic duct can be as high as 3.5 cm superior to the sternal notch and only 2 cm lateral to the midline.⁸ Furthermore, due to our patient undergoing reoperative neck dissection in the setting of recurrent regional thyroid cancer, the chylothorax may be caused by any type of injury of the chyle duct due to tissue fibrosis as a result of her initial surgery. Despite the fact that the central neck dissection is medial to the major neck vessels, the location of the chyle duct may have been altered, due to tissue distortion, thus increasing the risk of injury.

A second plausible mechanism of injury is from increased retrograde hydrostatic pressure in the thoracic duct resulting in increased intraluminal pressure. This combined with negative intrathoracic pressure causes a secondary rupture. Therefore, the point of extravasation is in the chest and not the neck. Based on the clinical picture and imaging, this likely resulted in chylothorax in our patient.

Morbidity from a chylothorax results from the loss of proteins, electrolytes, fluid, fat, fat-soluble vitamins and T cell lymphocytes.⁹ These losses can result in metabolic derangements, dehydration, nutritional deficiency and infections. Chyle in the pleural space can also cause cardiorespiratory compromise.⁹ Therefore, prompt diagnosis of a chylothorax and appropriate management is essential.

Thoracentesis and pleural fluid analysis can help confirm a chylothorax, with a pleural fluid triglyceride level of >110 mg/dL indicating a chyle leak.¹⁰ However, the gold standard for diagnosis is the presence of chylomicrons in the pleural fluid. Lymphangiography and lymphoscintigraphy are typically used to localise possible chyle leaks and assess thoracic duct patency. In addition, lymphangiograms and lymphoscintigraphy can be helpful in identifying aberrant thoracic duct anatomy and can even be therapeutic, with certain studies demonstrating that lymphangiography successfully occluded lymphatic leaks in 70% of patients when drainage was less than 500 mL/day.¹¹

Management of chylothorax ranges from conservative approaches with diet modification to assistance from

interventional radiology and surgical options. The most significant factor for the treatment of chylothorax is the volume of output of chyle. Chyle flow rates <500 mL/day are associated with favourable spontaneous closure rates and typically can be managed conservatively.⁷ Conservative management consists of drainage of chylothorax with lung re-expansion in addition to diet modification to a fat-free diet or providing total parenteral nutritional to reduce chyle production.

Some have used somatostatin or its analogues, octreotide, to reduce production of chyle. Somatostatin is a polypeptide that inhibits release of several hormones, including growth hormone and insulin, as well as inhibiting lymph fluid excretion.^{7 10} However, we are unaware of any controlled trials assessing the efficacy, dose or route of administration of octreotide to manage a chylothorax. Chyle leaks that are >1 L/day should be treated with surgical options, including direct thoracic duct ligation via video-assisted thoracoscopic surgery (VATS) or open surgery. In cases where conservative or VATS ligation of the thoracic duct fails, a pleuroperitoneal or pleurovenous shunt may be required as a last resort.

In summary, chylothorax is a rare, but serious complication that can occur after a central neck dissection. It should be included in the differential for postoperative dyspnoea, particularly in thoracic and thyroid surgeries. Analysis of the pleural fluid is critical for diagnosis, and appropriate management includes fluid drainage, nutritional support, lymphangiogram or surgical ligation of the thoracic duct if the chyle leak is severe.

Learning points

- ▶ Chylothorax is a rare, but serious complication that can occur after a central neck dissection. It should be included in the differential for postoperative dyspnoea, particularly in thoracic and thyroid surgeries.
- ▶ Analysis of the pleural fluid is critical for diagnosis, with elevated triglycerides or the presence of chylomicrons favouring a chylothorax.
- ▶ Appropriate management includes fluid drainage, nutritional support, lymphangiogram or surgical ligation of the thoracic duct if the chyle leak is severe.

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