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Case report

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Bilateral chylothorax after left neck lymphadenectomy for thyroid cancer: A case report

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ABSTRACT

Introduction: Chylothorax is caused by lymphatic chyle fluid leaking back through the thoracic duct and accumulating in the pleural cavity. It is related to a thoracic duct injury or occlusion. It is rare to have bilateral chylothorax after cervical lymph node dissection for thyroid cancer diagnosis.

Case report: A 28-year-old woman was admitted to our hospital with bilateral hypoechoic thyroid nodules and cervical lymph node abnormalities. She underwent thyroidectomy and lymphadenectomy but developed chylothorax 3 days after surgery. She was treated with bilateral thoracic drainage, electrolyte supplementation, and somatostatin, and was discharged 17 days post-treatment.

Conclusion: Bilateral chylothorax is a rare complication of thyroid cancer surgery. Early diagnosis and treatment, especially the detection of dyspnea, are key. Also, unobstructed bilateral thoracic drainage, improved surgical skills, and reduced thoracic duct injuries can help reduce complications.

1. Introduction

Chyle leakage (CL) is a rare but serious complication of cervical lymphadenectomy in thyroid cancer patients [1]. The incidence rate of CL is approximately 0.5–1.4% [2], which maybe due to the rupture of the lymphatic ducts and the failure to perform effective ligation [3]. Lateral lymphadenectomy-associated chylothorax is a rare condition [4]. In 1907, Stuart et al. [5] first reported bilateral chylothorax after neck dissection, and there are only a few case reports related to this topic. The release of a large amount of chylous fluid in the bilateral pleural cavity can compress the lungs and mediastinum, causing both respiratory and circulatory problems. Metabolic and immune dysfunctions are also considerable risks related to CL. Recently, a case of bilateral chylothorax after left cervical lymph node dissection was diagnosed with thyroid cancer and treated in our hospital, and the case is reported as follows.

2. Case report

A 28-year-old Chinese female was admitted to our hospital on May 17, 2022, with the unexpected discovery of thyroid nodules that had been present for more than a month. She had no remarkable discomforts, such as dysphagia, hoarseness, palpitations, or chest tightness, at admission. There was no family history of cancer or exposure to harmful carcinogens. A physical examination could not

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distinguish the obvious enlargement of the thyroid. However, thyroid ultrasound revealed a mass of about 1.3×1.0 cm in the right lobe, about 1.7×0.9 cm in the left lobe, and about 0.8×0.6 cm in the thyroid isthmus. These nodules were firm and had punctate calcifications. The Thyroid Imaging Reporting and Data System (TI-RADS) scoring of these nodules classified them as grade V thyroid carcinoma. At the same time, there were abnormal lymph nodes at levels III, IV, and VI of the bilateral neck. The preoperative ultrasound images of bilateral thyroid nodules and abnormal lateral cervical lymph nodes in this patient are shown in Fig. 1. In Fig. 1A and B show the ultrasound images of the right thyroid nodule in two sections, C shows the ultrasound images of the thyroid isthmus nodule, D shows the ultrasound images of the left thyroid nodule, E and F show the ultrasound images of the abnormal lymph nodes in level IV of the right and left neck, respectively. Also, a preoperative CT scan of the neck and chest showed low density and microcalcification in both thyroid lobes, as well as multiple abnormally enlarged lymph nodes in both necks. Moreover, a preoperative fineneedle aspiration biopsy (FNAB) of the right thyroid nodule confirmed papillary thyroid carcinoma (PTC). This patient was anemic due to iron deficiency, with a preoperative hemoglobin (Hb) value of 88 g/L, but she did not consent to an intraoperative allogeneic blood transfusion. Due to the long duration of the surgery and the possibility of bilateral neck lymph node dissection, the patient agreed to complete the surgical procedure in two stages: total thyroidectomy along with right cervical lymph node dissection in the first stage and left cervical lymph node dissection in the second stage. Therefore, the patient successfully underwent total thyroidectomy, bilateral central lymph node dissection, and lymph node dissection at levels II, III, IV, and V of the right neck on May 19, 2022. Postoperative pathological results identified classical PTC in bilateral lobes, a pathogenic BRAF^{V600E} mutation (mutations in other genes, such as Ret and TERT, could not be detected due to technical reasons), and lymph node metastasis (left central lymph nodes 6/ 11, right central lymph nodes 7/15, right neck II lymph nodes 1/14, right neck III lymph nodes 1/10, right neck IV lymph nodes 1/7, and right neck V lymph nodes 0/12). Postoperative recovery was uneventful, without obvious hoarseness or hypocalcemic convulsions. She was readmitted to our hospital on July 11, 2022, and underwent left cervical lymph node dissection at levels II, III, IV, and V under general anesthesia on July 13, 2022. Several enlarged (~1 cm in size) lymph nodes were discovered during the operation in the IV region of the left neck and the posterior medial of the internal jugular vein. Notably, some of these nodes were brown or black, adhered to each other, and tightly intertwined with the thoracic duct, so they could not be separated. Hence, the thoracic duct was ligated with a silk thread, and the lymph nodes in this region were completely removed. At the end of the procedure, the anesthesiologist changed the ventilation mode to manual control and increased airway pressure to temporarily increase venous pressure, and no lymphatic exudation or gelatinous object formation was observed. The patient was given a low-protein, low-fat diet after the surgery. On the postoperative first day, the patient's body temperature was about 38 °C but returned to normal after symptomatic treatment. The volume of fluid in the left neck drainage tube was about 20 mL on the first day, 20 mL on the second day, and 10 mL on the third day. On the third day, the patient reported a dull pain in the right upper abdomen, accompanied by chest tightness and dyspnea, which worsened after lying flat but was alleviated in the sitting position. The oxygen saturation level was about 95%, which was within the normal range. Since postoperative pain is very common, these symptoms were atypical and challenging to diagnose. To figure out the cause of the patient's lower abdominal pain, a chest and abdomen CT scan was performed immediately, and the results showed bilateral pleural effusion and bilateral lower lung inflation. So, the thoracic surgery department and respiratory medicine department were consulted, and the patient was transferred to the respiratory intensive care unit on the fourth day after the surgery. Bedside ultrasound positioning of pleural effusion and bilateral pleural drainage was performed, and a total of about 400 mL of milky fluid was drained from the bilateral pleural cavity. Thereafter, routine pleural effusion examination, biochemical tests, pleural effusion culture, and drug sensitivity tests were performed. Rivalta and chyle tests were positive, but the remaining results were negative. Based

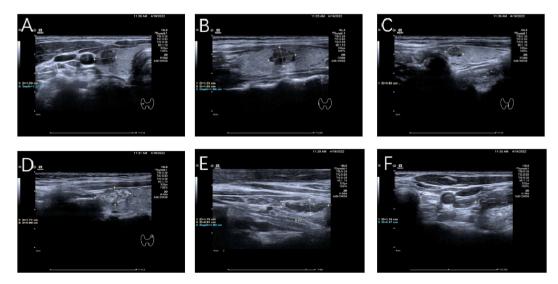


Fig. 1. Preoperative ultrasound images of the patient's neck (A), (B) the right thyroid, (C) isthmus of the thyroid gland, (D) the left thyroid, (E) metastatic lymph nodes in the right neck region IV, and (F) metastatic lymph nodes at the angle of the internal jugular vein in level IV of the left neck of this patient.

on these findings, combined with multidisciplinary consultation, the patient was diagnosed with bilateral chylothorax caused by neck dissection. Without timely diagnosis and effective treatment, bilateral chylothorax can lead to serious complications such as electrolyte disturbance, acid-base imbalance, and even death. Considering the chylothorax, drainage tubes were placed in both thoracic cavities, along with medications and diet control. The patient was given a low-fat diet, octreotide 0.1 mg Q8h subcutaneous injection, and supplementation with vitamins and electrolytes. On postoperative day 5, 600 mL of left thoracic drainage, 500 mL of right thoracic drainage, and 10 mL of neck drainage were reported. On postoperative day 6, the left thoracic drainage volume was reduced to 120 mL, and the right thoracic drainage volume to 400 mL; however, the neck drainage volume remained the same. Therefore, the neck drainage tube was removed, and the chest CT was reviewed. The bilateral pleural drainage volume was 20 mL on the 7th postoperative day. On day 8, 10 mL of left thoracic drainage and 8 mL of right thoracic drainage were obtained. On the 9th day, the volume of the left thoracic drainage fluid was 10 mL, and the right thoracic drainage tube had 5 mL of fluid. After pleural effusion localization, the right thoracic drainage tube was removed. The average volume of drainage fluid from the left chest on postoperative days 10-14 was approximately 5-8 mL per day. On day 14, the chest position and pleural effusion location were performed again, and no obvious effusion was found. At the same time, due to the patient's good compliance and tolerance, the left chest wall drainage tube was removed. The postoperative cervicothoracic drainage volume is shown in Fig. 2. Postoperative CT images of bilateral pleural effusions before and after the treatment are shown in Fig. 3. In Fig. 3A and B show the lung window and mediastinal window images of the patient's chest CT on the 3rd day after surgery, and C and D show the lung window and mediastinal window images of the patient's chest CT on the 6th day after surgery. The patient was discharged from the hospital on postoperative day 17. Routine pathological results of the left neck showed signs of lymph node metastasis (the left neck II area was 8/25, the left neck III area was 2/10, the left neck IV area was 1/20, and the left neck V area was 0/8). One month after her discharge, a chest X-ray showed no obvious abnormalities in the heart or lungs. The patient has been followed up for one year after the surgery, and there is no exudation or dyspnea in either lung.

3. Discussion

The incidence of chylous fistulas of the neck after cervical lymph node dissection is very low, about 1%–2% [6,7]. Notably, chylothorax, especially bilateral chylothorax, is even rarer. In general, chylous leakage after cervical lymphadenectomy does not cause pleural effusion or chylothorax. The exact mechanism of the development of chylothorax after cervical lymph node dissection remains poorly understood. Studies suggest that chylothorax pathogenesis may be related to any of the following mechanisms [8]: 1) intra-operative sudden abrupt increase in the thoracic duct pressure after ligation due to severe lymph node remodeling or unclear anatomical structure, which can build up negative pressure in the thoracic cavity during the inspiratory phase, or 2) during the cervical lymph node dissection, the main trunk and/or major branches of the thoracic duct may not be found on time, or the ligation line of the thoracic duct and its lymphatic duct may fall off, allowing the chyle fluid to flow directly into the mediastinum from the root of the neck and pressure-driven penetration into the pleura, or 3)when the integrity of the pleura is destroyed due to iatrogenic or trauma, and the lymphatic duct is broken, lymph fluid enters the thorax along the pleural tear, and then causes pleural effusion, that is, chylothorax [9].

In this case, we inflated the lung by overinflating the ventilator with the anesthesiologist during the operation. No obvious air leakage or exudation were observed in the operation area, indicating that the patient's pleura was not obviously damaged. The patient had less neck drainage after surgery, and there was no obvious neck swelling after the neck drainage tube was removed, and there was no significant increase in bilateral pleural drainage fluid. Therefore, it can be speculated that the mechanism of bilateral pleural effusion may be associated with abnormal pressure variation in the thoracic duct of the patient. Studies have reported that left cervical lymph node surgery in about 91.9% of thyroid cancer patients causes a small to moderate amount of pleural effusion after thoracic duct ligation, which is believed to be caused by the failure to establish immediate collateral circulation following the thoracic duct ligation [10], again supporting the first mechanism.

Chylothorax can affect the normal physiological functioning of vital thoracic organs such as the heart and lung, resulting in serious

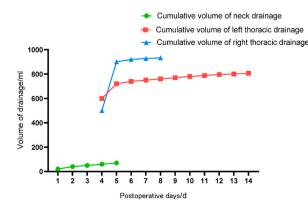


Fig. 2. Changes of cervicothoracic drainage volume over time.

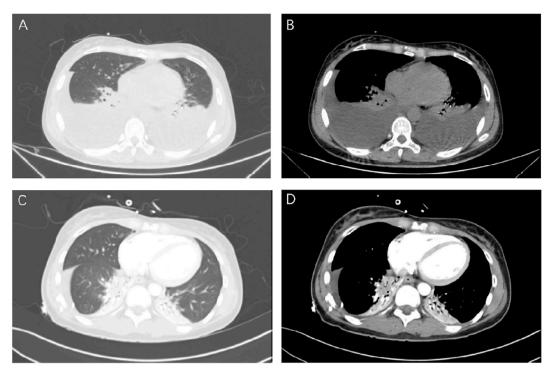


Fig. 3. Chest CT images before and after treatment. On the 3rd day after surgery, chest CT of lung window (A) and mediastinal window (B) showed massive bilateral pleural effusions. On the 6th day after surgery, chest CT images of lung window (C) and mediastinal window (D) exhibited significantly reduced bilateral pleural effusion.

health complications such as hypoxemia, dyspnea, and even heart failure. Massive extravasation of chyle fluid into the thoracic cavity can cause severe electrolyte imbalances and lipid metabolism disorders [11]. Therefore, in terms of diagnosis and prevention of chylothorax, the dissection of cervical lymph nodes, especially the left node, should be gentle, and the surgeon must be familiar with the anatomy of the thoracic duct to avoid unnecessary injury or unreliable ligation-related postoperative complications. After the dissection, the lungs are routinely inflated. The venous angle is carefully observed for lymphatic exudation or gelatin-like object formation and is covered with a gelatin sponge or hemostatic gauze. The volume, color, and nature of the neck drainage fluid should be observed over time following the surgery. Postoperatively, patients should be given a low-fat diet and taken care of to avoid any incidence of severe coughing. If the patient complains of chest pain, dyspnea, and other symptoms after surgery, lung auscultation and percussion should be performed on time, along with a chest X-ray or chest CT examination to understand the exact chest condition. Regarding invasive examinations, diagnostic thoracentesis is recommended for the removal of milky pleural fluid [12].

Regarding the effective treatment of chylothorax, the main aims are early detection and early drainage. Timely thoracic closed drainage or puncture aspiration can also achieve a good therapeutic effect. The course of the disease is about 7–14 days [13,14]. If we find patients with increasing postoperative white-colored neck drainage fluid volume, accompanied by pleural effusion, we should provide timely symptomatic treatments by first placing the drainage tube into the chest to clear pleural effusion, followed by biochemical examinations. To improve the condition of pleural effusion and atelectasis caused by breathing difficulties, the patient should be given parenteral nutrition, maintaining normal electrolyte balance and lipid metabolism. It can be beneficial to reduce chyle production through a low intake of dietary fat [15]. If the effect of conservative treatment is poor and the daily thoracic drainage volume exceeds 1000 mL, the thoracic surgery department should be consulted to perform thoracic duct ligation under the thoracoscope [16]. If the patient still presents infectious symptoms such as fever and chills after full drainage of bilateral pleural effusion or a higher white blood cell count in the effusion, there might be a chance of secondary complications, including pulmonary infection; therefore, antibiotics should be formulated according to the patient's condition, the nature of the microbial infection, and drug sensitivity. In such cases, the most commonly used antibiotics are penicillin, cephalosporin, aminoglycosides, quinolones, etc. Higher-grade antibiotics or combinations may be required for patients with severe infections or those infected with multi-drug-resistant bacteria.

Although the surgery was overall successful, the postoperative pain in her upper abdomen and difficulty breathing were unbearable. We acknowledge the seriousness and expertise of the doctor in charge of this patient in making the right decisions and conducting diagnostic tests that saved the patient's life with a successful treatment outcome.

4. Conclusion

Chylothorax is a relatively rare complication of cervical lymph node dissection in thyroid cancer patients and has a high probability

of being misdiagnosed and mistreated. Any injury to the main thoracic duct or lymphatic vessels must be avoided at all costs during surgery. If the patient exhibits certain postoperative symptoms, such as dyspnea and chest pain, the possibility of a chylothorax should be considered, and the chest X-ray or CT examination should be performed immediately. Once a chylothorax is diagnosed, a drainage tube should be introduced as soon as possible, followed by symptomatic treatments and surgical ligation of the thoracic duct, if necessary. In clinical practice, we should improve our understanding of such conditions, ensuring timely detection and treatment to avoid severe or fatal complications.

Data availability statement

The original contributions presented in the study are included in the article/Supplementary Material. Further inquiries can be directed to the corresponding author. Data will be made available on request.

Ethics statement

This study was approved by the Medical Ethics Committee of Henan Provincial People's Hospital. The medical research record number for this study is MR-41-22-011,497. We gained the written informed consent of the patient to use her clinical information and photographic material for the publication.

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CRediT authorship contribution statement

Wencong Sun: Writing – review & editing, Writing – original draft, Visualization, Software. **Xinhui Wang:** Writing – original draft, Visualization. **Chao Ding:** Data curation. **Yichen Wang:** Methodology. **Zijie Su:** Project administration. **Guoqing Li:** Writing – review & editing, Resources.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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