Endothoracic lymphatic plexus-hemiazygos vein anastomosis for chylothorax complicated with hepatocellular carcinoma: A case report

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Received May 8, 2023; Accepted January 24, 2024

DOI: 10.3892/etm.2024.12495

Abstract. For patients with hepatocellular carcinoma and cirrhosis, the rupture of thin lymphatic vessel walls leads to a profuse outflow of lymph fluid. Typically, chyloperitoneum tends to precede the development of chylothorax in patients with cancer. The present study describes the case of a male patient with hepatocellular carcinoma who developed chylothorax without chyloperitoneum. Computed tomography showed lymphatic system developmental abnormalities with a large volume of leaked lymph fluid. Multiple thoracic duct ligations (TDLs) failed, but a side-to-end lymphatic venous anastomosis (LVA) surgery resolved the symptoms. To the best of our knowledge, there are no reports of chylothorax occurrence after cirrhosis further complicated by congenital lymphatic abnormalities in the English-language literature. In conclusion, LVA could be appropriate to treat chylothorax when TDL is ineffective as a remedial or even prophylactic intervention.

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Abbreviations: TDL, thoracic duct ligation; LVA, lymphatic venous anastomosis; CT, computed tomography

Key words: chylothorax, hepatocellular carcinoma, TDL, LVA, lymphatic system abnormalities

Introduction

For patients with hepatocellular carcinoma and cirrhosis, venous hypertension can lead to lymph fluid stasis in the lymphatic plexus. The rupture of thin lymphatic vessel walls in some areas can lead to a profuse outflow of lymph fluid, eventually leading to chylothorax (1). Thoracic duct ligation (TDL) and thoracic duct interventional embolization are commonly used treatments for traumatic chylothorax (2). The present study details the unique case of a patient who developed chylothorax following cirrhosis, amidst congenital lymphatic system abnormalities. A chylothorax is the accumulation of chyle in the pleural space. This is most commonly seen following traumatic disruption of the thoracic duct and is typically diagnosed based on the milky appearance of fluid due to high-fat content. Most patients with chylothoraces require surgical exploration of the thoracic duct. Where TDLs fail, side-to-end lymphatic venous anastomosis (LVA) can offer an alternative. LVA reroutes lymphatic fluid directly into the bloodstream, bypassing the thoracic duct. The success of this procedure highlights LVA's potential as an effective treatment for chylothorax, especially in cases where traditional methods like TDL are unsuccessful. Each case of chylothorax requires an individualized approach, yet the findings of this study offer valuable insight into the expanding surgical options for this complex condition.

Case report

A 33-year-old man presented to Guangxi Medical University Cancer Hospital in January 2022. The patient presented with dull pain in the right upper abdomen without any other discomfort such as abdominal distension or diarrhea. Computed tomography examination previously performed in a local hospital indicated that the patient had massive liver cancer with multiple intrahepatic metastases in the right lobe of the liver. Furthermore, the examination also indicated that there was a tumor embolus formation (12.5x10.0x7.5 cm) in the right branch of the portal vein. The patient presented at Guangxi Medical University Cancer Hospital with an alpha fetoprotein level of 1,210 ng/ml. After 3 days, a surgical procedure,

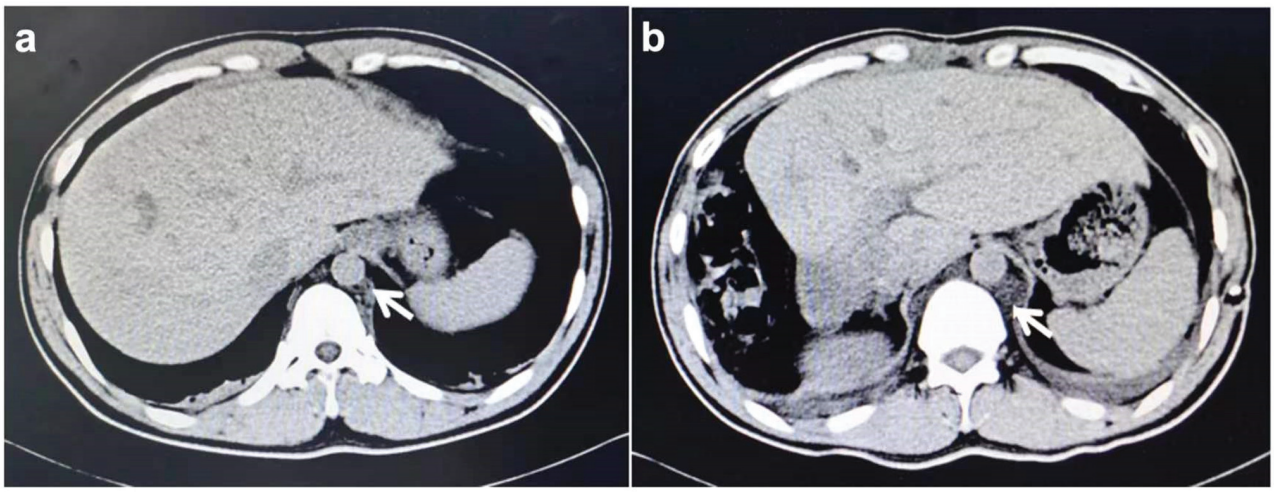

Figure 1. Dilated choroidal plexus can be found in hepatocellular carcinoma both (A) preoperatively and (B) post-right hemihepatectomy with portal vein thrombectomy. The white arrow indicates the dilated choroidal plexus.

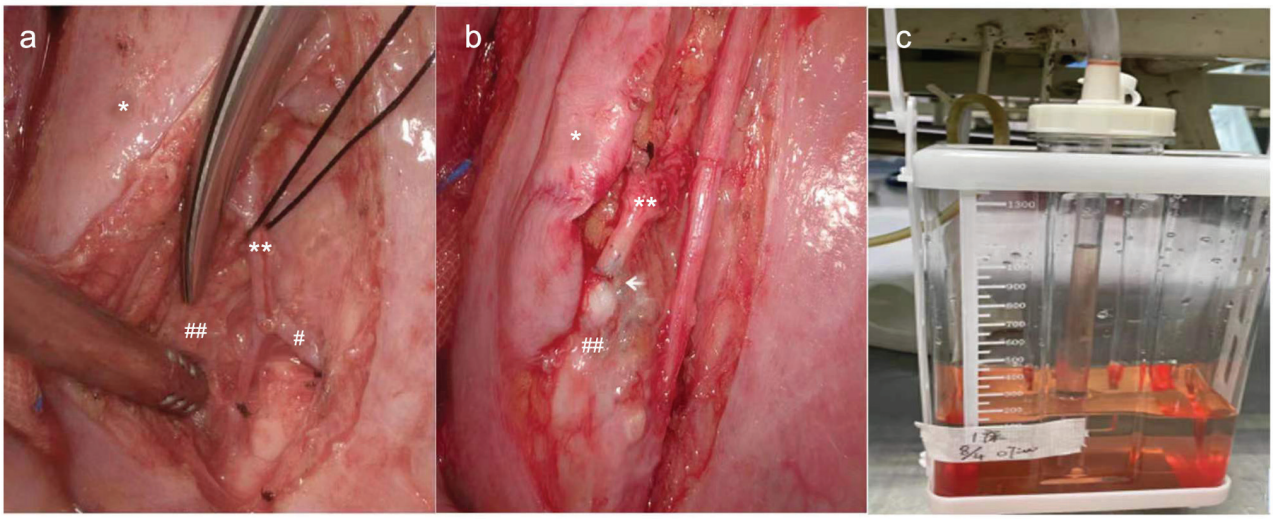

Figure 2. Side-to-end anastomosis of the lymphatic duct vein at the 8th intercostal level with a 9-0 suture. (A and B) Representative intraoperative images. *, Thoracic aorta; **, hemiazygos vein; #, intercostal branch of the hemiazygos vein; ##, dilated lymphatic plexus of the thoracic duct; white arrow, the anastomosis. (C) Change in drainage fluid post-anastomosis. Drainage fluid was transparent 1 day after the thoracic duct-hemiazygos vein anastomosis, amounting to 200 ml.

right hemihepatectomy with portal vein thrombectomy, was performed to treat primary liver cancer located in the right lobe of the liver. The cancer was complicated by portal vein thrombosis (BCLC Stage C and CNLC IIIB stage) (3,4). During the surgery, the medical team discovered that the cancer had invaded the right adrenal gland, which was then partially removed. A spontaneous chylothorax was identified in the left chest region postoperatively. In February 2022, the patient underwent a trans-thoracoscopic low TDL; however, their symptoms did not improve postoperatively, with daily chest drainage of chyle fluid ranging from 1,500 to 2,000 ml. The patient was assessed using computed tomography (CT) and then diagnosed with massive hepatocellular carcinoma in combination with cirrhosis. CT findings showed dilated choroidal plexus, and the presence of multiple cystic foci of vascular origin in the inferior vena cava and abdominal aorta (Fig. 1).

In April 2022, a high TDL was performed, following which aggravated the tortuous expansion of the thoracic duct, and multiple points of lymphatic fluid exudation were observed. A leak was found below the 8th rib, with exudation of a large amount of milky white lymphatic fluid. A hemiazygos vein and its bifurcations were observed next to the leak. Therefore, a side-to-end LVA at the location of the leak was performed to re-establish lymphatic circulation and reduce intra-thoracic pressure in the lymphatic vessels (Fig. 2A and B). After completion of the anastomosis, milky white lymphatic fluid was observed entering the vein, and the diameter of the previously dilated lymphatic plexus was reduced. No lymphatic fluid leaked from the original multiple exudation points after ~3 min. On postoperative day 1, the drainage fluid was clear (volume, 200 ml; Fig. 2C). The patient was discharged 3 days later and the chest drain was removed.

Discussion

Chyloperitoneum and chylothorax may result from surgical trauma or congenital lymphatic system abnormalities (5). In the present case report, the surgical procedure for treating liver cancer did not involve the thoracic region; therefore, chylothorax was not caused by the operation. To the best of our knowledge, no previous studies have reported spontaneous chylothorax without abdominal chyle in adults with cirrhosis. Additional research is required to determine whether a dilated non-venous plexus exists in patients with a history of cirrhosis and liver cancer who are undergoing chest and abdominal CT, as to the best of our knowledge this has not been previously reported. If so, it may be necessary to clarify whether the patient has congenital dysplasia of the lymphatic system. When performing the liver cancer-related treatment, clinicians may need to consider prophylactic thoracic duct-internal jugular vein anastomosis. Abnormal congenital development of the lymphatic system in the patient assessed in the present study was the cause of the chylothorax in the absence of abdominal chyle and the reason for the previous TDL failure. The presence of abnormal lymphatic system development should be suspected in patients with hepatocellular carcinoma when CT findings indicate abnormal vascular dilatation. LVA can be performed as a salvage surgery for patients who have undergone TDL and thoracic catheter interventional embolization with no postoperative improvement.

Acknowledgements

Not applicable.

Funding

This case report was supported by the Guangxi Medical and Health Suitable Technology Development and Popularization Application Project (grant no. S2020095).

Availability of data and materials

The data generated in the present study may be requested from the corresponding author.

Authors' contributions

XL and YJ designed and performed the surgical plan. CL, YH, YJ and NM followed-up the patient, drafted the article and all authors have read and approved the final version of the manuscript. XL and YJ confirm the authenticity of all the raw data.

Ethics approval and consent to participate

Informed consent was obtained from the patient.

Patient consent for publication

The patient provided written informed consent for the publication of this case report.

Competing interests

All authors declare that they have no competing interests.

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