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Case Report

Unusual Presentation of Bilateral Chylothorax and Chylous Ascites with Pancreatic Adenocarcinoma: A Case Report

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Keywords
Chylothorax · Chylous ascites · Pancreatic adenocarcinoma · Pancreatic cancer

Abstract
Chylothorax is a lymphatic fluid, which is contained in the pleural cavity. Chylothorax has a typical milky white fluid appearance and is diagnosed by triglyceride concentrations >110 mg/dL. The combination of chylothorax with chylous ascites is a rare condition that is difficult to manage. We present a case of a 60-year-old man who presented with dyspnea for 1 week and generalized abdominal distention. He was diagnosed with advanced-stage pancreatic adenocarcinoma and received palliative chemotherapy. On follow-up examination, he showed decreased breathing sounds in both lungs and marked ascites. Chest radiography showed bilateral pleural effusion, which was more prominent in the left hemithorax than the right hemithorax. Thoracocentesis and abdominal paracentesis were performed, and the finding of pleural and ascites fluid was compatible with bilateral chylothorax and chylous ascites. Although the patient was receiving dietary modification, he still required repeated thoracentesis. Bilateral chest tube insertion with medical pleurodesis with talc successfully treated his chylothorax. This treatment should be considered as an option for malignancy-associated chylothorax besides systemic cancer treatment to improve patients' quality of life.

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Introduction

Pancreatic cancer has a wide range of clinical presentations, such as early satiety, back pain, jaundice, and weight loss. The clinical course of pancreatic cancer is mostly subtle. Therefore, the prognosis is poor at the time of diagnosing this cancer owing to the advanced stage of the disease. Despite combination chemotherapy being the mainstay treatment of this catastrophic disease, other novel treatments including immune-checkpoint inhibitors, tyrosine kinase inhibitors such as poly-ADP-ribose polymerase inhibitors showed promising results in selected cases harboring actionable molecular alterations [1]. Concomitant chylothorax and chylous ascites are an infrequent presentation and challenging to manage. We report an uncommon presentation of bilateral chylothorax and chylous ascites caused by advanced-stage pancreatic adenocarcinoma and successfully treated the bilateral chylothorax with medical pleurodesis.

Case Presentation

A 60-year-old man was admitted to the hospital because of shortness of breath and worsening abdominal swelling for 1 week. Two months earlier, he was diagnosed with advanced-stage pancreatic adenocarcinoma with carcinomatosis peritonei. He received palliative combination chemotherapy regimen consisting of oxaliplatin, irinotecan, fluorouracil, and leucovorin (FOLFIRINOX).

On an examination, the patient had tachypnea. There were decreased breathing sounds in both lungs, and the abdomen showed marked distention. A chest radiograph showed that bilateral pleural effusion was more prominent in the left hemithorax than in the right hemithorax (Fig. 1a). Computed tomography of the chest and whole abdomen demonstrated new moderate bilateral pleural effusion without evidence of lung metastasis or intrathoracic lymphadenopathy. The pancreatic tail mass had a stable size of 2.4 × 1.9 cm but showed an increased size of multiple peritoneal carcinomatoses compared with that in the initial computed tomography scan. Moderate ascites was also observed (Fig. 2).

Bilateral thoracocentesis was performed. The pleural fluid showed a milky appearance (Fig. 3). Pleural fluid analysis showed a triglyceride (TG) concentration of 199 mg/dL, lactate dehydrogenase concentration of 272 U/L, protein concentration of 3.2 g/dL, and glucose concentration of 120 mg/dL, and lymphocytes were dominant. The serum protein

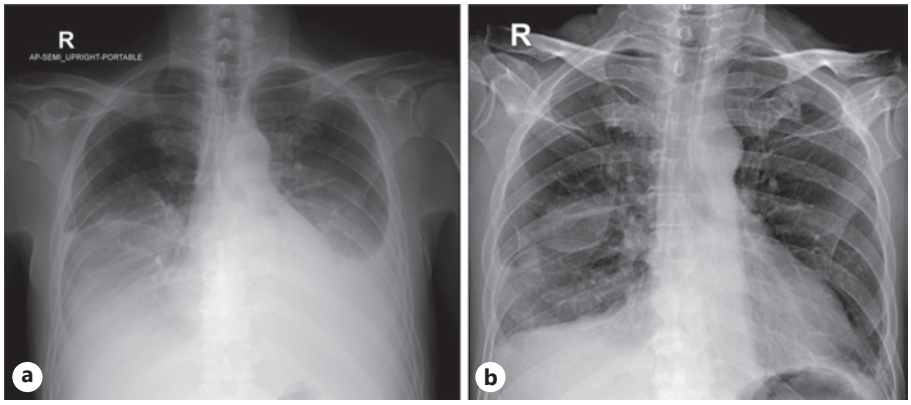

Fig. 1. Chest radiograph shows bilateral pleural effusion (**a**) and resolution of bilateral pleural effusion after medical pleurodesis (**b**).

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Fig. 2. Photograph showing a milky appearance of pleural fluid.

Fig. 3. Abdominal CT shows a pancreatic tail mass of 2.4 × 1.9 cm with a moderate amount of ascites.

concentration was 5.2 g/dL, and the lactate dehydrogenase concentration was 193 U/L. The pleural fluid was diagnosed as exudative on the basis of Light's criteria. The patient also received abdominal paracentesis. His ascites profile showed a milky appearance with an albumin concentration of 0.84 g/dL, protein concentration of 2.0 g/dL, and TG concentration of 199 mg/dL. A cytopathological evaluation and mycobacterial culture from ascites and pleural effusion were negative. These results indicated that the definitive diagnosis was bilateral chylothorax with chylous ascites. A multidisciplinary team comprising a nutritionist, medical oncologist, and pulmonologist consulted with our patient.

After 1 week of a dietary change with medium-chain TGs, the patient still required repeated thoracentesis to alleviate his dyspnea symptoms. Bilateral chest tubes were placed, followed by medical pleurodesis with talc. A chest radiograph after medical pleurodesis (Fig. 1b) showed markedly decreased bilateral pleural effusion. His chest symptoms improved and he was discharged home. Chylous ascites was also managed by repeated abdominal paracentesis, and chemotherapy was changed to weekly gemcitabine. Despite the change in chemotherapy, his condition still deteriorated, and he died 8 weeks later.

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Table 1. Summary of case reports of pancreatic adenocarcinoma with chylous ascites with or without chylothorax

Author, year	Sex	Age, years	Clinical presentation	Treatment
Kostiainen et al. [9]	Male	42	Bilateral chylothorax and chylous ascites	Thoracentesis, abdominal paracentesis, and palliative care
Tang et al. [10]	Female	67	Chylous ascites	Dietary restriction and palliative care
Wagayama et al. [11]	Female	60	Chylous ascites	Dietary restriction, furosemide, and peritoneovenous shunting with the Denver shunt for chylous ascites treatment
Ghafoor et al. [12]	Male	20	Chylous ascites	Dietary restriction, orlistat, abdominal paracentesis, and palliative chemotherapy
Satala et al. [13]	Male	76	Chylous ascites	Abdominal paracentesis and palliative care
Lutchmansingh et al. [14]	Female	86	Right chylothorax and chylous ascites	Palliative care

Discussion/Conclusion

A milky appearance of fluid is a clinical indication of chylothorax (pleural fluid) or chyloperitoneum (ascites). The presence of chylomicrons or elevated TG concentrations >100 mg/dL is considered the gold standard for the diagnosis of chylothorax [2]. Malignant chylothorax is the most common cause of nontraumatic chylothorax. In 84% of chylothorax cases, effusion occurs unilaterally, and half of them present in the right hemithorax [3]. These patients can present with a wide range of symptoms, such as cough, tachypnea, and chest discomfort.

The diagnosis of chylous ascites is made by the finding of TG concentrations >200 mg/dL, although some studies used a cut-off point of 110 mg/dL [4]. The TG concentration in our patient was 199 mg/dL, which is in the range of the criterion for diagnosing chylous ascites. Clinical presentations of chylous ascites are also varied, such as progressive abdominal distention, abdominal pain, and weight gain. Malignancy is still the most common cause of chylous ascites in adults, and lymphoma accounts for one third of all cases.

Chylothorax concomitant with chylous ascites is rarely encountered. This presentation can be caused by lymphatic duct disruption either from injuries or obstruction on multiple levels. Lymphatic duct disruption between the cisterna chyli and aortic hiatus leads to chylous ascites. Disruption of the lower thoracic duct causes right chylothorax, and disruption of the higher thoracic duct results in left chylothorax. However, we did not find any intrathoracic lymphadenopathy in our patient. The other mechanism of chylothorax with chylous ascites is transdiaphragmatic fluid migration from chylous ascites, which resembles hepatic hydrothorax or nephrotic syndrome. Pleural fluid TG concentrations in our case were identical to ascitic fluid TG analysis, which could be an explanation for the mechanism of chylothorax. An alternative method that can identify the source of chylothorax is an intraperitoneal injection of radioisotope (^{99m}Tc-sulfur colloid). The appearance of a radioisotope in the pleural space within 90 min can confirm this hypothesis [5], but we did not perform this method in our case.

Concomitant chylothorax and chylous ascites in patients with primary intra-abdominal malignancy has been reported in those with various types of cancer, such as uterine cancer [6], gallbladder cancer [7], and stomach cancer [8], most of which showed a grave prognosis.

Pancreatic adenocarcinoma with chylous ascites with or without chylothorax has also been reported [9–14] (Table 1). However, no cases of pancreatic adenocarcinoma had unique clinical characteristics as in our case.

The treatment of chylothorax and chylous ascites is challenging. Management of chylothorax genuinely requires a multidisciplinary team because of the need to treat the underlying etiology, adjust the diet, and cautiously follow up for electrolyte imbalance and malnutrition. Dietary modification using medium-chain TGs leads to decreased lymphatic flow, and the initiation of a somatostatin analogue can reduce chyle production, lymph flow, and intestinal fat absorption. Interventional treatment, such as pleural drainage via tube thoracostomy with pleurodesis, is recommended for chylothorax treatment in case of failed medical therapy or high-output chylothorax (≥1.1 L over 24 h or >1 L/day for more than 5 days) [3]. Early medical pleurodesis successfully controlled bilateral chylothorax in our case and helped alleviate dyspnea symptoms. This treatment should be considered as an option for malignancy-associated chylothorax besides systemic cancer treatment, to improve patients' quality of life.

Although dietary modification, a somatostatin analogue, orlistat, and repeated paracentesis are the initial approaches for chylous ascites, there is limited information on the best treatment. The treatment of primary cancer is also crucial in controlling malignant chylous ascites in which peritoneovenous shunting may be considered in selected cases [4, 15]. Despite the change in chemotherapy in our case, the patient still did not respond to treatment and became worse. Because of his poor performance status, the best supportive care was suitable for the patient.

In conclusion, we report an uncommon clinical presentation of bilateral chylothorax and chylous ascites in advanced-stage pancreatic cancer. The diagnosis was made by analysis of fluid, and management aimed to treat the underlying disease, which was cancer. The management of chylothorax and chylous ascites should combine medical treatment with dietary adjustment, including surgical treatment if conservative management fails. A multidisciplinary team approach is essential for successful treatment of these patients.

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Statement of Ethics

Written informed consent was obtained from the patient's next of kin for publication of the details of his medical case and any accompanying images. The study was approved by Chulabhorn Research Institute Ethic Committee (No. 001/2565).

Conflict of Interest Statement

All authors declare no conflict of interest.

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Author Contributions

Panisara Fangsaard and Jirapa Puriwekin drafted the manuscript. Nanthida Phattraprayoon reviewed and edited the manuscript. Teerapat Ungtrakul contributed to the manuscript's design, supervision, review, and editing.

Data Availability Statement

All data generated or analyzed during this study are included in this article. Further inquiries can be directed to the corresponding author.

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