Title:

Refractory chylothorax in HIV/AIDS-related disseminated mycobacterial infection

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Contributors:
All authors cared for the patient. TT wrote the report.

Competing interests:
None declared.

Patient consent:
Obtained.

Keywords: HIV, AIDS, NTM, chylothorax

Word count: 354 words
A 38-year-old man presented to a previous hospital in December 2011 with a high fever and acute abdominal pain. He was found to have abdominal lymphadenopathy and was diagnosed with human immunodeficiency virus (HIV) infection. His CD4 count was 25/mm³ and viral load was 490,000 copies/ml. He was transferred to our department for further investigations and treatment. Whole-body computed tomography revealed bulky, widespread lymphadenopathy, especially in the abdomen, but no ascites or pleural effusion (figure 1A, 1B). Stool, bone marrow, and an abdominal lymph node specimen were positive for acid-fast bacillus stains. A diagnosis of disseminated nontuberculous mycobacterial infection (NTM) with *Mycobacterium genavense* was confirmed by 16S rRNA gene sequence analyses of a sample of bone marrow and a lymph node specimen. Antiretroviral therapy with Raltegravir+Tenofovir+Emtricitabine anti-NTM treatment with Clarithromycin+Ethambutol+Rifabutin+Streptomycin, and prednisone 0.5 mg/kg/day to prevent immune reconstitution syndrome were introduced. However, bulky abdominal lymphadenopathy persisted despite treatment and led to recurrent intestinal obstruction and ileal strangulation requiring surgical intervention. The abdominal symptoms were markedly relieved after 8 months of treatment, though the bulky lymphadenopathy persisted.

Ascites and a chylous pleural effusion were identified 1 year after treatment.
initiation, in December 2012 (figure 1C, 1D, 1E, 1F). Obstruction or destruction of the thoracic duct by disseminated NTM infection was considered as a possible cause of the chylothorax. The bilateral pleural effusion had progressed further by April 2013 (figure 1G, 1H). Lymphoscintigraphy confirmed lymphatic obstruction and thoracic duct leakage (figure 2A, 2B, 2C).

Non-malignant acquired immune deficiency syndrome (AIDS)-related chylothorax has been reported in association with Kaposi’s sarcoma and tuberculosis,[1, 2] but no cases related to NTM have been reported. Continued loss of immunoglobulins and T lymphocytes into the pleural effusion leads to immunosuppression, and we anticipate that the survival prognosis in such cases will be poor, unless the chylothorax is controlled.[3] A number of additional interventions can be considered for the management of chylothorax, including pleural drainage, dietary modifications (fasting or reduced fat diet), pleurodesis, and thoracic duct ligation. However, specific guidelines are lacking, particularly in refractory cases. Long-lasting management guidelines for chylothorax are needed, especially in immunocompromised patients such as those with HIV/AIDS.
References


**Figure Legends**

**Figure 1.** (A) No pleural effusion was identified in 2011. (B) Bulky abdominal lymphadenopathy was identified in 2011. (C) Small amount of pleural effusion was identified in 2012. (D) Abdominal lymphadenopathy enlarged in 2012. (E, F) Chylothorax was confirmed. (G, H) Pleural effusion worsened in 2013.

**Figure 2.** Dynamic lymphoscintigraphy images. (A) Tc-99m lymphoscintigraphy after tracer injection in the lower limbs showed lymphatic obstruction at the abdominal level. (B) Thoracic-duct scintigraphy with oral I$^{123}$ β-methyl-iodophenylpentadecanoic acid showed tracer accumulation in the intestine, but not in the thorax, at 8 h after administration. (C) Tracer accumulation was identified in the thorax at 24 h after administration.