

# Bilateral Chylothorax. An Unusual Presentation in the Course of Immune Reconstitution Inflammatory Syndrome, HIV Infection and Kaposi's Sarcoma: A Case Report

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## Abstract

Immune reconstitution inflammatory syndrome (IRIS) is a term used to describe the paradoxical worsening of a pre-existing infection or the presentation of a previously undiagnosed condition in HIV-infected patients soon after commencement of antiretroviral therapy (ART). We report a case of a patient with HIV infection and Kaposi sarcoma, who developed bilateral chylothorax as a manifestation of IRIS, after the initiation of highly active antiretroviral therapy. Bilateral chylothorax is extremely rare and has been associated with significant morbidity and mortality. We present here the successful management with bilateral video-assisted thoracic surgery.

**Keywords:** Immune reconstitution inflammatory syndrome; Kaposi sarcoma; HIV infection; Chylothorax

## Introduction

Immune reconstitution inflammatory syndrome (IRIS) is a term used to describe the paradoxical worsening of a pre-existing infection or the presentation of a previously undiagnosed condition in HIV-infected patients soon after commencement of antiretroviral therapy (ART) [1]. Contemporary ART is both potent and tolerable for long periods. ART in HIV/AIDS patients leads to dramatic reduction in plasma viral load, improvement in CD4+ T cell counts and partial restoration of overall immune function [2]. It is unclear whether complete immune reconstitution ever occurs,

but it is clear that patients who commence ART when they are very immunodeficient are susceptible to immune reconstitution disorders [3].

Disseminated Kaposi's sarcoma (KS) is a common problem in patients with the acquired immunodeficiency syndrome. Intrathoracic involvement occurs clinically in as many as a fifth of patients with KS [4, 5], and has been reported in 11 of 23 consecutive patients in an autopsy series [6]. Pulmonary KS, usually, follows the appearance of characteristic lesion of the skin [7]. Its clinical and radiographic presentation may mimic pneumonia due to opportunistic infections, although nodular infiltrates and intrathoracic lymph node enlargement are commonly seen. Because pleural effusion only rarely occurs in patients with opportunistic infections, such as *Peunonocystis carinii*, the presence of pleural effusion in a patient with cutaneous KS suggests the diagnosis of pulmonary KS [8].

Chylous pleural effusion, on the other hand, has been reported previously in only a few patients with KS [9, 10]. Chylothorax represents chyle in the pleural cavity. The presence of chylomicrons and a triglyceride level > 110 mg/dL in the aspirated pleural fluid confirms the diagnosis of chylothorax [11]. We report a case of a patient with HIV and KS, who developed bilateral chylothorax as a manifestation of IRIS, after the initiation of highly active antiretroviral therapy.

## Case Report

A 37-year-old HIV-infected man was transferred to the thoracic surgery department due to bilateral chylothorax. The patient presented with respiratory distress and also malnutrition, hypovolemia and electrolyte imbalance. During his 30-day hospitalization, a central line and parenteral nutrition with medium chain triglycerides had been started in order to medically treat the condition. The diagnosis of chylothorax was documented based on bilateral thoracentesis with pleurocath and the appearance of a milky (chylous) pleural effusion with an elevated triglyceride level (> 110 mg/dL). One week after pleurocath insertion, a fever up to 38 °C complicated chylothorax. Antibiotics was started (teicoplanin

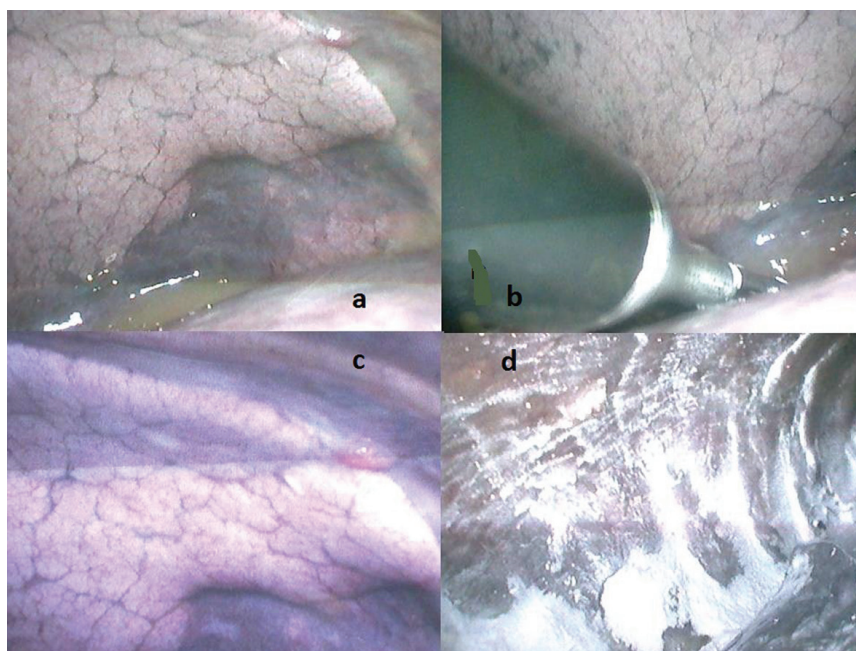
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**Figure 1.** (a) Initial intraoperative view. (b) Thoracoscopic clip ligation of the thoracic duct. (c) Cherry-like appearance of Kaposi's sarcoma (right upper corner). (d) Final view after talc insufflation, before lung re-expansion.

400 × 2, meropenem 1 × 3, itraconazole sir 10 × 2) along with antiretroviral treatment. The patient was treated with video-assisted thoracoscopic surgery (VATS) with clip ligation of the thoracic duct with talc pleurodesis at first at the right side (Fig. 1). Twenty days later, we operated on the left side. Postoperatively, in the second operation, the patient developed an acute respiratory distress syndrome and stayed for 24 h at the intensive care unit. The distress syndrome was probably a result of pulmonary edema due to lung re-expansion. Another possible explanation was an systemic reaction due to talc pleurodesis. The patient was extubated 20 h later and was discharged 10 days later in stable clinical condition. Three years later, the patient is stable in excellent clinical condition.

## Discussion

Chylothorax is caused by disruption or obstruction of the thoracic duct. An acute onset of pleural effusion and the presence of turbid fluid that does not clear when centrifuged or that contains at least 1.24 mmol/L (110 mg/dL) triglyceride establish the diagnosis [12]. In advanced HIV infection, chylothorax may also be caused by infiltration of the thoracic duct by KS [13].

Chylothorax, the presence of lymph in the pleural space, has remained a difficult clinical challenge for which the ideal treatment has not been well established. The general

approach to the problem varies in that some clinicians adopt early surgical intervention while others adopt a conservative approach to the problem.

The treatment of chyloous pleural effusions required prompt and aggressive measures. Although conservative approach may have a role to play in small chylothoraces, therapeutic thoracentesis and tube thoracostomy are the initial step in large chylothoraces that cause respiratory distress. During the period of excessive chyle leak, patients are generally advised nil by mouth or a diet rich in low fat, medium-chain triglycerides. Many follow guidelines that Selle and associates [14] suggested. They propose surgery in trauma, daily loss of chyle exceeds 1,500 mL in adults or > 100 mL/kg body weight per day in a child, when the output of chyle is not diminished over a 14-day period, or when nutritional complications seem imminent [15]. Graham et al recommend that surgical intervention is highly effective and seems warranted if the chylothorax has not responded after 5-7 day trial of medical therapy in both adults and children [16].

Our patient was treated with VATS with talc pleurodesis and the result was excellent. Its use specially for chylothorax has only a few case reports [17].

VATS offers the advantage of access to the entire hemithorax with excellent visualization of the mediastinal structures including the thoracic duct, without the morbidity of thoracotomy. It also allows application of clips to the thoracic duct at the aortic hiatus, or to thoracic duct injuries or pleural defects at other sites. Finally, it allows chemical

pleurodesis and application of talc or fibrin glue as required.

In the past, the mortality due to chylothorax was in excess of 50%. Currently, the morbidity and mortality have improved due to the more aggressive management strategies adopted. VATS offers the advantage of successful treatment of chylothorax while avoiding thoracotomy. The limited incisions of thoracoscopy produce less postoperative pain, less rehabilitation time and less recovery time, while accomplishing the same desired outcome. We strongly propose VATS for the management of chylothorax for these reasons.

### Conflict of Interest

None.

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