An Unusual Case of Chylothorax Complicating Childhood Tuberculosis

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Summary. Endobronchial tuberculosis (EBTB) and chylothorax are rare clinical disorders. The concurrence of these two disorders as manifestations of childhood pulmonary tuberculosis has not been reported. We report a 4-month-old boy presenting with chylothorax as the initial presentation of tuberculosis that has been successfully treated with octreotide, antituberculosis drugs and steroid therapy.


Key words: chylothorax; endobronchial tuberculosis; child.

INTRODUCTION

Chylothorax is the occurrence of chyle in the pleural space due to damage or blockage of the thoracic duct.1–3 Chylothorax may be caused by cardiac surgery, congenital malformations, trauma, malignant tumors, lymphangiomyomatosis, sarcoidosis, hepatic cirrhosis, and infection including tuberculosis (TB).4–11 Chylothorax, as the first manifestation of TB is rarely reported.5–14 Endobronchial Tuberculosis (EBTB) is defined as TB of the tracheobronchial tree and most often a complication of primary pulmonary TB.15–18 The concomitant appearance of chylothorax and EBTB in the same patient has not been previously reported. Here, we report a case of chylothorax with EBTB.

CASE REPORT

A 4-month-old boy was admitted to Marmara University Hospital with a history of shortness of breath for 3 days. There were no prior respiratory symptoms. His respiratory rate was 60 breaths/min. He did not have fever. Physical examination was unremarkable except for diminished breath sounds and dullness to percussion over the lower right hemithorax. The laboratory data revealed a white blood cell (WBC) count of 7,600/mm^3, a hemoglobin level of 9.0 g/dl, a platelet count of 515,000/mm^3 with an erythrocyte sedimentation rate of 23 mm/hr. Serum biochemistry profile and urinalysis were within normal limits. The chest X-ray (CXR) was suggestive of right pleural effusion. Thoracentesis of the right hemithorax disclosed milky pleural fluid with a glucose level of 87 mg/dl (Concomitant serum glucose level was 100 mg/dl), protein level of 9.0 g/dl, cholesterol level of 120 mg/dl, triglyceride level of 150 mg/dl, and WBC count of 8,800/mm^3 (67% lymphocytes). Because the triglyceride level was higher than 110 mg/dl, chylothorax was diagnosed and conservative management including chest tube, use of medium-chain triglycerides (MCT) and total parenteral nutrition (TPN) was initiated simultaneously on 2nd hospitalization day.

Tuberculin skin test (TST) and thorax computerized tomography (CT) were performed for differential diagnosis among congenital malformations, malignant tumors and TB. There were multiple mediastinal lymphadenopaties (LAP) with a size of 4 cm in addition to pleural effusion on thorax CT (Fig. 1). Tumor markers, CT of the abdomen and bone marrow aspiration results were normal. Direct smear of pleural fluid for M. tuberculosis and...
culture of other micro-organisms were negative. TST enduration was measured as 17 mm. Further attempts to clarify the family history revealed the previously unreported, recent paternal infection with TB and the father had been on anti-TB treatment for 3 months. Paternal reports suggested that his first smear was positive and sputum culture grew *M. tuberculosis* but drug resistance status was unknown. In the light of this new information and TST positivity, gastric aspirates of the patient were collected on three consecutive days. All samples were screened for acid-fast bacilli (AFB) with Ziehl–Neelsen (ZN) technique and cultured in Lowenstein–Jensen medium. Gastric aspirates microscopy by ZN stain revealed negative results. Flexible bronchoscopy (FB) was performed using the Olympus® BF 3C160, 2.8 to detect whether external compression of the bronchi by LAP or endobronchial lesions were present. The FB examination showed narrowing at the orifice of the right main bronchus. Microscopic examination of bronchoalveolar lavage (BAL) fluid revealed AFB. Anti-TB treatment with quadritherapy including isoniazid (INH), rifampicin (RFM), pyrazinamide and streptomycin was administered and methylprednisolone (2 mg/kg/24 hr) was added due to compression of the bronchial lumen >50% by LAP on day 9 of hospitalization.

Due to failure of conservative management including chest tube drainage, MCT, TPN and anti-TB treatment; intravenous infusion of octreotide, a somatostatin analogue, was administered at an infusion rate of 1 mcg/kg/hr for resolution of chylothorax on day 30 of admission. A marked reduction in pleural drainage was observed within 3 days of octreotide infusion, with complete cessation of drainage by day 36 of admission. Octreotide was discontinued after 8 days of continuous infusion, on day 40 of admission chest drains were removed. *M. tuberculosis* was cultured from BAL fluid and the bacilli were drug susceptible. The patient was discharged with anti-TB treatment. The dose of methylprednisolone was tapered gradually and discontinued at 8th week. Follow up FB was performed due to persistent atelectasis of the right middle lobe on CXR and thorax CT at 8th week of anti-TB treatment (Fig. 2). The FB examination showed a polypoid tumoral endobronchial mass lesion at the right bronchus intermedius occluding the lumen almost totally (Fig. 3). BAL microscopy by ZN stain revealed negative results for *M. tuberculosis*. Anti-TB treatment was continued as INH and RFM. Due to persistence of atelectasis at the 16th week, FB was repeated for a possible surgical resection. However, endobronchial lesion diminished in size and we continued with anti-TB treatment. Thorax CT showed improvement of atelectasis and disappearance of LAP at the 24th week. Anti-TB treatment was continued for 9 months with complete clinical, radiological and bronchoscopical improvement. The patient was followed up for 16 months after completion of therapy without any symptoms or radiological findings.

ABBREVIATIONS

EBTB endobronchial tuberculosis
BAL bronchoalveolar lavage
CXR chest X-ray
CT computerized tomography
FB flexible bronchoscopy
INH isoniazid
LAP lymphadenopathy
MCT medium-chain triglycerides
RFM rifampicin
TB tuberculosis
TPN total parenteral nutrition
TST tuberculin skin test
WBC white blood cell
ZN Ziehl–Neelsen

*Fig. 1. Chest computer tomography at diagnosis: paracardiac and subcarinal lymphadenopathy with a right sided pleural effusion.*

*Fig. 2. Right middle lobe atelectasis despite 8 weeks of anti-tuberculosis treatment.*
DISCUSSION

Chylothorax is a condition in which chyle leaks into the pleural cavity from the thoracic duct. The diagnosis is made by analysis of the pleural fluid. Triglyceride levels greater than 110 mg/dl are highly suggestive of a chylous effusion. Pseudochylothorax is a fluid which has a very high content of cholesterol. It can occur when a fluid has been present for a long time in the pleural space and the cholesterol level is permanently high in the patient (>200 mg/dl). In our case, because the triglyceride level was higher than 110 mg/dl and the cholesterol level was lower than 200 mg/dl, chylothorax, instead of pseudochylothorax, was diagnosed. In the treatment of chylothorax, conservative management with use of MCT and TPN is often effective. Octreotide can be administered if there is inadequate response to conservative management. In our case, because the drainage did not diminish with conservative management, treatment with octreotide was attempted and dramatic resolution in chylous effusion was observed.

*M. tuberculosis* as the cause of chylothorax has been only occasionally reported. The mechanism of tuberculosis for the development of chylothorax may be related to the enlarged lymph nodes obstructing the thoracic duct, or to the direct involvement of the lymphatic system by TB. In our case, there were multiple mediastinal LAP, therefore, the cause of chylothorax in our patient might be due to the enlarged lymph nodes obstructing thoracic duct or major lymphatic channels in the mediastinum by *M. tuberculosis* infection.

EBTB is most often a complication of primary pulmonary TB. Actual incidence is unknown because bronchoscopy is not routinely performed in all patients with TB. Sources of EBTB may include direct implantation of tubercle bacilli into the bronchus from an adjacent pulmonary parenchymal lesion, direct airway infiltration from tuberculous mediastinal LAP and erosion and protrusion of an intrathoracic LAP into the bronchus. EBTB can present as extrinsic bronchial obstruction by LAP or as endobronchial lesions such as mucosal inflammation and ulceration, caseification, granuloma formation and polypoid-tumoral mass lesion. It has been reported that some children with EBTB had initial worsening of airway compression and an increase in the size of lesions after the initiation of treatment of TB. This is believed to be the result of a hypersensitivity reaction. Treatment with corticosteroids together with anti-TB drugs has been shown to be effective for the resolution of EBTB in several studies. Many experts recommend steroids for the treatment of pediatric patients with EBTB. In this case, the first presentation of EBTB was extrinsic bronchial obstruction by LAP, and polypoid-tumoral lesion was seen at the 8th week and finally resolved at 8th month. EBTB resolved by anti-TB and methylprednisolone therapy without sequela. Since the duration of anti-TB therapy for endobronchial lesion is uncertain, we continued therapy until disappearance of the lesion. Surgery is the choice of therapy when medical treatment fails. It is advised to use FB to monitor the response of patients to treatment and to decide whether surgery is necessary. In this case, consecutive FBs were performed in order to follow the response to treatment.

In conclusion, a patient with chylothorax and EBTB is described in this report. The concomitant appearance of chylothorax and EBTB, in the same patient has not been previously reported. After the addition of octreotide treatment, chylothorax resolved. As the prevalence of tuberculosis is increasing worldwide, we hope this report will increase physicians’ alertness to the complications of *M. tuberculosis* infection.

REFERENCES


