Chylothorax due to sarcoidosis: A unique occurrence

Sir,

Chylothorax is a rare condition that results from the thoracic duct damage with chyle leakage from the lymphatic system into the pleural space. Sarcoidosis[1] is an unconventional cause for chylothorax, and high index of suspicion is required to diagnose it. We herein report a rare case of sarcoidosis presenting as chylothorax in a young female.

A 22-year-old female, student by occupation nonaddict, symptomatic for 15 days with complaints of cough and dyspnea with insignificant history had been reported. Chest radiograph showing right-sided effusion and intervened with implantable cardioverter-defibrillator (ICD) following which she was referred to us for further management. On examination, vitals were stable and respiratory system examination revealed movements decreased on the right side with volume loss signs with reduced breath sounds in the right lower lobe areas with ICD in situ draining milky white fluid. Pleural fluid triglycerides level was 234 milligrams per deciliter (mg/dl), pleural fluid cholesterol level was 5.6 mg/dl, chylomicrons were present, and no cholesterol crystals were seen. Contrast-enhanced computed tomography (CT) thorax showed right-sided hydropneumothorax with the right middle lobe consolidation with pericardial effusion with subtle lung nodules [Figure 1]. She underwent Cope’s percutaneous closed needle pleural biopsy, and both fluid and biopsy were negative for Mycobacterium tuberculosis (MTB). CT-guided biopsy of the right middle lobe consolidation lesion was performed which showed noncaseating granuloma [Figure 2] on histopathology and microbiologically did not detect MTB. She also underwent bronchoscopy, where tracheobronchial tree was normal, and bronchial washing again turned out be negative for MTB. Her serum angiotensin-converting enzyme level was normal, and tuberculin skin test was negative. Her spirometry was suggestive of a restrictive abnormality with forced vital capacity (FVC) of 0.71 L and 31% predicted. Her two-dimensional echocardiography showed nontappable pericardial effusion along with bicuspid aortic valve labeled as congenital heart disease. In view of the multisystemic involvement, a congenital syndromic association like Turner’s or Noonan syndrome were contemplated but were ruled out on karyotyping. Direct nodal CT lymphangiography showed obstruction of the thoracic duct due to left inguinal adenopathy. Hence, a final diagnosis of sarcoidosis leading to thoracic
per day in a child, leaks chyle >1 l/day x 5 days or has a persistent chyle flow for >2 weeks or severe electrolyte imbalance or severe malnutrition. Thus, in our case, it was the inflammation associated with sarcoidosis and the adenopathy led to insult to the thoracic duct and the small lymphatic channels in the lungs, which subsequently manifested as chylothorax.

A systematic search of the PubMed database (from inception till August 1, 2018) with the following search terms was undertaken: ("Chylothorax" or "Sarcoidosis" or "Sarcoidosis-related chylothorax"). The search yielded 31 citations, of chylothorax which were identified to be associated with sarcoidosis. Most of the search emphasized genetic mutation involved in the disease and recent advances in the treatment of the disease.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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Letters to Editor

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