

Simultaneous Chylothorax and Chylous Ascites Due to Burkitt lymphoma

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Abstract- Lymphoma of burkitt can commonly present as chylothorax and rarely as chyloperitenium. Here we are presenting a case of a 14-years-old boy with bilateral chylothorax and chylous ascites all together finally diagnosed to have lymphoma of Burkitt as the etiology. To the best of our knowledge, it has been reported very infrequently.

Index Terms- Burkitt lymphoma, chylothorax, chylous ascites, malignant effusion

INTRODUCTION

Chylothorax and chyloperitenium is uncommon condition characterized by accumulation of chyle in pleural and abdominal cavities respectively [1]. Chyle is a milky appearing lymph fluid consisting of lymphocytes, electrolytes, and triglycerides [2]. The loss of it causing nutritional, immunologic, metabolic, and respiratory complications [3].

Simultaneous accumulation of chyle in cavities, though rare, can be associated with nontraumatic etiologies. By far, malignancy causes (especially lymphoma) represents the most frequent underlying cause [1]. We present a case of both chylous ascites and bilateral chylothorax due to follicular lymphoma of 14 years old boy.

CASE REPORT

A 14-year-old was admitted for abdominal pain associated with abdominal distention and dyspnea, polyadenopathy, all evolving in a context of altered general condition. On admission, body temperature was 37.4 C, heart rate was 117/min, breathing frequency 28/min. On chest auscultation, breathing sound was decreased on both lower lung fields. Abdominal examination revealed shifting dullness. The liver is palpable 2 cm below the costal margina at the midclavicular line. The spleen is not palpable.

Initial laboratory results were as follows: white blood cell count 9.77 103/mm3 Lymphocytes: 12.2 % 1/19 103/mm3, Neutrophils: 77.1% 7. 43 103/mm3, Eosinophil 0.2 103/mm3, hemoglobin 10.7 g/dl, thrombocytes 494 103/mm3, serum total protein 62 g/l and albumin 47.4. C- Reactive Protein (CRP), Procalcitonin 0.236, LDH 948 UI/l, Beta- 2 Microglobulin : 1.25

mg/l. Profile of the serum protein electrophoresis (SPEP) was compatible with an inflammatory syndrome.

Chest and abdominal CT scan showed a magma of adenopathies associated with thickening of the ileal coves, massive peritoneal and pleural bilateral effusion.

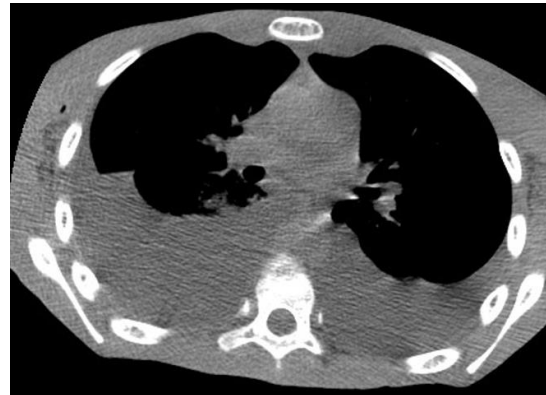


FIGURE 1. Positron emission tomography computed tomography (PET/CT), chest CT scan at the time of diagnosis showing bilateral pleural effusion.

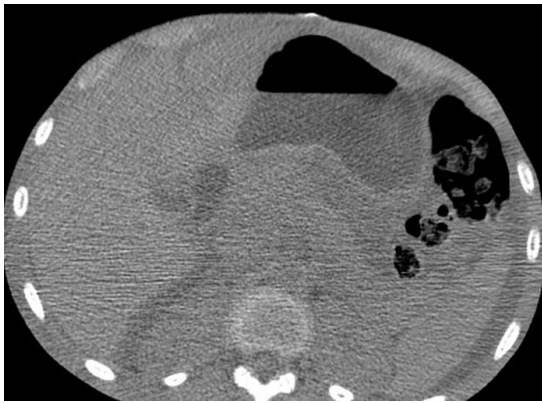


FIGURE 2. Abdominal CT showing ascites.

Thoracentesis revealed a whitish colored pleural fluid, exudate with protein level or content: 34.0 g/l, lactate dehydrogenase: 2 356 UI/LU I/L, total leukocyte counts 11 400 /mm³ predominantly lymphocytic 95 %. Paracentesis find a milk-like and turbid in appearance fluid, exudate with protein content 38.0 g/l, lactate dehydrogenase: 1702 UI/L. Direct smear and PCR of pleural fluid and ascites for acid fast bacilli (AFB) was negative. Bacteriologic findings are normal. We diagnosed chylothorax and chylous ascites because the triglyceride level of pleural fluid and ascites were 1.511 g/L and 3.27 g/l respectively.

Gastric fibroscopy finds moderate and follicular chronic antropoditis, slightly atrophic and mildly active with absence of lymphocytic gastritis lesion or dysplasia and metaplasia. Colonoscopy observes nodular ileitis with subacute interstitial ileitis on biopsies.

A minilaparotomy revealed a magma ganglionnaire and milky fluid. We realisation biopsies of adenopathy and specimen of invaded peritoneum. Pathological analysis revealed a high grade lymphoma B of burkitt. Immunophenotyping: Tumor cells express: CD20, CD10 and BCL6. They do not express: CD5, TDT, CD 99, BCL2, NSE, CK ae1-ae3myogenin, WT1, Desmin, EBV, C myc. -100% of tumor cells express KI67. Myelogram showed a reactive marrow.

Pet Scan demonstrate a metabolically active lymphomatous disease above and below the diaphragm with pleural and peritoneal extension. The diagnosis of burkitt's lymphoma, stage C was retained.

Two and half liters of chylous ascites and 2 L of pleural fluid was drained to reduce respiratory symptoms. Our patient is put on a low fat, medium chain triglyceride diet.

He was transferred to hematoly department to receive chemotherapy. The following were administered one cycle of COP and 2 cycles of RCOPADEM RCYVE then 2 cycles of RCYVE. The re-evaluation later found a complete response and

chylothorax and chyloperitoneum have completely resolved. He is currently undergoing maintenance treatment.

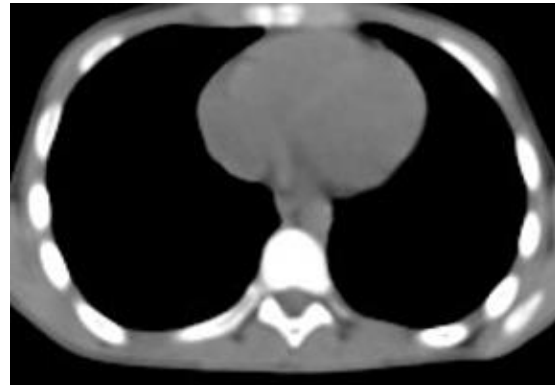


FIGURE 3. Chest CT showing complete resorption of pleural effusion after chemotherapy.

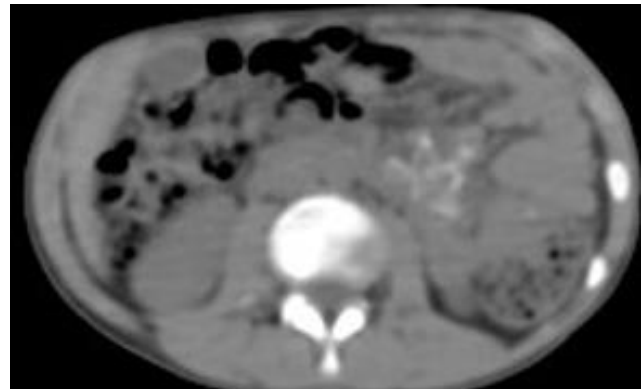


FIGURE 4. Abdominal CT showing resorption of ascites after chemotherapy.

DISCUSSION

Chylothorax and chylous ascites is characterized by chyle in the pleural and peritoneal cavities produced by obstruction and disruption of the lymphatic channel [4]. Diagnosis is confirmed when the triglyceride levels are >110 mg/dl [5] (equivalent to 1.24 mmol/L) in effusions. In cases where triglycerides range between 50 mg/dL and 110 mg/dL, a diagnosis of chylothorax can be made using lipid electrophoresis to detect the presence of chylomicrons [6,7].

The common causes of chyle effusions are malignancy, congenital anomalies, trauma and thoracic surgery. More than 50% of the cases of chylothorax are associated with malignancy, among which, majority are due to lymphoma [8,9]. The reported incidence of the combined occurrence of chylothorax and

chylous ascites has varied from 9% to 55% of chylous effusion [10,11]. To our knowledge, no case describe the association of chylous ascites and bilateral chylothorax secondary to burkitt lymphome has been published to date.

Chyle effusions are associated with high mortality due to severe malnutrition and immunodeficiency due to excessive loss of fat, proteins, vitamins lymphocytes and immunoglobulins [12].

Regarding the management of the chylothorax, it is based on fluid evacuation by setting a chest tube or occasionally drainage by thoracentesis. Decrease lymph production by a low-fat dietary regimen, which contains medium-chain triglyceride (directly absorbed into the blood via the portal system) or to use total parenteral nutrition (TPN) [13]. Somatostatin or octreotide are also reported to have been effective in decreasing lymph production [14,15].

Surgery is performed in case of chylothorax of surgical origin with a flow exceeding 1 L/d, or after failure of medical treatment and doing magnetic resonance lymphangiography IRM in case of chylothorax of medical origin [16].

The surgical strategy is based on reinforced elective suture of the leak, possibly completed by ligation of the thoracic duct in case

of surgical cause or by pleural symphysis by talcage in case of medical cause.

When the chylothorax is related to a specific lymphatic pathology, this one can be treated electively knowing that an excessive gesture of lymphatic resection or ligation may aggravate the situation, or even decompensate a chylous ascites.

At most, a pleuroperitoneal shunt may be indicated in case of refractory high flow chylothorax. Embolization of the thoracic duct is an attractive alternative to its surgical ligation, but remains complex and not widely available [17].

Treatment of the underlying lymphoma with chemotherapy led to a significant improvement of chylothorax and chylous ascites in this case and did not require any surgical intervention or radiation therapy.

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