Case Report

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Spontaneous bilateral chylothorax in both vena cava thrombosis; cisterna chyli ligation

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ABSTRACT

Chilothorax, a rare clinical condition characterized by high triglyceride white fluid in the thoracic cavity; when the literature is examined, it is seen because of trauma, malignancy, complication of surgery, and rarely central catheter-related thromboplasty. In the treatment, diet change, nutrition, drainage with tube thoracostomy, conservative method, surgical procedure with thoracotomy are applied. In our case there was no response to nutritional changes, drainage, thoracotomy and somatostatin treatments. Cisterna chyli ligation was performed with laparotomy. We present a case of spontaneous bilateral chylothorax associated with superior and inferior vena cava thrombosis (without central catheter relationship).

Keywords: Spontaneous, Chylothorax, Vena cava thrombosis, Surgical, Cisterna chyli ligation

INTRODUCTION

Chylothorax is a rare clinical condition characterized by a white fluid with increased triglyceride levels in pleural effusion. It is often seen as trauma, malignancies and complications to surgery. Prolonged catheterization of the subclavian vein or vena cava superior is among other causes of chylothorax. When the literature was examined, it has been observed that bilateral chylothorax cases are rare and those associated with the superior vena cava usually occur after thromboplasty secondary to the central catheter.

Current study was aimed to present a case of bilateral chylothorax due to superior vena cava thrombosis, which developed spontaneously and did not have a history of catheter and trauma.

CASE REPORT

An 18 year old male patient was admitted with gradually increasing shortness of breath. Bilateral lung breath sounds were decreased on physical examination. Other system examinations were considered normal. No pathological finding was found in routine laboratory tests. Pleural effusion was observed on the right side on chest X-ray and bilateral pleural effusion, pericardial effusion and superior vena cava thrombosis were detected on thorax CT (Figure 1A-C, Figure 2A). There was no mediastinal tumor or intrathoracic mass lesion. Right tube thoracostomy was performed because the color of the fluid obtained by right thoracentesis was milky white. Laboratory analysis of white fluid drained from the right thorax; LDH 76, TG 316, cholesterol 31, albumin 12, total protein 20, glucose 113. It was evaluated as compatible with chylothorax. The patient, who drank only water as an oral diet, was given peripheral parenteral

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nutrition support with high calorie and protein content but low in fat (central catheter was not applied due to the presence of superior vena cava thrombosis).

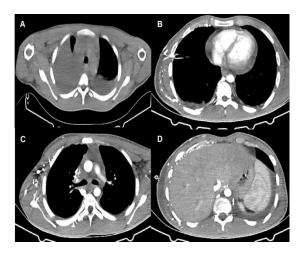


Figure 1: A) bilateral pleural effusion tomography image, B) pericardial effusion tomography image, C) tomography image of superior vena cava thrombosis and D) tomography image of inferior vena cava thrombosis.

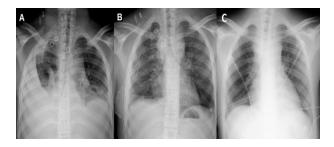


Figure 2: A) first chest radiography, B) chest radiography after thoracotomy and C) chest radiography after laparotomy.

Daily 1200-1600 ml/day drainage was performed from the thorax tube. ECHO examination reported EF 60 and 1.5 cm pericardial effusion. No findings consistent with malignancy were found in the FDG PET examination. The patient with left leg pain and abdominal pain on day 3; partial thrombosis (16x10 mm) was reported at the inferior hepatic level of the vena cava in doppler examination. Subtotal thrombosis was detected in the left lower extremity common femoral vein, superficial femoral vein and popliteal vein. Upon detection of inferior vena cava thrombosis on intraabdominal CT angiography (Figure 1D), percutaneous vena cava filter was applied at the renal vein level and compression stockings were put on the left lower extremity and upgraded.

When the daily thoracic tube drainage continued at 1200-1600 ml, right thoracotomy was performed on the patient and the area where the chylous fluid came from distal to the right vena subclavia was sutured, and then no fluid was seen (Figure 2B). Pericardial window was opened in

the same session. Postoperative percutaneous left thoracic catheter thoracostomy was performed. Left pleural fluid laboratory analysis; LDH 94, TG 201, cholesterol 27, albumin 19, total protein 28, glucose 108. The diagnosis of bilateral chylothorax was confirmed in the presence of superior vena cava and inferior vena cava thrombosis. Oral diet was started on the postoperative seventh day of the patient who had a mean right 400-600 serous and left 200-300 serous drainage from the thorax tube. Following the oral regimen, the pleural fluid turned into chylosis and the right 1300, left 600 drainage was seen, and the oral regimen was closed again and somatostatin treatment at a dose of 3.5 µg/kg/hour was started with parenteral nutrition support. Liquid electrolyte treatments were administered to the patient who was found to have hypoalbuminemia, hypokalemia, hyponatremia and hypocalcemia. Although somatostatin treatment was increased to 5 µg/kg/hour on the fifth day of the treatment, an average of 1100 left 300 pleural drainage per day continued. Pathology of bilateral pleural fluid was reported as benign and microbiological examination as culture negative. Electrolyte imbalance of the patient, whose daily drainage was continuing, gave clinical findings. The patient, who was provided with liquid and electrolyte support with appropriate parenteral peripheral nutrition, was consulted with the general surgery clinic, since no significant decrease was observed in the daily drainage amount. Cisterna chile ligation was performed with laparotomy operation (Figure 3).

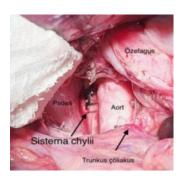


Figure 3: Surgical image of cisterna chyli ligation.

Oral diet was started on the postoperative third day, and the left thoracic catheter and abdominal drain were removed due to reduced drainage during follow-up. The patient, who had an average of 300-400 serous drainage per day from the right thoracic tube, was discharged on the thirty-third day of his hospitalization, leaving his right thoracic tube to chronic drainage due to the absence of electrolyte imbalance and normal vital (Figure 2C) and discharged after policlinic controls were recommended. The thorax tube was removed on the twentieth day in the controls.

DISCUSSION

Chylothorax is a disease characterized by the accumulation of lymphatic fluid in the pleural cavity after the anatomical integrity of the ductus thoracicus is

broken. This fluid originates mostly from the intestines, some of it from the lungs, liver, abdominal wall and extremities.²

The thoracic duct enables the transfer of lymphatic fluid digested fats and proteins outside the vascular system into the system's circulation.3 Cholesterol/triglyceride ratio of <1 in pleural fluid is one of the most important criteria for the diagnosis of chylothorax. Triglycerides >110 and milky white color in fluid analysis make the diagnosis of chylothorax. According to these criteria, presented case is compatible with chylothorax. The first step in the treatment of chylothorax is the drainage of chylous fluid, providing nutrition and reducing lymph flow. A low-fat diet with high calories and protein is given for nutrition. If the lymphatic fluid loss is mild to moderate (<0.25 ml/kg/ hour), it can be waited up to 1 month for drainage interruption. However, if drainage is severe, such as 2 ml/kg/hour, surgical treatment is required without waiting.⁴ Our case had 1200-1600 (0.8-1.2 ml/kg/hour) drainage initially. Malnutrition, dehydration, metabolic acidosis and immunodeficiency may develop in patients with chylothorax due to the loss of necessary substances for the body.⁵ Despite supportive treatments, electrolyte imbalance and subsequent serious clinical deterioration were observed in our patient. The patient's condition was observed and the decision for surgery was made. However, it was observed that drainage continued when the postoperative oral diet was opened despite surgical treatment with right thoracotomy. In most studies, the use of somatostatin therapy in the range of 2-12 µg/kg/hour has been shown to be effective in the treatment of chylothorax.6

Somatostatin acts by decreasing intestinal blood flow, chylomicron synthesis and fat absorption. Since chylous drainage continued after thoracotomy, somatostatin was used initially 3.5 and then 5 in our case. However, there was no significant response to somatostatin treatment. We observed it with drainage and parenteral nutrition for a while, but there was severe clinical worsening and significant electrolyte loss. The loss continued despite albumin, electrolyte, and fluid replacements. Clinical findings compatible with electrolyte deficiency such as tremor, cramps and spasm were observed, mainly ECG changes.

In addition to electrolyte, fat, protein and vitamin loss in chylothorax, T cell loss is also seen. Post-lymphopenia infection and malnutrition are the main causes of death in chylothorax. Intra-abdominal ligation of the thoracic duct or cisterna chyli was available as a treatment option for interrupting drainage and preventing losses. Cisterna chyli ligation was performed with laparotomy. It was observed that fluid loss significantly decreased in the early postoperative period and clinical findings improved. The patient's chylothorax treatment was completed with this method.

CONCLUSION

The most important health problem in chylothorax patients is due to losses from the lymphatic system. The treatments applied are diet change, drainage, conservative and surgical methods. The main thing is to replace electrolyte, fat, protein, vitamin and fluid losses during the course and treatment of the disease. Due to the loss of T cells in lymph fluid, patients on long-term therapy are likely to have immunodeficiency and malnutrition. Since the main causes of death in chylothorax are malnutrition, infection and immunodeficiency, it should be treated immediately. If there is no response to treatment methods, cisterna chyli ligation, which is one of the surgical methods, should be kept in mind.

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