

Chylopericardium: A Two Cases Report

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I. Introduction:

Chylopericardium is a rare clinical entity that is characterized by the presence of chyle in the pericardial cavity [1]. Its association with lymphangiomas, cystic hygromas, thoracic and cardiac surgery, trauma, radiation and malignancy is well reported [2, 3]. However, primary chylopericardium, in which there is absence of known precipitating factors, is rarely encountered in clinical practice. The severity of clinical manifestations of chylopericardium is varied and may range from the complete absence of symptoms to cardiac tamponade, and thus a high index of suspicion is required for the prompt recognition of the underlying diagnosis and management of its sequelae [2].

In this report, we present two cases of chylopericardium for two patients who had hospitalized in cardiology department at Chu ibn roched of Casablanca city.

II. Case Presentation:

FIRST CASE:

A 32-year-old man was admitted with no cardiovascular risk factor of pericardium effusion no signe of systemic disease, or past history tuberculosis except for a thoracic trauma 3 years before with no complications. He complained dyspnea of NYHA class II, evolving for 18 months and decreasing at an anteflexed position. The worsening symptoms during the two last months led patient to seek medical consultation. Chest X- ray was performed showing cardiomegaly and echocardiography revealed liquid pericardial effusion of 16 mm in front of the right ventricle, 18 mm in front of the right atrium and 28 mm in front of the left ventricle without any sign of compression. His blood pressure was 120/80 mmHg and heart rate 95 Bpm. There was no jugular vein distension or other signs of tamponade. ECG showed a sinus rhythm, with a low voltage and electrical alterans. A computed tomography scan was performed showing pericardial effusion with no other abnormalities(Fig.1)

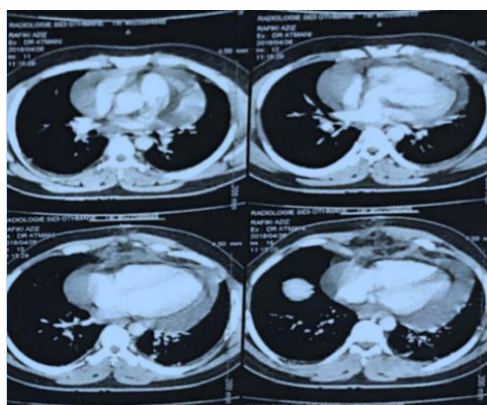


Figure 1: Axial Tomodensitometric Section passing through the anterior Mediastinum objectifying a Pericardial Effusion of Average Abundance.

Blood count, electrolytes, kidney function, and liver function tests had normal findings. Immunological assessment, thyroid function tests and tuberculosis had negative results. Lipid profile demonstrated cholesterol of 5.4mmol/l, triglycerides of 1.9mmol/l, low density lipoprotein (LDL)-Cholesterol of 2.8mmol/l and high density lipoprotein (HDL) - Cholesterol of 1.4 mmol/l. A pericardial biopsy puncture was performed and 800 mL of thick milky chylous fluid was drained. The chylous nature of the fluid was confirmed by high level of triglycerides (10mmol/L) and a cholesterol– triglyceride ratio of 0.3 (below 1). Cytology demonstrated

abundance of lymphocytes and no evidence of malignant cells. The culture for bacteria had negative result and anatomic-pathological study showed a chronic fibrous pericarditis with no sign of malignancy. A lymphoscintigraphy was performed, on a GE 630 Gamma Camera to look for a breach in thoracic duct or a communication with the pericardium, It confirmed a normal lymphatic drainage in the upper and lower limbs with the absence of scintigraphic argument that may explain chyloous pericardial effusion.

The patient received lipid lowering treatment and anti-inflammatory dosage of aspirin, colchicine 1mg for 6 months and assigned to low fat diet with no improvement. Therefore, an anti-bacillary treatment was started for 6 months, since the patient was in an endemic country for tuberculosis. 3 months later, the patient came back with recurrence of the pericardial effusion without any sign of compression or tamponade. The inferior vena cava measured 21 mm. Therefore, the patient was candidate for a pleuro-pericardial window with a favorable evolution for 3 months till writing the manuscript.

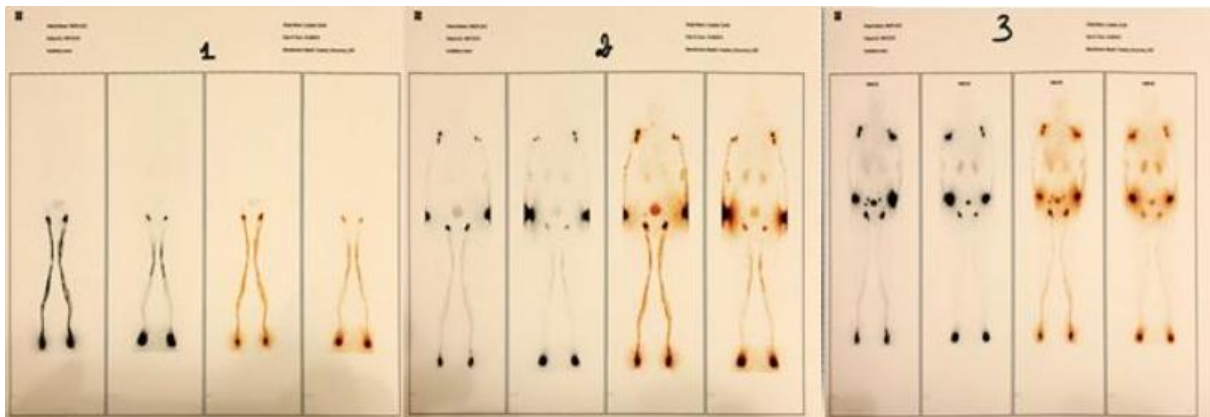


Figure 2: Lymphoscintigraphic Images showing Normal Lymphatic Drainage.

SECONDE CASE:

We present the case of 47-year-old man heavy smoker 20 parquets by year, he was neither diabetic nor hypertensive, initially presented to emergency room with a five-day history of chest pain and dyspnea. The chest pain started the day of admission; however, he noted worsening shortness of breath and a productive cough over the course of several days. He also noted a 10-kilogram weight loss occurring over a three-month period.

Cardiology was consulted and during their physical exam displayed distant heart sounds, jugular venous distention, Kussmaul's sign, superior vena cave syndrome. Vitals at presentation included a temperature of 37.3°, tachycardia of 130, respiratory rate of 24 c/m, blood pressure 10/50mmHg, and an oxygen saturation of 95%. An echocardiography revealed liquid pericardial effusion measuring 30 mm in front of the right ventricle, 20 mm in front of the right atrium and 15 mm in front of the left ventricle with signs of tamponade (specially the IVC measuring 21mm, with latterly collapsed during inspiration. diastolic collapse of the RV and systolic collapse of the RA was noted). Thus, Initial pericardiocentesis was performed in urgently and 1200ml of milky white fluid drained (Figure 3). Body fluid studies revealed a high level of triglycerides (7.13mg/dL). Cytology with no evidence of malignant cells. The culture for bacteria had negative result. A pericardial biopsy puncture was performed an anatomic-pathological study showed a congestive pericardium with no sign of malignancy or tuberculous granuloma.



Figure 3: milky pericardium fluid drained

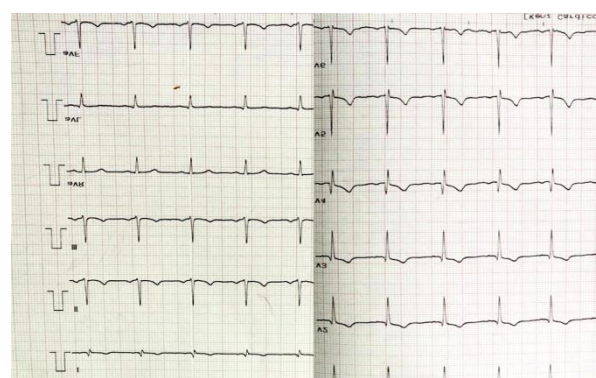


Figure 4: His EKG

In this instance, an EKG was obtained showing sinus rhythm with widespread concave ST elevation and no electrical alternans (Figure 4). He was found to have a normal blood count and electrolytes. His kidney, thyroid and liver function was found normal. During his hospitalization, chest X-ray was performed revealing a large pleural effusion; therefore. CT thoracic angiogram was performed demonstrating mediastinum pulmonary masse and superior vena cave thrombosis extend to the jugular vena (Figure 5 and 6). Extension tests showed multiples metastasis sites in particular; thoraco-abdomino-pelvic CT revealed a metastatic lesion in hepatic and adrenal glands, cerebral CT demonstrated left temporal secondary brain tumor measuring 14x13x14.7 mm (Figure 7) and bone scintigraphy revealed multiple bones hyperfixation in particular his proximal right humerus, sternum body, hemi pelvis, right femoral diaphysis and trochanters and left calf bone in favor of metastasis lesions (Figure 8).

In reason of recurrent pericardial effusion, Cardiothoracic surgery was consulted and the patient was taken for an emergent pericardial window. A left pericardial window was created with drainage to the left pleural space. Approximately 800 mL of chylous pericardial fluid and serou-hematic pleural fluid was drained. Intraoperatively, Biopsies were taken however no specific lesions was found. The patient was brought to the ICU and he was clinically stable. On evolution, we had been programmed him for bronchoscopy to have diagnostic track but unfortunately the was dead by covid-19 infection two days before.

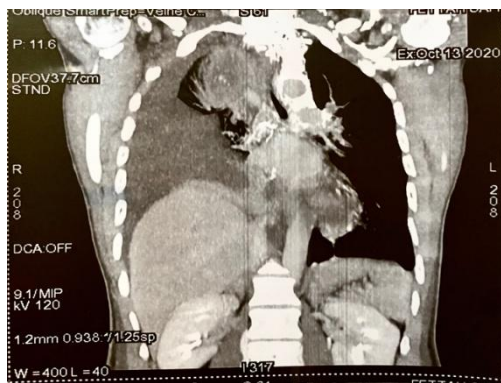


Figure 5: CT thoracic angiogram demonstrating mediastinum pulmonary masse and superior vena cave thrombosis

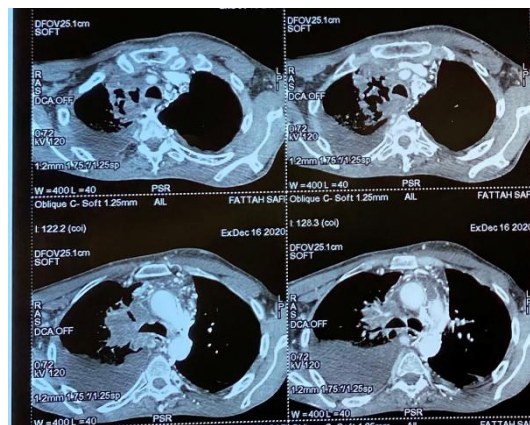


Figure 6: CT thoracic angiogram demonstrating mediastinum pulmonary masse

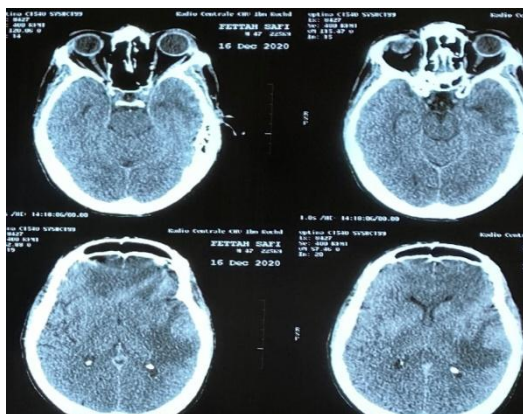


Figure 7: cerebral CT demonstrated left temporal secondary brain tumor

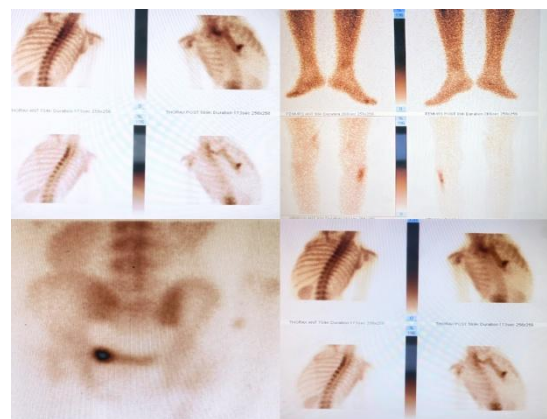


Figure 8: Bone scintigraphy revealed multiple bones hyperfixation

III. Discussion

Chylopericardium is an abnormal accumulation of chylous fluid containing triglycerides in high concentrations in the pericardium [4]. Only few cases are described in literature; the first one was by Hasebrock in 1888 [5] and then it was reviewed in other studies. Sagristà-Sauleda et al. reported only one case of CP among 461 patients with a large pericardial effusion [6]. C. Dib et al. reported 33 patients with CP in a large cohort of 10 years [6]. Yater et al. found 3 cases among 100 non-traumatic patients with chylothorax [7]. the latest one in Beijing, china which analyzed 65 years of 92 English and Chinese studies reported 104 patients with CP in the literature.

The association of chylopericardium with trauma, previous thoracic or cardiac surgery, congenital lymphangiomas, radiation and malignancy is well recognized [2-3-11-12]. However, primary chylopericardium is a clinical entity that is more rarely encountered and only diagnosed if all known precipitants have been excluded [6–8].

Its pathophysiology remains controversial, and is thought to be related to either elevated pressure in the thoracic duct, or to abnormal communications between the thoracic duct and the lymphatics of the pericardium [3, 13]. Clinical manifestations are varied as it is the age spectrum of patients affected, with reports of chylopericardium in neonates through to older adult [15].

The imaging modalities that can be used to ascertain the underlying cause, with varying specificity and sensitivity, include lymphangioscintigraphy, lymphangiography, monitoring of chest radioactivity after oral intake of I-triolein and observation of the distribution of Sudan III dye in the pericardial cavity after ingestion [2, 3]. However, the diagnosis still relies on the cytology, chemistry and microbiology of pericardial aspirate, obtained via pericardiocentesis.

The management of this condition though controversial has two main therapeutic aims. The first is the prevention of cardiac tamponade. This can be achieved via pericardiocentesis or surgically with the formation of a pericardial window/partial pericardiectomy, which can be combined with ligation of the thoracic duct and by conventional open methods or via video assisted thoracoscopic surgery (VATS). The second aim is the avoidance of metabolic, nutritional and immunological compromise, resulting from the loss of chyle, via a diet rich in medium chain triglycerides or, if necessary total parenteral nutrition [2, 16, 17].

IV. Conclusion :

Chylopericardium is a rare entity. Presentation may be acute, chronic, or even subclinical. Diagnosis always requires pericardiocentesis when the color of the fluid characteristics, triglyceride content, cytologic examination, and negative culture suggests the diagnosis. Patients should undergo chest computed tomography followed by lymphangioscintigraphy/lymphangiography to rule out secondary causes. Treatment of the underlying cause should always be the therapy of choice. Even though, many causes can be found after investigations, idiopathic or primary CP is still a rare condition with only around 100 cases reported worldwide to date, and even less in Morocco.

Ethical approval:

Consent written informed consent for the publication of this manuscript was obtained from the patient. publication was approved by the ethics committee of our institution.

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