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Pseudochylothorax in a Patient with Rheumatoid Arthritis – Case Report

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Abstract

Background: Pseudochylothorax is usually associated with chronic inflammatory disorders like tuberculosis or rheumatoid arthritis, but there are few reports of Rheumatoid Arthritis-associated pseudochylothorax. It usually occurs during the course of a previously diagnosed RA but is occasionally seen contemporaneously with or preceding the onset of other arthritic signs and symptoms.

Case: A 49-year-old female patient presented to our hospital with an undiagnosed pleural effusion. She had a history of cough, difficulty breathing, fatigue, profuse sweating, and morning stiffness for a period of 12 months. On admission she was afebrile, but presented signs and symptoms of chronic inflammation of the joints. On CT-scan angiography were observed left sided minimal pleural effusion. The pleural fluid was confirmed as pseudochylothorax due to high cholesterol and low triglyceride concentrations (300 mg/dL and 60 mg/dL, respectively). Cholesterol/ triglyceride ratio was >1 and the presence of cholesterol crystals along with elevated levels of anti-cyclic citrullinated peptide antibody and rheumatoid factor suggested the diagnosis of Rheumatoid Arthritis origin.

Conclusion: We encountered a rare case of a pseudochylothorax occurring contemporaneously with the onset of other arthritic signs and symptoms.

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INTRODUCTION

Pleural effusion may occur in patients with rheumatoid arthritis (RA), typically as an exudate but in rare occasion has features of pseudochylothorax. Although this uncommon type of cholesterol-rich pleural effusion is linked more frequently to tuberculosis (54%), it may be present in around 9% of patients with RA (1) (2). Usually unilateral pleural effusion is seen, however cases with bilateral effusion have been also reported (3). We report a case involving a 49-year-old female with arthritic symptoms who was diagnosed with RA-related pseudochylothorax by anamnestic information, imaging examinations, laboratory examinations (including blood and pleural fluid examinations) and follow-up after starting the therapy for rheumatoid arthritis.

CASE REPORT

A 49-year-old female patient refers a history of 12 months of cough, difficulty breathing, fatigue and profuse sweating, morning stiffness that last longer than 20 minutes. She was admitted to the hospital with the diagnosis: Left sided pleural effusion to be determined. On physical examination, it was observed on auscultation weakened basal respiration on the left hemithorax. Left and right metacarpophalangeal joints and proximal interphalangeal joints with increased tenderness during palpation. Left and right knee joint with increased tenderness on extension and flexion, bilateral talocrural joints with increased tenderness. In past medical history she referred: Spinal discal herniation and hypothyroidism (under Levothyroxine). A series of radiological and routine hematological investigations were performed. Complete blood count and biochemical blood test parameters were normal. Hormone panel blood test showed increased level of TSH but normal level of FT4 and FT3. Rheumatologic panel showed increased level of Anti CCP (19UI/ml) and RF (35UI/ml). Microbiological examination for mycobacterium tuberculosis resulted negative. On CT scan angiography there was no evidence of pulmonary tuberculosis, interstitial pneumonia, or other disease in the lung fields. Heart size was normal. The mediastinal structures had normal configuration. There was pleural effusion on the left side (Figure 1).

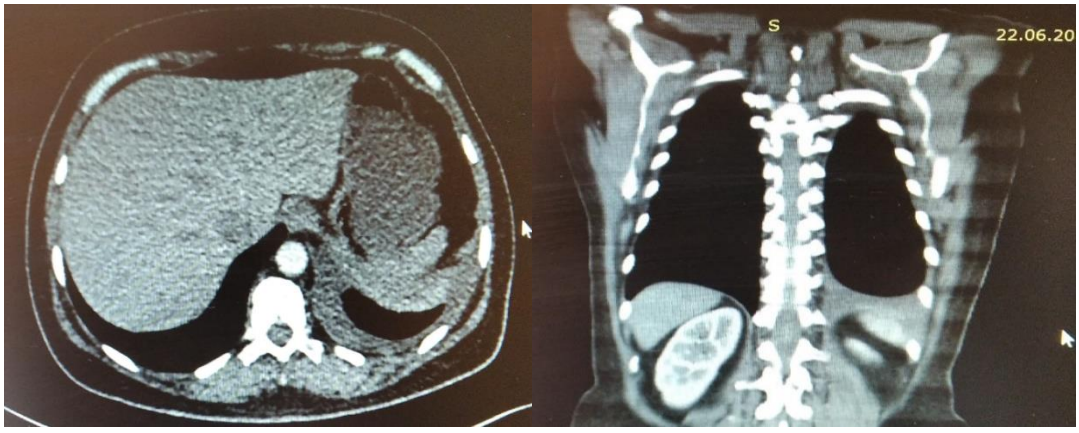

Figure 1. Chest CT scan showing left pleural effusion.

There was no evidence of lymphadenopathy. There was no evidence of pulmonary embolism. Macroscopical examination showed cloudy pleural effusion (Figure 2).

Figure 2. Macroscopical examination shows cloudy pleural effusion.

As the result of Light Criteria, the nature of pleural effusion was exudate. The pleural fluid was confirmed as pseudochylothorax because it had high cholesterol and low triglyceride concentrations (300 mg/dL and 60 mg/dL, respectively) also with the presence of cholesterol crystals on microscopy. Low glucose (2.0 mg/dl) and high lactate dehydrogenase (LDH) (2444U/l) were noted. Pleural effusion cell profile showed 30% neutrophils, 55% lymphocytes and 15% monocytes. There were seen spumous mononuclear cells, excessive cytolysis and presence of cholesterol crystals on microscopy. No malignant cells were found in cytological examination of the pleural fluid. Mononuclear cells were observed in moderate amount.

Treatment and course

The patient was treated as *seropositive* rheumatoid arthritis, third stage, with:

Prednison 30 mg daily, methotrexate 10 mg weekly, folic acid 10 mg weekly, hydroxychloroquine 200 mg daily, pantoprazol 40 mg daily. After 45 days of treatment the health condition of the patient improved, presenting with mild morning stiffness, no cough, neither difficulty breathing, nor profuse sweating. We performed a chest ultrasound and there was no pleural effusion (Figure 3).

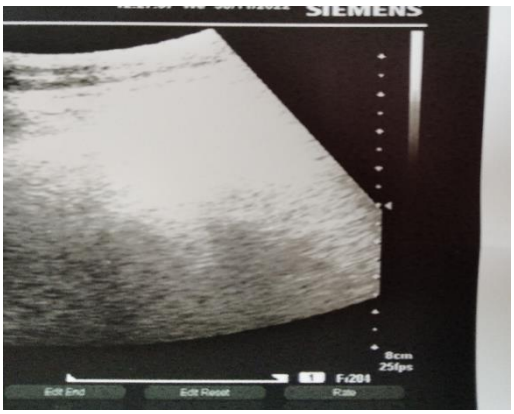

Figure 3. Chest ultrasound showing no pleural effusion.

DISCUSSION

This case is special because rheumatoid pleurisy was presented as pseudochylothorax and because is a rare case of a pseudochylothorax occurring contemporaneously with the onset of other arthritic signs and symptoms. The most common cause of pseudochylothorax is tuberculosis followed by rheumatoid arthritis, both

contributing to 88.5% of total cases (4). The other rare causes include paragonimiasis, echinococcosis, malignancy and trauma (4) (5). The malignant causes are mainly associated with hematological malignancies (5). It can be also idiopathic (6) (7). There are only a few reports of arthritis associated pseudochylothorax in the literature (8), while in the albanian medical practice this is the first case reported. We suspected the possibility of rheumatoid pleurisy based on the finding of pseudochylothorax and the additional blood tests which were positive for RF and anti-CCP antibody. Anti-CCP antibody is regarded as the most reliable serologic marker of RA (9).The characteristic accumulation of turbid or milky white pleural fluid associated with pseudochylothorax is due to a high lipid content. Typically, the pleural fluid cholesterol level will be ≥ 200 mg/dL with a triglyceride level of <110 mg/dL(always with a cholesterol/triglycerides ratio >1, in this case greater than 5) and often with cholesterol crystals seen on microscopy (10) (7). Since pleural biopsy has a lower diagnostic value in predicting etiology, it should not be considered in the first place (11). Most effusions associated with RA pleurisy are asymptomatic and do not require specific additional treatment despite that of RA. Some patients respond to corticosteroids, but others do not. Symptomatic relief with thoracentesis and management of the underlying disease remains the mainstay of treatment (3).Invasive procedures, such as decortication/pleurectomy, may be considered when there is symptomatic worsening or recurrent pleural effusion (12).

Because cartilage damage frequently occurs within the first two years of disease, we have started the therapy with DMARD-s (Disease modifying antirheumatic drugs) early in the course of disease and the result was impressive. Ultrasonography is more sensitive than CT for detecting complications within the effusion and can be used to assess its characteristics. After 45 days of treatment we performed a chest ultrasound and there was no evidence of pleural effusion.

CONCLUSION

We encountered a rare case of a pseudochylothorax occurring contemporaneously with the onset of other arthritic signs and symptoms.

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Conflict of Interest Statement: The authors declare that they have no conflict of interest.

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