Pseudochylothorax Secondary to Rheumatoid Arthritis: Case Report

Romatoid Artrite Bağlı Psödoşilotoraks

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Yazışma Adresi/Correspondence: Handan İNÖNÜ, MD Gaziosmanpasa University, Faculty of Medicine, Department of Pulmonary Diseases, Tokat, TÜRKİYE/TURKEY handaninonu@gmail.com **ABSTRACT** A 61-year-old women was referred to our clinic because of bilateral pleural fluid in her chest X-ray. Her history revealed that swelling and stiffness in her joints was diagnosed as rheumatoid arthritis five years ago. On admission, the patient had no complaints, appeared well, and her vital signs were normal. Lung examination revealed that breath sounds were diminished in both lung bases. We did thoracentesis which yielded a milky-odorless fluid. Biochemical characteristics of the fluid was consistent with exuda; trigliseride: 20 mg/dl, cholesterol: 286 mg/dl. Direct smear and culture for acid fast bacilli were negative. Thoracic computerized tomography scans revealed bilateral pleural fluid, volume loss of the left lung with ipsilateral shifting of mediastinum. According to clinicoradiological and laboratory findings, we concluded that pseudochylothorax was the result of chronic rheumatoid pleurisy. Since pseudochylothorax is an uncommon entity detected during the analyses of pleural fluids, we intended to present our rarely seen case with pseudochylothorax.

Key Words: Arthritis, rheumatoid; pleurisy; chylothorax; cholesterol; triglycerides

ÖZET Altmış bir yaşında bayan hasta, akciğer grafisinde bilateral plevral efüzyon nedeniyle kliniğimize yönlendirilmiş. Özgeçmişinde; beş yıl önce eklemlerinde şişlik-tutukluk şikayeti olmuş, romatoid artrit tanısı konulmuş. Kliniğimize başvurusunda yakınması yoktu, fizik muayenesinde genel durumu iyi, vital bulguları stabildi. Bilateral akciğer bazallerinde solunum sesleri hafif azalmıştı. Torasentezle alınan mayi sütsü görünümde, kokusuz, biyokimyasal incelemesi eksüda karakterinde, trigliserid: 20 mg/dl, kolesterol: 286 mg/dl idi. Sıvının asit rezistan bakteri teksif ve kültür incelemesi negatifti. Bilgisayarlı toraks tomografisinde bilateral plevral efüzyon, sol akciğerde volüm kaybı, kalp ve mediastende sola deviasyon tespit edildi. Mevcut klinik-laboratuvar ve radyolojik bulgularla kronik romatoid plöreziye bağlı psödoşilotoraks kabul edildi. Psödoşilotoraks plevra sıvıları içinde nadir görüldüğünden olgu sunulmuştur.

Anahtar Kelimeler: Artrit, romatoid; plörezi; şilotoraks; kolesterol; trigliseridler

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Pseudochylothorax, which is also termed as chyliform pleural effusion or cholesterol pleural effusion is an entity that develops as a result of increase in the amount of cholesterol in fluids with long-standing exudative characteristics and resembles to chylothorax. It is generally observed in pleural effusions lasting more than five years. Up to 1999, a total of 29 publications and 172 case reports regarding pseudochylothorax which is a rare form of pleural effusions have been cited in the literature. The most common causes of pseudochylothorax are tuberculous pleurisy, chro-

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Thoracic Diseases İnönü et al

nic rheumatoid pleurisy and therapeutic pneumothorax.³⁻⁵ In this case report, we present a patient who developerd pseudochylothorax secondary to rheumatoid arthritis (RA).

CASE REPORT

A 61-year-old woman was referred to our clinic because of bilateral pleural effusion detected radiologically. Five years ago she was diagnosed with RA in the Department of Rheumatology after complaints of swelling and stiffness in her joints, and then prednisolone and leflunomide were given. Five months ago, she had undergone chest tube thoracostomy in the Department of Thoracic Surgery.

Since medical history indicated presence of pleural effusion at that time, her medical records were requested from the medical center concerned, and they were examined. Her fiberoptic bronchoscopy was reported to be normal, while her histopathology report indicated "chronic non-specific pleuritis". Serum and pleural fluid RF levels were reported as 73.2 IU/ml (N: 0-20 IU/ml) and 316 IU/ml, respectively. At the time of diagnosis, on plain chest X-ray (Figure 1) pleural effusion was seen on the left, and on thoracic computerized tomography (CT) there were nodules in the parenchyma of both lungs and pleural effusion on the left side. When serial X-rays taken during radiologic controls were examined, bilateral effusions were seen starting after the first year of diagnosis. In the medical center concerned, pleural effusion was reported to be secondary to RA, and maintenance of her treatment was recommended.

On her referral to our clinics, she had no complaints and she was under RA therapy. Her general health status was good with stable vital signs. Respiratory sounds were slightly diminished bilaterally at lung bases. Thoracentesis of the left lung yielded a milky- odourless fluid with exudative characteristics. Biochemical examination revealed following: triglyceride: 20 mg/dl (N:30-150 mg/dL), cholesterol: 286 mg/dl (N:1-200 mg/dL), cholesterol/triglyceride ratio: 11.3, HDL: 77 mg/dl (N:32-65), glucose: 6 mg/dl, total protein 7.59 g/dl, density: 1010. Tests for acid-fast bacilli and antibiograms yielded negative results. Results of routine



FIGURE 1: Posteroanterior chest X-ray shows pleural effusion in the left hemitorax.



FIGURE 2: Posteroanterior chest X-ray demonstrates bilateral pleural effusions.

tests, WBC, biochemical analyses, sedimentation rate (10 mm/hr [N: 9-20 mm/hr]) and C-reactive protein levels (3.19 mg/L [N:0-5 mg/L]) were within normal limits. Pulmonary Function Test values, FEV1: (2.14 [87.6%]), FVC: (3.00 [104.4%]), FEV1/FVC ratio (71.42), were within physiologic limits. The patient had bilateral pleural effusions on posteroanterior chest X-ray (Figure 2). Her thoracal CT revealed bilateral pleural effusions (Figure 3) more prominent on the right side, pleural consolidation, volume loss on the left side, shift of the heart and mediastinum towards left, and millimetric nodules localized in the parenchyma and

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FIGURE 3: Postcontrast computed tomography demonstrates bilateral pleural effusions more prominent in the right hemitorax and high attenuation values in the atelectatic lower lobes. Pleural thickening and enhancement are present in the left lower lobe.



FIGURE 4: Computed tomography shows a milimetric peripheral nodule in anterior segment of the right upper lobe.

subpleural space of both lungs (Figure 4). The patient was diagnosed with pseudochylothorax secondary to chronic rheumatoid pleurisy in the light of available clinical, laboratory and radiological findings, and we did not perform any interventions due to lack of any respiratory complaints.

DISCUSSION

Pseudochylothorax is a rarely seen form of pleural effusions.^{2,3} Pseudochylothorax develops as a result of increase in the amount of cholesterol in fluids with long-standing exudative characteristics and resembles chylothorax. Although its pathoge-

nesis is not fully known, it is accepted that the absorption process of lipids synthetized in pleural tissue is prevented because of thickened pleura resulting in accumulation of fluid inside pleural cavity. The origin of lipids and cholesterol in pleural fluid is not completely elucidated. It is thought to originate from degenerated erythrocytes or leukocytes. Hypercholesterolemia or any disorder in cholesterol metabolism is not a predisposing factor. 5

Pseudochylothorax is seen in chronic pleural effusions with a mean latent period of five years and a time interval of 11-15 years.⁷ In all disorders that lead to pleural consolidation and fibrosis, pseudochylothorax can develop, however the most frequently encountered three etiologies are tuberculosis, RA and therapeutic pneumothorax.¹ Other causes of pseudochylothorax include hemothorax, malignancy, syphilis, alcoholism, diabetes, paragonimiasis, undertreated empyema, lung cancer, hydatid cyst, Hodgkin disease, heart failure and nephrotic syndrome. It can be also idiopathic.^{1,4,5} Our case was diagnosed with RA five years ago, and the presence of pleural effusion since that time was verified.

In a patient with pleural effusion, the differential diagnosis of cloudy or milky fluid aspirated through thoracentesis includes empyema, chylothorax and pseudochylothorax. In empyematous fluids, leucocytes give the fluid a cloudy appearance, and after centrifugation or waiting, while cells precipitate at the bottom. If turbidity persists after centrifugation, a differential diagnosis between chylothorax and pseudochylothorax should be made through analyses of the pleural fluid. In pseudochylothorax the density of the fluid is under 1010, while triglyceride levels are under 200 mg/dL.1 The levels of triglycerides can be sometimes elevated, however cholesterol/triglyceride ratio should be always above 1. The fluid has exudative characteristics, with a protein concentration of 4-7 g/dl, glucose levels < 60 mg/dL and pH < 7.20. On microbiological examination neutrophils are dominant cell types.7 Since pleural biopsy has a lower diagnostic value in predicting etiology, it should not be considered in the first plaThoracic Diseases İnönü et al

ce. Results of the biochemical analysis of the pleural fluid of our case were completely in accordance with the diagnosis of pseudochylothorax. All microbiological test results were negative for tuberculosis.

In cases with established diagnoses of pseudochylothorax, thoracal CT should be requested for detailed examination of pleura. Thickened and calcified pleural membranes, and loculated pleural effusion are seen. Usually unilateral pleural effusion is seen, however cases with bilateral effusion have been also reported. Thorax CT of our case revealed bilateral pleural effusions. Findings indicating a chronic process were also observed which included volumetric loss of the left lung, leftward shifting of heart and mediastinum, and pleural thickening. Millimetric nodules were detected localized in parenchyma and subpleural space.

Usually scarce-moderate amounts of pleural fluid is detected in cases with pseudochylothorax. Clinically most of the patients are asymptomatic.⁸ This entity does not require treatment because of

its benign course. However the presence of large amounts of fluid resulting in symptoms necessitate therapeutic thoracentesis. In symptomatic patients, if the underlying lung tissue is thought to be functional, decortication can be performed to achieve marked improvement in respiratory functions. Pseudochylothorax can result in complications such as respiratory failure, reactivation of tuberculosis, nonspecific infections, fungal infections (especially aspergillus), bronchopleural and/or pleurocutaneous fistulas. We did not perform additional intervention on our case due to lack of any respiratory complaints and normal pulmonary functions.

As a conclusion, when a cloudy or milky fluid is aspirated through thoracentesis, empyema, chylothorax and pseudochylothorax should be considered in the differential diagnosis and the levels of cholesterol and triglyceride should be measured. Since pseudochylothorax is a rarely seen type of pleural effusion, a case with this pathology has been presented.

REFERENCES

- Tabak L. [Chylothorax and pseudochylothorax]. Çavdar T, Ekim N, editörler. Plevra Hastalıkları; Toraks Kitapları. 4.Baskı. İstanbul: Turgut Yayıncılık; 2003. p.285-94.
- Güzel A, Özgül A, Kadakal F, Küçük B, Yılmaz V. [A rare case of pseudochylothorax]. Solunum Hastalıkları 2006;17(2):95-8.
- Garcia-Zamalloa A, Ruiz-Irastorza G, Aguayo FJ, Gurrutxaga N. Pseudochylothorax: Report of 2 cases and review of the literature. Medicine (Baltimore) 1999;78(3):200-7.
- Hillerdal G. Chylothorax and pseudochylothorax. Eur Respir J 1997;10(5):1157-62.
- Ferguson GC. Cholesterol pleural effusion in rheumatoid lung disease. Thorax 1966; 21(6): 577-82.
- Light RW. Chylothorax and pseudochylothorax. Pleural Diseases. 4th ed. Philedelphia: Lippincott, Williams and Wilkins; 2001. p.284-98.
- Agrawal V, Sahn SA. Lipid pleural effusions. Am J Med Sci 2008;335(1):16-20.
- Solmaz S, Hanta İ. [A rare pleural effusion: Pseudochylothorax]. Turkiye Klinikleri J Med Sci 2007;27(3):478-81.
- Goldman A, Burford TH. Cholesterol pleural effusion: A report of 3 cases with a cure by decortication. Chest 1950;18(6):586-94.
- Erkan ML, Atıcı AG, Fındık S. [A case of pseudochylothorax]. Turkish Respiratory Journal 2001;2(1):23-5.