UNUSUAL PRESENTATION OF PULMONARY SARCOIDOSIS

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INTRODUCTION

Sarcoidosis is a systemic granulomatous disease of unknown etiology, with ambiguous presentation. Pulmonary sarcoidosis is rare and its presentation is atypical. In this paper we present a case of pulmonary sarcoidosis with unspecific presentation. In 1995, a 36-year-old pilot was presented with no previous history of any notable illness and without any symptoms. Chest radiography showed a lesion and Ziehl-Neelsen staining showed the presence of acid-resistant bacteria, after which antituberculotic therapy started. After multiple hospitalizations, in 2011, radiography showed changes in lungs that resemble sarcoidosis or tuberculous sequelae. In 2016, tuberculosis was excluded and revised pathohistological findings of granulomatous inflammation in 2003, clinical presentation and radiography findings suggested sarcoidosis with slow progression and fibrous indurative changes in upper lung lobes with the presence of low-grade lymphocyte alveolitis.

Key words: sarcoidosis; tuberculosis; lung diseases, interstitial.

CASE REPORT

In 1995, a 36-year-old pilot came to an annual check-up at the tertiary medical center, with no previous history of any notable illness and without any symptoms. He was an ex-smoker. Laboratory results, spirometry and gas analysis were in normal range. Chest radiography showed a lesion in the right upper pulmonary lobe. He was hospitalized in October 1995 at this institution with the duration of 23 days. During that time, lung tuberculosis was excluded and revised pathohistological findings of granulomatous inflammation in 2003, clinical presentation and radiography findings suggested sarcoidosis with slow progression and fibrous indurative changes in upper lung lobes with the presence of low-grade lymphocyte alveolitis. In 2016, tuberculosis was excluded and revised pathohistological findings of granulomatous inflammation in 2003, clinical presentation and radiography findings suggested sarcoidosis with slow progression and fibrous indurative changes in upper lung lobes with the presence of low-grade lymphocyte alveolitis.
was suspected, but sputum examinations with direct bacilloscopy did not show acidoresistant bacilli (ARB) and Lowenstein–Jensen (Low) cultures were negative. CT scan showed an infiltration surrounded with satellite lesions in the right lung apex. Transbronchial biopsy was done and pathohistological finding (PH) was bronchitis and granulomatous peribronchitis. Ziehl-Neelsen staining showed the presence of acid-resistant bacteria, after which triple antituberculotic (ATL) therapy (isoniazid (H), rifampicin (R), pyrazinamide (Z)) was started at the Special Hospital for Pulmonary Diseases "Ozren" Sokobanja where he was referred. The treatment lasted for 2 months, during which Low cultures and direct microscopy remained negative. Two years later, he was hospitalized for the second time at the Special Hospital for Pulmonary Diseases "Ozren" Sokobanja, for 3 months, during which time new lesions were presented in left lung and right apex and showed poor radiological regression. An open lung biopsy was made and pathohistological findings were in favor of chronic granulomatous inflammation of tuberculous nature. The treatment was started with four ATL drugs (H, R, ethambutol (E) and streptomycin (S)), with an extension phase with three ATL drugs (H, R, E) in Ozren. In August 1997, Low culture was positive (7 colones) after which it remained negative. Resistance test showed sensitivity to all ATL. The patient was treated for a total of 9 months.

In 2002, the patient came with an elevated body temperatures up to 38°C, cough, increased sweating and fatigue during exercise. He was hospitalized at the Special Hospital for Pulmonary Diseases "Ozren" Sokobanja for 3 months. Chest radiography showed progression of lung lesions (Figure 1). Two samples of sputum examined by direct microscopy were positive. Relapse treatment was started and conducted with 5 ATL drugs. Pathohistological samples from 1995 and 1997 were revised after which granulomatous inflammation was seen and sarcoidosis was suspected.

In 2003, chest radiography showed progression and new lesions in the upper left lobe. Laboratory results were in the normal range, bronchoalveolar lavage showed lymphocytic alveolitis. Samples from 1995 and 1997 were revised and showed granulomatus inflammation, most likely within the framework of sarcoidosis. Immunosuppressive therapy (with chemioprofilaxys eutison – izoniazide + vitamin B6) was indicated and the treatment lasted for 6 months.

In 2008 and 2010 two CT scans of thorax were made. The first one showed fibrous indurative changes in both upper pulmonary lobes and the CT from 2010 (Figure 2) showed progression of lung lesions with mediastinal lymphadenopathy which suggested chronic sarcoidosis.

In 2011 the patient was admitted to the other institution. He was presented with cough, fatigue and edema of right ankle joint. Laboratory analysis were within the limits of reference values. Radiography of feet and hands showed initial degenerative changes.

Bronchial findings were normal: bronchoalveolar lavage (BAL), tranbronchial needle aspiration (TBNB) on trachea, transbronchial biopsy (TBB) as well as aspirate and cytology, ARB and Low. BAL showed in the sediment: erythrocytes, cylindrical cells, macrophages.
48%, seg.35%, eoynophilis 5% and Ly 12%. ARBs were not seen from the bronchial aspirate in the direct preparation. PH finding from TBNA and TBB was non-specific. Pulmonary function tests showed mixed ventilation disorder of a mild degree. Carbon monoxide (CO) diffusion was normal. Saturation was 98%. CT scan of thorax was performed, and signs of specific inflammation, sarcoidosis or neoplasm were not found. Radiography showed changes in lungs that resemble sarcoidosis or tuberculous sequelae.

In 2016 a new set of examinations was performed due to the presence of symptoms within the last 6 months (fatigue, shoulder and pelvis pain, with limited mobility and right ankle edema). The patient denied respiratory symptoms. Bronchial examination was normal (BAL and aspirate). Spirometry showed mild obstructive disorder. CO diffusion was decreased (75.5%) and saturation was 99%. Thoracic CT scan was also performed (Figure 3). Immunosuppressive treatment continued.

In the absence of the expected radiological regression of lung lesions, in addition to adequately applied ATL therapies, tuberculosis was excluded. Revised PH finding of granulomatous inflammation in 2003, clinical presentation and radiography findings suggested sarcoidosis with slow progression and fibrous indurative changes in upper lung lobes with the presence of low-grade lymphocyte alveolitis.

DISCUSSION

Sarcoidosis is a condition that includes both lung findings as well as other locations. Its finding in the lungs is not specific for sarcoidosis and it is very diverse, which is why it can resemble a great number of pulmonary diseases. Because of that, diagnosing sarcoidosis in patients with only pulmonary findings can be very challenging. Imaging has an important role in the diagnosing process, even though sarcoidosis has variable appearance (7-9).

Our patient was presented only with pulmonary findings which were the only imaging finding for almost 16 years of the treatment period. After that, degenerative changes were seen in feet and hands. This was not the specific finding due to the patient’s age at that time. But it should be also noted that arthritis is seen in 18% of patients as an extra-pulmonary finding, which is why doctors should pay close attention to its signs and symptoms (4, 10). Our study showed no correlation between the incidence of extra-pulmonary findings and duration of the disease, which is also described in the literature (4). Our study also showed that the diagnosis of sarcoidosis can take time and even though laboratory results at one point suggested tuberculosis, the final diagnosis was sarcoidosis, thanks to the revision of previous samples. The revised pathohistological finding, clinical presentation and radiography supported that diagnosis.

CONCLUSION

Sarcoidosis is a disease with ambiguous presentation. Pulmonary sarcoidosis is rare and its presentation is untypical. It is important for patients and doctors to stay determined in the process of treatment due to the fact that time needed for the diagnosis of sarcoidosis can be extended and in most cases unnecessary treatment and diagnostic tests are performed.

ABBREVIATIONS

computed tomography (CT), acidoresistent bacilli (ARB), Lowenstein–Jensen (Low), pathohistological (PH), antituberculotic (ATL), isoniazid (H), rifampicin (R), pyrazinamide (Z), ethambutol (E), streptomycin (S), bronchoalveolar lavage (BAL), transbronchial needle aspiration (TBNA), transbronchial biopsy (TBB), Carbon monoxide (CO)
REFERENCES