Case Report

A rare simultaneous condition: Pulmonary brucellosis in patient with hypogonadotropic hypogonadism

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Abstract

Brucellosis is an important zoonotic disease that remains a world-wide public health problem. Pulmonary involvement in the course of brucellosis is an extremely rare condition reported about 1-5% of the cases. Here, we present a case with pulmonary brucellosis, and hypogonadotropic (secondary) hypogonadism referred our tertiary medical center for further evaluation about eunuchoidism and delayed puberty when he was applied to the primary care center with the complaint of only non-productive cough.

Clinicians should be considered to the possibility of brucellosis being the cause of respiratory symptoms, even in the absence of other frequent symptoms of brucellosis particularly osteoarticular problems.

Introduction

Brucellosis is an important zoonotic disease that remains a world-wide public health problem (1). Annually, there are 5,00,000 new cases reported in the world, and the incidence is 0.59/1,00,000 persons per year in Turkey (2). Pulmonary involvement in the course of brucellosis is an extremely rare condition reported about 1-5% of the cases (3,4). Pulmonary infections of brucellosis transmitted by respiratory tract can be presented in different manifestations including bronchopneumonia, consolidation, pleural effusion, lung abscess, empyema, solitary nodules, granulomas, and hilar and paratracheal lymphadenopathy (5,6). Here, we present a case with pulmonary brucellosis, and hypogonadotropic (secondary) hypogonadism referred our tertiary medical center for further evaluation about eunuchoidism and delayed puberty when he was applied to the primary care center with the complaint of only non-productive cough.

Case Report

A 21-year-old male patient presented with a dry cough was diagnosed with a common cold and, symptomatic treatment had been given by the primary care center. In addition to this and due to the presence of clinical findings compatible with eunuchoidism and delayed puberty in the case, the patient was referred to the Endocrinology Department of Haydarpaşa Training Hospital, Gulhane School of Medicine, Istanbul. The patient told that he had not developed secondary sex characteristics since childhood, and he had erection and ejaculation problems. The patient had no headache, nausea, vomiting, loss of vision, double vision, loss of smell, weight loss, or fever in a detailed clinical history. However, the patient had an ongoing dry cough for about a month. He had a history of penicillin derivative
antibiotics in addition to symptomatic treatments due to these complaints. He stated that he worked as a shepherd for 8 years until last month, and he had been drinking 2 packs of cigarette a day.

He was conscious, with vital signs of arterial blood pressure 110/70 mmHg, the heart rate 104 beats per minute, body temperature 37.2°C, respectively. In the respiratory system examination, there were sounded crackles over the left lower lobe at the end of inspiration. The patients’ secondary sexual features were significantly slow, and Tanner stage was consistent with G1P1, and there was eunuchoidal appearance. Other systemic examination was normal.

Chest X-ray was compatible that appearance of suspicious nodular opacities on the right lung lower lobe and suspicious infiltrative shadow on the left lung lower lobe. Thorax computerized tomography (CT) scans were obtained to confirm, and ground-glass opacity around the nodular lesion was seen on the right lung, and also, pneumatic infiltration shadows were observed at the subpleural area on the left lung (Figure 1). Acute-phase reactants such as erythrocyte sedimentation rate (ESR 41 mm/h, 0-15 mm/h) and C-reactive protein (CRP 14.1 mg/L, 0-8 mg/L) were elevated. Advanced tests were planned for etiology as the patient was considered having pneumonia. Sputum acid-fast staining was negative. Due to a history of shepherding, Brucella examination was performed, Rose Bengal was positive, and standard tube agglutination test was negative. Coombs tests were reported as positive (1/160 titer). On the 3rd day, two sets of blood cultures yielded Brucella melitensis. Skeletal scintigraphy was negative in terms of bone and joint involvement. The patient was diagnosed as brucellosis with pulmonary involvement and started on oral doxycycline (200 mg/day) and rifampin (600 mg/day) for 6 weeks. Hormone panels of the patient due to hypogonadism etiology were quite low (follicle stimulating hormone 0.23 mIU/ml; luteinizing hormone 0.03 mIU/ml; total testosterone 18.79 ng/dl). Other pituitary and thyroid hormone panel and pituitary magnetic resonance imaging were normal. Testicular volumes were showed significantly lower by scrotal ultrasound (right testicle 1.1 cc; left testicle 1 cc). Idiopathic hypogonadotrophic hypogonadism was evaluated, and human chorionic gonadotropin therapy (pregnyl 1500 IU, 3 times a week) was started. After the specific antibiotic treatments, respiratory symptoms were improved, laboratory values were returned to normal (ESR 13 mm/h; CRP 3.69 mg/L), and blood culture was negative. In control, thorax and lung lesions were observed to disappear (Figure 2). The treatment related to hypogonadism was recommended to continue since the patient was getting better.

Discussion

Brucellosis that transmitted to humans by contact with animals as directly or indirectly is an important zoonotic disease. As osteoarticular involvement is the most common presentation, pulmonary involvement is extremely rare in brucellosis (3,4,7). Pneumonia may also be the sole presentation of brucellosis (8). While the certain mechanism of pulmonary involvement is not clear, inhalation of contaminated aerosols and hematogenous spread are a most common route for pulmonary brucellosis (9). Most of the patients have applied as an acute infection with respiratory symptoms such
as cough, fever, dyspnea, productive sputum, pleuritic chest pain, and crackles for up to 1 month before hospital admission (10,11). There was a strong reason which explained the source of brucellosis because of the patient who was a shepherd in his medical history. However, the detection of pulmonary brucellosis as an extremely rare condition in our case who has become interesting, While the patient was evaluated for hypogonadism and given treatment of common cold. In addition, there was only seen the pulmonary involvement of brucellosis in the case in the absence of the most frequent finding as well as bone-joint involvement.

Pulmonary infections of brucellosis can be presented in different manifestations including bronchopneumonia, consolidation, pleural effusion, lung abscess, empyema, solitary nodules, granulomas, and hilar and parastracheal lymphadenopathy (5,6). The parenchymal nodule and abscess are the rare pulmonary manifestations. The prevalence of parenchymal nodule is found 7.5% in the largest study about pulmonary brucellosis (12). In our case, there was a pulmonary nodule which improved after medication and shown by chest roentgenogram and thorax CT (Figures 1 and 2).

Diagnosis depends on the presence of clinical findings suggestive of brucellosis and serological tests with or without positive blood culture (5). The percentage of brucellosis with positive blood cultures ranges from 15 to 70 (13). Positivity ratio of blood cultures is decreased with the previous use of antibiotics and with subacute and chronic forms of brucellosis (14). A variety of serologic methods is available for serodiagnosis of brucellosis such as standard tube agglutination test, ELISA, Rose Bengal agglutination, Coombs test, and Immunocapture agglutination (15). Rose Bengal agglutination test is often used as a rapid screening test with very high sensitivity (>99%) and low specificity (16). The serum agglutination test (SAT) is the most widely used and practical test to the definition of brucellosis (17). Because of the SAT titer may be decreased or negative, due to blocking antibodies in subacute or chronic forms of brucellosis, a Coombs test must be performed to demonstrate incomplete antibodies (18). Optimal therapy of brucellosis remains based on traditional combinations of doxycycline, with the addition of either streptomycin or rifampicin (19). The results of patients’ two different blood cultures were positive in spite of the use of antibiotic in the recent weeks. Nevertheless, in our case that defined as an acute brucellosis the Coombs test was found positive even though the SAT titers were negative. When the patient was re-examined after the treatment with the standard dose of doxycycline and rifampin, clinical findings, laboratory, and radiological findings were recovered.

Although a patient with primary hypogonadism (hypergonadotropic hypogonadism) due to brucellosis was reported in the past, to our knowledge there were not published any case of secondary hypogonadism as coexisting or associated with brucellosis. In this case, we have shown coincidentally two independent and different entities including brucellosis and secondary hypogonadism coexistence.

**Conclusion**

Pulmonary involvement in brucellosis is a rare condition. In general, respiratory symptoms have been revealed for 1 month before admission for Brucella pneumonia. Clinicians should be considered to the possibility of brucellosis being the cause of respiratory symptoms, even in the absence of other frequent symptoms of brucellosis particularly osteoarticular problems.

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