CASE REPORT

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Incidentally Detection of the Swyer-James Syndrome With Opposite Side Pneumonia

Guner Demir[1], Mehmet Aslan[2*]

[1]Ozel Agri Yasam Hospital, Department of Radiology, Agri, Turkey
[2]Yuzuncu Yil University, Medical Faculty, Department of Internal Medicine, Van, Turkey

ABSTRACT

Swyer-James Syndrome (SJS) is a rare disease that is also known as unilateral hyperlucent of the lung. This disease may lead to atelectasis and pulmonary hypoplasia. The viral and bacterial respiratory system infections in the childhood are considered as predisposing factors of the chronical bronchial inflammation that leads to SJS. It is characterized by obliteration of the bronchias, air-trapping and lung hypoperfusion. In this report, we describe the case of a 50-year-old male with pneumonia and incidentally detected SJS in the other side of the thorax.

Key Words: Swyer-James Syndrome, unilateral hyperlucent lung, pneumonia, adults

Introduction

Swyer-James Syndrome (SJS), a unilateral hyperlucent lung, was described by Swyer and James in 1953 (1). Childhood respiratory infections such as bronchiolitis and pneumonia were thought as predisposing factors of this disease (2). The radiological findings of the SJS are attenuated vascular appearance of the bronchias and a slight hilar shadow due to diminished arterial supply (3). Most of the patients are asymptomic but dyspnea, recurrent respiratory infections. The severe infectional disease of the bronchias may lead to atelactasis and obliteration (4).

It has been reported that SJS may be associated with low respiratory tract infections such as pneumonia and bronchitis in the childhood (5). However, to our knowledge, this syndrome is asymptomic in the adults and suspection is a key point to diagnosis this disease. In this paper, we aimed to demonstrate an adult SJS patient with opposite side pneumonia whose had no respiratory problem in his history.

Case Report

A 50-year-old male patient admitted to emergency service with the complaints fatigue, weakness, productive cough and fever. No systemic symptomic such as night sweat and weight loss were observed. He had no systemic disease. There was a 20 pack-years smoking history but he quit smoking five years ago.

The vital signs was normal except for body temperature of 38.5 ^{0}C. On the physical examination, breath sounds were diminished in the left chest and the expirium was prolonged. Furthermore, there were inspiratory crackles in the right chest.

The white blood cell count was 13.200 cells/mm3 (neutrophilic predominance); C-reactive protein was 7.8 mg/dL; erythrocyte sedimentation rate was 42 mm/hour; the other haemogram levels, blood glucose, liver function tests and kidney function tests were within normal limits (Table 1).

Unilateral increased radiolucency and markedly decreased broncovascularity in the mid and low zone of the left lung, and increased opacity in low zone of the right lung were detected in the chest X-ray (Fig. 1). Increased density increased in the lower lobe of the right lung and significant air-trapping sign with the emphysema in the lower lobe of the left lung according to the left upper lobe were observed in the high resolution computed tomography (HRCT) of the thorax (Fig. 2). Beside this, there was a significantly lower main artery diameter in the left side compared to right side (Fig. 2). Finally, the diagnosis was considered as SJS of the left lung and bacterial pneumonia of the right lung. The patient was treated with sefuroxime axetil 500 mg for 14 days and the clinical findings dramatically regressed. At the end of the first month, the patient had no respiratory complaint. Unilateral left lung hyperlucency without bronchiectasis was demonstrated in the HRCT at the 6th month follow-up (Fig. 3).

***Corresponding Author:** Dr. Mehmet Aslan, Van Yuzuncu Yil University, Medical Faculty, Department of Internal Medicine Van, Turkey
E-mail: m.aslan301@mynet.com, Fax: +90 (432) 216 75 19, Phone: +90 (432) 215 04 73
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Table 1. Blood parameters of the patient

WBC (x10³/μl)	13.200 (normal range:4.000-10.000)
Absolute neutrophil count (x10³/μl)	8.820 (normal range:1.100-7.000)
Hb (g/dL)	17.17 (normal range:12-18)
MCV (fL)	91.6 (normal range:80-100)
Platelet number (x10³/ μl)	325 (normal range:150-350)
CRP (mg/dL)	7.8 (normal range:0-5)
ESR (mm/h)	42 (normal range:0-20)
Creatinine (mg/dL)	0.66 (normal range:0.7-1.3)
AST (U/L)	15 (normal range 0-31)
ALT (U/L)	18 (normal range:0-31)
Glucose (mg/dL)	85 (normal range:70-105)

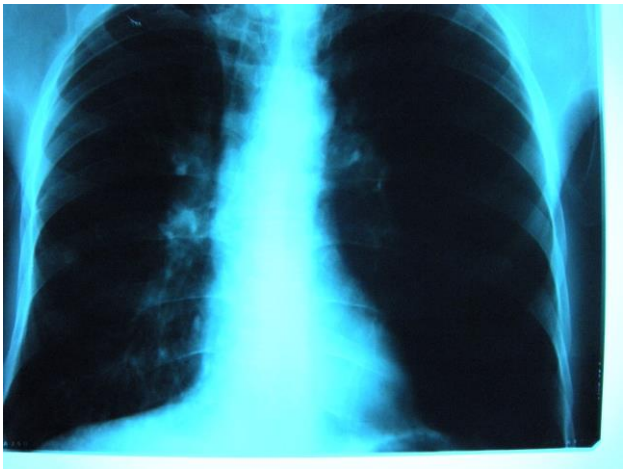

Fig. 1. Increased radiolucency and markedly decreased in signs broncovascularity in mid and low zone of the left lung, and increased opacity in low zone of the right lung in the X-ray

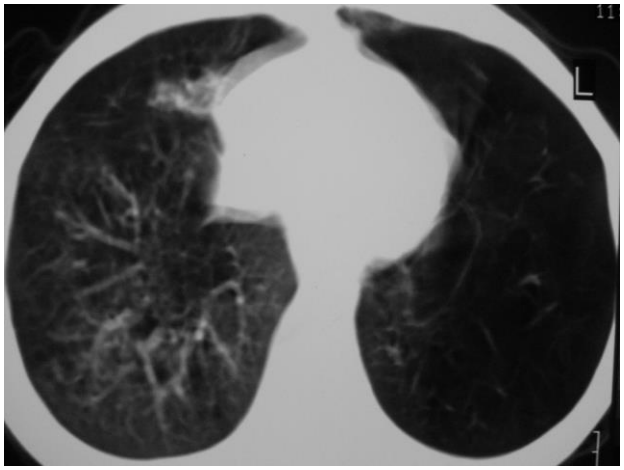

Fig. 2. Increased density in the lower lobe of the right lung and markedly air-trapping in the lower lobe of the left lung according to the left upper lobe in thorax HRCT

Discussion

SJS is characterized by an unilateral hyperlucency small lung (4). The pathological findings of this disease are presence of bronchiolitis obliterans, destruction of the alveolar structures, air-trapping sign and lung hypoperfusion.

The SJS and congenital absence or hypoplasia of a pulmonary artery is considered as the major causes of the unilateral hyperlucent lung (4). At first, SJS was considered as congenital disease, and associated with hypoplasic pulmonary artery (4). It is now considered as a complication of the respiratory system infections of the childhood and the narrowing of the small airways that leads to air-trapping and pulmonary artery hypoplasia (4). Several respiratory infections have been implicated in the cause of SJS, especially adenovirus types 3, 7, and 21, measles virus, respiratory syncytial virus, whooping cough by bordetella pertussis, tuberculosis, influenza virus A and mycoplasma pneumonia (6).

There are different studies that demonstrate the relationship of the bacterial infections and SJS. Wong et al. (5) described the case report of SJS as a complication of the atypical pneumoniae in 5 year-old girl. Stokes et al. (7) reported the case report of SJS after severe bacterial pneumoniae in 11-year-old girl. Okabe et al. (8) reported a hyperlucency area in the right lower lung in chest X-ray and bronchiectasis in the HRCT of a 25-year-old man who had Mycobacterium avium infection. Similarly, Okabe et al. (8) reported hyperlucency in the right lung in chest X-ray of a 65-year-old woman who admitted to hospital with dyspnea.

Chest radiography, HRCT, ventilation/perfusion (V/Q) scintigraphy and magnetic resonance angiography were described as imaging methods to diagnosis the SJS (9). Decreased perfusion and ventilation was observed in the patients with SJS (7).

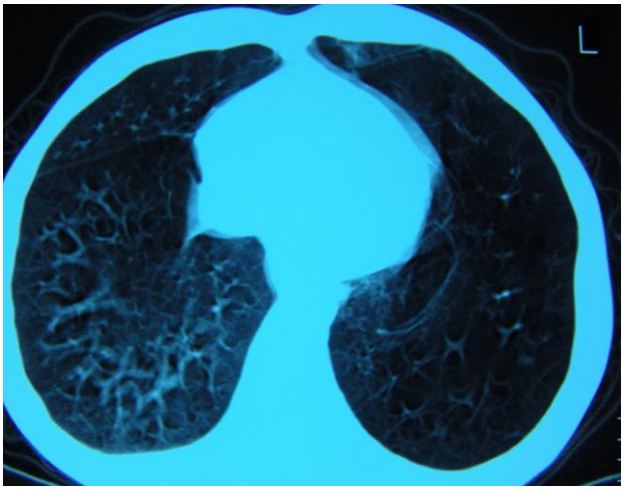

Fig. 3. Severe air-trapping in the affected lung tissue and unilateral left lung hyperlucency without bronchiectasis in thorax HRCT

The V/Q defect is also one of the findings of the chronic obstructive pulmonary disease and pneumothorax (10). Decreased pulmonary circulation may be the cause of the hypoplasia of the affected lung in the patients with the SJS. On the other hand, the clinical presentation may be the hyperinflated lung that was herniated to other side (11). In a case report, SJS was diagnosed with a coronary artery abnormality (12). According to clinical and radiological findings, treatment methods have to be evaluated at a wide perspective. It could be conservative treatment, eradicating active infectional disease or resection of the affected area of the lung (6,13).

In our report, we incidentally detected the presence of the SJS while diagnosing the opposite lung. As we mentioned before, SJS may be associated with hypoperfusion and hypoplasia of the affected lung. Because of this reason, the current situation of the opposite lung is become more important in case of lung disease. For the continuation of the respiratory system, predominance of the non-affected lung is an expected situation. In this situation, while there is SJS on one side, the disease on the opposite side should be eradicated immediately. Luckily, our patient had responded antibiotherapy to eradicate the typical bacterial pneumonia.

In conclusion, we presented a case of SJS along with opposite side pneumonia and the patient was treated with antibiotherapy. Considering both sides of the chest when evaluating a lung disease may be of vital importance for predicting the prognosis of the disease.

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