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Case report

A case of familial hot tub lung

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ABSTRACT

Hot tub lung is a lung disease caused by *Mycobacterium avium* complex. We report the first case of familial hot tub lung appearing simultaneously in a husband and wife. Our case supports the consideration that hot tub lung is a hypersensitivity pneumonitis rather than an infectious lung disease. It also suggests that the state of hot tub lung changes seasonally depending on temperature variations, in a manner similar to summer-type hypersensitivity pneumonitis. This case demonstrates similarities between hot tub lung and summer-type hypersensitivity pneumonitis in regards to familial occurrence and seasonal changes in the disease state.

1. Introduction

It has recently been recognized that a hypersensitivity pneumonitis-like lung disease, which has been termed hot tub lung, can occur with exposure to *Mycobacterium avium* complex (MAC), composed of *M. avium and M. intracellulare* [1]. Despite an increasing number of reported hot tub lung cases, a familial case has never been reported [2,3]. Here, we describe the first case of familial hot tub lung occurring simultaneously in a husband and wife.

2. Case reports

A 79-year-old man and his 72-year-old wife were admitted to the National Hospital Organization Kure Medical Center and Chugoku Cancer Center (Kure, Japan) on 23 October 2013 with a chief complaint of dyspnoea on effort and cough, which had been present for one month. Although the husband had been treated for diffuse panbronchiolitis with a macrolide antibiotic and his wife had been treated for atypical pneumonia with a quinolone antibiotic, their symptoms did not improve. Their house was mouldy and wooden, and they had no pets. The husband had a history of hypertension and hyperuricemia and his wife had a history of colon cancer.

On admission, the husband's and wife's temperatures were 36.4 °C and 36.6 °C, respectively. Both patients had bilateral coarse crackles on auscultation of the lungs, an increased respiratory rate (30 breaths/min), and increased respiratory accessory muscle activity. The husband was hypoxemic with a percutaneous oxygen saturation (SpO_2) of 88% while the wife maintained a SpO_2 of 96% on room air. Bilateral ground glass opacities were observed on chest radiography and computed tomography in both patients (husband, Fig. 1a and b; wife, Fig. 2a and b). In addition, nodular shadows were present in both lung fields in the husband (Fig. 1a and b), and in right upper lung lobe (right S^2) in the wife (Fig. 2a and b).

The serum Krebs Von Den Lungen-6 (KL-6) levels were elevated at 839 U/mL and 721 U/mL in the husband and wife, respectively (normal range, <500 U/mL) (Fig. 3) [4]. The possibility of summer-type hypersensitivity pneumonitis was considered based on their symptoms, physical findings, radiological findings, and elevations in serum KL-6 levels [5,6]. However, the serum immunoglobulin G antibody against *Trichosporon asahii* was negative in both patients with a corrected absorbance index of 0.03 and 0.00 in the husband and wife, respectively.

Transbronchial lung biopsy revealed a small noncaseating granuloma with a lymphocytic infiltration of the alveoli. *M. intracellulare* was isolated from the bronchoalveolar lavage fluid of the husband, which was obtained from the right upper lung lobe (right S^3a) where the nodular shadow existed; however, MAC species were not isolated from the wife's bronchoalveolar lavage fluid obtained from the right S^2 segment where the nodular shadow existed. Their symptoms improved without any intervention within ten days after admission. Chest computed tomography findings at three weeks after admission showed improvement of the bilateral ground glass opacities in both patients (husband,

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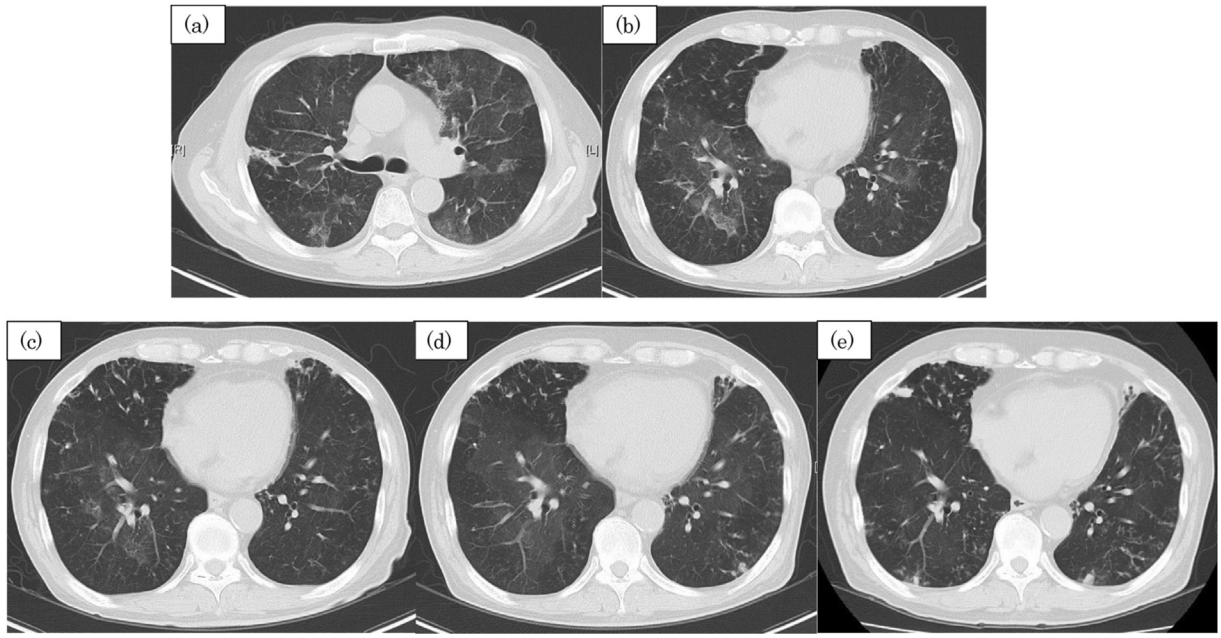

Fig. 1. Serial computed tomography (CT) findings in the husband. (a), (b); CT scan at first admission in October 2013 shows bilateral ground glass opacities and nodular shadows in both lung fields. (c); CT scan 3 weeks after the first admission shows remarkable improvement in the ground glass opacities. (d); CT scan at the second admission in June 2014 once again shows bilateral ground glass opacities. The bilateral nodular shadows have increased in comparison to the first admission and bronchiectasis has appeared in the left lingual lobe. (e); CT scan from August 2015 shows minimal persistence of the bilateral ground glass opacities. Diffuse bilateral nodular shadows persist.

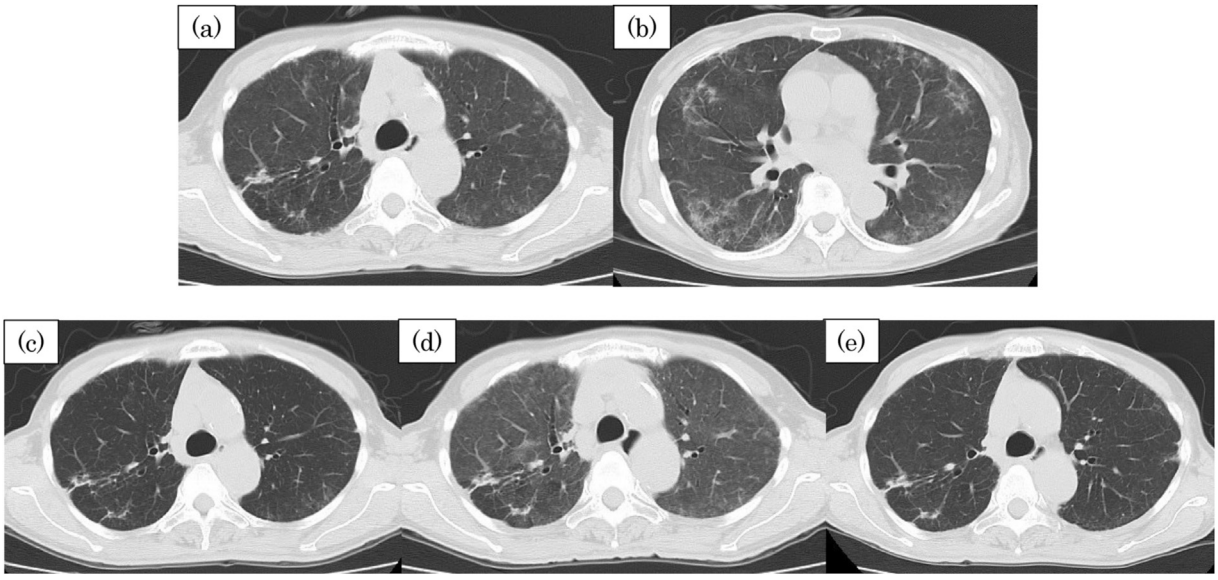

Fig. 2. Serial computed tomography (CT) findings in the wife. (a), (b); CT scan at first admission in October 2013 shows nodular shadows in the right upper lung lobe (right S^2) in addition to bilateral ground glass opacities. (c); CT scan 3 weeks after the first admission shows remarkable improvement in the ground glass opacities. (d); CT scan at the second admission in June 2014 once again shows the presence of bilateral ground glass opacities. (e); CT scan from August 2015 show minimal persistence of the bilateral ground glass opacities. The nodular shadows in the right S^2 have remained stable throughout the entire clinical course.

Fig. 1c; wife, Fig. 2c). Serum immunoglobulin A antibodies to the glycopeptidolipid core, derived from the *M. avium* complex by enzyme-linked immunosorbent assay (Capilia™ MAC Ab ELISA, TAUNS Laboratories, Inc., Izunokuni, Japan), were positive in the husband (corrected absorbance index >10.0), but not in his wife (corrected absorbance index <0.05) [7]. Although we recommended that the patients clean their house or move to a new house because of the suspicion of hot tub lung, they remained in the same house and did not clean it.

During the winter and spring, they had no respiratory symptoms, such as dyspnoea on effort or cough, and serum KL-6 levels declined (Fig. 3). However, on 18 June 2014, they were re-admitted to our hospital due to dyspnoea on effort and cough. Bilateral ground glass opacities were observed once again on chest