

Hypersensitivity pneumonitis due to unclean continuous positive airway pressure equipment

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Abstract

Hypersensitivity pneumonitis is a potentially fatal immunological lung disease caused by occupational or environmental exposure to specific antigens. Here, we report on an obstructive sleep apnea patient whose hypersensitivity pneumonitis was aggravated because of the use of unclean continuous positive airway pressure equipment. This report shows that careful history taking is important when diagnosing hypersensitivity pneumonitis. Sleep specialists should be aware of the risks related to unclean continuous positive airway pressure equipment use, and obstructive sleep apnea patients should be instructed and monitored in the regular cleaning of their equipment.

KEYWORDS

continuous positive airway pressure, hypersensitivity pneumonitis, obstructive sleep apnea

1 | INTRODUCTION

Hypersensitivity pneumonitis (HP) is a potentially fatal immunological lung disease caused by occupational or environmental exposure to specific antigens. A number of etiological factors of HP have been reported, including contaminated forced-air systems and water reservoirs, but the type and source of offending environments are sometimes difficult to identify from clinical history.¹

Wind instruments such as saxophones and trombones have been documented as potential causes of HP.^{2–5} The first 2 documented cases, reported in 1988 and 2010, were those

of saxophone players, who showed improvement after routinely cleaning their saxophones.^{2,3} The third case, reported in 2011, was that of a trombone player whose HP worsened when he neglected to clean his trombone.⁴ In April 2017, a bagpipe player was reported to have HP related to his bagpipe use. The cause was not identified in time, and the patient died of repeated exacerbations of HP.⁵

Recently, clinicians identified a new type of ‘wind instrument’ as a potential cause of HP if it is inadequately cleaned and remains moist. Continuous positive airway pressure (CPAP) equipment, widely used for the treatment of obstructive sleep apnea (OSA), has a forced-air system and heated

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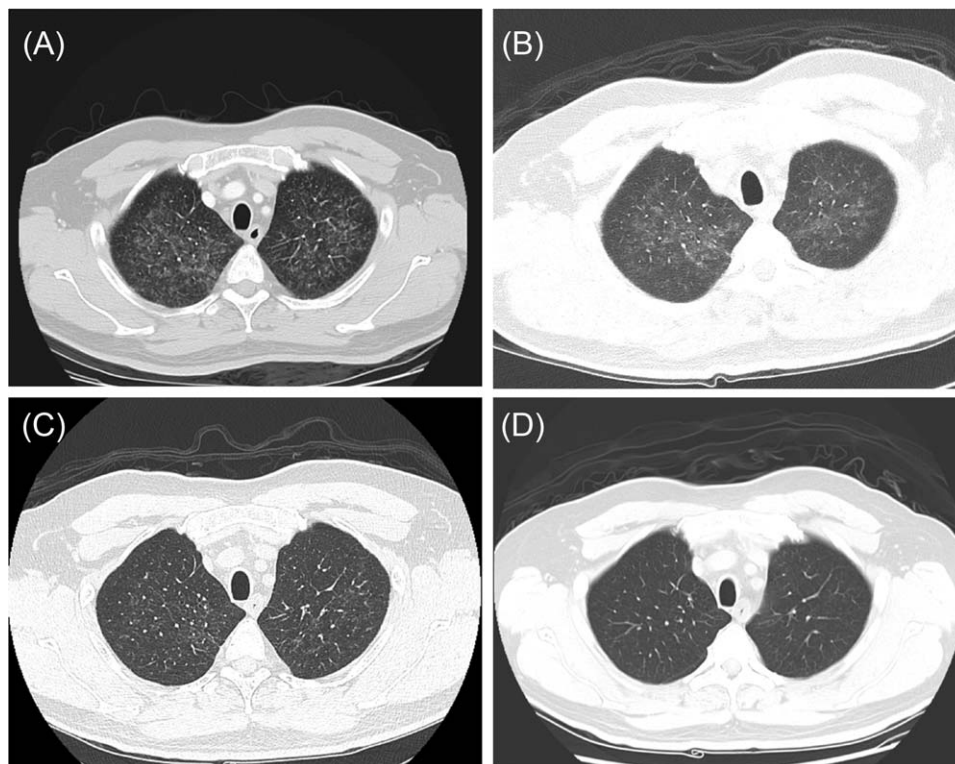


FIGURE 1 Series chest CT results showed the relationship between unclean CPAP and his hypersensitivity pneumonitis. A, The chest CT showed numerous poorly defined centrilobular nodules and some ground-glass opacities with centrilobular concentrations, predominately in upper and middle lung at January 2016. B, At April 2016, the chest CT showed partial regression with prednisolone use but continuous using foul CPAP. C, At May 2016, the chest CT dramatically improved after he stopped to use his unclean CPAP. D, At November 2016, the chest CT revealed completely regression after resumed his own clean CPAP

humidifier. Because CPAP equipment provides a warm and moist environment, it may be a trigger for HP. Musicians play wind instruments by blowing air into the instrument, but some players inhale air from their instrument or mouthpiece during inspiratory phases, leading them to inhale contaminated air that may trigger HP. However, OSA patients must inhale and exhale air from CPAP equipment during daily use. Because of this, HP related to use of unclean CPAP equipment might be an even more pressing issue. However, CPAP-related HP has not been reported previously.

Here, we report an OSA patient whose HP was aggravated because of the use of unclean CPAP equipment. The patient had already provided informed consent for the publication of this report. The Institutional Review Board of Taipei Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation had proved this report.

2 | REPORT OF CASE

A 48-year-old man, a certified securities investment analyst, had been regularly treated for diabetes and hypertension since 2007. He was diagnosed with severe OSA and had

maintained regular CPAP use since May 2011. Heavy smoking was noted, but he took our advice and quit smoking.

In November 2015, he had a cough with white sputum, but no fever. Allergic rhinitis was noted, and an antihistamine was prescribed for 2 months. However, the cough and sputum worsened, and haemoptysis developed in January 2016. A chest radiograph (CXR) showed poorly defined, small opacities in both lungs with sparing on the bases. The chest computed tomography (Chest CT) showed poorly defined centrilobular nodules and some ground-glass opacities with centrilobular concentrations, predominately in the upper and middle lung (Figure 1A)

HP was highly suspected. Clinical history was taken, but no offending factor could be identified. His wife declared their house was clean, without mould or contaminated water. We decided his CPAP was a possible culprit. However, the patient declared he cleaned the mask and tubing regularly according to the user guide. We suggested he return his CPAP equipment to the manufacturer to be checked, but he refused.

Bronchoscopy was arranged, but no fungi, tuberculosis, cytomegalovirus, *Pneumocystis jiroveci* or bacteria were found. Autoimmune disease was checked for, but the results of all serologic tests were normal, including anti-nuclear

antibody, rheumatoid factor, anti-Ro/La antibody and anti-neutrophil cytoplasmic antibody. Serum *Cryptococcus* antigen and human immunodeficiency virus screening yielded negative results. Although no obvious environmental exposure could be identified, prednisolone 20 mg daily was prescribed for 3 months.

In April 2016, his cough and haemoptysis were still persisting. The chest CT showed only partial regression of his HP (Figure 1B). It was supposed that the causal exposure remained. His wife admitted he really did not clean and dry the CPAP or the pieces of apparatus regularly. He had only wiped the mask with wet napkins occasionally in the past 5 years, and the mask and tubing had much secretion and smut. His wife also clarified that his symptoms always presented after CPAP use in the morning while awakening and were absent while the CPAP was not in use.

Therefore, CPAP was confirmed as the cause of HP. He was forced to discontinue CPAP use and return the equipment to the manufacturer for maintenance and cleaning. The manufacturer provided him a new CPAP for 1 month and monitored his daily maintenance and cleaning of the equipment every week. This time, the patient refused prednisolone treatment.

In May 2016, the chest CT revealed dramatic improvement after he had stopped using the unclean CPAP for 1 month (Figure 1C), and he was allowed to use his own CPAP again. In the following 6 months, he did not experience cough or haemoptysis.

In November 2016, the chest CT revealed complete resolution of all image abnormalities (Figure 1D). The HP has not recurred.

3 | DISCUSSION

To our knowledge, this is the first report of CPAP-related HP. Daily CPAP treatment was initially considered to be a potential trigger in the development of the disease, but the possibility was excluded because the patient claimed the equipment was clean. Later, the relationship between his symptoms and unclean CPAP use became clear and was proved from his wife. Prednisolone treatment with continuing unclean CPAP use failed to improve his symptoms, suggesting that the causative factor of his HP remained. Only when the causative factor was withdrawn (unclean CPAP, without prednisolone use), his symptoms and radiological abnormalities improve. Continued CPAP use mimicked a challenge test and helped confirm the diagnosis in the absence of pathological proof. This reinforces that assessments of HP patients should include inquiries related to home-based health care equipment, especially for OSA patients who use CPAP equipment regularly.

Recently, new environments such as hot tubs and wind instruments have attracted physicians' interest because they provide ideal environments for HP development. The first case of wind-instrument (saxophone) induced HP was reported by Lodha and Sharma in 1988.² In 2010, Metzger et al reported the second case of a wind-instrument player who developed HP due to inhalation of *Candida albicans* from his saxophone. His HP improved 2 months later after a month of methylprednisolone treatment and regularly cleaning and drying his saxophone.³ In 2010, Metersky et al reported a trombone player who developed HP based on clinical and radiological findings.⁴ His symptoms also showed the same temporal relationship as described in our case, as symptoms improved on cessation of playing the trombone for 2 weeks. Another case was recently reported by King et al in April 2017 in *Thorax*.⁵ In this report, a bagpipe player died from repeated exacerbations of HP because the offending factor, the unclean bagpipe, was not recognized in time. It was suggested that preventative measures to ensure a period of abstinence and regular cleaning of the wind-instrument should be emphasized and monitored.

The best treatment of HP is contact avoidance.¹ Currently, the only accepted medicine for HP is corticosteroid, which is suggested for severe cases or when the offending factors cannot be completely removed.¹ Wind-instrument players can be forced to stop playing for a period, but for some professional players, it would be difficult to stop playing completely. It was difficult for our patient to stop CPAP use, so, we forced him to stop using his unclean CPAP and asked the seller to borrow him a clean CPAP.

This report shows that careful history taking is important when diagnosing HP. Inquiries should include questions about occupational or environment triggers, hobbies and even home-based health care equipment. With the increasing prevalence of OSA, sleep specialists should be aware of the risks related to unclean CPAP use, and OSA patients should be instructed and monitored in the regular cleaning of their equipment.

PRIVACY AND INFORMED CONSENT

The patient had already provided informed consent for the publication of this report. All authors are responsible for ensuring that their manuscript, figures comply with the Health Insurance Portability and Accountability Act (HIPAA)

CONFLICT OF INTEREST

The authors have stated explicitly that there are no conflicts of interest in connection with this article. All authors affirm that it is an original manuscript, is unpublished work, and is not under consideration elsewhere.

ETHICS

The Institutional Review Board of Taipei Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation had proved this report (Protocol No: 06-CR05-064).

AUTHOR CONTRIBUTIONS

All authors read and approved the final version of the manuscript and declare that there was no off-label or investigational use.

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