

World Journal of *Clinical Cases*

World J Clin Cases 2023 July 16; 11(20): 4734-4965



MINIREVIEWS

- 4734 Inflammatory myofibroblastic tumor of the distal common bile duct: Literature review with focus on pathological examination
Cordier F, Hoorens A, Ferdinande L, Van Dorpe J, Creytens D
- 4740 Probiotics and autoprobiotics for treatment of *Helicobacter pylori* infection
Baryshnikova NV, Ilina AS, Ermolenko EI, Uspenskiy YP, Suvorov AN
- 4752 Plant-based diet and its effect on coronary artery disease: A narrative review
Mehta P, Tawfeeq S, Padte S, Sunasra R, Desai H, Surani S, Kashyap R

ORIGINAL ARTICLE

Clinical and Translational Research

- 4763 Identification of survival-associated biomarkers based on three datasets by bioinformatics analysis in gastric cancer
Yin LK, Yuan HY, Liu JJ, Xu XL, Wang W, Bai XY, Wang P
- 4788 High expression of autophagy-related gene *EIF4EBP1* could promote tamoxifen resistance and predict poor prognosis in breast cancer
Yang S, Hui TL, Wang HQ, Zhang X, Mi YZ, Cheng M, Gao W, Geng CZ, Li SN
- 4800 Delineation of fatty acid metabolism in gastric cancer: Therapeutic implications
Fu Y, Wang B, Fu P, Zhang L, Bao Y, Gao ZZ
- 4814 Mechanical analysis of the femoral neck dynamic intersection system with different nail angles and clinical applications
Wang Y, Ma JX, Bai HH, Lu B, Sun L, Jin HZ, Ma XL

Retrospective Cohort Study

- 4824 Development and validation of a predictive model for spinal fracture risk in osteoporosis patients
Lin XM, Shi ZC

Retrospective Study

- 4833 Risk prediction model for distinguishing Gram-positive from Gram-negative bacteremia based on age and cytokine levels: A retrospective study
Zhang W, Chen T, Chen HJ, Chen N, Xing ZX, Fu XY
- 4843 Sudden death in the southern region of Saudi Arabia: A retrospective study
Al-Emam AMA, Dajam A, Alrajhi M, Alfaiji W, Al-Shraim M, Helaly AM

- 4852** Diagnostic value of preoperative examination for evaluating margin status in breast cancer

Liu P, Zhao Y, Rong DD, Li KF, Wang YJ, Zhao J, Kang H

Prospective Study

- 4865** Defining the awareness and attitude of the clinicians through pharmacovigilance in Turkey

Aydin OC, Aydin S, Guney HZ

- 4874** Predictive value of the trans-perineal three-dimensional ultrasound measurement of the pubic arch angle for vaginal delivery

Liang ZW, Gao WL

CASE REPORT

- 4883** Microwave ablation of solitary T1N0M0 papillary thyroid carcinoma: A case report

Dionísio T, Lajut L, Sousa F, Violante L, Sousa P

- 4890** Acute spinal subdural haematoma complicating a posterior spinal instrumented fusion for congenital scoliosis: A case report

Michon du Marais G, Tabard-Fougère A, Dayer R

- 4897** Subacute osteomyelitis due to *Staphylococcus caprae* in a teenager: A case report and review of the literature

Vazquez O, De Marco G, Gavira N, Habre C, Bartucz M, Steiger CN, Dayer R, Ceroni D

- 4903** ABCB4 gene mutation-associated cirrhosis with systemic amyloidosis: A case report

Cheng N, Qin YJ, Zhang Q, Li H

- 4912** Metagenomic next-generation sequencing in the diagnosis of neurocysticercosis: A case report

Xu WB, Fu JJ, Yuan XJ, Xian QJ, Zhang LJ, Song PP, You ZQ, Wang CT, Zhao QG, Pang F

- 4920** Drug-coated balloons for treating *de novo* lesions in large coronary vessels: A case report

Zhang ZQ, Qin YR, Yin M, Chen XH, Chen L, Liang WY, Wei XQ

- 4926** Pretreatment with a modified St. Thomas' solution in patients with severe upper limb injuries: Four case reports

Sun ZY, Li LY, Xing JX, Tong LC, Li Y

- 4932** Unexpected diffuse lung lesions in a patient with pulmonary alveolar proteinosis: A case report

Jian L, Zhao QQ

- 4937** Contrast-induced ischemic colitis following coronary angiography: A case report

Qiu H, Li WP

- 4944** Posterior pedicle screw fixation combined with local steroid injections for treating axial eosinophilic granulomas and atlantoaxial dislocation: A case report

Tu CQ, Chen ZD, Yao XT, Jiang YJ, Zhang BF, Lin B

- 4956** Antithrombin III deficiency in a patient with recurrent venous thromboembolism: A case report

Luo JQ, Mao SS, Chen JY, Ke XY, Zhu YF, Huang W, Sun HM, Liu ZJ

- 4961** Laryngospasm as an uncommon presentation in a patient with anti-N-methyl-D-aspartate receptor encephalitis: A case report

Wang L, Su HJ, Song GJ

ABOUT COVER

Editorial Board Member of *World Journal of Clinical Cases*, Kengo Moriyama, MD, PhD, Associate Professor, Department of Clinical Health Science, Tokai University School of Medicine, Tokai University Hachioji Hospital, Hachioji 1838, Tokyo, Japan. osaru3moving@gmail.com

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The primary aim of *World Journal of Clinical Cases* (WJCC, *World J Clin Cases*) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

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Unexpected diffuse lung lesions in a patient with pulmonary alveolar proteinosis: A case report

Li Jian, Qi-Quan Zhao

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Li Jian, Department of Endocrinology, Dazu Hospital of Chongqing Medical University, The People's Hospital of Dazu, Chongqing 402360, China

Qi-Quan Zhao, Department of Respiratory and Critical Care Medicine, Dazu Hospital of Chongqing Medical University, The People's Hospital of Dazu, Chongqing 402360, China

Corresponding author: Qi-Quan Zhao, MMed, Department of Respiratory and Critical Care Medicine, Dazu Hospital of Chongqing Medical University, The People's Hospital of Dazu, No. 1073 2nd Ring South Road, Tangxiang Street, Dazu District, Chongqing 402360, China. 316318057@qq.com

Abstract

BACKGROUND

Pulmonary alveolar proteinosis (PAP) often presents nonspecifically and can be easily confused with: (1) Idiopathic interstitial lung fibrosis; (2) alveolar carcinoma; (3) pulmonary tuberculosis; and (4) other lung diseases such as viral pneumonia, mycoplasma pneumonia, and chlamydial pneumonia.

CASE SUMMARY

Diagnosis: In this case, a patient was diagnosed with PAP through transbronchial cryobiopsy (TBCB) and quantitative metagenomic next-generation sequencing, which confirmed the impairment of surfactant turnover as the underlying cause of PAP. **Interventions:** High-volume total lung lavage was performed for this patient. **Outcomes:** The patient's clinical condition had improved significantly by the 6-month follow-up, with a 92% finger oxygen saturation. A repeat chest computed tomography scan revealed scattered patchy ground-glass shadows in both lungs, which was consistent with alveolar protein deposition but with a lower density than in the radiograph from October 23, 2022.

CONCLUSION

TBCB has unique advantages in diagnosing atypical alveolar protein deposition, particularly for enabling the early detection of PAP. This information can help patients take preventive measures to prevent or halt PAP development by avoiding dusty environments and seeking treatment with total lung lavage and inhaled granulocyte macrophage colony-stimulating factor.

Key Words: Diffuse lung lesions; Pulmonary alveolar proteinosis; Quantitative metagenomic next-generation sequencing; Transbronchial cryobiopsy; High-volume double lung

lavage; Case report

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Core Tip: Pulmonary alveolar proteinosis (PAP) is frequently misdiagnosed due to the absence of typical map-like and pavement-like manifestations in the lungs or the absence of typical pathological findings. Transbronchial cryobiopsy has unique advantages in diagnosing atypical alveolar protein deposition, particularly for enabling the early detection of PAP. This information can help patients take preventive measures to prevent or halt PAP development by avoiding dusty environments and seeking treatment with total lung lavage and inhaled granulocyte macrophage colony-stimulating factor.

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INTRODUCTION

Pulmonary alveolar proteinosis (PAP) often presents nonspecifically and can be easily confused with: (1) Idiopathic interstitial lung fibrosis; (2) alveolar carcinoma; (3) pulmonary tuberculosis; and (4) other lung conditions such as viral pneumonia, mycoplasma pneumonia, and chlamydial pneumonia. PAP is frequently misdiagnosed due to the absence of typical map-like and pavement-like manifestations in the lungs or the absence of typical pathological findings. Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

CASE PRESENTATION

Chief complaints

Cough, sputum production, and shortness of breath for 2 mo.

History of present illness

After catching a chill 2 mo ago, there was a cough with white frothy sputum. Shortness of breath occurred after physical activity but improved with rest. Other accompanying symptoms include chills, fever (exact temperature unknown), headache, and dizziness.

History of past illness

The patient had no medical history.

Personal and family history

A 40-year-old female who works in a furniture factory with a history of significant dust exposure.

Physical examination

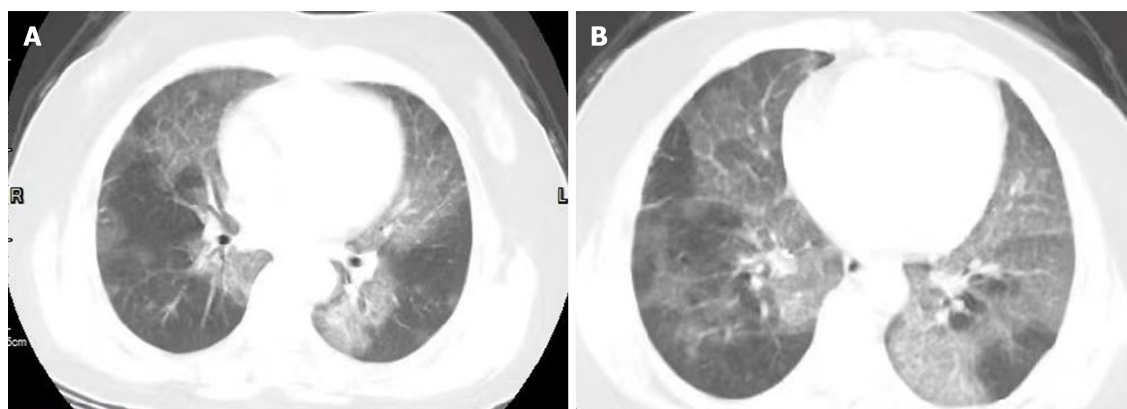
Temperature: 37.2 °C; Pulse: 90 beats per minute; Respiration rate: 20 breaths per minute; Blood pressure: 108/72 mmHg; Oxygen saturation: 93%. The patient is conscious and cooperative during the physical examination. No significant abnormalities were found during the examination of the lungs, heart, and abdomen.

Laboratory examinations

Further investigation through electron bronchoscopy and quantitative metagenomic next-generation sequencing did not reveal any significant abnormalities. Transbronchial cryobiopsy (TBCB) was performed to obtain 3 pieces of lung tissue. Pathological examination of tis tissue showed localized fibrous tissue hyperplasia, a thickening of some alveolar septa, and an eosinophilic structureless material in the focal alveolar lumen. The histological findings were combined with the clinical findings to obtain the diagnosis. Special staining results were as follows: Periodic Acid Schiff (+), Periodic Acid Schiff Diastase (±), and Congo red (-).

Imaging examinations

The patient had a finger oxygen saturation of 75%, and a chest computed tomography (CT) scan revealed diffuse infectious lesions in both lungs, suggesting possible opportunistic pneumonia (Figure 1).



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Figure 1 Chest computed tomography scan shows diffuse infectious lesions in both lungs with the possibility of opportunistic pneumonia, and it was recommended to combine these findings with the findings of the clinical and treatment follow-up. A: The right middle lobe, left lingula, and both lower lobes of the lungs exhibit diffuse ground-glass opacities; B: Both basal segments of the lower lobes of the lungs show diffuse ground-glass opacities.

The patient underwent high-volume double lung lavage. At the 6-mo follow-up, a repeat chest CT scan revealed scattered patchy ground-glass shadows in both lungs, which was consistent with alveolar protein deposition but with lower density than the October 23, 2022 radiograph (Figure 2).

FINAL DIAGNOSIS

The final diagnosis was PAP.

TREATMENT

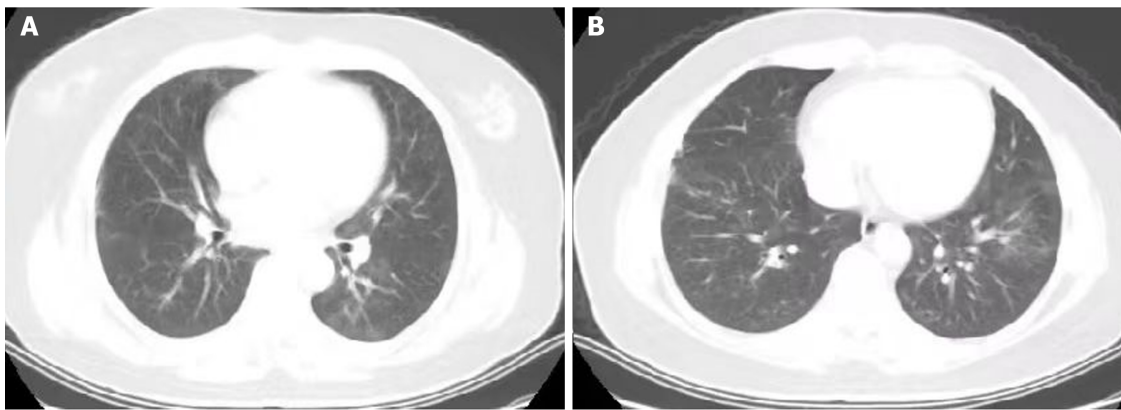
High-volume double lung lavage was performed on the patient.

OUTCOME AND FOLLOW-UP

The patient's clinical condition had improved significantly by the 6-mo follow-up, with a 92% finger oxygen saturation.

DISCUSSION

PAP is a rare lung disease characterized by the accumulation of phospholipid proteins within the alveoli[1,2]. It often presents insidiously, with typical symptoms including exertional dyspnea progressing to dyspnea at rest, accompanied by cough, white sputum, fatigue, and weight loss. PAP can be classified into the following three types[2-4]: Primary PAP is the most common type and is typically associated with abnormalities in the production of granulocyte-macrophage colony-stimulating factor (GM-CSF) antibodies. These antibodies inhibit the function of macrophages, leading to the obstruction of proteinaceous material clearance within the alveoli. Secondary PAP is caused by other diseases or factors such as certain infections, malignant tumors, immunodeficiency, and so on. Congenital PAP is a rare genetic disease caused by mutations in the genes encoding pulmonary surfactant proteins. The treatment approach depends on the type of PAP[5]: For primary PAP, whole-lung lavage is a common therapeutic method used to remove proteinaceous deposits from the alveoli. In some cases, GM-CSF therapy may also be employed. The treatment of secondary PAP involves addressing the underlying condition. The management of congenital PAP is more complex and typically includes surfactant replacement therapy, antibiotic treatment for infections, and supportive care. In some refractory cases, lung transplantation may be the only viable treatment option. PAP is more common in middle-aged and young adults and is approximately three times more common in men than in women. Dust exposure, particularly exposure to silica dust, can cause PAP. It is believed that PAP may be a nonspecific response to certain stimuli, leading to the breakdown of alveolar macrophages and the production of PAS-positive proteins[3,6-8]. PAP is a challenging disease to diagnose clinically, and a definitive diagnosis is usually made through histopathological analyses[9]. The primary methods of obtaining tissue specimens include transbronchial forceps biopsy (TBFB), percutaneous biopsy, and surgical lung biopsy (SLB). TBFB has limited diagnostic efficacy due to the small size and poor quality of the specimens obtained, making it difficult to meet



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Figure 2 Chest computed tomography scan shows scattered patchy ground-glass shadows in both lungs, which was consistent with alveolar protein deposition, with lower density than the October 23, 2022 film. A: The diffuse ground-glass opacities in the right middle lobe, left lingula, and both lower lobes of the lungs have significantly resolved compared to previous findings; B: The diffuse ground-glass opacities in both basal segments of the lower lobes of the lungs have significantly resolved compared to previous findings.

pathological analytical requirements. Percutaneous lung puncture biopsy is also associated with the problem of small specimens often not meeting pathological analytical requirements and carries the risk of pneumothorax and hemopneumothorax. SLB is less commonly used due to its high invasiveness, high cost, and limitations in patients with reduced cardiopulmonary function.

CONCLUSION

PAP is often nonspecific and can be easily confused with other lung diseases and conditions, such as idiopathic pulmonary fibrosis, lung cancer, tuberculosis, viral pneumonia, mycoplasma pneumonia, and chlamydial pneumonia. The aim of management is to improve symptoms and quality of life. TBCB is a safe and effective technique for lung tissue biopsy and is used primarily for the etiological diagnosis of diffuse lung disease but also for biopsying localized lesions in the lung periphery. TBCB is associated with less trauma, larger and higher-quality specimens, fewer complications, and significantly lower costs than SLB. TBCB has unique advantages in the diagnosis of atypical alveolar protein deposition, particularly for enabling the early detection of PAP. This information can help patients take preventive measures to prevent or halt PAP development by avoiding dusty environments and seeking treatment with total lung lavage and inhaled GM-CSF.

FOOTNOTES

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ORCID number: Qi-Quan Zhao 0000-0003-1255-4660.

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