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Pulmonary Alveolar Proteinosis Revealed by Chronic Respiratory Distress in a Young Adult: A Case Report

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Abstract **Case Report**

Pulmonary alveolar proteinosis (PAP) is a rare disease caused by the intra-alveolar accumulation of lipoproteinaceous material resulting from impaired surfactant clearance. It mainly affects young adults and often presents with progressively worsening dyspnea, sometimes contrasting with severe hypoxemia. We report the case of a 32-year-old man with no medical history, admitted for progressively worsening respiratory distress over 3 months, in an afebrile context. On admission, he was conscious and tachypneic at 38 breaths/min, with an oxygen saturation of 60% on room air, corrected to 97% with 6 liters of oxygen, intercostal retraction, and hemodynamic stability. Chest radiography showed symmetrical bilateral opacities, and chest computed tomography showed diffuse ground-glass opacities without associated cardiac signs, suggesting PAP. Bronchoalveolar lavage (BAL) returned a milky fluid, confirming the diagnosis of idiopathic PAP. The course after whole-lung lavage was favorable, with marked clinical improvement within 3 days. PAP should be considered in the presence of progressive dyspnea associated with diffuse ground-glass opacities in the absence of a cardiac cause. BAL guides the diagnosis, and whole-lung lavage remains the reference treatment, allowing rapid improvement.

Keywords: Pulmonary alveolar proteinosis; Ground-glass opacities; Bronchoalveolar lavage; Whole-lung lavage.

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INTRODUCTION

Pulmonary alveolar proteinosis (PAP), first described by Rosen *et al.,* in 1958 [1], is a rare diffuse lung disease characterized by the intra-alveolar accumulation of surfactant-rich lipoproteinaceous material, secondary to impaired clearance by alveolar macrophages [2,3]. Its prevalence is in the order of a few cases per million inhabitants [3].

Three forms are classically distinguished: the autoimmune form, also called idiopathic, which accounts for the great majority of cases and is related to the presence of autoantibodies against granulocyte-macrophage colony-stimulating factor (GM-CSF) [3,4]; secondary forms, associated in particular with hematologic disorders, infections, or inhalation of mineral dust; and congenital forms, related to genetic mutations [3]. The disease mainly affects young or middle-aged adults and most often presents with progressively worsening dyspnea, sometimes contrasting with the severity of hypoxemia [2,5,6].

We report the case of a 32-year-old patient with no medical history, admitted for chronic, progressively worsening respiratory distress, in whom imaging and bronchoalveolar lavage (BAL) led to the diagnosis of PAP, with a favorable outcome after whole-lung lavage.

CASE REPORT

This retrospective clinical observation concerns a 32-year-old man with no medical history, admitted for respiratory distress that had been worsening progressively for 3 months, in an afebrile context. On admission, he was conscious and tachypneic, with a respiratory rate of 38 breaths/min and a pulse oxygen saturation of 60% on room air, corrected to 97% with 6 liters of oxygen. Examination showed intercostal retraction. The patient was hemodynamically stable.

A chest radiograph was performed first and showed symmetrical bilateral opacities (Figure 1).



Figure 1: Frontal chest radiograph showing symmetrical bilateral opacities

It was completed by a chest computed tomography (CT) scan, which showed diffuse ground-glass opacities without associated cardiac signs (Figure 2). This picture suggested PAP.

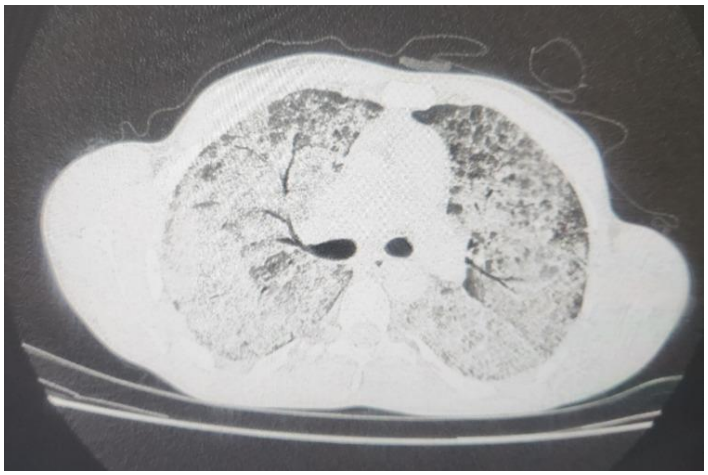


Figure 2: Axial chest CT scan (lung window) showing diffuse bilateral ground-glass opacities

Bronchoalveolar lavage was then performed and returned a milky fluid (Figure 3), confirming the diagnosis of idiopathic PAP.

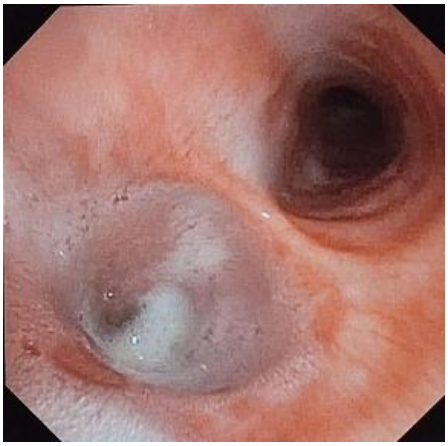


Figure 3: Endoscopic image during bronchoscopy with bronchoalveolar lavage

The patient underwent whole-lung lavage. The clinical course was favorable, with marked clinical improvement within 3 days.

DISCUSSION

PAP is a rare disease whose pathophysiology rests on defective surfactant catabolism by alveolar macrophages. In the autoimmune form, this defect is related to the presence of neutralizing autoantibodies against GM-CSF [3,4], a form that accounts for about 90% of adult PAP cases [3]. In our patient, the diagnosis of the idiopathic form was retained. His profile, that of a young adult with no medical history, is compatible with the usual presentation of the disease, whose age at diagnosis most often lies between the fourth and fifth decades [5,6].

The clinical presentation is often insidious, dominated by progressive exertional dyspnea and cough, with fever typically absent in the absence of superinfection [2,6]. Hypoxemia results from alveolar filling, which causes a shunt effect and ventilation/perfusion mismatch [2]. Our patient stands out by the severity of his presentation on admission, with a saturation of 60% on room air, tachypnea at 38 breaths/min, and intercostal retraction, after 3 months of progressive worsening. The absence of fever and the duration of the course argued in favor of a chronic process rather than an acute infectious cause.

Chest radiography typically shows bilateral alveolar opacities, often symmetrical and predominantly perihilar [6,8], which is consistent with the bilateral and symmetrical pattern observed in our patient (Figure 1). Chest CT is the key imaging examination: it classically shows areas of ground-glass opacity associated with thickening of the interlobular septa, producing the "crazy-paving" pattern, with sharp margins between affected and spared parenchyma [7]. In our patient, CT showed diffuse ground-glass opacities (Figure 2). This appearance is however not specific and may be seen in pulmonary edema, infectious pneumonia, notably Pneumocystis jirovecii pneumonia, alveolar hemorrhage, or acute respiratory distress syndrome [7,8]. The absence of associated cardiac signs made it possible here to rule out a cardiogenic origin and pointed toward the diagnosis of PAP.

Bronchoalveolar lavage (BAL) is the examination that guides, and may even confirm, the diagnosis. It classically returns a milky, opaque fluid, whose cytological examination shows acellular material positive on periodic acid-Schiff (PAS) staining and foamy macrophages [2,8]. Combined with a suggestive CT appearance, it most often avoids the need for lung biopsy [3,8]. Measurement of serum anti-GM-CSF antibodies, which is highly sensitive and highly specific, allows confirmation of the autoimmune form [3,4]. In our patient, the milky appearance of the lavage fluid confirmed the diagnosis (Figure 3).

Whole-lung lavage (WLL) remains the reference treatment for symptomatic forms, particularly in cases of significant hypoxemia [3,10]. Under general anesthesia with selective intubation using a double-lumen tube, one lung is lavaged with large volumes of warmed saline, administered in aliquots, until the returning fluid is clear, while the contralateral lung is ventilated [10,11]. It allows clinical, functional, and gas-exchange improvement in the majority of cases, with a benefit that may last from several months to several years; relapses may nevertheless occur and justify repeat lavages [10]. In our patient, the course was favorable, with marked clinical improvement within 3 days. Other approaches, notably inhaled GM-CSF, have shown benefit in the autoimmune form [12,13], but their availability varies between countries.

This report has the limitations inherent to a single case. Serum anti-GM-CSF antibody measurement is not part of our observation, and the reported follow-up, 3 days after lavage, does not allow assessment of the durability of the response or of the risk of relapse, which requires clinical, blood-gas, and radiological follow-up. Nevertheless, this case illustrates that PAP should be considered in the presence of progressive dyspnea associated with diffuse ground-glass opacities in the absence of a cardiac cause, and that an approach combining imaging and BAL, followed by WLL, allows rapid improvement.

CONCLUSION

PAP is a rare disease that should be considered in the presence of progressive dyspnea associated with diffuse ground-glass opacities, in the absence of a cardiac cause, including in a young adult with no medical history. Bronchoalveolar lavage guides the diagnosis through the milky appearance of the recovered fluid, and whole-lung lavage remains the reference treatment, allowing rapid improvement. This case underlines the value of early diagnosis in the presence of severe hypoxemia of non-cardiac origin.

Conflict of Interest

The authors declare that there is no conflict of interest.

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