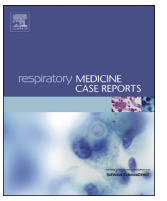
Respiratory Medicine Case Reports 48 (2024) 101996

Contents lists available at ScienceDirect

Respiratory Medicine Case Reports

journal homepage: www.elsevier.com/locate/rmcr



Case Report

Pulmonary siderosis complicated with severe mycoplasma pneumoniae pneumonia: A case report

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ARTICLE INFO

Keywords:
Pulmonary heme-siderosis
Mycoplasma pneumoniae pneumonia
Computed tomography

ABSTRACT

Idiopathic pulmonary hemosiderosis (IPH) is a rare and fatal lung disease. Mycoplasma pneumoniae pneumonia (MPP) is the main community-acquired pneumonia among children aged 5 and above in China. We report the following case of IPH complicated with severe mycoplasma pneumoniae pneumonia(SMPP). An 8-year-old boy with cough and fever was diagnosed with IPH for 3 years and his chest computed tomography showed bilateral bronchopneumonia, lobular consolidation and subpleural interstitial fibrosis. As far as we know, IPH related to SMPP is rarely reported. In the high incidence period of MPP, clinicians and radiologists should be alert to the co-occurrence of IPH and SMPP.

1. Introduction

Idiopathic pulmonary hemosiderosis (IPH) is a rare and fatal pulmonary disease, which is more common in children, and the prognosis is poor [1]. Mycoplasma pneumoniae pneumonia (MPP) is the leading community-acquired pneumonia in children aged 5 years and older in China [2] and is more prevalent during epidemics [3]. Recently, there has been a high incidence of MPP in many parts of China. Since September, the number of children with MPP admitted to many hospitals across the country has soared. However, few cases of IPH combined with MPP have been reported in previous studies. We herein summarize and discuss a case of IPH combined with severe mycoplasma pneumoniae pneumonia (SMPP).

2. Case presentation

An 8-year-old boy had a nonproductive cough and fever with a maximum temperature of 39.0 °C after contact with a classmate 8 days ago, and went to the outpatient department of Chengdu Second People's Hospital for treatment. After symptomatic treatment, he still had a severe cough. The patient returned to our hospital and was admitted due to severe lung lesions revealed by imaging findings： a chest x-ray (CXR) revealed a left lower lung infiltration，a chest computed tomography (CT) scan showed changes in bronchiolitis, especially in the left inferior lobe, with substantial consolidation. At the same time, the corresponding manifestations of IPH can be seen in both lungs. He has a 3-year history of IPH and has been admitted annually for pulmonary infection since he was diagnosed with IPH at another hospital due to the discovery of hemosiderin-containing macrophages in bronchofiber-based alveolar lavage fluid and clinical and radiographic findings, while antinuclear antibodies(ANA) and antinuclear cytoplasmic antibody tests were negative. Physical examination at the time of admission showed that the body temperature was 36.8 °C, the pulse rate of 122/min, the breathing rate of 40/min, the breathing sounds in both lungs were thick, and moderate to fine moist rales could be heard. Af-

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https://doi.org/10.1016/j.rmcr.2024.101996
Received 30 November 2023; Received in revised form 5 February 2024; Accepted 16 February 2024
Available online 17 February 2024

ter admission, the patient tested positive for mycoplasma pneumoniae (MP) nucleic acid. Furthermore, fiberoptic bronchoscope(FOB) was completed. According to the etiological results, the patient was diagnosed as SMPP (resistant to macroliones), and azithromycin was replaced with doxycycline anti-mycoplasma. After treatment, the child's cough has been significantly reduced, and his condition is stable at present.

Other related major auxiliary examinations that patient has carried out since the onset of the disease are as follows. Four days before admission, the laboratory tests revealed mild anaemia. The hemoglobin (HGB) was 111g/L and the hypersensitive C-reactive protein (HSCRP) was 15.05mg/L. White blood cell count and the percentage of neutrophils were normal at 5.79 × 109/L and 60.1%, respectively. The re-examination results did not change significantly on the day of admission. Meanwhile, the serum procalcitonin (PCT) level was 0.108ng/ml. Moreover, the child received bronchoalveolar lavage fluid (BAL), and MP was detected in bronchoalveolar lavage fluid and positive for drug-resistant Mycoplasma genes, which was detected by next generation sequencing (NGS).

3. Discussion

IPH is the cause of a rare diffuse pulmonary hemorrhage, the main pathology of which is repeated alveolar hemorrhage with abnormal accumulation of hemosiderin in the lung interstitial, leading to subsequent pulmonary fibrosis [4,5]. The clinical triad is the triad of respiratory symptoms (i.e.,dyspnea, cough and hemoptysis), anemia, and diffuse pulmonary infiltrates on chest images, but not often simultaneously [6,7]. The golden criteria of diagnosis of IPH is lung biopsy, which is an invasive procedure that is not practiced in children [7]. At present, bronchoscopy technology is widely used in pediatrics, and the examination of alveolar lavage fluid has become the preferred choice for the confirmation of IPH [4]. IPH can be diagnosed with clinical manifestations, such as bleeding, fine particle shadow or ground glass shadow on chest imaging, and hemosiderin cells found in gastric fluid, sputum or bronchoalveolar lavage fluid, except for other factors causing pulmonary bleeding [6,7]. It should be noted that IPH has a variety of clinical manifestations and is often missed and misdiagnosed [1]. Therefore, a detailed and comprehensive medical history is imperative.

The imaging features of IPH are closely related to the amount of pulmonary bleeding and the degree of pulmonary fibrosis, and chest CT is more sensitive than CXR: (1) In acute or subacute stage of pulmonary hemorrhage, chest CT shows active bleeding with focal or diffuse pulmonary infiltration; (2) During the interval of pulmonary hemorrhage, infiltrative changes are gradually absorbed. With the prolongation of the course of the disease, interstitial changes appear, manifested as diffuse or focal reticular opacities, granular opacities, nodule opacities, cystic lesion, pleural thickening, etc [1].

On the chest CT of this patient, we found diffuse ground-glass opacities (GGOs) throughout the lungs (Fig. 1A), which were considered to be caused by pulmonary hemorrhage. At the same time, the multiple subpleural honeycomb-like radiolucent areas in both lungs were also seen (Fig. 1B), considering the pulmonary interstitial fibrosis changes. In addition, in the later stage of IPH development, pulmonary heart disease may be caused by severe pulmonary fibrosis [1]. The chest CT of this child found a slight thickening of the pulmonary artery (Fig. 1C), which requires further investigation of pulmonary hypertension. The patient had recurrent cough, mild anemia, and chest CT showed the above changes. We concluded that after the previous diagnosis of IPH, the patient had recurrent episodes and gradually became chronic, developing subpleural interstitial fibrosis of both lungs. The etiology and pathogenesis of IPH has not been clearly defined and is prone to recurrent attacks [5], so it is also necessary to prevent infection during quiescent period [4]. As in this case, after diagnosis of IPH, the patient was treated with oral prednisone at a dosage of 2 mg/kg/d for 2 weeks, and then treatment was slowly tapered until a daily dose of 10mg is reached, and treatment was maintained for 2 years. The patient has had recurring lung infections for the past three years, in which case SMPP is more likely to occur. Such a low immune status may be related not only to the pre-existing disease in the lungs, but also to the side effects of long-term glucocorticoid therapy.

The main clinical symptoms of MPP are high fever, cough and wheezing [8]. MP culture is the gold standard for the diagnosis of MPP [2], but the conditions of MP culture are harsh and slow growing, which lacks the value of early diagnosis [9]. According to clinical and imaging findings, combined with either or both of the following: (1) Single serum MP antibody titer 1:160; double serum MP antibody titer increased by 4 times or more during the course of the disease; (2) MP nucleic acid test: MP-DNA or RNA positive, MPP can be diagnosed [2]. As for imaging findings, CXR or chest CT at the early stage of MPP mainly showed thickening of lung texture or

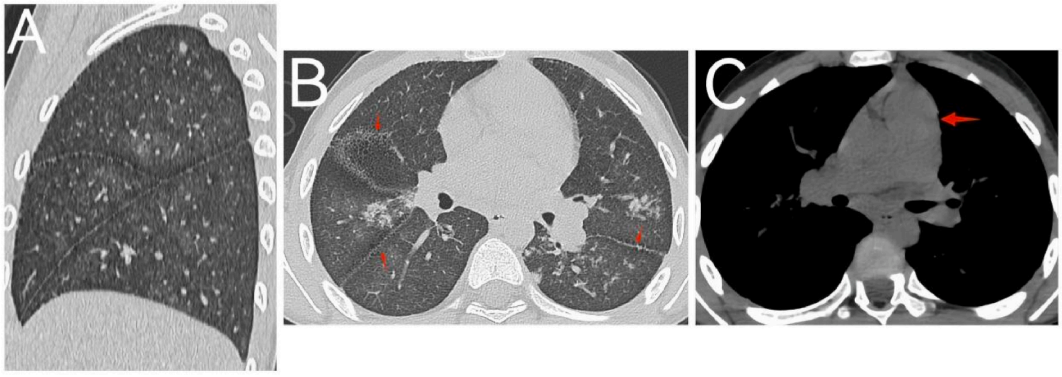

Fig. 1. Imaging findings more closely related to IPH. A: GGOs of both lungs in sagittal position B: Subpleural honeycomb-like radiolucent areas in both lungs(↑) C: Slightly thick pulmonary trunk(↑).

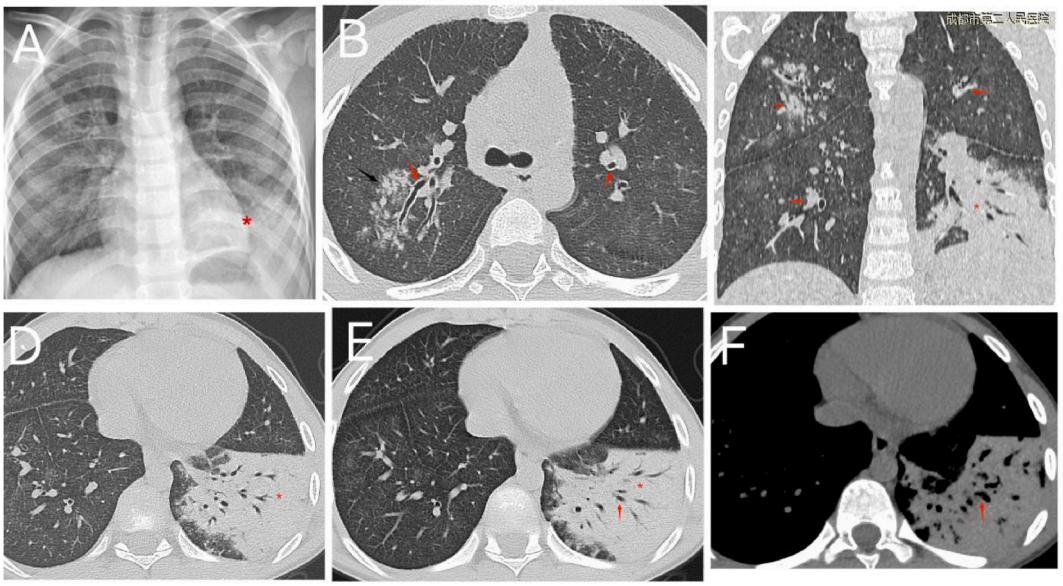

Fig. 2. Chest radiograph and computed tomography findings. A:Patchy area of increased attenuation in the left lower field(*) B:Thickened bronchial walls(↑)and small nodular opacities (↑) C:Small nodular(↑) and patchy(*) opacifications in the coronal position D、E、F: large patchy consolidation(*) with air bronchograms(↑)in the inferior lobe of left lung.

bronchial vessels, thickening of bronchial wall, GGOs, "tree bud sign", thickening of interlobular septum, reticulonodular opacities, etc. However, alveolar inflammatory changes vary according to the range of alveolar involvement, which can have GGOs, patchy, segment and even lobar consolidation. In severe cases, it can be combined with pleural effusion, unilateral lesions are more common than bilateral lesions, and can be accompanied with or without air bronchograms in the lesion [2].

The patient presented with fever, cough, positive mycoplasma DNA, CXR demonstrated patchy area of increased attenuation in the lower field of the left lung (Fig. 2A), which is an infiltration of the left lung. Chest CT also showed increased and blurred bronchial tracts in both lungs, thickening of bronchial wall (Fig. 2B), small nodular opacities (Fig. 2B and C), large patchy consolidation with air bronchograms in the inferior lobe of left lung (Fig. 2D、 E and F), which met the diagnostic basis of MPP. Furthermore, two-thirds of a single lung lobe (left lower lobe) is involved, and more than two lung lobes are involved, which should be considered as severe MPP (SMPP) [2].

There are no clear diagnostic criteria for SMPP, but reference can be made to the "Diagnosis and treatment guideline of community acquired pneumonia in children" (2019 edition) [10]and the "Diagnosis and treatment for Mycoplasma pneumoniae pneumonia in children" (2023 edition) of the National Health Commission [2]. The reported incidence of SMPP has been increasing in China over the past decade [11]. Early assessment of SMPP and timely application of bronchoscopy can effectively alleviate complications and limit sequelae progression [12]. FOB and BAL can rapidly detect pathogens and identify drug-resistance genes, which are conducive to the precise treatment of severe infections [3] and reduce the irrational use of antibiotics [10].

4. Conclusions

IPH is a rare disease with a lack of clinical specificity, and the report suggested that doctors should attach great importance to the careful judgment of the patient's medical history and the follow-up observation of patients with recurrent episodes when diagnosing IPH. During periods of frequent MPP, it is worth noting that IPH patients have MPP and even SMPP. We have presented the clinical journey and potential factors of a pediatric patient with IPH who developed SMPP, highlighting in particular the imaging features of IPH complicated with SMPP. It's strongly recommended that radiologists and clinicians should pay attention to the CT imaging manifestations of IPH in different periods and the diagnostic criteria of MPP and SMPP, which is conducive to timely identification, early and effective treatment, and reduce the occurrence of serious cases and sequelae.

Funding sources

None.

CRediT authorship contribution statement

Zhen Huang: Writing – review & editing, Writing – original draft. **Tao Cheng:** Writing – review & editing. **Guangwen Chen:** Writing – review & editing, Conceptualization.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this manuscript.

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