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Case report

An uncommon cause of pneumonia: The golden diagnosis

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ABSTRACT

A 28-year-old sub-Saharan African Italian non-smoker male presented with signs and symptoms of pneumonia and respiratory failure. Despite antibiotic treatment he experienced a significant worsening of respiratory conditions and admission to intensive care unit. He thus underwent chest computed tomography followed by fiberopict bronchoscopy with bronchoalveolar lavage whose macroscopic examination led to the diagnosis of acute chest syndrome. A brief literature review was conducted to discuss the first manifestation of this disease.

1. Clinical case

A 28-year-old sub-Saharan African Italian non-smoker male presented in late November 2016 to the Emergency Room of the University Hospital of Modena for the gradual onset of shortness of breath, fever and mild chest pain. The past medical history was silent. At the physical examination, bilateral posterior crackles were detected. Arterial blood gas analysis was performed and revealed acute hypoxic respiratory failure. He underwent a chest X-ray showing posterior lower bilateral consolidations (Fig. 1A). He was then admitted to the Respiratory Diseases Unit where antibiotic course with amoxicillin/clavulanate and oxygen therapy were started. During hospital stay he experienced a significant worsening of the hypoxic respiratory failure, thus chest Computed Tomography (CT) scan was performed. It showed bi-basal posterior consolidation with attenuate air bronchogram and mild pericardial effusion (Fig. 1B). He was admitted to the Respiratory Intensive Care Unit (ICU) where he underwent fiberoptic bronchoscopy (FOB) with bronchoalveolar lavage (BAL) whose sample presented with a light yellow appearance (Fig. 1C). The peculiar appearance of the BAL fluid, associated to the patient ethnicity suggested the hypothesis of a pulmonary manifestation of sickle cell disease. We performed a hemoglobin (Hb) electrophoresis test and a heterozygosis condition for β S globin chain with C Hb was reported; thus the diagnosis of sickle cell-Hb SC disease was made and hydroxyurea was started with rapid improvement of respiratory failure and symptoms.

2. Discussion

Acute chest syndrome (ACS) is a major cause of hospitalization and ICU admission in patients with sickle cell disease [1]. It requires clinical awareness because of its potentially life-threatening complications, which can lead to refractory hypoxemia and death [2]. Different pathologic mechanisms affecting sickle cell have been hypothesized as causal factors for the occurrence of ACS. While lung infections were initially recognized as a major cause for the onset of respiratory failure and symptoms in sickle cell patients, further investigations identified parenchymal infarction related to in-situ thromboembolism, hypoventilation due to painful chest wall lesions, intravascular overload and fat embolism syndrome as important factors in determining and precipitating ACS [3]. The presence of a yellowish plasma-like coloration of the BAL fluid, namely known as “golden BAL fluid”, is a pathognomonic sign of ACS. The nature of this particular appearance was commonly attributed to the presence of bilirubin in alveolar spaces due to injury-related increased capillary endothelial permeability [4,5]. More recently Contou et al. analyzed the BAL fluid of 3 patients experiencing ACS showing that the yellowish stain was related to an intense exudative process with increased levels of proteins and lactate dehydrogenase rather than to bilirubin content [6]. What seems peculiar in our case was that ACS appeared to be the first manifestation of the underlying hemoglobinopathy. Moreover it is worth to say that the incidence of ACS is lower for Hb SC genotype

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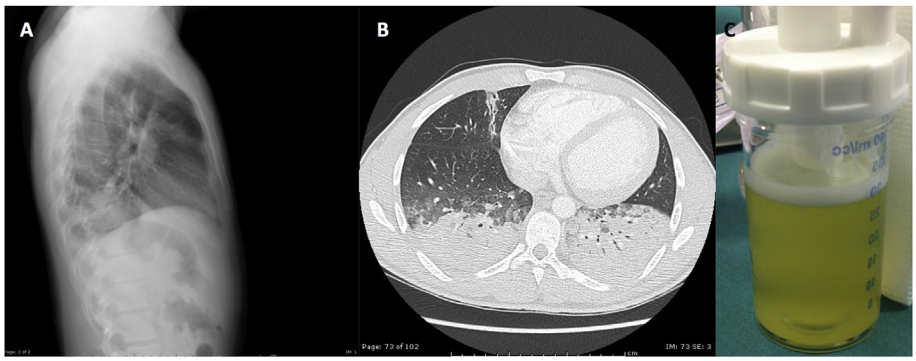

Fig. 1. **A**. Chest X ray in lateral view showing posterior basal consolidations. **B**. Chest CT scan depicting bilateral lung base consolidation. **C**. Light yellow stain bronchoalveolar lavage fluid.

as compared to Hb SS or Hb S ß^0-thalassemia [5,7]. In conclusion we would like to stress the attention on an uncommon cause of pneumonia, whose bronchoalveolar manifestation could be of paramount importance for its correct diagnosis.

Compliance with ethical standards

Funding

None.

Conflict of interest

Roberto Tonelli declares that he has no conflict of interest, Laura Fabbri declares that she has no conflict of interest, Alessandro Andreani declares that she has no conflict of interest, Ivana Castaniere declares that she has no conflict of interest, Riccardo Fantini declares that he has no conflict of interest, Alessandro Marchioni declares that he has no conflict of interest, Enrico M Clini declares that he has no conflict of interest.

Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent

Informed consent was obtained from all individual participants included in the study.

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