PEDIATRICS INTERNATIONAL Official Journal of the Japan Pediatric Society

Pediatrics International (2022) **64**, e14991 doi: 10.1111/ped.14991

Photos In Pediatrics

Idiopathic pulmonary hemosiderosis: A mimic of severe COVID-19 pneumonia

Tomohiro Hirade,[1] Aisa Moriyama,[2] Keisuke Wada,[2] Shigeki Nakashima[2] and Takeshi Taketani[2]

Department of Pediatrics [1]Shimane Prefectural Central Hospital, [2]Faculty of Medicine, Shimane University, Shimane, Japan

A 2-year-old boy was admitted to our hospital with lethargy. He had been treated for iron-deficiency anemia and failure to thrive 1 year earlier. On examination, he appeared pale and had tachypnea and tachycardia, without abnormal breath or heart sounds. His temperature was 36.7°C, pulse rate 180 beats per minute, respiratory rate 56 breaths per minute, oxygen saturation 76%, and blood pressure was 101/58 mm Hg. Despite the low oxygen saturation level, he could speak and walk independently. Laboratory tests revealed a white blood cell count of 13.7 × 10^9/L, a hemoglobin concentration of 8.9 g/dL, and a C-reactive protein level of 15.3 mg/L. The chest X-ray showed bilateral diffuse pulmonary infiltrates (Fig. 1). A computed tomography (CT) scan of the chest revealed diffuse alveolar infiltrates with ground-glass opacities (GGOs) (Fig. 2). Considering the increased number of patients with coronavirus disease 2019 (COVID-19) in our area, our provisional diagnosis was COVID-19 pneumonia. However, the reverse transcription-polymerase chain reaction testing for severe acute respiratory syndrome coronavirus 2 was negative. The patient was intubated following a gradual worsening of the respiratory state after admission. Despite no history of hemoptysis, bloody sputum was aspirated from the tracheal tube. Sputum cytology with Prussian blue iron staining revealed hemosiderin-laden macrophages, called siderophages (Fig. 3). Following the exclusion of other pulmonary hemorrhagic diseases, the patient was diagnosed with idiopathic pulmonary hemosiderosis (IPH). He underwent two courses of pulse steroid therapy and was discharged after one-and-a-half months.

Idiopathic pulmonary hemosiderosis is a rare, recurrent diffuse alveolar hemorrhagic disease that occurs primarily in children. The estimated prevalence is 0.24–1.23 cases per million.[1] The etiology of IPH remains unknown. The classic triad consists of pulmonary infiltrates, iron-deficiency anemia, and hemoptysis. However, young children can hardly expectorate sputum. Therefore, hemoptysis does not always present.[2]

Correspondence: Tomohiro Hirade, MD, PhD, Department of Pediatrics, Shimane Prefectural Central Hospital, 4-1-1, Himebara, Izumoshi, Shimane 693-8555, Japan.
Email: hirade5525@gmail.com

Received 30 July 2021; revised 2 September 2021; accepted 13 September 2021.

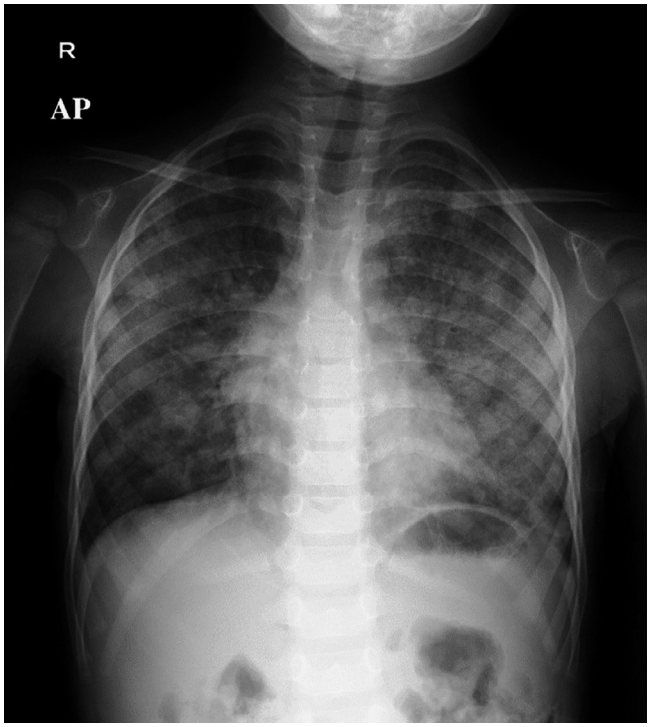

Fig. 1 Chest radiograph showing bilateral diffuse pulmonary infiltrates.

The chest X-ray findings of IPH are non-specific. CT scan exhibits diffuse alveolar infiltrates with GGOs.[1–3] Interlobular septal thickening and intralobular lines superimposed on GGOs, known as the "crazy-paving" pattern, may be observed. The opacities on the CT scan usually decrease after several days, and fibrotic changes appear, reflecting recurrent episodes of alveolar hemorrhage.[1,3] CT findings in COVID-19 pneumonia typically demonstrate bilateral, peripheral, multifocal, and lower lobe predominant GGOs, with or without consolidation or the "crazy paving" pattern.[4,5] Therefore, imaging findings during an acute episode of IPH are similar to those of COVID-19 pneumonia, and the distinction between these two diseases is difficult without clinical history.[5]

The early diagnosis of IPH is important because delay in treatment causes progression to pulmonary fibrosis. However, the rarity and variety of non-specific symptoms often delay

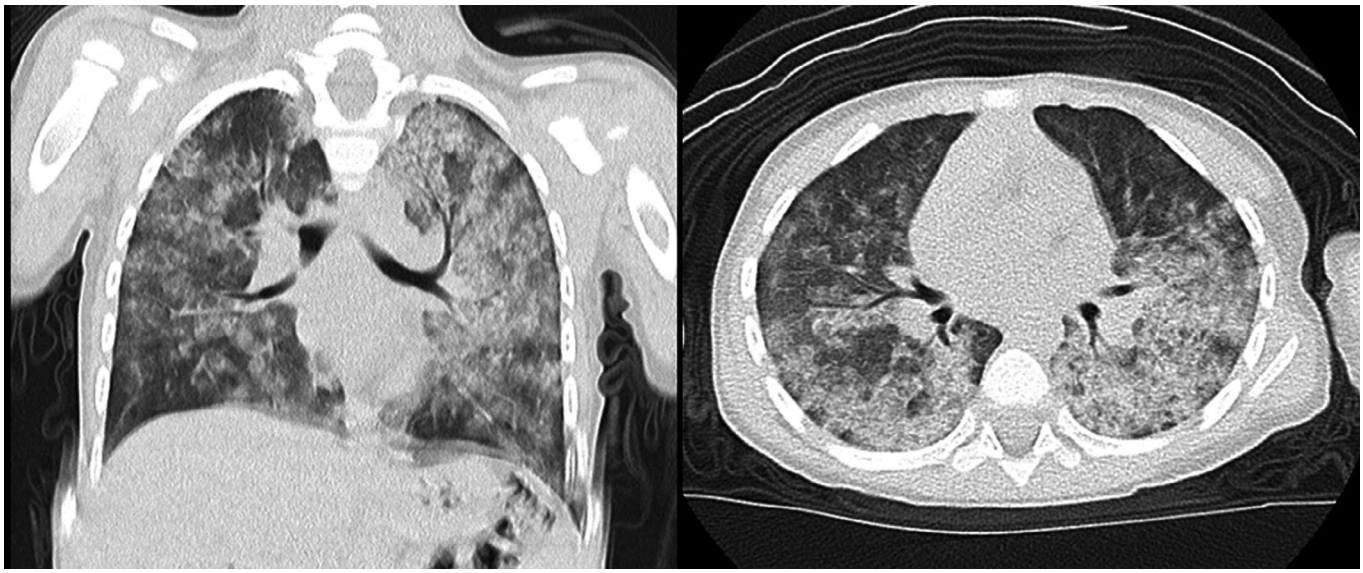

Fig. 2 Computed tomography scan of the chest showing diffuse alveolar infiltrates with ground-glass opacities.

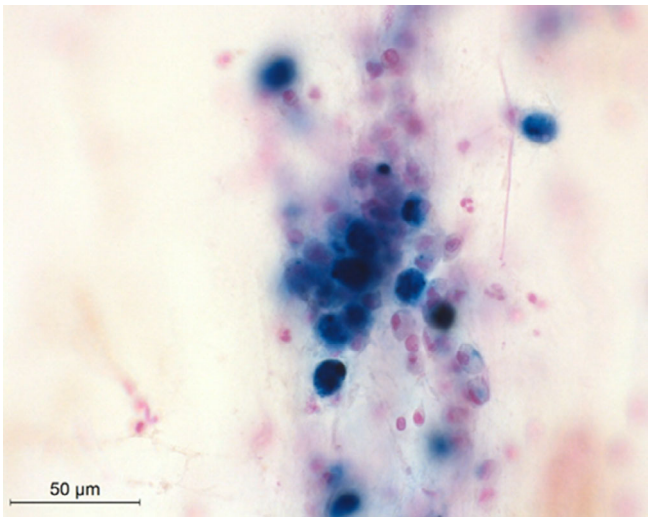

Fig. 3 Sputum cytology with Prussian blue iron staining showing hemosiderin-laden macrophages (400× magnification).

the diagnosis. Refractory anemia and failure to thrive are suggestive of IPH.[6] Other than conducting a lung biopsy to diagnose IPH, identification of siderophages in the sputum, bronchoalveolar lavage, or gastric aspirate is helpful for diagnosis if other pulmonary hemorrhagic diseases are excluded.[6] Systemic corticosteroid treatment improves IPH prognosis.[7]

Acknowledgments

We thank Dr Hiroshi Watanabe, Watanabe Children Women Clinic, Shimane, Japan, for taking care of the patient.

Disclosure

The authors declare no conflict of interest.

Author contributions

T.H. is the main author and wrote the manuscript; A.M., K.W., and S.N. were responsible for diagnosing and treating the patient; T.T. critically reviewed the manuscript and supervised the study process. All authors read and approved the final manuscript.

Ethics and consent statement

This report was reviewed and approved by the appropriate ethics committee. Written informed consent was obtained from the parents of the patient for publication of this report.

References

1 Saha BK. Idiopathic pulmonary hemosiderosis: A state of the art review. *Respir. Med.* 2021; **176**: 106234.
2 Taytard J, Nathan N, Blic J *et al*. New insights into pediatric idiopathic pulmonary hemosiderosis: The French RespiRare® cohort. *Orphanet. J. Rare Dis.* 2013; **8**: 161.
3 Reisman S, Chung M, Bernheim A. A review of clinical and imaging features of diffuse pulmonary hemorrhage. *Am. J. Roentgenol.* 2021; **216**(6): 1500–9.
4 Shelmerdine SC, Lovrenski J, Caro-Domínguez P, Toso S. Coronavirus disease 2019 (COVID-19) in children: A systematic review of imaging findings. *Pediatr. Radiol.* 2020; **50**(9): 1217–30.
5 Parekh M, Donuru A, Balasubramanya R, Kapur S. Review of the chest CT differential diagnosis of ground-glass opacities in the covid era. *Radiology* 2020; **297**: E289–302.
6 Nevin MA. Pulmonary hemosiderosis. In: Kliegman RM, St Geme JW, Blum NJ (eds). *Nelson Textbook of Pediatrics*, 21st edn. Elsevier, Philadelphia, PA, 2020; 2306–8.
7 Doi T, Ohga S, Ishimura M *et al*. Long-term liposteroid therapy for idiopathic pulmonary hemosiderosis. *Eur. J. Pediatr.* 2013; **172**(11): 1475–81.