



A case of *Strongyloides* hyperinfection syndrome with elevated IgG4

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ARTICLE INFO

Keywords:

IgG4
Respiratory failure
HTLV-1

Case illustrated

A 73-year-old woman was referred to our department for acute respiratory failure. She had a 10-year history of chronic abdominal pain, and immunoglobulin G4 (IgG4)-related gastrointestinal disease had been diagnosed through colonoscopy 4 months earlier. She had been treated with 30 mg/day of prednisolone (PSL) 2 months; however, respiratory failure developed, and she was referred to our department. On physical examination, she had a temperature of 38.1 °C, heart rate of 115 beats/min, blood pressure of 86/49 mmHg, respiratory rate of 24 breaths/min, and oxygen saturation of 88 % on 2 L/min nasal O₂. Tenderness was observed in the left lower abdomen. Laboratory tests showed a white blood cell count of 9600 cells/μL with 87 % neutrophils, a hemoglobin level of 7.2 g/dL, and C-reactive protein level of 4.74 mg/dL. The patient's IgG4 levels increased from 495 mg/dL to 675 mg/dL (normal range, 11–121 mg/dL) before hospitalization. Chest computed tomography (CT) revealed bilateral ground-glass opacity, interlobular septal thickening, and multiple random granular shadows (Fig. 1). Bronchoscopy was performed, and *Strongyloides* larvae were detected in the BALF smears and cytology (Fig. 2). The larvae were also found in the stool, and stool culture showed colony formation in which the larvae

had migrated on bromothymol blue lactate agar (Fig. 3). Thus, *Strongyloides* hyperinfection syndrome was diagnosed. The patient developed septic shock due to strongyloidiasis and secondary bacterial infections. She was treated with oral ivermectin 200 μg/kg daily and 6 g/day meropenem for strongyloidiasis and bacterial infection, respectively. Ivermectin was administered for 3 weeks until the *Strongyloides* larvae disappeared from her stool. The patient's IgG4 levels rapidly decreased to 200 mg/dL after treatment of strongyloidiasis.

Strongyloides larvae migrate from the capillary bed into the alveoli, resulting in hemorrhage in the alveolar space and interstitium [1]. Random granular shadows occur when large numbers of pathogens are disseminated [2]. In this case, disseminated larvae may have affected the pulmonary circulation.

She tested positive for human T-lymphotropic virus 1 (HTLV-1) antibody. HTLV-1 co-infection is associated with shifting of the immune response from Th2 to Th1, attenuating the ability of the immune system to control *Strongyloides* larvae [3]. In chronic infection, interleukin-10 produced by Tregs promotes IgG4 production to suppress the reaction to larvae [4]. This causes immune tolerance toward the larvae and persistent infection of the host.

Parasitic diseases should be considered in cases of IgG4-related

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<https://doi.org/10.1016/j.idcr.2023.e01739>

Received 13 February 2023; Accepted 5 March 2023

Available online 6 March 2023

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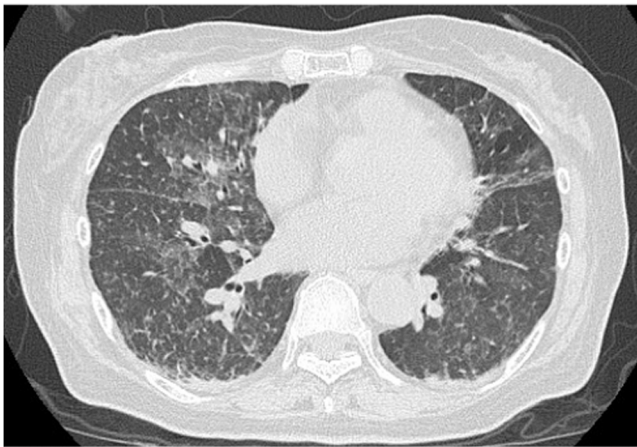


Fig. 1. Chest computed tomography (CT). Chest CT shows bilateral ground-glass opacity, interlobular septal thickening, and multiple random granular shadows.



Fig. 2. Bronchoalveolar lavage fluid cytology. Bronchoalveolar lavage fluid cytology shows *Strongyloides* larvae (magnification, $\times 400$; papanicolaou stain).

disease refractory to corticosteroid treatment or with unexplained IgG4 elevation.

CRediT authorship contribution statement

Ryota Takao: Data curation, Investigation, Resources, Visualization, Writing – original draft. **Yuya Ito:** Data curation, Investigation, Resources, Visualization, Writing – review & editing. **Yasuhiro Tanaka:** Resources, Visualization, Writing – review & editing. **Nobuyuki Ashizawa:** Investigation, Resources, Writing – review & editing. **Kazuaki Takeda:** Resources, Writing – review & editing. **Shotaro Ide:** Resources, Writing – review & editing. **Naoki Iwanaga:** Resources, Writing – review & editing. **Masato Tashiro:** Resources, Writing – review & editing. **Takahiro Takazono:** Conceptualization, Methodology, Project administration, Writing – review & editing. **Takeshi Tanaka:** Writing – review & editing. **Motohiro Sekino:** Investigation, Resources, Writing – review & editing. **Akitsugu Furumoto:** Writing – review & editing. **Shinji Okano:** Resources, Visualization, Writing – review & editing. **Tetsuya Hara:** Writing – review & editing. **Koichi Izumikawa:** Writing – review & editing. **Katsunori Yanagihara:** Resources, Visualization, Writing – review & editing. **Hiroshi Mukae:** Supervision, Writing – review & editing.

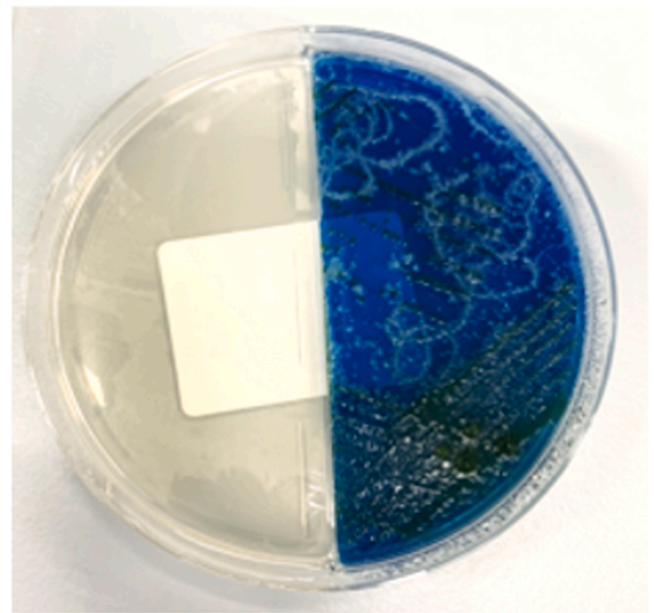


Fig. 3. Stool culture on bromothymol blue lactate agar. *Strongyloides* larvae migration accompanied by enteric bacteria, forming colonies in a linear pattern.

Funding

This case illustrated did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethical approval

N/a.

Consent

Written informed consent was obtained from participants for publication of the details of their medical case and any accompanying images.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Data Availability Statement

Not applicable

Acknowledgments

None.

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