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CASE REPORT

# Pulmonary Lophomoniasis in a pediatric patient with primary immunodeficiency: a case report

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Lung diseases,
Parasitic.

## Abstract

**Introduction:** Protozoosis are rare infections, strongly associated with imunodeficiency and lung problems. **Objective:** To describe a pediatric case of Lophomoniasis, a rare pulmonary disease caused by a protozoan, emphasizing its clinical manifestations and the importance of diagnostic suspicion and appropriate therapy. **Case report:** Male pacient, inicially with tachydyspnea at 2 months old, presenting respiratory distress and worsening of the respiratory pattern at 6 months old, requiring oxygen therapy. In the initial evaluation, chest X-ray and computed tomography revealed a diffuse lung disease, with bilateral interstitial pattern. Subsequent investigation revealed the presence of the protozoan Lophomonas spp. in bronchoalveolar lavage, leading to a diagnosis of Pulmonary Lophomoniasis, associated with combined primary immunodeficiency. **Discussion:** Pulmonary lophomoniasis is a disease caused by the multiflagellated protozoan Lophomonas spp., an endocommensal parasite that inhabits the intestines of cockroaches and termites. Antibiotic therapy is the key treatment. Knowledge of this disease and its manifestations is crucial for accurate diagnosis and appropriate treatment, minimizing therapeutic errors and potential complications for the patient. There are few reports in the literature of similar cases, but there has been a significant increase in cases in recent years.

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## INTRODUCTION

Although rare, protozoal infections are strongly associated with immunodeficiencies and lung diseases. Most cases of these infections have been predominantly reported in adults in Asia[1-4]. In recent years, the number of protozoan infections in the general population has increased; this rise can be attributed to the growing number of individuals with compromised immune systems linked to poor hygiene conditions, particularly in developing countries.[1,2]

Lophomonas spp. is a multiflagellate endocommensal parasite that inhabits the guts of cockroaches and termites[1]. Infection by this protozoan primarily affects the respiratory tract, although it can also be found in other systems[1,5]. Patients present with nonspecific signs and symptoms such as cough, fever, and respiratory distress; approximately one-third show eosinophilia[2,5,6]. Infected cockroaches or termites in domestic dwellings increase the risk of inhaling the protozoan and developing a lung infection[2].

Patients undergo various treatments and complementary exams before being diagnosed with pulmonary lophomoniasis via bronchoscopy and bronchoalveolar lavage [4-6]. After diagnostic confirmation, treatment is administered with metronidazole, and reports suggest an improvement in symptoms following the completion of treatment[2].

## CASE REPORT

A 7-month-old male patient was born after a full-term pregnancy in São José, located in the metropolitan area of Florianópolis, SC. The newborn did not require oxygen therapy at birth, and no neonatal complications were reported. At two months of age, he developed tachypnea with no other associated symptoms. He was taken to the emergency room at six months as his condition worsened, leading to respiratory distress. During the emergency assessment, the patient exhibited tachydyspnea and was afebrile, requiring oxygen therapy due to desaturation. An imaging exam was performed, and a chest X-ray showed a bilateral diffuse interstitial infiltrate pattern. On physical examination, the patient appeared in good general condition, afebrile, with only mild tachydyspnea and no changes noted on pulmonary auscultation.

During hospitalization, further investigations were conducted through laboratory and imaging tests, which revealed the following: a nasal swab viral panel positive for rhinovirus; a blood count indicating anemia with a hemoglobin level of 10.6 and a hematocrit of 33; leukocytes at 9,970 with eosinophilia (658 total) and C-reactive protein within the reference range; a transthoracic echocardiogram demonstrated a patent ductus arteriosus without hemodynamic repercussions; and a chest computed tomography (CT) scan showed a pattern of diffuse lung disease, with significant bilateral interstitial involvement (Figure 1). Considering the clinical signs and symptoms along with the images suggestive of interstitial lung disease, the investigation was expanded. Changes were noted in cellular (CD19 and CD8 < p10) and humoral immunity (IgG and IgA < p3 and IgM > p 97). HIV infection was ruled out.

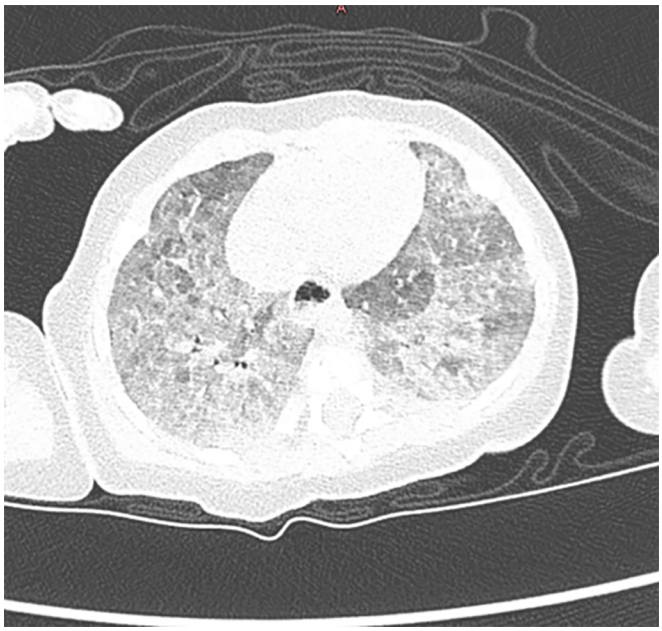


**Figure 1.** Chest CT: A chest CT scan showing a bilateral interstitial involvement pattern, with extensive ground-glass attenuation and smooth septal thickening, diffusely present in both lungs, forming a crazy-paving pattern.

Initially, the patient required oxygen therapy through a high-flow nasal cannula, which was later switched to a simple nasal cannula. Weaning proved to be challenging. During his hospitalization, he received azithromycin (10 mg/kg/day) for five days and intravenous human immunoglobulin as directed by the immunology team, with infusions every three weeks due to the initial suspicion of humoral immunodeficiency.

In the following days, an expanded investigation was conducted using bronchoscopy and bronchoalveolar lavage, initially identifying a protozoan later confirmed as the parasite Lophomonas spp (Figure 2). Bacterioscopy and culture for typical germs, PCR and AARB for Mycobacterium tuberculosis, and mycological examination and culture for fungi returned negative results. The team discussed the case and initiated treatment with intravenous metronidazole at a dose of 40 mg/kg/day for 10 days. A CT scan was ordered after ten days of metronidazole treatment.

The patient's tachydyspnea improved slowly and gradually since he started intravenous metronidazole. He was progressively weaned off oxygen therapy until he could tolerate room air. After 13 days of antimicrobial treatment, the patient had only partially improved and still exhibited mild

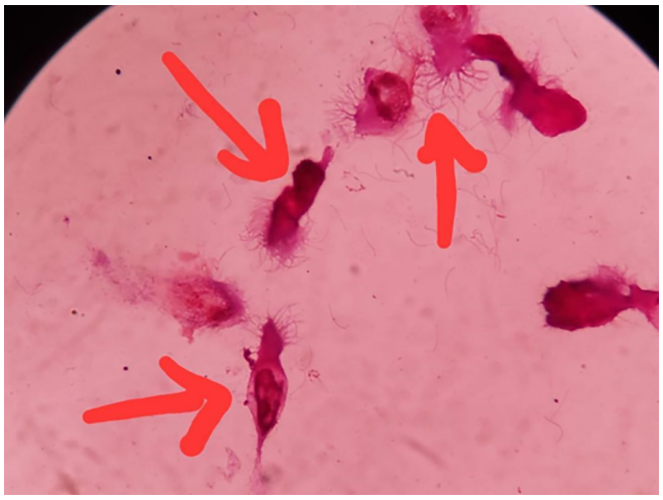


**Figure 2.** Microscope image. Mobile structures of ciliated protozoa observed on a slide under direct microscopic visualization.

tachydyspnea; a chest CT scan showed the same interstitial pattern. The team decided to continue the metronidazole treatment for 21 days and perform a lung biopsy to investigate further, which revealed changes in the lung parenchyma, suggesting a chronic inflammatory process with a predominantly lymphocytic interstitial pattern and nonspecific immunohistochemical markers. It was not possible to rule out associated infectious etiological agents. During hospitalization, whole exome sequencing was also conducted, confirming the diagnosis of primary combined immunodeficiency (CD40 ligand deficiency or X-linked Hyper IgM Syndrome) and excluding other genetic diseases that could cause tachydyspnea and diffuse interstitial lung disease in infants. After receiving the results, prophylaxis with trimethoprim-sulfamethoxazole was initiated, and follow-up care was arranged with the pediatric immunology and pulmonology team.

## DISCUSSION

A search of the main databases (PubMed, SciElo, Google Scholar) uncovered a lack of articles on pulmonary lophomoniasis and only a few case reports, totaling 35 published works globally, with seven involving children. The majority of adult cases were reported in China and Iran, whereas pediatric cases of the disease were noted in Peru, Mexico, and Afghanistan[4-6].

There are approximately 300 cases documented worldwide, and according to research, 73 of these cases involved children. In Brazil, only one case was described in an adult, linked to a COVID-19 infection and tuberculosis, with no cases reported in the pediatric population[3]. Most of the identified cases were in male patients, as indicated in this report[1-8]. The literature emphasizes that the parasite is found in the guts of cockroaches and termites; however, the patient described had no reports of such insects in their home or in environments they frequented.

The clinical signs and symptoms of pulmonary lophomoniasis are nonspecific. Ding Q and Shen K evaluated 53 infected children and demonstrated that the main symptom was cough (74.4%), followed by dyspnea (18.9%); nearly 50% of the patients had fever[1]. Our patient exhibited tachydyspnea and a leukogram indicating eosinophilia as the primary manifestation. He did not develop a fever. It is worth noting that the association with immunodeficiency may explain our patient's absence of fever[7].

Upon arriving at the emergency room, the patient needed oxygen therapy due to desaturation and respiratory distress. In the literature, few reports exist of children requiring supplemental oxygen during the first days of hospitalization. Our patient displayed more pronounced symptoms, possibly due to co-infection with rhinovirus and a later-diagnosed immunodeficiency.

Most patients with lophomoniasis exhibit alterations in imaging exams, including bilaterally scattered nodular or linear opacities that may be migratory, ground-glass opacities, and irregular consolidations[1,7]. Our patient's X-ray images displayed a bilateral diffuse interstitial infiltrate pattern. A subsequent chest CT scan revealed a pattern of diffuse lung disease with bilateral interstitial involvement with ground-glass opacities and septal thickening, consistent with findings reported in the literature. Xue et al. analyzed the imaging patterns of 67 chest CT scans from 131 cases of pulmonary lophomoniasis and found that ground-glass opacities were the second most frequent finding (32.8%)[7]. Another study showed involvement of the interstitium and/or ground-glass opacities in seven out of 50 children (14%) with pulmonary infections caused by Lophomonas[1].

Bronchoscopy and bronchoalveolar lavage are crucial for confirming the presence of protozoa in the respiratory tract[1,5,9,10]. Diagnosis relies on directly observing the mobile structures of ciliated protozoa under a microscope, as demonstrated in our case (Figure 2). One limitation of this article is the inability to perform metagenomic detection due to the small quantity of parasite material for DNA extraction.

The standard treatment for pulmonary myeloma is metronidazole, administered at 30 mg/kg/day for 3-4 weeks in the pediatric population. The medication was initially given intravenously and later transitioned to oral administration for home treatment[4,7]. Our patient received intravenous metronidazole for 21 days, resulting in a partial improvement in symptoms. He was gradually weaned off oxygen therapy until he could tolerate room air. He was discharged a few days after the discontinuation of metronidazole and was scheduled for outpatient follow-up with a multidisciplinary team.

According to the analyzed studies, immunodeficiencies (both primary and secondary) are the most common comorbid conditions associated with Lophomonas infection[9]. Our patient was diagnosed with Hyper IgM syndrome, a primary combined immunodeficiency that impairs the function of T and B cells, allowing for opportunistic infections, primarily in the airways

[11,12]. It is important to emphasize that pulmonary lophomoniasis can occur in patients without associated comorbidities[3]. A Chinese study found that 49% of the disease cases affected otherwise healthy children[1].

It is essential to remain vigilant about opportunistic infections caused by protozoa, such as Lophomonas species, in patients with pulmonary infections that are unresponsive to standard treatments. This is particularly important in cases suspected of an underlying immunosuppressive disease and in areas with inadequate hygiene and sanitation conditions.[3,5] Bronchoscopy and bronchoalveolar lavage should be performed as early as possible in such cases.

The treatment for lophomoniasis is highly effective, and early diagnosis is crucial for initiating appropriate treatment and enhancing outcomes. As it is a rare condition with limited reports in the literature, further studies are necessary to understand the disease's progression better, raise awareness, and refine diagnostic methods to facilitate early treatment and monitoring.

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