progressive dyspnea. He had lost 8 kg weight in 2 months, but had no other complaint. His oxygen saturation in room air was 96%. Laboratory findings were normal. Chest CT revealed numerous bilateral small nodules, with some coalescence in the posterior and lower lung regions (Fig. 1A, B). On day 8, he developed a maculopapular (purpuric) rash (Fig. 1C). Bronchoalveolar lavage (BAL) performed during flexible fiberoptic bronchoscopy revealed the presence of *S. stercoralis* larvae (Fig. 1D). BAL findings for bacteria, fungus, and acid-fast bacteria were negative. Blood cultures were also negative. A stool sample demonstrated numerous larvae, as well as a few adult organisms. The patient was negative for human immunodeficiency virus (HIV), but positive for human T-cell lymphotropic virus (HTLV) 1/II quantiative antibodies. A diagnosis of Strongyloides hyperinfection syndrome was made, and treatment with ivermectin and albendazole was started.

Strongyloidiasis, an infection caused by the nematode *S. stercoralis*, is prevalent in tropical and subtropical countries. In the setting of severe immunosuppression, the worm may disseminate, causing severe life-threatening syndromes such as hyperinfection and dissemination, showing massive infection. These syndromes are associated with significant morbidity and mortality. The clinical diagnosis is often delayed because the clinical and radiographic findings are nonspecific. The most important risk factor for the development of Strongyloides infection is residence in or visit to an endemic area. *S. stercoralis* hyperinfection is generally fatal, as it is normally associated with immunosuppression, either iatrogenic (e.g., caused by systemic corticosteroid use) or to underlying illness (e.g., HIV infection, HTLV-1 carriage, or organ transplantation).

Hyperinfection syndromes manifest clinically in a nonspecific manner, with gastrointestinal and pulmonary symptoms being the most common findings. The pathognomonic rash of Strongyloides infection is a serpiginous and urticarial petechial purpuric eruption over the abdomen and proximal thigh. Pulmonary symptoms and signs consistent with adult respiratory distress syndrome or intra-alveolar hemorrhage may be seen. Radiographic changes may include nodular, reticular, and airspace opacities, with distribution ranging from multifocal to lobar. Blood eosinophilia is seen in more than 75% of patients with chronic Strongyloides infection, but may be absent in immunocompromised patients with hyperinfection syndrome. Detection of a large number of larvae in stool and/or BAL fluid or sputum is hallmark of hyperinfection. Therefore, the diagnosis rests mainly on the recognition of the organism’s morphology in pathology specimens. In conclusion, in endemic areas, Strongyloides hyperinfection should be included in the differential diagnosis of pulmonary miliary lesions.

References


Bruno Hochegger,a Gláucia Zanetti,b Edson Marchiori,b,c

a Irmandade Santa Casa de Misericórdia de Porto Alegre, Rio Grande do Sul, Brazil
b Department of Radiology, Federal University of Rio de Janeiro, Rio de Janeiro, Brazil
c Corresponding author.
E-mail address: edmarchiori@gmail.com (E. Marchiori).

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Pulmonary Disease Caused by Mycobacterium szulagai

*Enfermedad pulmonar producida por Mycobacterium szulagai*

To the Editor,

*Mycobacterium szulagai* is slow-growing, rarely isolated environmental non-tuberculous mycobacterium (NTM). It accounted for less than 0.2% of isolated strains in a study of over 36,000 NTM samples from 14 countries, including Spain. Like other NTM, it can be present in dust, soil, water, plants, and animals. Isolation from the respiratory tree does not always imply disease, so the American Thoracic Society and the Infectious Diseases Society of America have produced a series of diagnostic criteria in an attempt to establish the pathogenic role of this and other NTM when isolated from biological samples.

We report the case of a 49-year-old woman, employed as a cleaner, smoker of 35 pack-years, chronic alcoholic with moderate COPD, who was transferred by ambulance to the emergency department in coma (Glasgow scale 3), shock (BP 50/30 mmHg) and respiratory failure (SO2 75% with FiO2 0.21), where she was intubated and mechanical ventilation was applied, with administration of vasoactive amines and admission to the ICU. Two months previously she had reported general malaise, 10 kg weight loss, and in the last week, fever of up to 39°C, cough and mucopurulent expectoration.

Examination showed that she was severely underweight (BMI 14.2), with global loss of breath sounds in both pulmonary fields. Analytical parameters on admission were 16,200 leukocytes/mm3 (81% neutrophils, 9% lymphocytes, and 10% monocytes), ESR 11 mm/h, blood glucose 174 mg/dl, AP 133 IU/L, gamma GT 231 IU/L, LDH 392 IU/L, and high-sensitivity C-reactive protein 9 mg/dl. Other parameters – complete blood count, hemostasis and biochemistry – were normal, including procalcitonin and immunoglobulins. Urine sample was negative for *Streptococcus pneumoniae* antigen and positive for *Legionella pneumophila* serogroup 1 antigen, so the initial empirical treatment with ceftriaxone and azithromycin was switched to levofloxacin.

Chest radiograph revealed fibrocavitary tracts in both upper fields with signs of air trapping, confirmed on CT (Fig. 1). No particular findings were revealed in the head or the abdomen. Fiberoptic bronchoscopy was performed, showing signs of inflammation of the bronchial mucosa, and bronchial aspirates were obtained for standard and mycobacterial culture. Standard culture techniques showed growth of saprophytic flora, with no *Legionella* spp. or fungal strains. No alcohol-acid resistant bacilli (AARB) were observed on Ziehl-Neelsen staining. A serology study for HIV and IgM antibodies against *Mycoplasma pneumoniae*, *Coxiella burnetti*, *Chlamydia pneumoniae*, *Legionella pneumoniae* and

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anti-Chlamydia psittaci indirect immunofluorescence were negative. Blood cultures and Mantoux were also negative.

One week after admission to the ICU, bronchoscopy was repeated, with similar findings and negative PCR for Mycobacterium tuberculosis (GeneXpert® , Cepheid). After extubation, a Stage IVA epidermoid carcinoma at the base of the tongue was diagnosed. Three weeks later, AARB growth was observed in 2 bronchial aspirates obtained on bronchoscopy. Solid phase hybridization was performed using GenoType® Mycobacterium CM/AS (Hain Life-science, Germany), but the strain could not be identified, so it was sent to the Mycobacteria Genetic Group of the Universidad de Zaragoza, where it was identified as Mycobacterium szulgai (M. szulgai). Treatment began with rifampicin 600 mg, ethambutol 600 mg, and azithromycin 250 mg/day, and continued for 10 months until she died due to tumor progression. Initially, despite treatment for her cancer with chemotherapy and radiation therapy, the patient’s clinical and radiation therapy progress was good, and no M. szulgai growth was encountered in follow-up sputums.

According to the ATS/IDSA criteria, isolation of M. szulgai is associated with real lung disease to a greater extent than other NTM.\(^5\)\(^-\)\(^6\) Although it has been described in immunosuppressive states and with the administration of drugs, cancer, and HIV infection, this association is considerably rarer than with other NTM.\(^7\)-\(^13\) It is highly unusual for a diagnosis of M. szulgai to be the first manifestation of an underlying tumor, but the patient presented other risk factors for developing the disease, such as chronic alcoholism, smoking, and COPD.

It is more common in men (>85% of patients reported), and can appear at any age, although most individuals are, on average, around 50–60 years old. In over 2 thirds of patients, disease is limited to the respiratory system, and the radiological and clinical picture is indistinguishable from that of M. tuberculosis and other NTM. Extrapulmonary disease and disseminated disease have also been described in immunocompromised patients.

In line with the ATS/IDSA guidelines, Philley and Griffith recommend a combination of at least 3 oral drugs: ethambutol 15 mg/kg/day, rifampicin 600 mg/day or rifabutin 150–300 mg/day, and azithromycin 250 mg/day or clarithromycin 500 mg/12 h, or moxifloxacin 400 mg/day, for at least 12 months after a negative culture: most patients respond well.\(^1\)\(^,\)\(^1\)\(^4\)

The patient met ATS/IDSA criteria for pulmonary disease due to M. szulgai, since she had consistent radiological lesions on Rx and CT, M. szulgai was isolated in 2 samples of bronchial aspirate obtained 1 week apart, and the presence of other mycobacteria, other pathogens, and lung cancer were ruled out. Clinical and radiological progress of the infection with rifampicin, ethambutol, and azithromycin treatment was good, even though the patient later died due to progression of the tumor at the base of the tongue.

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References


Ana Milagro Beamonte,a Elena Briz Muñoz,b Luis Torres Sopena,a,* Luis Borderías Claub

a Servicio de Microbiología, Hospital San Jorge, Huesca, Spain
b Servicio de Neumología, Hospital San Jorge, Huesca, Spain

* Corresponding author.
E-mail address: lt@torres@salud.aragon.es (L. Torres Sopena).

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