



## Case Report

# Bacterial Meningitis and *Strongyloides* Hyperinfection Syndrome in an Immunocompetent Adult: A Case-Based Review

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## Abstract

*Strongyloides* hyperinfection syndrome (SHS) is caused by an accelerated autoinfection cycle of the parasite, frequently observed in immunocompromised hosts. However, it has rarely been described in immunocompetent patients. Penetration of intestinal mucosa by filariform larvae facilitates the transport of enteric organisms into the bloodstream, leading to extraintestinal bacterial infection such as meningitis. We present a case of a 79-year-old Bolivian immunocompetent male with SHS and meningitis due to *Lactobacillus rhamnosus*, and a literature review of the few cases reported of bacterial meningitis associated with SHS in immunocompetent patients. To our knowledge, this is the first case of *Lactobacillus* meningitis in association with SHS described in the literature.

**Keywords:** *Lactobacillus* meningitis; *Strongyloides* hyperinfection; *Strongyloides stercoralis*; Strongyloidiasis.

**Abbreviations:** CT: Computerized tomography; CSF: Cerebrospinal fluid; PCR: Polymerase Chain Reaction; b.i.d: bis in die - twice a day; HIV: Human Immunodeficiency Virus; HTLV-1: human T-cell lymphotropic virus type 1; SHS: *Strongyloides* hyperinfection syndrome

## Case Report

A 79-year-old Bolivian male with diabetes (glycated hemoglobin 7.5%) was admitted to the emergency department because of cephalgia and confusion for two days. Three years ago, he had been diagnosed with interstitial lung disease with no evident cause (Figure 1). The studies performed showed normal pulmonary function tests, IgE increase > 5000UI/mL and oscillating eosinophilia. The patient did not accept the performance of bronchoscopy or lung biopsy. He also referred a 2-year history of diarrhea and pruritus. Previous outpatient

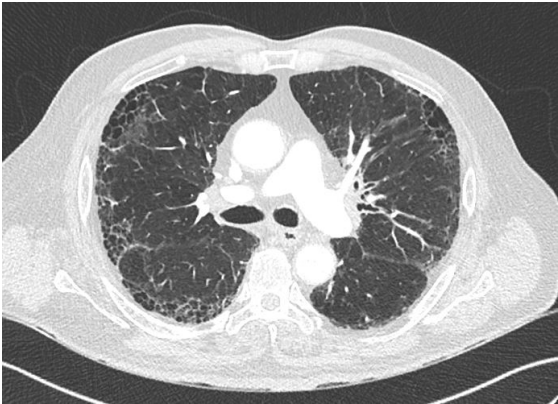
investigations included a negative stool culture, 1 of 3 parasitic stool tests positive for *Dientamoeba fragilis* (treated with metronidazole), a normal abdominal computerized tomography (CT), and a colonoscopy showing active mild chronic colitis. He did not receive any immunosuppressive treatment for the last 18 months (previously, he had been treated with oral corticosteroids for an acute bronchitis). He had immigrated from Bolivia to Spain 18 years earlier, not travelling abroad since then.

Physical examination revealed a hemodynamically stable febrile patient. He was confused and lethargic, with no other neurological abnormalities. Significant laboratory findings included leukocytosis (12.48x10<sup>9</sup>/L) with neutrophilia but normal eosinophil count, hemoglobin 117g/L and mild thrombocytosis (409x10<sup>9</sup>/L). Brain CT showed previous lacunar strokes. The cerebrospinal fluid (CSF) analysis was consistent with acute bacterial meningitis (leukocyte count of 2,070/mm<sup>3</sup> of which 84% were polymorphonuclear cells, glucose level of 45mg/dL, proteins at 2.14 g/L). He was started on dexamethasone 12mg intravenous, cefotaxime 300mg/kg/day, and ampicillin 250mg/kg/day. On the

next day, a multiplex Polymerase Chain Reaction (PCR) assay (Meningitis/Encephalitis FilmArray® panel) was performed in the CSF, discarding the most frequent pathogens of cerebral nervous system infection, including viruses, bacteria, and yeast. Ampicillin and corticoid were stopped, and cephalosporin was switched to ceftriaxone 2g b.i.d. Blood cultures were negative, but in CSF culture grew cephalosporine-resistant *Lactobacillus rhamnosus*. A lumbar puncture on day eleven showed a CSF examination with leukocyte count of 123/mm3 (99% polymorphonuclear cells), glucose level of 85mg/dL, proteins at 0.83 g/L.

		Age	Country of origin	Eosino-philia	Comorbidities	CSF analysis	CSF culture	Blood cultures	Diagnosis Strongyloides	Outcome
F. Reyes et al., IDCases. (2017)		76	-	-	Hypertension, type 2 DM, dyslipidemia, hypothyroidism, ischemic stroke, asthma	Raised white cell count (85% neutrophils).	<i>Citrobacter koserii</i>	<i>Citrobacter koserii</i>	Necropsy (gastrointestinal tract, respiratory tract and in the meninges).	Died
Vandebosch et al., Acta Gastroenterol Belg (2008)		49	Congo	No	Hypertension, type 2 DM	-	Five episodes of meningitis in one patient ( <i>Streptococcus anginosus</i> yield in one).	-	Duodenal biopsies	Discharged
Hager et al., W V Med J (2007)		66	USA (rural community)	No	-	Raised white cell count (74% neutrophils) and protein (706mg/dL), and low glucose (23mg/dL)	<i>Streptococcus bovis</i>	<i>Streptococcus bovis</i>	Stool	Discharged
Smallman LA et al., J Clin Pathol (1986)	Case 1	38	Jamaica	No	-	Described as pus (Diagnostic of meningitis by necropsy)	<i>Escherichia coli</i>	<i>Escherichia coli</i>	Biopsy of distal ileum, stool, and nasogastric, pharyngeal, and tracheal aspirates.	Died
	Case 2	68	West Indian	No	-	Raised white cell count (neutrophils, polymorphs), low glucose concentration.	<i>Escherichia coli</i>	-	Necropsy (biopsy of duodenum, jejunum, and ileum).	Died
CSF Cerebrospinal fluid										

**Table 1:** Literature review of bacterial meningitis associated with SHS in immunocompetent patients.



**Figure 1:** CT Scan showing interstitial lung disease.



**Figure 2:** Agar plate culture showing larvae of *S. stercoralis*.

Concurrently, screening serologic tests were performed, detecting IgG antibodies to *Strongyloides stercoralis*, and test stool samples were collected for parasitological examination to study the chronic diarrhea. Microscopic examination was performed after concentration techniques (Parasep®SF) and showed the presence of rhabditiform larvae of *S stercoralis*, which were also observed with the agar plate culture method for diagnosis of *S stercoralis* (Figure 2). The diagnosis of SHS with meningitis due to *L rhamnosus* was made, so ivermectin 200µg/kg/day and linezolid 600mg b.i.d were added.

HIV and HTLV-1 serology were negative, and no nutrient deficiencies were found. An induced sputum was obtained for agar plate culture of *S stercolaris* and direct microscopic examination, and no larvae were found. However, the sputum sample was collected when the patient was under ivermectin therapy. He received ivermectin for 14 days, after which stool examination did not show parasites. CSF examination on day 17 showed leukocyte count of 43/mm<sup>3</sup> of which 100% were mononuclear cells, glucose level of 84mg/dL, proteins at 0.64 g/L and negative culture. He was discharged with resolution of neurological symptoms, pruritus, and diarrhea.

Discussion

*Strongiloydes stercolaris* infection is more frequent in tropical and subtropical regions. However, because of migration and immunosuppressive therapies, increasing number of cases are being detected in developed countries. This intestinal nematode can penetrate the duodenal mucosa or perianal skin to access venous circulation (autoinfection). This phenomenon is unique to this parasite, and it is seen in both asymptomatic and symptomatic hosts, where continuous cycles may perpetuate the infection for decades. However, in some immunocompromised hosts the parasite begins to replicate through an accelerated autoinfection cycle, causing more severe forms of the disease, known as *Strongyloides* hyperinfection syndrome (SHS). Penetration of intestinal mucosa by filariform larvae facilitates the transport of enteric organisms into the bloodstream, leading to extraintestinal bacterial infection such as meningitis. CSF can exhibit an aseptic meningitis or bacterial meningitis characteristics. CSF cultures are usually positive for enteric organisms, such as *E. coli*, *P. mirabilis*, *K. pneumoniae* or *E. faecalis* [1]. To our knowledge, this is the first case of *Lactobacillus* meningitis in association with SHS described in the literature. The most common risk factors for the SHS are coinfection with HTLV-1 or HIV, treatment with corticosteroids, transplants, alcoholism, or malnutrition [1]. It is infrequently described in immunocompetent adults [2]. In our patient we did not identify any immunological alteration other than diabetes. This chronic disease is present in a few case reports of disseminated strongyloidiasis [2,3] and has been associated with treatment failure [4], but increased risk of SHS has not been reported. However, the association between diabetes and *Strongyloides* infection remains controversial with both positive and negative associations demonstrated in previous studies [5-7]. Few cases of bacterial meningitis associated with SHS in immunocompetent patients have previously been described in the literature (Table 1) [8-11]. As a whole, most patients came from regions where *S stercolaris* is more prevalent. Eosinophilia was not observed in any case, as in our patient. Comorbidities were described only in two cases, and diabetes was present in both.

In conclusion, although it is more frequent in immunocompromised hosts, SHS can occur in immunocompetent individuals. This case highlights the need for suspecting SHS in an enteric meningitis in a patient with a consistent epidemiological background, despite the absence of immunological alterations.

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## References

1. Vasquez-Rios G, Pineda-Reyes R, Pineda-Reyes J, Marin R, Ruiz EF, et al. (2019) *Strongyloides stercoralis* hyperinfection syndrome: a deeper understanding of a neglected disease. J Parasit Dis. 43:167-175.
2. Chan FLY, Kennedy B, Nelson R. (2018) Fatal *Strongyloides* hyperinfection syndrome in an immunocompetent adult with review of the literature. Intern Med J. 48: 872-875.
3. Murali A, Rajendiran G, Ranganathan K, Shanthakumari S. (2010) Disseminated infection with *Strongyloides stercoralis* in a diabetic patient. Indian J Med Microbiol. 28: 407-408.
4. Hays R, Esterman A, McDermott R. (2015) Type 2 diabetes mellitus is associated with *Strongyloides stercoralis* treatment failure in Australian aboriginals. 9.
5. Mendonça SCL, Gonçalves-Pires M do RF, Rodrigues RM, Ferreira A, Costa-Cruz JM. (2006) Is there an association between positive *Strongyloides stercoralis* serology and diabetes mellitus? Acta Trop. 99:102-105.
6. Hays R, Esterman A, Giacomini P, Loukas A, McDermott R. (2015) Does *Strongyloides stercoralis* infection protect against type 2 diabetes in humans? Evidence from Australian Aboriginal adults. Diabetes Res Clin Pract. 107: 355-361.
7. McGuire E, Welch C, Melzer M. (2019) Is *Strongyloides* seropositivity associated with diabetes mellitus? A retrospective case-control study in an East London NHS Trust. Trans R Soc Trop Med Hyg. 113: 189-194.
8. Smallman LA, Young JA, Shortland-Webb WR, Carey MP, Michael J. (1986) *Strongyloides stercoralis* hyperinfestation syndrome with *Escherichia coli* meningitis: Report of two cases. J Clin Pathol. 39: 366-370.
9. Hager C, Abaaba C, Kerns F. (2007) *Streptococcus bovis* meningitis and sepsis associated with Strongyloidiasis in an immunocompetent patient. W V Med J. 103: 19-21.
10. Vandebosch S, Mana F, Goossens A, Urbain D. (2008) *Strongyloides stercoralis* infection associated with repetitive bacterial meningitis and SIADH: A case report. Acta Gastroenterol Belg. 71: 413-417.
11. Reyes F, Singh N, Anjuman-Khurram N, Lee J, Chow L. (2017) *Strongyloides* Hyperinfection Syndrome causing fatal meningitis and septicemia by *Citrobacter koseri*. IDCases. 10:102-104.