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## Case Report

# Late onset of *Strongyloides stercoralis* meningitis in a retired Belgian miner

Y. Pypen<sup>1,2</sup>, E. Oris<sup>2</sup>, J. Meeuwissen<sup>3</sup>, M. Vander Laenen<sup>4</sup>, F. Van Gompel<sup>5</sup>, G. Coppens<sup>2</sup>

<sup>1</sup>Laboratory Medicine, University Hospital Leuven, Belgium, <sup>2</sup>Laboratory Microbiology, Hospital East Limburg, Genk, Belgium, <sup>3</sup>Infectiology, Hospital East Limburg, Genk, Belgium, <sup>4</sup>Intensive Care, Hospital East Limburg, Genk, Belgium, <sup>5</sup>Department of Clinical Sciences, Institute of Tropical Medicine, Antwerpen, Belgium

We report a rare case of *Strongyloides stercoralis* meningitis in an immunocompromised patient treated for a lung carcinoma. Despite his Belgian origin, he was infected with *S. stercoralis* due to his former work as a miner. Although mostly prevalent in (sub)tropical areas, there are temperate regions where this nematode can occur.

**Keywords:** *Strongyloides stercoralis*, Strongyloidiasis, Meningitis, Parasitology

## Case Report

A 57-year-old male presented at our emergency department for general malaise, nausea, fever and chills and a light headache since 2 days. About 2 months earlier, he was diagnosed with a non-small-cell lung carcinoma T2aN2M0 subtype squamous-cell carcinoma, treated with radio-chemotherapy. Except for inhalation corticosteroids, no other medication was taken at home. Clinical investigation showed a diaphoretic conscious patient with minimal neck stiffness without photophobia. Further clinical investigation showed no abnormalities nor did standard blood analysis, except for a minor increase of CRP with a value of 6.5 mg/l (<5) and a discrete decrease of sodium with a value of 130 mmol/l (135–145). There was no peripheral eosinophilia. The known lung carcinoma was visible on the chest radiograph without any other abnormalities. Computed tomography of the brains was normal. After lumbar puncture, results of spinal fluid were as follows: glucose 34 mg/dl (40–70), proteins 109.1 mg/dl (15.0–45.0), lactate 5.4 mg/dl (1.10–2.40), erythrocyte count: 8 cells/ $\mu$ l, white cell count: 747 cells/ $\mu$ l (0–5) with the following differentiation: 87% neutrophils (2 $\pm$ 4), 2% lymphocytes (60 $\pm$ 20) and 11% monocytes (30 $\pm$ 15). In a cytocentrifuge Gram stain preparation of the spinal fluid, we found many neutrophils and one larva suspicious for *Strongyloides stercoralis* (Figure 1). Since this is extremely exceptional

in this kind of sample, we asked the patient some more questions about his background.

He had worked in the ‘André Dumont’ colliery nearby Genk (Belgium) from 1976 to 1987. He never had made exotic travels and recalled that in the past that he once was treated preventively for ‘Miner’s worm’.

After this information, a stool sample was collected for ova and parasite examination. This was positive for multiple mobile *S. stercoralis* larvae.

Bacteriological culture of the cerebrospinal fluid (CSF) was also positive for *Enterococcus faecalis*. The diagnosis of disseminated *S. stercoralis* hyperinfection with meningitis due to *S. stercoralis* and *E. faecalis* was made. For completeness, HTLV-1 infection, a known possible risk factor related to disseminated strongyloidiasis, was excluded by a negative serology.

The initial empirical treatment for bacterial meningitis with ceftriaxone 2  $\times$  2 g I.V. was switched to amoxicillin 6  $\times$  2 g I.V. for 14 days. For the strongyloidiasis, we started a treatment with ivermectin 200  $\mu$ g/kg/day (or 5  $\times$  3 mg/day) during 5 days, followed by 2 days of therapy (200  $\mu$ g/kg/day) after 2 weeks. As long as the patient underwent radiotherapy and chemotherapy, he received a 2 days therapy of ivermectin (200  $\mu$ g/kg/day) every 3 months. After treatment with antibiotics and ivermectin, there was a good recovery from the strongyloidiasis and the bacterial meningitis. Unfortunately, the patient died of his malignancy a few months later.

\*Correspondence to: Yolande Pypen, Laboratory Microbiology, Hospital East Limburg, Genk, Belgium. Email: yolandepypen@gmail.com



**Figure 1** Gram stain of cerebrospinal fluid showing a *Strongyloides stercoralis* larva

## Discussion

*S. stercoralis* is a soil-transmitted intestinal nematode that infects tens of millions of persons worldwide. *S. stercoralis* is common in tropical and subtropical areas (e.g. Southern Europe), but there are also endemic temperate regions where the nematode can occur.<sup>1</sup> In Belgium, it is known that the nematode lived in the formerly André Dumont colliery in Waterschei (Genk).<sup>2</sup> Ground soil is the primary source of the nematode, and under warm moist conditions, it is found in two forms: infective filariform larvae and free-living adults.<sup>1</sup>

After contact with contaminated soil, the filariform larvae can penetrate the skin from where they will eventually migrate to the small intestine. First, they enter the circulatory system and will migrate to the lungs, where they will penetrate the alveolar spaces. They will be carried to the trachea and pharynx, swallowed and will reach the small intestine, where they become adults. The female adults will deposit eggs into the intestinal mucosa. These eggs will hatch into larvae within the mucosa and rhabdiform larvae will either be excreted in stool or will develop into infective filariform larvae that can cause autoinfection by penetrating the intestinal mucosae or the perianal skin. Once infected and without treatment, the parasite may stay present in the human body for several decades, perhaps even lifelong.<sup>1</sup> In case of any breakdown in the immune defense, a rapid increase in the worm-burden results in hyperinfection.

The clinical syndromes of *S. stercoralis* infection encompass a spectrum of different symptoms.

### Acute strongyloidiasis

The clinical manifestation in this stage can be associated with the path of the larval migration to the small intestine. Infected patients may experience symptoms like a local

skin reaction starting at the site of larval entry hence occurring around the anus but may be seen anywhere on the trunk (larva currens), followed by pulmonary symptoms (dry cough, tracheal irritation, respiratory distress) and eventually gastrointestinal symptoms (diarrhoea, constipation, abdominal pain, nausea ...).<sup>1</sup>

### Chronic uncomplicated strongyloidiasis

Chronic strongyloidiasis is most often asymptomatic. A broad spectrum of vague symptoms dependent on the localisation of the infection can occur (e.g. irritation of the skin, respiratory distress, dry cough, abdominal pain, diarrhoea ...), especially during autoinfection, but mostly these symptoms are ambiguous and not recognised as a chronic strongyloidiasis.<sup>1</sup>

### Hyperinfection syndrome

Hyperinfection describes the syndrome of accelerated autoinfection, generally the result of an alteration in immune status. In our case report, the hyperinfection occurred after radiochemotherapy. The diagnosis of hyperinfection syndrome implies the presence of signs and symptoms attributable to increased larval migration. Development of rapidly progressing linear urticarial tracks (larva currens), gastrointestinal or pulmonary symptoms and the detection of increased number of larvae in the stool and/or sputum may be seen with hyperinfection syndrome.<sup>1</sup>

### Disseminated infection

In this situation, the larvae migrate away from the lungs and gastrointestinal tract into other organs. Larvae may carry microorganisms from the gastrointestinal tract into the bloodstream or to other organs causing bacterial sepsis or other infections.<sup>1</sup> This explains the *E. faecalis* meningitis in our case report.

The diagnosis of strongyloidiasis is often difficult and delayed or overlooked due to the vague ambiguous non-specific symptoms. Patients with chronic strongyloidiasis usually have a low parasitic load. Therefore, it is not easy to detect a carrier of the parasite. There are several diagnostic techniques available, but all these methods have their limitations concerning sensitivity, specificity or availability.

Laboratory findings are usually non-specific, and in some cases, there is an intermittent peripheral eosinophilia. In severe complicated strongyloidiasis, the eosinophilia disappears and is an indication of poor prognosis.<sup>1</sup>

Routine ova and parasite testing of stool and sputum has poor sensitivity for detection of strongyloidiasis. Concentration methods, such as the Bearmann technique, will increase the sensitivity of stool examination, but because of the low parasite load in chronic infection, it is more effective in acute infection, hyperinfection and in

disseminated infection. Not only are these methods inadequate for screening, they also require experienced laboratory technicians and they are labour-intensive: multiple examinations are often necessary. The stool agar culture, where we observe 'larval tracks' of enteric organisms on the plate, is a preferred method because of its high sensitivity and ease of implementation in standard microbiology laboratories.<sup>1</sup>

Serological methods are now widely available and have a better sensitivity and specificity, respectively, 83–93 and 95–98% for enzyme-linked immunosorbent assay (ELISA) and 97 and 100% for luciferase immunoprecipitation system (LIPS). Enzyme-linked immunosorbent assay remains positive long time after treatment and is prone to cross-reactions with other nematodes. The more recently developed LIPS that uses two very specific antibodies against *S. stercoralis* shows no cross reaction and turns negative after treatment. Both methods have a window of (false) negativity before immunological response of the patient following an acute infection.<sup>1</sup>

A real-time PCR method has been developed to detect *S. stercoralis* with the use of 18S rRNA gene sequencing. This technique is very specific and may be promising for screening. However, it is not available everywhere, in particular less in the (sub)tropical regions where the infection is most prevalent.<sup>1</sup>

*S. stercoralis* should be treated whether or not the infection is giving rise to symptoms. The goal of treatment is total eradication of the parasite, otherwise the infection will be preserved causing generalised severe infections when defenses of the body break down.

Oral ivermectine 200 µg/kg/day is the treatment of choice. The duration of treatment depends on the stage of the infection. For acute/chronic infections, a therapy of 1–2 days with a repeat after 1–2 weeks usually covers the infection, but in case of hyperinfection or disseminated infection, therapy should last until 2 weeks after negative stool samples. An alternative for acute/chronic infection is orally albendazole 400 mg/day for 3–7 days. Thiabendazole has been completely abandoned because of the sometimes severe neuropsychological side effects. In some cases, where oral medication was not possible, subcutaneous ivermectine was safely administered with success.<sup>1</sup>

Finding larvae of *S. stercoralis* in CSF is very exceptional and only a few papers report on this phenomenon.<sup>3,4</sup> While most patients in these case reports did not survive, our patient recovered completely from his *S. stercoralis* infection after therapy, perhaps thanks to a fast but fortuitous diagnosis after a Gram stain of the CSF, in which the larva of *S. stercoralis* was well visible and directly recognised.

Bacterial meningitis caused by a disseminated *S. stercoralis* infection is more frequently described.<sup>4</sup> In these cases, the vague gastrointestinal and/or pulmonary complaints accompanying the meningitis were not solved after the treatment of bacterial meningitis. Further investigation eventually led to *S. stercoralis* infection as the initial cause of the disease.

Most case reports on *S. stercoralis* hyperinfection describe a various range of vague symptoms and complaints.<sup>4</sup> Diagnosis is often difficult and with a certain delay, whereas early recognition is an important prognostic factor. To anticipate the development of *S. stercoralis* hyperinfection, awareness of a possible *S. stercoralis* infection is necessary in travellers or residents from endemic areas (prevalent in tropical South America, China and Southeast Asia, but also documented in France, Portugal, Suisse, the Balkan, Italy and Poland) with eosinophilia, urticarial skin symptoms or abdominal pain, especially if these patients have an immunosuppressive condition or if they need an immunosuppressive therapy (chemotherapy, transplantation, corticosteroids). Some reports,<sup>3–5</sup> together with our case report, suggest that retired coal miners who worked in the mines of Limburg (Belgium, the Netherlands) and especially in the mine of André Dumont Waterschei (Genk) may have a certain risk. It has been assumed that *S. stercoralis* had been introduced into the mines by migrant workers coming from Silesia (now Western Poland) in the years between the World Wars. The larvae were probably introduced into the soil by infected miners defecating in the mine. Because of the warm temperature and the humidity in the mine, it was possible for the larvae to remain viable in the soil. Infection of other miners was acquired due to contact of bare skin with the faecal-contaminated soil.<sup>3–5</sup>

This case report underlines the importance of early diagnosis of strongyloidiasis, especially in the immunocompromised patient. Any adult patient presenting with enteric organism meningitis should make one think of the possibility of disseminated strongyloidiasis. Before the administration of immunosuppressive therapy, especially corticosteroids, the presence of larva currens, eosinophilia and a positive serology for strongyloidiasis should be excluded. In case of any suspicion of strongyloidiasis, empirical therapy should be started before the administration of immunosuppressive therapy.

## Disclaimer statements

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