


A Fatal Case of Systemic Strongyloidiasis and Review of the Literature

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Systemic strongyloidiasis is a rare but serious complication of intestinal strongyloidiasis. The condition occurs mainly in immunosuppressed patients and has a significant mortality rate. A case of systemic strongyloidiasis is described in a patient who received systemic steroid treatment, and a short review of the literature is given. The increased use of immunosuppressive and cytotoxic treatment necessitates increased awareness of this infection. HIV-infection, however, does not appear to increase the risk of developing systemic strongyloidiasis. Patients from endemic areas and travellers to such areas, even in the remote past, should be examined for strongyloidiasis before being given immunosuppressive treatment. Awareness of the possibility of systemic strongyloidiasis is essential if such a patient develops gastrointestinal or pulmonary symptoms or has repeated episodes of unexplained gram-negative infections while undergoing immunosuppressive treatment.

*Strongyloides stercoralis* was first identified in 1876 as the aetiological agent of “Cochin China diarrhoea” in French troops returning from Southeast Asia (1). The parasite is endemic in tropical and subtropical zones (2–4). It is also widespread in countries in East Europe such as Hungary, Romania, Poland and the former Soviet Union (2, 3). Due to increased travel and immigration the infection is now also seen increasingly in non-endemic areas, and has been found in 0.6–11.5 % of refugees (5). The intestinal infection may disseminate in case of immunosuppression. The widespread and increased use of immunosuppressive and cytotoxic treatment makes strongyloidiasis an important human parasitic infection (5–7). More than 500 cases have been reported. This

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paper presents a case of fatal systemic strongyloidiasis and a review of the literature.

**Case Report.** A 27-year-old woman from Bangladesh with lepromatous leprosy treated with dapsone, clofazimine and rifampicin developed an erythema nodosum leprosum reaction and was treated with systemic steroids at a dosage of 40–60 mg daily. She was admitted to hospital in Bangladesh seven weeks later with headache, pulmonary symptoms, stomach pain and constipation. One week earlier she had suffered severe diarrhoea for some days. She was mentally alert, but had severe dyspnoea and signs of ileus. A chest radiograph showed diffuse bilateral pulmonary infiltrates. The leucocyte count was initially 5.7 x 10^9/l with 4 % eosinophils, and dropped to 1.3 x 10^9/l with 3 % eosinophils. Many rhabditiform and filariform Strongyloides larvae were found in fresh specimens of stool and sputum, and a diagnosis of systemic strongyloidiasis was made. No serological tests were done. *Mycobacterium tuberculosis* was not found on direct microscopy of the sputum or on culture. She was treated with oral thiabendazole 25 mg/kg x 2 daily and with parenteral ampicillin and gentamicin. Initially her condition improved, but on the fourth day her condition deteriorated markedly. She developed pulmonary insufficiency, ileus and meningitis. A chest radiograph showed progressive diffuse bilateral pulmonary infiltrates. A lumbar puncture showed a slightly elevated protein concentration of 0.64 g/l in the cerebrospinal fluid but no other abnormal findings. The patient died within a few hours, with signs and symptoms of multiple organ failure. Permission for an autopsy was refused by the family.

**Discussion.** *Strongyloides stercoralis* is a small intestinal nematode which can infect man and other vertebrates when the filariform larvae penetrate intact skin or mucous membranes, usually causing a benign infection of the duodenum and proximal jejunum (4, 8-10). The hermaphroditic adult females burrow into the submucosa, where they produce small and irregular amounts of eggs daily (2, 9, 11). These rapidly hatch out into non-infectious rhabditiform larvae. Most rhabditiform larvae are passed in the stools, resulting in a heterogenic cycle with free-living male and female worms in the soil. Here the potential female rhabditiform larvae can develop into the filariform larvae, which can infect a new host (2, 4, 9-12). Some rhabditiform larvae, however, metamorphose into the filariform larval stage in the colon and can penetrate the colonic mucosa or the peri-anal skin whereby a new cycle is started in the same host (2, 4, 9-11). Autoinfection is unique to *Strongyloides stercoralis*. The infection can persist for decades through autoinfection. The parasite burden may increase within the infected host without exogenous reinfection.

Strongyloidiasis presents as an immunological spectral disease, the manifestations ranging from self-cure to hyperinfection syndrome and disseminated strongyloidiasis (7, 8, 13). This is also seen in filariasis, leishmaniasis and leprosy.

Autoinfection normally remains at a low level (6, 9). When the host-parasite balance is disturbed, a hyperinfection syndrome can develop due to accelerated autoinfection (2, 4, 11, 13, 14). It is characterized by a heavy worm burden and a disproportionate transformation of rhabditiform larvae to filariform larvae. This metamorphosis occurs in the proximal small intestine, normally occupied only by rhabditiform larvae and adult worms. There is an increased amount of adult worms in the upper bowel, increased penetration of the filariform larvae through the colonic mucosa and migration through the lungs. Gastrointestinal and pulmonary haemorrhages are seen, either due to physical damage caused by the large amount of penetrating larvae or due to immunological reactions. Disseminated strongyloidiasis is a further complication with widespread invasion of the filariform larvae into organs which are not normally involved in the parasitic life cycle (2, 4, 11, 13, 14). The filariform larvae can invade the lungs, liver, brain, heart, pancreas, kidneys, adrenal glands, thyroid gland and central nervous system. Here the adult worms develop and eventually reproduce with development of extraintestinal rhabditiform larvae.

Ninety per cent of cases of systemic strongyloidiasis are seen in immunosuppressed patients including patients on systemic steroid treatment due for instance to rheumatological, dermatological or obstructive pulmonary diseases (2, 4, 6, 15–20); patients on cytotoxic treatment due to malignant diseases, especially leukaemia and lymphoma (2, 4, 6); patients with chronic renal diseases, especially renal transplant recipients, receiving immunosuppressive treatment (2–4, 11, 21, 22), although cyclosporin A may have a protective role due to its parasitocidal activity (23); and patients with severe malnutrition or chronic alcoholism (2, 4, 6).

Seventy-seven per cent of the cases described in detail have been associated with immunosuppressive or cytotoxic treatment (6, 7), 8 % had an un-