# Rare Case of *Strongyloides stercoralis* Hyperinfection in a Greek Patient with Chronic Eosinophilia

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Case Report

## ABSTRACT

Strongyloidiasis is a disease characterized by a diverse spectrum of unspecific manifestations that complicate its diagnosis. Although, the course of its chronic form is usually benign, in cases of immunosuppression, iatrogenic or not, it can evolve to a hyperifection syndrome with even fatal complications. Herein, we report a case of *Strongyloides stercoralis* hyperinfection in a Greek patient receiving corticosteroid treatment for chronic eosinophilia and angioedema. The case represents an extremely rare case of autochthonous strongyloidiasis in Greece and underlines the importance of the early diagnosis of the disease's uncomplicated forms in order to prevent its severe sequelae.

**Keywords:** Angioedema, corticosteroids, eosinophilia, hyperinfection, *Strongyloides stercoralis*

## INTRODUCTION

Strongyloidiasis is caused by the soil transmitted nematodes *Strongyloides fuelleborni* and *Strongyloides stercoralis*. While the former infects mainly non-human primates with human infections occurring sporadically in Africa and Southeast Asia *S. stercoralis* is a common human pathogen with a cosmopolitan distribution. Although mostly prevalent in the tropics and subtropics, it is also endemic in temperate regions including industrialized countries in Europe and USA. Through autoinfection, the parasite may silently persist for decades within the host and even when symptomatic the uncomplicated intestinal cases commonly present with non-specific symptoms. Therefore, the infection tends to be underdiagnosed and underreported with the current estimate of 30-100 million human cases being too low. Nevertheless, in conditions associated with immunocompromise, especially corticosteroid treatment, uncontrolled autoinfection may lead to hyperinfection and even fatal dissemination. In Greece, autochthonous human infection is extremely rare, therefore unspecific findings in a setting of negative travel history, paired with the poor sensitivity of stool examinations might delay clinical suspicion. Here, we present a rare case of *S. stercoralis* hyperinfection in a Greek patient receiving chronic corticosteroid treatment for persistent eosinophilia.

## CASE REPORT

A 66-year-old Greek male was admitted to our institution with weakness, fatigue and epigastric pain of 8 days duration. During the past 8 years, he had frequent outpatient allergiologic follow up because of recurrent episodes of angioedema and persistent eosinophilia and had been under treatment with per os prednisolone. Within that period, the patient developed a high titer of *Echinococcus* specific IgG, as well as a pulmonary cyst which was surgically removed, but hydatid disease was histologically excluded. The patient also reported multiple episodes of epigastric discomfort during the last 2 years. He had a negative travel history and no clonality had been identified as an underlying cause of his eosinophilia. He reported a recent febrile period of two days with a temperature of 38°C that remitted after antibiotic treatment. Physical examination upon admission revealed an afebrile, ill looking patient with epigastric tenderness and few minor ecchymoses. Auscultation demonstrated bilateral crepitations, with the rest of the examination being unremarkable. Laboratory investigations revealed leucocytosis with moderate eosinophilia ($1.98\times10^9$/L), hyponatraemia (125 mmol/L), hypoalbuminaemia (4.8 g/dl) and C-reactive protein of 55.3 mg/L (normal values <6 mg/L), with the rest of the laboratory data being within the normal range. At the time of admission, the patient was receiving 40 mg of per os prednisolone. Despite the absence of fever, the urine culture from the sample obtained at admission was positive for *Escherichia coli* >100.000 cfu/ml. Serological investigation was negative for CMV, HSV (I, II), EBV, *Toxoplasma gondii* and *Echinococcus* but total serum level for IgE was tenfold higher than normal limits, at 1,450 IU/ml. On the second day after admission the patient developed diarrhoea and the microscopic stool examination revealed filariform larvae of *S. stercoralis.* A diagnosis of *Strongyloides* hyperinfection syndrome was made for which he received per os albendazole 400 mg per day and cessation of steroid therapy was planned by gradual dosage decrease. Cerebrospinal fluid examination was normal, while chest and abdominal computerized tomography scan revealed a left pleural effusion without further pathological findings. Oesophagogastroduodenoscopy was performed and the duodenal and gastric biopsies obtained demonstrated evidence of parasitic infection with findings of chronic granulomatous inflammation, numerous eosinophils and remnants of parasitic ova. On the 4th day of therapy the patient's clinical picture improved and the eosinophilic count normalized. Nevertheless, in view of persistent positive stool samples ivermectin treatment was planned, with continuation of albendazole until the former would be available. Following the first negative stool sample on the 17th day of albendazole, the patient received per os ivermectin for two days and was discharged on the 21th day after admission, symptom free and with negative stool samples for *S. stercoralis*.

## DISCUSSION

*S. stercoralis* has a unique life cycle compared to other nematodes due to the ability of the non-infective rhabditiform larvae to mature directly into infective filariform larvae while still within the host. This feature has important clinical implications, as it is responsible for the persistence of infection. In cases of intact immune system, this autoinfection is under control and the parasitic burden is low, however in cases of immunosuppression it accelerates and the increased number of parasites may lead to hyperinfection, when limited to the respiratory and gastrointestinal tracts or to disseminated infection when more organ systems are involved. Although, strongyloidiasis has a very wide range of manifestations, most cases are clinically inapparent. Even when present, manifestations are usually protean and may mimic a wide range of diseases.[1] Angioedema for instance, which was a prominent feature of the presented case, exhibits a wide differential diagnosis, therefore, although commonly associated with strongyloidiasis, it seldomly raises suspicion towards this direction.[2] The diagnostic challenges are further enhanced by the low sensitivity of stool specimens and the limited success of serologic assays.[1,3] On the other hand, eosinophilia is present in the majority of cases of uncomplicated strongyloidiasis and is highly sensitive and specific in high risk populations, therefore should be followed by serologic investigations and multiple stool examinations in order to achieve definite diagnosis.[1,3] In non-endemic regions however, where the degree of suspicion is low, infections

even when accompanied by eosinophilia might be misdiagnosed or diagnosed with a delay. In case of iatrogenic -as in our case- or disease related immunocompromise, strongyloidiasis may evolve to hyperinfection or even disseminated disease, both of which are associated with severe complications and mortality rates up to 87%.[3] In such cases, eosinophilia is usually absent but when present it is considered as a good prognostic factor.[1] In Greece, autochthonous strongyloidiasis is extremely rare, with very few cases reported in literature. This fact has been attributed to the probable existence of strains adapted to complete the indirect cycle in the environment without yielding the infective filariform/L3.[4,5] This poses a diagnostic challenge in cases of individuals with negative travel history to endemic regions presenting with eosinophilia and unspecific symptoms. When strongyloidiasis is not suspected, corticosteroid management of the eosinophilia, as in our case, might trigger hyperinfection, even in low doses and in short courses.[1] Although, detection of *S. stercoralis* is easier in hyperinfection due to increased worm output, prognosis is less favorable. Therefore, even in non-endemic regions and non-target populations, strongyloidiasis should always be placed within the physician's differential diagnostic process of eosinophilia, particularly when associated with recurrent angioedema, and should be excluded prior to corticosteroid therapy. We should keep in mind that early detection of chronic uncomplicated strongyloidiasis is the optimal prevention of the more severe forms of the disease.

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