Case report

Travel-associated human trichinellosis in Portugal

Francisco Vale, telma azevedo, maria lima, Inês oliveira, mário parreirA, margarida anes, rui bajanca, maria joão gargate, josé poças

A Department of Infectious Diseases, Hospital de São Bernardo, Centro Hospitalar de Setúbal, R. Camilo Castelo Branco 175, 2910-548, Setúbal, Portugal
b Department of Medical Oncology, Portuguese Institute of Oncology Francisco Gentil, R. Prof. Lima Basto, 1099-023, Lisbon, Portugal
c Department of Internal Medicine, Hospital de São Bernardo, Centro Hospitalar de Setúbal, R. Camilo Castelo Branco 175, 2910-548, Setúbal, Portugal
d Department of Dermatology, Hospital de São Bernardo, Centro Hospitalar de Setúbal, R. Camilo Castelo Branco 175, 2910-548, Setúbal, Portugal
* National Reference Laboratory of Parasitic and Fungal Infections, Department of Infectious Diseases, National Institute of Health Dr. Ricardo Jorge (INSA), Av. Padre Cruz, 1649-016, Lisboa, Portugal

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A B S T R A C T

Trichinellosis is an emerging or re-emerging foodborne parasitic zoonotic disease caused by nematodes of the genus Trichinella. It represents a global food safety problem and a public health hazard. This case report aims to describe the first case of human trichinellosis in Portugal since the creation of the European Union surveillance system. Infection by Trichinella spp. is a rare cause of hypereosinophilia and hospital admission, but it can cause high morbidity. Clinical detailed history is crucial to obtain a correct diagnosis.

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I N T R O D U C T I O N

Trichinellosis is a foodborne parasitic zoonotic disease caused by nematodes of the genus Trichinella and represents a global food safety problem and a public health hazard [1]. Migration, different food practices, international traveling, and the illegal importation of non-controlled meat from endemic to non-endemic countries has resulted in outbreaks and isolated reports of this disease [2,3]. Humans acquire the disease by consuming raw or undercooked meat that contains cysts with viable Trichinella spp. larvae. The prevalence of Trichinella spp. infection is unknown, and their cases are significantly underreported [4]. In Portugal, the last notified case was in 1987 [5]. This article describes the first case of human trichinellosis in Portugal since the beginning of the European Union (EU) surveillance system.

C A S E   R E P O R T

In 2017, a 70-year-old male patient was hospitalized with a history of one-month complaints characterized by progressive pruritic and desquamative lesions on the scalp, scapular and cervical regions associated with myalgia, muscle weakness, arthralgia, and fatigue. The patient denied fever, weight loss, enlarged lymph nodes, or diarrhea. His medical history was not pertinent. The patient had traveled abroad two months before the symptoms started to Paris, France, where he ingested an undercooked meal whose contents he could not specify. He denied contact with farm animals, consumption of undercooked meat in Portugal, similar complaints in his close relatives or taking any medication or medical supplements.

Physical examination revealed a maculopapular, confluent, and pruritic rash at the scalp, face, scapular, cervical, chest and back regions (Fig. 1), purpuric maculopapular lesions on both hands as well as bilateral hand hypoesthesia. Laboratory tests showed anemia with a hemoglobin of 12.2 g/dL (reference range, 13–17), high white blood cell (WBC) count 20.200/µL (4.500–11.400) with marked eosinophilia 12.100/µL (59.7 %), c-reactive protein 2.42 mg/dL (<0.5), erythrocyte sedimentation rate 30 mm/1 h (1–14), elevated levels of IgE 958 kU/L (<120), with normal levels of creatine phosphokinase (CPK) 51 U/L (30–200) and lactate dehydrogenase (LDH) 225 U/L (125–230).

A diagnostic workup was performed for etiological diagnostic of hypereosinophilia which included blood cultures and autoimmunity markers that were all negative, there were no ova or parasites in the stools, computed tomography scan of the body did not have suspicious images of neoplasia, bone marrow biopsy and myelogram had no evidence of clonal neoplastic and gene fusion FIP1L1/PDGFRα analysis was negative. A skin biopsy showed an eosinophilic dermatosis with granuloma and eosinophilic spongiosis (Fig. 2), but muscular biopsy demonstrated no degenerative
Discussion

Human trichinellosis is a worldwide anthropozoonosis and has been documented in 55 countries, particularly those with well-established food behavior that includes consuming raw or undercooked meat [4]. A literature review from 1986 to 2009 revealed 65,818 cases of trichinellosis and 42 deaths reported from 41 countries [6]. In 55 countries where trichinellosis occurs autochthonously, the yearly total number of clinical trichinellosis was estimated to be 10,000 cases, with a mortality rate of about 0.2% [4]. However, it is estimated that 11 million humans worldwide are infected by Trichinellasp. [7,8]. In the EU/European Economic Area (EEA), in 2017, the overall notification rate was 0.03 cases per 100,000 population. Bulgaria, Croatia, and Romania accounted for 73.8% of confirmed cases [9].

In France, human trichinellosis incidence is low, with only 68 cases reported from 2001 to 2016 in which more than half were related to the consumption of infected meat abroad or illegally imported meat [10]. Recently, Barruet et al. (2020) reported an outbreak of 20 cases in Paris, France, in 2017 [11].

In Portugal, the first outbreak occurred in Sabugal in 1881, followed by an outbreak described in Penamacor in 1951 [12]. Additionally, two reports are described (five cases in 1962 and one case in 1987) [13]. The last reported case by Portuguese surveillance system was in 1987 [5]. Ferreira et al. (2014) conducted a seroprevalence study in 273 high risk individuals in Évora and Portalegre districts and they revealed a positivity by western blot for T. spiralis between 4%–5.5%. The authors question the non-existence of this infection in Portugal, probably due to asymptomatic disease or atypical presentation, thus leading to this infection being underdiagnosed and, therefore, underreported [14]. Of note, trichinellosis is a mandatory notifiable disease in Portugal.

Clinically, *Trichinella* spp. infection in human hosts can be divided into an intestinal phase and a muscular phase, with clinical manifestations ranging from asymptomatic infection to fatal disease [1,8]. The intestinal phase is characterized by gastrointestinal symptoms like malaise, mild transient diarrhea, nausea, vomiting and abdominal pain that can persist from the first to the third week [1,6,8]. This phase was asymptomatic in our patient. During the migration of *Trichinella* spp. which starts in one week and may last for several weeks, the symptoms detected usually include myalgia, high fever, chills, periorbital and facial edema, conjunctivitis, pain, and skin rashes. The muscular phase is characterized by myalgias, arthralgia, headache, periorbital, and facial edema. Although fever, diarrhea and facial edema are very typical signs of trichinellosis, our patient never presented with these findings, however, he had severe myalgia, muscle weakness and arthralgia, and an exuberant maculopapular rash. Chronic stage of trichinellosis occurs after four weeks or later, and can present complications such as meningoencephalitis, myocarditis, and secondary infections [1,8,15].

Laboratory studies usually revealed a moderate increase in WBC count and circulating eosinophilia appears early after infection and increases between the second and the fifth week. Elevated levels of muscle enzymes such as LDH and CPK in the blood, which indicate myositis, are found in 75%–90% of infected patients [1,8]. Despite clinical findings, our patient did not present an elevated level of both enzymes, and muscular biopsy demonstrated no inflammatory or degenerative changes and did not show parasitic eggs or cysts. *Toxocara* spp., *Fasciola* spp., and *Strongylodes stercoralis* serologies were negative by enzyme-linked immunosorbent assay but for *Trichinella* spp. was strongly positive. An immunoblotting test confirmed *T. spiralis* infection. A diagnosis of trichinellosis, probably acquired abroad, was made. The patient was treated with albendazole and prednisolone. He was discharged with full clinical recovery, regression of cutaneous lesions, and normalization of laboratory tests.

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and vasculitis like Churg-Strauss syndrome. Neoplastic diseases manifested by paraneoplastic eosinophilia include varied adenocarcinomas, some forms of Hodgkin’s disease, T cell lymphoma, and mastocytosis. Hypereosinophilic syndromes comprise a heterogeneous group of uncommon disorders that should make part of differential diagnosis [1,16].

The definitive diagnosis of trichinellosis must be based on three main criteria: clinical findings, laboratory parameters, and epidemiological research. Indirect diagnostic methods involve detecting antibodies against *Trichinella* spp. and direct diagnostic methods include the detection of *Trichinella* spp. larvae in muscle biopsy. Antiparasitic and corticosteroid drugs are the cornerstone of severe trichinellosis treatment [1,8,15].

**Conclusion**

Trichinellosis continues to affect the health of both animals and humans worldwide. It is crucial to increase veterinary control to ensure food safety, epidemiological research, and population education about prevention. Another important highlight is the increasing number of international travelers who acquire *Trichinella* spp. infection while traveling and subsequently develop the disease after returning home. Epidemiology and detailed clinical history from travelers are essential to optimize patients’ clinical management.

**Author’s contributions**

FV was responsible for the conception and design of the article, acquisition of data, wrote the first draft, and approved the final version. TA, ML, IO, MP, MA, RB, MJG and JP analyzed and interpreted the data, critically revised the manuscript, and approved the final version.

All authors read and approved the final version of manuscript.

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**Informed consent**

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

**Declaration of Competing Interest**

The authors declare no conflicts of interest.

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